Xanthogranulomatous Cholecystitis Mimicking Gallbladder Cancer: Report of a Case

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Summary: A 61-year-old woman was admitted to our hospital with abnormal findings of abdominal computed tomography. Whereas she had neither fever nor abdominal pain, a cholecystitis was suspected. Ultrasonography showed a mass in the gallbladder with several stones, and an unclear border between the gallbladder and liver. Computed tomography showed a large mass in the gallbladder with findings that seemed to indicate hepatic invasion and para-aortic lymph node metastasis. On the basis of these findings, we made a diagnosis of gallbladder cancer associated with hepatic invasion and lymph node metastasis. We treated this gallbladder tumor by hepatic arterial infusion chemotherapy via catheter with cisplatin and 5-fluorouracil. Four weeks after administration of the anti-cancer drugs, the tumorous lesion of the gallbladder could not be detected by abdominal imagings, and the gallbladder wall revealed no irregular findings. During laparotomy, the gallbladder showed signs of chronic cholecystitis, and a cholecystectomy was performed. Findings of the resected specimens showed severe inflammation, fibrosis, and bleeding in the gallbladder wall with infiltration by many foamy cells. Histopathological diagnosis was xanthogranulomatous cholecystitis. We report here a case of xanthogranulomatous cholecystitis mimicking gallbladder cancer and review the literature.

Key words xanthogranulomatous cholecystitis, gallbladder cancer, diagnosis, treatment

INTRODUCTION

Xanthogranulomatous cholecystitis (XGC) is an uncommon inflammatory disease of the gallbladder. In severe cases, the inflammation extends to adjacent structures, and XGC is sometimes confused with a malignant neoplasm. We recently experienced a case of XGC which had been diagnosed preoperatively as an advanced gallbladder carcinoma. We discuss this case and review the literature.

CASE REPORT

A 61-year-old woman was admitted to our hospital with abnormal findings of abdominal computed tomography (CT) during a work up for common cold-like symptoms. She had neither fever nor abdominal pain, which suggested acute or chronic cholecystitis. On admission, the patient was afebrile and had no palpable mass in her abdomen, and laboratory data including tumor markers were nearly normal. Ultrasonography (US) showed a mass in the gallbladder with several stones, and an unclear border between the gallbladder and liver. Abdominal CT showed a large mass in the gallbladder with findings which seemed to indicate hepatic invasion (Fig. 1a) and a para-aortic lymph node metastasis. Hepatic arteriography revealed an irregularity of the cystic artery, and showed a hypervascular area of the gallbladder in the venous phase.

On the basis of these findings, we made a diagnosis of gallbladder cancer with hepatic invasion and
Fig. 1. a: Abdominal computed tomography showed a large mass in the gallbladder with suspected findings of hepatic invasion. b: Abdominal computed tomography also revealed no abnormal lesion of the gallbladder.

Fig. 2. a: Macroscopic findings of the resected specimen showed a markedly thickened wall and destructive mucosa. b, c: Microscopic findings show severe inflammation, fibrosis, and bleeding in the gallbladder wall (H & E, ×50) with infiltration by many foamy cells. (H & E, ×100)

lymph node metastasis, stage IVb according to the general rules for surgical and pathological studies on cancer of the biliary tract [1]. Therefore, we treated this gallbladder tumor by hepatic arterial infusion chemotherapy using cisplatin (CDDP) and 5-fluorouracil (5-FU). Four weeks after administration of the anti-cancer drugs, the tumorous lesion of the gallbladder could not be detected by US or CT (Fig. 1b), and the gallbladder wall revealed neither irregularity nor abnormal findings. The para-aortic lymph node seemed to be in the same condition as before chemotherapy.

During laparotomy, the gallbladder showed signs of chronic cholecystitis with severe adhesion to surrounding tissues, and an intraoperative examination of frozen sections from the gallbladder
showed chronic cholecystitis without malignancy. Cholecystectomy with liver bed resection was performed because we could not completely deny the possible coexistence of a cancerous lesion. Findings of the resected gallbladder showed severe thickening of the wall without tumorous lesion (Fig. 2a). A histopathological diagnosis of XGC was made based on the findings of severe inflammation, fibrosis, bleeding, and a large number of foamy cells infiltrating the gallbladder wall (Fig. 2b, c).

DISCUSSION

The term xanthogranulomatous cholecystitis was initially proposed by Goodman and Ishak [2] in 1981 in a review of 40 cases from the Armed Forces Institute of Pathology. XGC is found in a small proportion of cholecystectomy specimens, ranging from 0.7% to 1.8% in the United States [3,4], 9.3% to 13.2% in India [5,6] and 1.2% to 10% in Japan [7,8]. Although the pathogenesis of XGC remains elusive, obstruction of the cystic duct by gallstones and bile stasis are important etiologic factors [9]. The current hypothesis suggests that the lesion starts as gallbladder wall inflammation and obstruction. Bile then enters the stroma of the gallbladder through ulcerations in the surface of the mucosa, or through ruptured Aschoff-Rokitansky sinuses. Macrophages gather at the site of inflammation and ingest the bile lipids to form large, round, pale xanthoma cells [4,7]. Edlund and Olsson [10] and Kitagawa et al. [11] reported that the time between the onset of symptoms of acute cholecystitis and granuloma formation is 3 or 4 weeks.

XGC can be found unexpectedly during elective cholecystectomy and is often mistaken for gallbladder cancer. The coexistence of XGC and carcinoma of the gallbladder is another problem [2,12]. The frequency of such coexistence is nearly 10% [4,11] and most of the reported cases were discovered by histologic examination of the cholecystectomy specimen [12,13].

Since serum carbohydrate antigen (CA19-9) levels increase both in gallbladder cancer and XGC, the tumor marker CA19-9 is not useful for the diagnosis of coexisting carcinoma or for the distinction between XGC and carcinoma [14-16].

The preoperative diagnosis of XGC is difficult, and an intraoperative diagnosis from frozen sections is often needed to distinguish it from carcinoma [4,7,12,17]. According to Parra et al. [18], the presence of hypoechoic nodules or bands in the gallbladder wall on US or of a hypodense band around the gallbladder on CT, is highly suggestive of this disease. In our case, we made a diagnosis of gallbladder carcinoma because of abnormal findings of CT, US and angiography. Irregularity and stenosis of the cystic artery strongly suggested carcinoma. Gallbladder cancer was suspected at first, however, in the clinical course after chemotherapy, there were no abnormal findings of the gallbladder by US or CT. Therefore, cholecystectomy was performed under the preoperative diagnosis of chronic cholecystitis. Operative findings often reveal a more severe picture than that seen in ordinary cholecystitis. Adhesion to surrounding tissue is often encountered, as in our case, resulting in technical difficulties and prolonged operating time, and complete resection of the gallbladder is not always possible [19].

In conclusion, despite its characteristic histologic appearance, the clinical and radiological presentation of XGC is nonspecific and difficult to differentiate from other forms of cholecystitis and sometimes from gallbladder cancer. Therefore, it is difficult to make a preoperative diagnosis of XGC. If XGC can be diagnosed by some modality, such as percutaneous fine needle aspiration biopsy [6,20], it may be possible to avoid an extensive operation. Total, and not subtotal, cholecystectomy should be done if possible, because the incidence of gallbladder cancer accompanying XGC is higher than that with either ordinary cholecystitis or gallstones [2,4,12,15]. In our case, it was impossible for us to distinguish between XGC and gallbladder cancer before chemotherapy.

REFERENCES