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CASE REPORT

Ampullary Xanthogranulomatous Inflammation Mimicking Periampullary Cancer: Report of a Case

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ABSTRACT

Context Xanthogranulomatous inflammation commonly affects the gallbladder. To date, there have been no reports of xanthogranulomatous inflammation of the ampulla.

Case report A 48-year-old female presented to us with fever, jaundice and a palpable gallbladder. Evaluation revealed features of periampullary malignancy. The patient underwent a Whipple's pancreaticoduodenectomy. Histopathology revealed a xanthogranulomatous inflammation affecting the ampulla and the gallbladder.

Conclusion Xanthogranulomatous inflammation should be added to the differential diagnosis of patients presenting with a suspected periampullary lesion accompanied by a thick-walled gallbladder.

INTRODUCTION

Xanthogranulomatous inflammation can affect the gallbladder, kidney, bone, ovary, pancreas, lymph nodes and soft tissues, the gallbladder being the commonest site among these. Commonly, the disease is correctly identified on histopathological examination. Fine needle aspiration cytology can also diagnose the condition. Patients with xanthogranulomatous cholecystitis can present with obstructive jaundice which is seen in up to 23 % of cases [1]. The causes of obstructive jaundice include common duct stones, extrinsic compression of the common bile duct or associated malignancy. We report a case of obstructive jaundice due to xanthogranulomatous inflammation affecting the ampulla of Vater, a clinical situation hitherto undescribed.

CASE REPORT

A 48-year-old female was referred to our centre complaining of fever and jaundice. Six weeks before presenting to us, she had experienced recurrent attacks of high-grade fever up to 38.9°C associated with jaundice, dark urine, pale stools and pruritus. She was initially treated by a local physician with antibiotics and her fever subsided temporarily, but the jaundice gradually progressed. She did not experience any pain, weight loss or anorexia. Clinical examination showed deep jaundice and scratch marks all over the body. Abdominal examination revealed a 3 cm firm hepatomegaly and a tense palpable gallbladder. She had an ultrasonogram which showed bilobar intrahepatic biliary dilatation with dilatation of the common bile duct extending to the ampullary region. The gallbladder was distended with a single calculus of 28 mm and the gallbladder wall was 5 mm thick. No mass was seen at the lower common bile duct or the

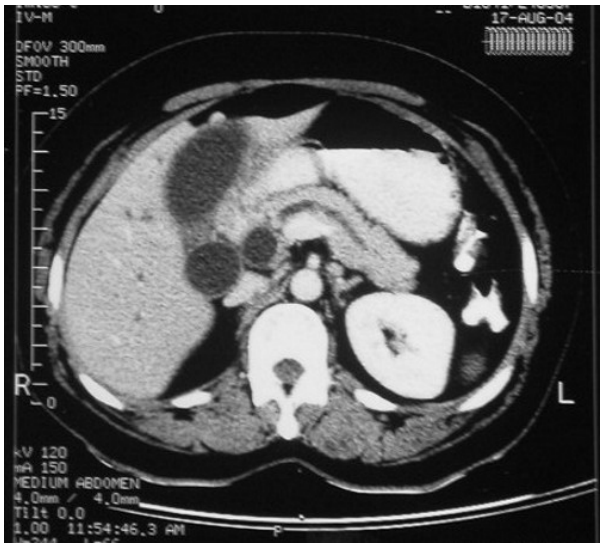

Figure 1. Contrast-enhanced CT scan (CECT) showing a dilated common bile duct, a dilated pancreatic duct, and a thick walled gallbladder. There is no mass lesion.

pancreas on ultrasonogram. At the time of referral, she had a bilirubin level of 7.9 mg/dL (reference range: 0.10-1.30 mg/dL), alkaline phosphatase of 1,901 IU/L (reference range: 35-150 IU/L), SGOT of 187 IU/L (reference range: 5-40 IU/L), and SGPT of 284 IU/ L (reference range: 5-40 IU/L). With a diagnosis of malignant biliary obstruction, she was referred to us for further management.

She underwent a contrast-enhanced CT scan (CECT) of the abdomen. It revealed bilobar intrahepatic biliary radical dilatation and a dilated common bile duct extending to the lower portion. The pancreatic duct was also dilated. No mass was seen on CECT (Figure 1). A periampullary malignancy was suspected. During this period, she had another attack of cholangitis which did not respond to broad spectrum antibiotics, and her bilirubin level rose to 14.8 mg/dL. Her cholangitis was managed with an endoscopic papillotomy and drainage. Endoscopy did not reveal any ampullary lesion (endoscopic picture not available). A common bile duct bulge was seen at the medial wall of the second part of the duodenum. Multiple biopsies were taken from the ampullary region which did not show malignant cells or any other specific pathology. With a strongly suspected but unproven diagnosis of periampullary malignancy, she underwent a pancreaticoduodenectomy (PD) two weeks later.

At laparotomy, a hard growth was palpable at the ampullary region. A few soft retropancreatic lymph nodes (the largest measuring 2 cm) were also present. The gallbladder was distended and thick-walled and there was a 30 mm calculus in the lumen. There was also intense pericholedochitis involving the first part of duodenum. The head and uncinate process of the pancreas were firm and fibrotic. A classical Whipple's PD was carried out. Postoperative recovery was uneventful and the patient was discharged on the 8th postoperative day.

Gross examination of the resected PD specimen revealed an irregularly thick-walled gallbladder with a maximum thickness of 10 mm. There was a necrotic area of 30x25 mm at the fundus of the gallbladder. A grayish white area of thickening measuring 20x5 mm was seen at the ampulla of Vater. The common bile duct wall was 3 mm thick. Microscopy of the ampulla revealed ulceration and dense panmural inflammatory cell infiltrate comprised of plasma cells, lymphocytes and sheets of foamy histiocytes with focal antral metaplasia of the lining epithelium (Figure 2). These changes extended from the gallbladder through the common bile duct to the ampulla. At the ampulla, the inflammatory cell infiltrates

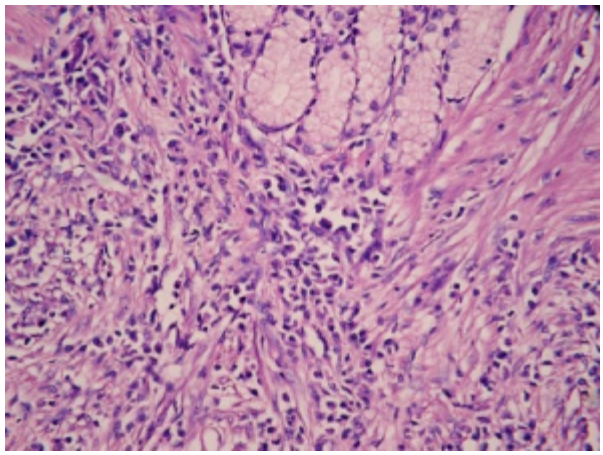

Figure 2. Histopathological examination of the ampullary region showing Brunner's glands with mixed inflammatory cell infiltrate (plasma cells, lymphocytes and histiocytes) consistent with xanthogranulomatous inflammation.

extended transmurally from the duodenal mucosa to the superficial pancreas. No evidence of malignancy was seen. All the lymph nodes showed reactive hyperplasia. A diagnosis of xanthogranulomatous inflammation was made.

DISCUSSION

Xanthogranulomatous cholecystitis is a destructive form of chronic cholecystitis characterized by the presence of grayish nodules or streaks in the gallbladder wall, mainly caused by lipid laden macrophages. The disease can mimic gallbladder cancer and can be locally advanced with involvement of adjacent organs which may necessitate an enbloc resection because of the difficulty in distinguishing it from a carcinoma by pre-operative investigations [2]. Involvement of the common bile duct by the inflammatory process can result in obstructive jaundice. Common causes of jaundice include the Mirizzi syndrome due to extrinsic compression of the common bile duct, primary involvement of the common bile duct by the inflammation (xanthogranulomatous choledochitis) or associated gallbladder malignancy [1, 3, 4, 5]. Xanthogranulomatous inflammation presenting with obstructive jaundice due to the involvement of the lower bile duct or the head of pancreas is rarely reported [5, 6]. As most periampullary lesions are malignant, patients with this clinical presentation are usually subjected to a pancreaticoduodenal resection.

From January 1989 to December 2003, 4,800 cholecystectomies were performed at the authors' institution. A xanthogranulomatous inflammation was seen in 453 cholecystectomy specimens (9%). Forty three out of 453 (9%) patients had a total bilirubin greater than 3 mg/dL. The predominant causes for an elevated bilirubin level included choledocholithiasis in 23 (53%) patients, associated gallbladder cancer in 7 (16%) and Mirizzi syndrome in 9 (21%). In 214/453 (47%) patients, the gallbladder wall was thick (greater than 4 mm on ultrasonography).

Xanthogranulomatous inflammation of the ampulla causing obstructive jaundice has not previously been reported. In the experience reported with xanthogranulomatous choledochitis and xanthogranulomatous pancreatitis, pancreaticoduodenectomy was performed [5, 6]. In our patient, it is the involvement of the ampulla with this inflammation which resulted in this peculiar clinical presentation. Repeated biopsies taken from the ampulla usually will fail to reveal malignancy. It is probable that the use of intraductal sonography, particularly in combination with per-oral cholangioscopy, could give a better anatomy of the lesion and under their guidance, fine needle aspiration cytology could accurately diagnose the condition pre-operatively [7]. We do not have any experience of this modality at our centre. However, given a tissue diagnosis of ampullary xanthogranulomatous inflam-mation, the absence of malignancy could still prompt the same surgical treatment because of the known association between this pathology and malignancy. Although the disease is rare, it should be added to the differential diagnosis of patients presenting with a suspected periampullary lesion and a thick-walled gallbladder.

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Keywords Ampulla of Vater; Common Bile Duct; Pancreaticoduodenectomy

Abbreviations PD: pancreaticoduodenectomy

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