Aggressive Xanthogranulomatous Cholecystitis Mimicking Gallbladder Cancer

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1. Abstract
Xanthogranulomatous cholecystitis (XGC) is a benign uncommon variant of chronic cholecystitis. Perioperative findings may lead to confusion with gallbladder cancer, as the inflammation frequently extends to adjacent organs. In this report, we present a 50-year-old woman with a 2-years history of chronic abdominal pain and imaging suggestive of gallbladder carcinoma. The pathological examination of the resected specimen is consistent with xanthogranulomatous cholecystitis.

2. Introduction
Xanthogranulomatous cholecystitis (XGC) is a benign variant of chronic cholecystitis. Its incidence rate varies between different geographic regions from 0.7 – 9 % and the mean age at presentation ranges from 44 to 63 years [1]. XGC is characterized by chronic inflammation of the gallbladder. It is thought to be caused by an inflammatory response to extravasated bile from blocked or ruptured Rokitansky-Aschoff sinuses, leading to a proliferation of fibrotic tissue [2, 3]. XGC may be confused with gallbladder cancer as the inflammation frequently extends to adjacent organs most likely the liver and duodenum [4, 5]. Furthermore, it is unusual to reach the diagnosis of XGC before the postoperative pathological examination [6]. This challenging in the differentiation between benign and malignant disease before the surgery and the risk of metastasis of cholangiocarcinoma with the biopsy complicates the planning for surgery.

Here, we report a diagnostically challenging case of XGC in which perioperative findings were highly suggestive of malignancy, although the postoperative histopathology was ultimately benign.

3. Case Presentation
A 50-year-old female presented to our center with chronic epigastric pain associated with fever, and anorexia for the past 2 years. The patient was admitted as a case of biliary colic based on clinical and radiological findings. Her abdominal ultrasound showed a contracted gallbladder over multiple intraluminal stones, with neither cholecystitis nor biliary dilatation. The patient's physical examination was unremarkable. Laboratory investigations were normal. An abdominal computed tomography (CT) scan showed a suspicion of GBC. There was an ill-defined uncalcified 6x3x2.5 cm nodular enhancing mass, involving the gallbladder and hepatic segment IV and extending to the gastric antrum and pylorus with associated stranding and nodularity of the surrounding peritoneum and omentum (Figure 1). Tumor markers CEA, CA 19.9, and AFP did not have significantly high values. A chest CT scan revealed no distant metastasis. A diagnostic laparoscopy was performed due to the suspicion of gallbladder cancer with carcinomatosis. A mass at the gallbladder fundus was attached to the adjacent structures, namely liver segment IV, the falciform and the stomach. Peritoneal fluid for cytology had revealed reactive mesothelial cells, lymphocytes, histiocytes, and neutrophils, with no malignancy.

As there was no extension, we proceeded with a secondary curative surgery. A radical cholecystectomy and en bloc resection of liver segment III, IVb and V (partial) with excision of the retroportal lymph...
nodes were performed (Figure 2). The gastric antrum was free of direct invasion and there were no peritoneal deposits. A frozen section of the cystic duct stump was negative for malignancy. The histopathological findings were suggestive of XGC. Macroscopically, the gallbladder was filled with multiple stones, pus, and necrotic material. It had a thick wall surrounded by fibrosis, necrosis, and sloughed mucosal lining. Microscopically, the mucosa was ulcerated with extensive chronic lymphoblastic inflammatory infiltrate, foamy histocytes with multinucleated giant cells, fibrin deposition and fibrosis. Sections from the liver showed reactive periportal and lobular lymphoplasmacytic inflammation and inflammatory infiltrate in liver parenchyma. The cystic duct and falciform ligament were normal and the reactive LN was negative for malignancy.

Postoperatively, the patient was clinically well with normal investigations. The patient was discharged on postoperative day five.

**Figure 1:** Abdominal computed tomography images showing the gall bladder mass with invasion of surrounding structures.

**Figure 2:** The resected specimen

4. Discussion

XGC is a variant of chronic cholecystitis. Its clinical presentation is variable, ranging from GB inflammation to mass formation and the involvement of adjacent organs, resulting in a diagnostic challenge and a therapeutic dilemma [7, 8]. As reported in (Nacif LS, et al) XGC and GBC can have similar clinical presentations [9]. Abdominal pain was found to be the most common presenting symptom of XGC, and less likely to present with GBC which usually present with anorexia and weight loss [10]. Elevated liver function test, and elevated CA 19.9 can both be present in GBC and XGC. Thus, these are not considered to be reliable markers [11]. Furthermore, neither CT nor magnetic resonance imaging (MRI) are useful in differentiating XGC from GBC [12]. Our patient presented with chronic abdominal pain and radiological findings of mass-forming cholecystitis infiltrating the adjacent structures. Thus, GBC was suspected and diagnostic laparoscopy was performed to assess the disease extension.

Simple cholecystectomy is a sufficient therapy when the diagnosis is clear intraoperatively [13]. However, it has been suggested that intraoperative frozen-section analysis may be useful when the diagnosis is in doubt, as this avoids an unnecessarily aggressive intervention [13, 14]. However, this approach is problematic, as GBC may co-exist with XGC in up to 31% of cases and may provoke outflow obstruction. Moreover, GBC could be missed due to a sampling error if both XGC and GBC are present simultaneously. Also, opening a potentially cancerous gallbladder to examine the mucosa risks cutting across tumor and disseminating malignant tissue [14].

For cases of aggressive XGC with preoperative diagnostic uncertainty and a high suspicion of GBC, the best option is to perform a radical resection by expert surgeons. In a previous case series, a radical surgical excision was performed as a definitive treatment for three cases of aggressive XGC [9]. In our case, all investigations were highly suggestive of GBC. Thus, a radical cholecystectomy and en bloc resection of liver segments III, IVb and V was performed.
5. Conclusion
Aggressive XGC mimics the presentation of GBC. Surgical resection and the final histopathological analysis is the only method to confirm the diagnosis.

References

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