Case Report

A Case Report of Bronchobiliary Fistula and Literature Review

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Bronchobiliary fistula (BBF) is one of the very rare disease. In this report, we described a BBF case. The case was diagnosed by endoscopic retrograde cholangio-pancreatography (ERCP) and percutaneous transhepatic cholangial drainage (PTCD) examinations, and treated properly. From the diagnosis of this BBF case, a patient with cough, biliptysis, fever and pain, should be considered for diagnosis of BBF.

Key words: Diagnosis; Bronchobiliary fistula; ERCP; PTCD; Misdiagnosis

Bronchobiliary fistula (BBF) is one of the very rare clinical conditions. It is caused by a pathological communication between the biliary channels and the bronchial tree.¹ BBF is easily misdiagnosed. In this paper, we report one case of BBF in Qinhuangdao No.3 Hospital and review the papers related to BBF in the literature.

CASE REPORT

A 48-year-old male patient was admitted to our department with a two-day history of cough, yellow-green sputum and fever along with chills. Physical examination revealed moderate jaundice of skin, an abdominal scar, absent breath sounds in the right lower lobe and a biliary drainage tube with no bile. His past medical history revealed that he had been diagnosed as HCC (hepatocellular carcinoma) by Beijing Youan Hospital 38 months ago, and had undergone TACE (transhepatic arterial chemotherapy and embolization) at that time. Five months ago, he came to our hospital due to yellow sclera and yellow urine for 7 days, and abdominal pain for 2 days. He was diagnosed as "gall stones" and undergone a cholecystectomy in our hospital. After one month, he occurred obstruction of biliary tract and was treated by endoscopic retrograde cholangio-pancreatography (ERCP) and percutaneous transhepatic cholangial drainage (PTCD) in Beijing Chaoyang Hospital. The drainage after operation worked well till 2 days before admission. His blood test on presentation showed increased bilirubin levels (total bilirubin of 228.9 mmol/L and conjugated bilirubin of 76.7 mmol/L), and sputum test revealed a total bilirubin level of 144 μ mol/L, conjugated bilirubin level of 78 μ mol/L. Thoracic computed tomography (CT) revealed

that the right lung had patchy shadows, middle lobe atelectasis and pleural effusion. Upper abdominal CT showed no bile in bile duct and subphrenic abscess occurred. The patient was treated with intravenous antibiotics. Intensive supportive therapy also included high calorie infusion. His body temperature returned to normal after two days. However, other symptoms still continued. ERCP was performed which showed intra-hepatic tract deformation with the contrast agent appearing in the bronchi. Therefore, the case was diagnosed as a "broncho biliary fistula (BBF)". As this patient with BBF also had malignant disease, palliation treatment was considered as alternative treatment and establishing an unobstructed biliary drainage was taken as the preferred choice. After this, he recovered rapidly and complained of less cough. The total amount of biliptysis also reduced rapidly.

DISCUSSION

BBF is very uncommon and requires a high degree of clinical suspicion. Peacock reported the first case of BBF in 1850 which was caused by liver hydatidosis.² BBF is caused by several reasons including hereditary causes and non-hereditary causes (Table 1). Wu et al reported that approximately over 50 percent BBF were due to biliary tract diseases, which may cause hepatopostema. Furthermore, hepatopostema is the major cause of BBF.³ While this case was caused by liver and biliary tree tumor.

The theories of mechanisms resulting in BBF have not been confirmed. It is said that it might be due to different reasons leading to subdiaphragmatic abscess and an inflammatory reaction into the bronchial tree, which subsequently creates a fistula between the biliary and the bronchial system. BBF often presents with cough, biliptysis, pain and fever, and

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Table 1. Causes of bronchobiliary fistula

Hereditary causes	Non-hereditary causes
Congenital malformation	Hydatid cyst, trauma, postsurgical states, liver or biliary tree tumor (primary tumor & metastatic tumor), chronic pancreatitis, biliary stenosis, subphrenic abscess, hepatic hydatidosis, syphilis gummosa, uterine leiomyosarcoma with hepatic metastases, cholangiolithiasis, mucinous adenocarcinoma, acute cholecystitis in a suprahepatic gallbladder

this case presented all the symptoms except pain.⁴ The common tool to evaluate the pathologies of lung and liver are sputum analysis and CT images. MR (magnetic resonance) cholangiography may help to investigate the pathology of a communication between the biliary tree and the bronchial tree. Zhou et al argued that fiberoptic bronchoscopy may not help to visualize a BBF; therefore, it is not suggested to be carried out routinely.⁵ BBF can be an aggressive disease with a high fatality rate. Normally it needs to be treated urgently by endoscopic or open surgical method. The different modalities of treatment include surgery, ERBD (endoscopic retrograde cholangio-pancreatography), PTC, endoscopic sphincterotomy alone, endoscopic stone extraction and sphincterotomy, endoscopic sphincterotomy and stone extraction with percutaneous drainage. Correct treatment depends on the individual cause of BBF and other associated conditions. This patient had a short life expectancy due to liver and bile malignancy and a combined approach of biliary drainage with intensive supportive therapy which provided satisfactory palliation.

CONCLUSIONS

BBF is very rare and caused by different conditions. Therefore, the diagnosis of BBF requires a high degree

of clinical suspicion when patients occurred cough, bilioptysis, fever and pain. Only several cases were reported in China and abroad. Till now, there is no standardized regimen to follow. In our opinion, as each patient condition is rather diversified, suitable management of BBF should be tailored to their individual condition.

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