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A complication of xanthogranulomatous cholecystitis with Mirizzi syndrome

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Abstract. – A patient had right upper quadrant pain with sclera was transferred from emergency room to the hospital, she was proposed to have acute cholecystitis, gallstones, obstructive jaundice, and a four-year history of gallbladder stones. The NMR results showed that the gallbladder was significantly enlarged and the gallbladder wall was thickening irregularly. The liver morphology was not abnormal except with extensive intrahepatic bile duct dilatation. The MRCP results demonstrated that the intrahepatic bile ducts were significant expanded. The ERCP results showed that duodenal stenosis and extrahepatic bile duct stenosis. We placed a plastic stent of 8.5Fr and 12 cm in length in the hepatic duct, and after biliary plastic stent placement, jaundice was rapidly reduced and liver function was improved significantly. A surgery was performed and the final pathologic diagnosis is a complication of Xanthogranulomatous cholecystitis with Mirizzi syndrome. After the surgery of cholecystectomy and a bile duct repair were performed, the patient was recovered well. Conclusively, if a patient was diagnosed as biliary stricture, a biliary metal stent should not be placed until pathological diagnosis of malignancy.

Key Words:
Xanthogranulomatous cholecystitis, Mirizzi syndrome.

Case Report

A 72-year-old female patient had right upper quadrant pain with yellow skin and sclera for 10 days, and one day later the symptom had been severe. Transferred from the emergency room to the hospital, she was proposed to have acute cholecystitis, gallstones, obstructive jaundice, and a four-year history of gallbladder stones.

The patient's blood sample contains WBC 13.69 × 10^ 9/L, neutrophils 91.8%. Liver function parameters were measured, total bilirubin 169.1 umol/L, alanine aminotransferase 146 U/L, aspartate aminotransferase 184 U/L, alkaline phosphatase 813 U/L, and γ-glutamyl transpeptidase 486.1 U/L. B Ultra results indicated a possible cholecystitis, and multiple clumps occupying the gallbladder, possibly gallstones. It was also observed that the intrahepatic bile ducts were overwhelmingly dilated.

After admission to the hospital, the patient was fasted and I.V. injected saline with antibiotics to protect the liver from further deterioration. However, liver damage continued to increase, i.e., total bilirubin 236.68 μmoL/L, conjugated bilirubin 138.60 μmoL/L, bilirubin 98.1 μmoL/L, alanine aminotransferase 129 IU/L, aspartic acid aminotransferase 126 IU/L, alkaline phosphatase 814 IU/L, γ-glutamyl transferase 487.29 IU/L, the tumor markers carbohydrate antigen 19-9 > 1000.0 U/mL, alpha-fetoprotein (AFP) 3.06 ng/mL, carcinoembryonic antigen (CEA) 1.81 ng/ml.

From the NMR results (Figure 1), the gallbladder was significantly enlarged, containing multiple round weak signals with different sizes up to about 3x2 cm. The gallbladder wall was thickening irregularly. The liver morphology was not abnormal with a clear surface and a normal proportion, except that extensive intrahepatic bile duct dilatation was observed with no abnormal enhancement. The spleen has a proper size and the internal signals were evenly distributed.

The results of Magnetic Resonance Cholangiopancreatography (MRCP) demonstrated that no gallbladder was displayed and the intrahepatic bile ducts were significant expanded. The top end of the common bile duct in the segment display is unclear, and the bottom end of the common bile duct was not dilated. The hepatic duct was no clearly displayed.

We reached the conclusion that the patient developed multiple gallbladder stones, cholecystitis, and wait for the clinical results to exclude the possibility of gallbladder cancer lesions. As the upper section of the common bile

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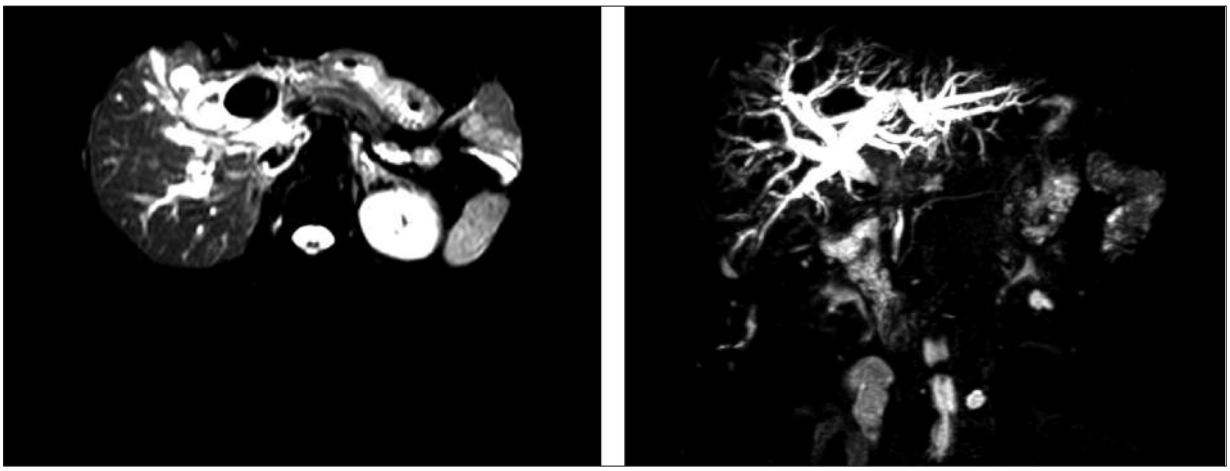

Figure 1. NMR results show that the gallbladder was significantly enlarged and with different sized multiple round weak signals.

duct is unclear from NMR (Figure 1) and the fact of overwhelming dilatation of the intrahepatic bile duct, the patient was recommended for further examination.

The results of endoscopic retrograde cholangiopancreatography (ERCP) showed that duodenal stenosis was developed due to external pressure, but still wide enough for duodenoscopy (Figure 2). The extrahepatic bile duct stenosis in the segment eccentric was developed for about 2 cm long, with a significant dilatation of the top bile duct. Within the gallbladder, there was a filling defect in an oval duct of 3 cm in diameter.

We placed a plastic stent of 8.5 Fr and 12 cm in length in the hepatic duct, with the other end outside of the nipple (Figure 3). After biliary plastic stent placement, jaundice was rapidly reduced and liver function was improved significantly. The results of ERCP and endoscopic retrograde biliary drainage (ERBD), performed after eight days of stent placement, suggested dramatic improvement of liver function, i.e., total bilirubin 56.53 μmoL/L, direct bilirubin 27.93 μmoL/L, bilirubin 28.6 μmol/L, alanine aminotransferase 25 IU/L, aspartate aminotransferase 38 IU/L, alkaline phosphatase 542 IU/L, γ-glutamyl transpeptidase 420.11 IU/L.

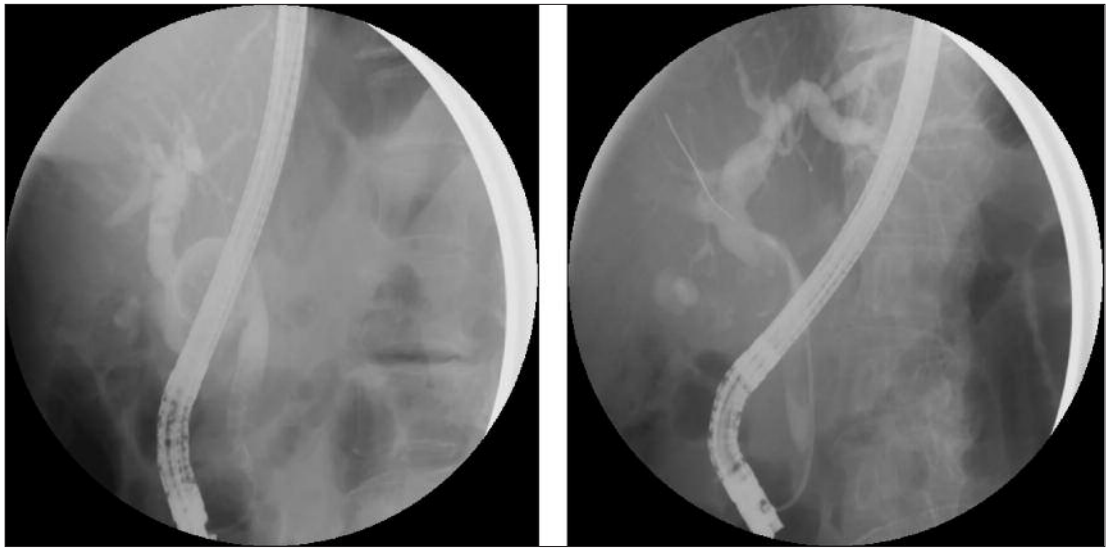

Figure 2. ERCP results show that the duodenal stenosis was developed due to external pressure, but still wide enough for duodenoscopy.

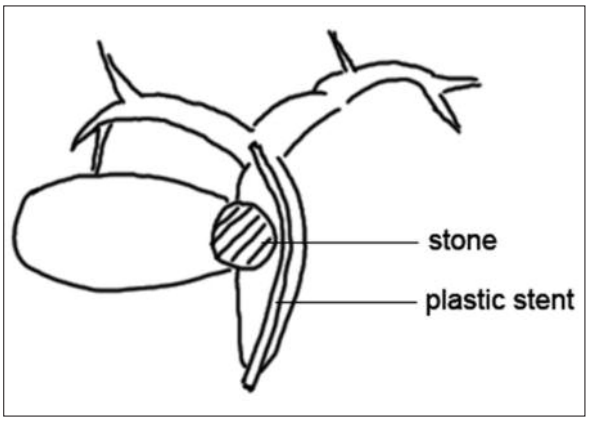

Figure 3. A schematic showing the position of plastic stent and the gallblander stone

After 13 days of ERCP, a surgery was performed. The patient was diagnosed as acute cholecystitis and gallstones. Meanwhile, the tumor cells of gallbladder had invaded into bile duct and duodenum, indicating that the patient may have a Mirizzi syndrome.

During the surgery, it was observed that the gallbladder was densely intermixed with omentum and duodenum dense adhesions, with severe edema. The gallbladder had a size of 7 × 5 × 5 cm and wall thickness of 1 cm, with white purulent bile in the gallbladder. The gallbladder was consolidated and hard at the bottom, with a size of 3 × 3 × 3 cm. The gallbladder and hepatic duct together form a gallbladder-hepatic duct fistula, and the cystic duct disappeared. The fistula has a round impacted stone with a 3 cm diameter, which squeezed the hepatic duct and led to biliary obstruction. The common bile duct, about 1 cm in diameter, had a plastic biliary stent in place but no stones. The gallstone squeezed the ball part of duodenum. A pathology analysis indicated an acute cholecystitis with local granuloma formation. We performed the surgery of cholecystectomy in the gallbladder neck, removed the stones, and repaired the damaged common bile duct wall with the gallbladder wall.

As shown in Figure 4, the pathological diagnosis is that the limitations of the gallbladder wall structure was damaged, and was replaced with granulomatous structure. The central of gallbladder was filled with necrotic tissues, and the outer layer was full of lipid-filling foam cells and proliferative fibroblasts. All these symptoms indicated the patient had a yellow granulomatous cholecystitis. Therefore, with all these data, the patient was finally diagnosed as a yellow granulomatous cholecystitis, incarcerated gallstones and type IIb Mirizzi syndrome. After a week of the surgery, the patient was recovered well.

Discussion

Xanthogranulomatous cholecystitis is an inflammatory lesion based on chronic inflammation of the gallbladder, with the formation of a yellow granuloma, severe fibrosis and foamy cells. Xanthogranulomatous cholecystitis caused continuous thickening of the gallbladder wall, and adhesions with the surrounding organs and tissues to form lumps, leading to unclear boundaries of adjacent organs. The special cholecystitis may spread to adjacent liver tissue, which would otherwise have seemed to be gallbladder tumors metastatic to the liver if only based on imaging result. It was very difficult to distinguish between the two[1,2]. It was reported that 54.05 percent of xanthogranulomatous cholecystitis patients had increased carbohydrate antigen 19-9, which was difficult to be diagnosed before a surgery and only can be confirmed by pathological analysis of biopsy obtained from surgery[3].

Mirizzi syndrome (Mirizzi syndrome) refers to a series of syndrome composed of cholangitis and obstructive jaundice, caused by obstruction of the common liver duct or bile duct in varying degrees. The obstruction was a combinatory effect of stone impaction in the gallbladder neck or cystic duct, and (or) compression of other nearby benign diseases or inflammation. Mirizzi syndrome is actually a complication of cholelithiasis, rather than one single disease[4,5]. Mirizzi syndrome is also associated with increased carbohydrate antigen 19-9[6]. Incarceration of gallstones may squeeze the common liver duct or bile duct, and cause biliary strictures, which is similar to gallbladder tumor invasion of the bile duct. It is difficult to distinguish solely based on the radiographic data[7].

In the preoperative examination, we documented the following data. First, MRI showed that irregular thickening of the gallbladder wall and interruption of the bile duct, which may lead to a seemingly correct conclusion that tumorigenesis of gallbladder invaded bile duct. Second, ERCP showed that there was external pressure duodenal stenosis and extrahepatic bile duct had eccentric stenosis, supporting the seemingly correct diagnosis that gallbladder tu-

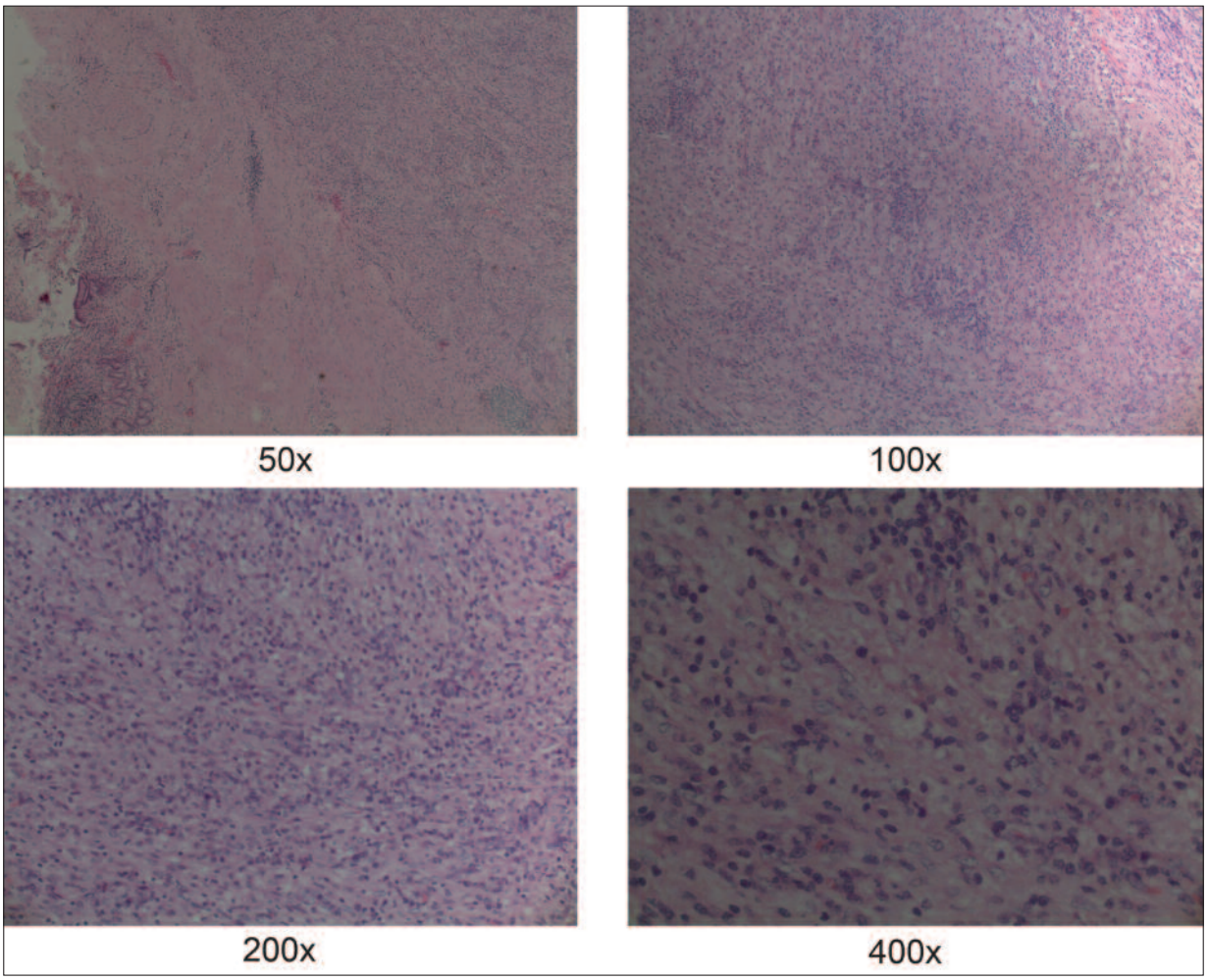

Figure 4. Immunohistochemistry of the paraffin biopsy shows that the limitations of the gallbladder wall structure was damaged, and was replaced with granulomatous structure.

morigenesis violated duodenal and extrahepatic bile ducts. Third, the tumor markers carbohydrate antigen 19-9 > 1000.0 U/mL also supported the diagnosis of gallbladder cancer. If solely looking from the MRI result, the gallbladder tumor was unresolvable, and in order to relieve biliary obstruction, doctors would often have installed permanent biliary metallic stent. In fact, the patient just developed a benign disease with expected long lifespan. If installed metal stent, the biliary sludge would be clogging the biliary metal stent after one year, leading to recurrent cholangitis and jaundice. The wrong diagnosis would need another ERCP exam and a plastic stent eventually would have to be placed in the metal bracket to clear through the bile duct. In the worst case, the biliary sludge would gradually spread up to the intrahepatic bile ducts and cause cholangitis. Doctors would have to replace periodically the plastic stent, making worse complications.

However, the final pathologic diagnosis, in this case, is a benign disease, i.e., yellow granulomatous cholecystitis. As long as a cholecystectomy and a bile duct repair surgery were performed, the patient was recovered.

Conclusions

If a patient was diagnosed as biliary stricture, a biliary metal stent should not be placed without further careful examination. It should only be placed after pathological diagnosis of malignancy.

Conflict of Interest

The Authors declare that there are no conflicts of interest.

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