Case Report

Recurrent cholangitis caused by common bile duct compression due to hepatic artery variation

Wu Tian1*, Qingrong Yao2*, Zeyan Duan1, Chunlai Li1, Deyuan Huang1

¹Department of Hepatobiliary Surgery, The First People's Hospital of Guiyang, Guiyang 550002, Guizhou, China; ²Ultrasonic Center, The People's Hospital of Guizhou Province, Guiyang 550002, Guizhou, China. *Equal contributors and co-first authors.

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Abstract: Many different benign and malignant diseases can cause extrahepatic biliary duct obstruction. Common causes include calculus of the bile duct, stenosis, and tumors. Bile duct obstruction is often complicated bybacterial infection due to acute cholangitis, leading toan increase in the bile duct pressure and entry ofa large number of bacteria and toxins into the blood circulation. Consequently, a severe infection of the biliary tractoccurs, resulting in acute abdomen; this infectionis a major benign biliary tract disease and the most common direct cause of death. In biliary surgery, anatomic variation is often noted in the biliary ductarea, and anatomic variation of the gallbladder artery or hepatic artery is common. However, extrahepatic bile duct compression by the proper hepatic artery has rarely been reported. A 56-year-old woman was admitted to our department with a 2-year history of repeated upper abdominal pain associated with fever. Four years before her admission, she had undergone laparoscopic cholecystectomy. She experienced recurrent abdominal pain every 1 or 2 months, often accompanied by fever (maximum temperature, 40.5 °C) and the passage of dark-yellow urine. Gastroscopy showed chronic non-atrophic gastritis. Abdominal ultrasonic examination and computed tomography did not show any abnormality. However, magnetic resonance cholangiopancreatographyshowedan angulated common bile duct with no stone, tumor, or other disease. Because the cause of the biliary obstruction was unclear, surgical exploration was decided fora clear diagnosis and determination of the appropriate treatment.

Keywords: Vascular malformation, obstructive jaundice, cholangitis

Introduction

Many different benign and malignant diseases can cause extrahepatic biliary duct obstruction. A rather serious complication of biliary duct obstruction is cholangitis, which leads to acute cholangitis. Anatomic variations in the biliary ductregion are common; however, extrahepatic bile duct compression by the gallbladder artery or the proper hepatic artery has rarely been reported [1].

In the case reported here, because the cause of biliary obstruction was unclear, surgical exploration was decided for clear diagnosis and determination of the appropriate treatment.

Case report

A 56-year-old woman was admitted to our department with a 2-year history of repeated

upper abdominal pain associated with fever. Four years before her admission, she had undergone laparoscopic cholecystectomy. She experienced recurrent abdominal pain every 1 or 2 months, often accompanied by fever (maximum temperature, 40.5°C) and the passage of dark-yellow urine. Gastroscopy repeatedly showed chronic non-atrophic gastritis. Repeated abdominal ultrasonic examination and computed tomography did not show any obvious abnormality in the liver, bile tract, or pancreas. She was diagnosed with gastritis or postcholecystectomy syndrome at different hospitals and was administeredanti-inflammatory or anti-acid treatment, but the outcomes were unsatisfactory. The patient's weight had reduced by approximately 5-8 kg over the previous 2 years. The results of the physical examination after admission indicated normal body temperature without jaundice and epigastrium



Figure 1. MRCP image.



Figure 2. The retention suture and the CBD, with the proper hepatic artery lying superiorly.



Figure 3. The T-tube placed into the CBD, with the proper hepatic artery lying superiorly.

tenderness. Liver functiontest results indicated increased alanine transaminase (82U/L) and aspartate transaminase (104 U/L) levels. The total bilirubin level was 31.2 mmol/L, and the conjugated bilirubin level was 22.9 mmol/L. Gastroscopy showedchronic non-atrophic gas-

tritis. Abdominal ultrasonic examination and computed tomography did not show any abnormality. However, magnetic resonance cholangiopancreatography showedan angulated common bile duct (CBD) (Figure 1), with no stones. tumors, or other diseases, caused by the obstruction. An exploratory laparotomy through a right upper abdominal rectus incision was performed. After careful dissection, the proper hepatic artery (PHA) was found to be markedly ectatic, with a diameter of 10 mm, but not obstructed. The dilated PHA adhered to and compressed the CBD, resulting in the deviation of the CBD from its normal path toward the left and its angulation. The diameter of the CBD was 9 mm (Figures 2, 3). The distal end of the PHA did not show obstruction or compression. The variation of the dilated PHA in the case of this patient was considered congenital. The CBD compression was caused by the effects of adhesion after laparoscopic cholecystectomy. For the complete release of the adhesion between the PHA and CBD, the CBD was dissected and incised, and biliary tract endoscopy was performed. Choledolithiasis was not detected, and the CBD was unobstructed. A type-T drainage tube was placed in the CBD during the operation. The patient recovered satisfactorily, and the drainage tube was withdrawn after cholangiography showed normal findings at 2 weeks after the operation. At the 3-year follow-up, the patient did not experience abdominal pain or show jaundice; she showeda good appetite, with a weight gain of 10 kg.

Discussion

Extrahepatic bile duct obstructionis caused by several factors, including benign conditions such as stones, Mirizzi's syndrome, postoperative stricture, polyps, and anatomic variations and anomalies [2]; it is also caused by malignant conditions such as tumors of the CBD, pancreas head, or Vater. The functions of some tubularorgans such as the esophagus, duodenum, or ureter can be deteriorated because of compression of the adjacent blood vessels. In the rare case presented here, the CBD was compressed by the passage of the PHA. The CBD was divertedand angulated and led to an incomplete obstruction. Bile stasis proximal to the site of CBD obstruction may have triggered the symptoms of cholangitis, such as abdominal pain, fever, and jaundice. Over the course of

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time, recurrent cholangitis could result in fibrosis of the bile duct wall, and, along with the bile stasis, could facilitate the formation of choledolithiasis. Surgery is aneffective therapy to relieve CBD compression through restoration of the normal anatomic structure of the CBD and prevention of the formation of choledolithiasis. Vascular malformations should be considered as a possible cause of extrahepatic biliary obstruction. Computed tomography angiography may be helpful in the identifi cation of these malformations [3], but ma-Iformation sare usually overlooked before surgery. Identification of malformations is a common clinical challenge, and hence, the present case is rare in clinical practice. Therefore, in the future, in cases where preoperative examination before surgery cannot help determine the exact cause of extrahepatic biliary obstruction, an anatomic variation with vascular compression of the extrahepatic bile duct should be considered and computed tomography angiography should be performed.

Disclosure of conflict of interest

None.

Address correspondence to: Qingrong Yao, Ultrasonic Center, The People's Hospital of Guizhou Province, Guiyang 550002, Guizhou, China. Tel: +86-18508516119; Fax: +86-21-64085875; E-mail: yaoqingrong1980@126.com

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