



Case report

Bile in bronchi: A case report

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ABSTRACT

Background: The biliary bronchial fistula is rare and difficult to treat. Here we report a 49-year-old woman diagnosed with biliary bronchial fistula due to cough with yellow-green sputum.

Case presentation: this is a typical case of the biliary bronchial fistula with typical symptoms. The position of the abscess cavity below the diaphragm could not be catheter drainage. After anti-infection treatment, yellow-green sputum was reduced. Follow-up showed a good prognosis.

Conclusion: biliary bronchial fistula is rare in the clinic, combined with chest and abdomen infection.

1. Background

The biliary bronchial fistula (BBF) is a rare condition characterized by abnormal communication between the biliary tract and the bronchial trees [1]. Peacock [2] in 1850 described the first case of BBF in a patient with hepatic hydatidosis. Since then, many studies have been carried out to improve the management of BBF. Here, we present a case of biliary - bronchial fistula diagnosed and treated in our department. We further describes the etiology, clinical manifestations, diagnosis and treatment of biliary - bronchial fistula to provide a reference for future diagnosis and treatment.

2. Case presentation

A 49-year-old Chinese female patient was admitted to hospital on April 12, 2019, after presenting with a cough that had persisted for 10 days. The patient was also coughing up about 100–200 mL of a yellow-green sputum per day. However, the patient did not present any other characteristic symptoms such as fever, coughing of phlegm and chest pain. Upon seeking medical help at the local hospital, there was no significant improvement in symptoms. The patient then visited our department and received High-resolution Computed tomography (HRCT) of the chest and upper abdomen. HRCT revealed that an abscess in the right lower lobe broke through the diaphragmatic muscle and affected the lower diaphragm but the impact on the liver was unclear. In

addition, cholangitis was evident and there were multiple stones in the intrahepatic bile duct with an accumulation of gas and dilatation. Magnetic Resonance Cholangiopancreatography (MRCP) showed intrahepatic and external bile duct dilatation and multiple intrahepatic bile duct stones. In 2006, the patient underwent cholecystectomy, choledocholithotomy, and choledocholithostomy due to the presence of intrahepatic and extrahepatic bile duct stones and cholecystolithiasis. In May 2018 and March 2019, the patient presented with chills, and high fever of up to 39.5 °C. The symptoms improved after drug treatment. Based on the medical history, symptoms and imaging data (Fig. 1), the patient was diagnosed with BBF. Upon admission, laboratory examination indicated the presence of inflammatory indicators, high bilirubin levels in sputum, low protein levels and significantly increased tumor marker CA125 and normal CA199 and CEA (Table 1). The patient had a pulmonary infection and biliary tract infection which was treated with Cefmetazole. On April 13, 2019, a chest ultrasound evaluated the abscess cavity of the lung and puncture catheter drainage was planned to fully control the pulmonary infection. Ultrasound showing right chest exploration and mixed echo group close to the spine makes it impossible to perform catheterization. Pathogenic examination and detection index changes should be recommended by a physician after evaluation, and a catheter used to cover anaerobic bacteria and cocci for anti-infection treatment and to improve bronchoscopy examination where necessary. Thereafter commencement of biliary fistula treatment followed by a re-evaluation of the biliary leakage if cured or if surgical treatment is

Abbreviations: BBF, Broncho biliary fistula; HRCT, High-resolution computed tomography; MRCP, magnetic resonance cholangiopancreatography; ERCP, endoscopic retrograde cholangiography; PTCD, percutaneous transhepatic cholangial drainage.

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needed. This is because the current operations are risky and the patients may develop postoperative complications. In this case, after treatment, the patient was gradually relieved from the symptoms of cough and sputum which disappeared without bronchoscopy examination. Chest ultrasound evaluation was performed on April 23, 2019, with no obvious abscess cavity found in the right chest cavity. Furthermore, a plain chest CT scan showed that the original lesion in the lower lobe of the right lung had been absorbed, and the intrahepatic bile duct pneumatosis was reduced. The diagnosis of the biliary bronchial fistula was clear, and the pulmonary and abdominal infection lesions were controlled using drug treatment. Surgical treatment in the presence of edema which is part of the inflammatory process is not suitable, therefore, pulmonary infection and biliary inflammation should continuously be treated and controlled. Anti-infection treatment was offered in the outpatient department. The operation time and mode were assessed after returning to the hospital on May 19, 2019, and upon further investigations fiber bronchoscopy was found to have improved, suggesting inflammatory changes, but no obvious fistula was found. Subsequently, liver parenchyma was incised and removed for lithotomy and cholangioscopy to detect fistula resection and diaphragmatic repair respectively. The patient was successfully discharged after surgery, and followed up in the outpatient department during a four-month period and reported a good recovery path.

3. Discussion and conclusion

According to its etiology biliary bronchial fistula can be divided into two; congenital and acquired. Congenital biliary bronchial fistula is relatively rare and was first reported by Neuhauser [3] in 1952, as a rare malformation of the digestive tract. On the contrary, the causes of acquired biliary bronchial fistula are diverse, including local infection factors caused by a liver abscess or subdiaphragmatic abscess, obstruction factors caused by intrahepatic and external biliary stones or tumors, tumor invasion factors of liver and biliary tract near the diaphragm, chest and abdomen trauma and iatrogenic injury etc [4,5]. Eryigit [4] reported that hepatic hydatidosis was the main cause of acquired biliary bronchial fistula, however other factors, especially iatrogenic injuries complicated biliary bronchial fistula. More recently, hepatobiliary surgery and endoscopic operation, have increased significantly [6]. The most common symptoms of BBF are sputum, fever, abdominal pain, jaundice, and dyspnea. In the current case, the patient reported the main symptom as daily coughing of up to 100–200 ml of yellow-green sputum. Combined with previous biliary tract surgery, there was increased risk in the formation of bile duct stones, and the possibility of

Table 1

Laboratory tests at admission, CA = carbohydrate antigen.

Laboratory measures	Reported values	Normal range
White blood cell count	$14.58 \times 10^9/L \uparrow$	$3.50-9.50 \times 10^9/L$
Neutrophil count	$12.07 \times 10^9/L \uparrow$	$1.80-6.30 \times 10^9/L$
Total bilirubin	13.3	$3.40-20.50 \mu\text{mol/L}$
Direct bilirubin	7	$0.00-6.80 \mu\text{mol/L}$
Albumin	31.4 ↓	$40.0-55.0 \text{g/L}$
Aspartate transaminase	48	$13-40 \text{U/L}$
Alanine aminotransferase	47	$7-45 \text{U/L}$
Alkaline phosphatase	138	$35-100 \text{U/L}$
Procalcitonin	0.511 ↑	$0.00-0.05 \text{ng/ml}$
Hypersensitive c-reactive protein	$>10.00 \uparrow$	$0.00-1.00 \text{mg/L}$
C-reactive protein	$>192.00 \uparrow$	$0.00-10.00 \text{mg/L}$
Carcinoembryonic antigen	1.99	$0.00-5.00 \text{ng/ml}$
CA-199	20.19	$0.00-37.00 \text{U/ml}$
CA-125	122.7 ↑	$0.00-35.00 \text{U/ml}$

BBF was considered. Hepatolithiasis is associated with bile duct stones, strictures and inflammation. Severe infection increased the pressure in the bile duct resulting in its bursting on the liver surface, forming a subdiaphragmatic abscess. The abscess then broke through the diaphragm and spread to the lower lungs to form a biliary duct – bronchial fistula through the bronchus.

One of the most prominent clinical manifestations of BBF is the expulsion of the bile in the original bile duct from the sputum through pathological channels, namely bilie-like sputum, and this is a specific symptom of BBF. The possibility of biliary fistula should be considered if the patient develops a yellow or yellow-green bile-like sputum and has potential risk factors of biliary fistula (such as biliary obstruction and infection). In addition, the detection of bilirubin in sputum can be used in the diagnosis of BBF. CT, MRCP and ultrasound examination are also of great significance in the diagnosis of BBF as they possess the ability to show the primary lesions of the liver and lung, evaluate the need for puncture catheter and stent drainage, however, it is difficult to directly find fistula [7]. Other diagnostic methods include Endoscopic Retrograde Cholangiopancreatography (ERCP), biliary angiography and bronchoscopy. ERCP and bronchoscopy play both diagnostic and therapeutic roles in controlling local infected lesions and in follow-up treatment. In this case, bilirubin was detected in sputum, and CT scan showed clear primary lesion of intrahepatic bile duct stones, obvious sinus tract between bile duct and bronchus, and this was a clear diagnosis of BBF.

BBF is often associated with pulmonary and abdominal infection. Therefore, sensitive antibiotics can be used to control local inflammation and actively deal with the primary disease. The main principle in

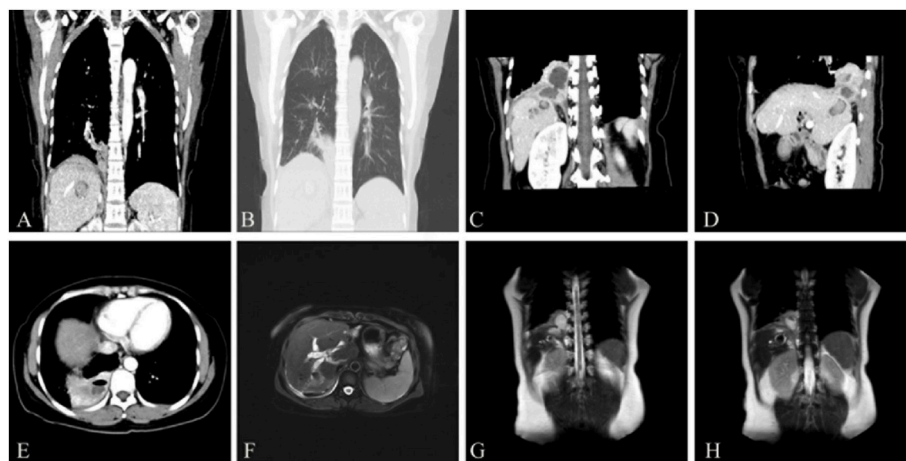


Fig. 1. Image data, (A, B) chest high-resolution CT showed patchy high-density shadows in the lower lobe of the right lung, (C, D, E) chest and upper abdomen enhanced CT showed pulmonary abscess in the lower lobe of the right lung involving the right diaphragm and the lower diaphragm, and unclear affinity with the liver. (F,G,H) MRCP, intrahepatic bile duct calculi, and biliary tract dilation.

BBF treatment is to first eliminate an obstruction, smoothen the drainage, remove the lesion and close the fistula. Surgical treatment is the classical treatment method for BBF, including drainage of the sub-diaphragmatic abscess, resection of fistula and liver and lung tissues, and reconstruction of bile to duodenum drainage [5]. For recurrent acute suppurative cholangitis of biliary calculi disease, the post-operative effect in the acute stage is not good. Therefore, surgery should be avoided at the acute stage, and selective imaging examination (ultrasound, Percutaneous Transhepatic Cholangial Drainage (PTCD) or ECRP should be conducted after disease remission to first understand the biliary tract system lesions, and achieve a thorough treatment in a single operation. Rokni Yazdi H [8], Pinsker N [9] and Honrubia Lopez R [10] reported cases of successful treatment of BBF with endoscopic embolization injection, while Na KJ et al. [11] reported cases of successful treatment of BBF with thoracoscopic assistance. Minimally invasive treatment is preferred for BBF with benign biliary tract obstruction caused by infectious diseases and also in patients with poor general health and surgery intolerance [12,13]. It is also suitable for palliative treatment of advanced malignant tumor associated with BBF [14]. In terms of prognosis, Sachdev et al [15] summarized and analyzed 27 cases of congenital biliary bronchial fistula, among which 4 patients died and the fatality rate was 4.8%. Liao [5] retrospectively analyzed 68 cases of acquired biliary bronchial fistula, and reported a cure rate of 87.7%. The onset time for this patient was relatively short, and the clinical symptoms improved after short-term anti-infection treatment and were successfully cured by surgery.

BBF is rare in clinical practice and most of the lesions are located in the chest and abdomen. The prognosis of the biliary bronchial fistula is closely related to the primary disease and the severity of disease complications. Therefore, selection of an optimal treatment modality for BBF requires full consideration of the specific cause, conditions and medical levels, as well as individualized treatment and this, calls for multidisciplinary collaboration.

Author's contribution

The HYF compiled the data and wrote the draft. LTY, MST operates and collects data. CZJ also assists in analyzing and interpreting data. HCY, YXP, and PT participated in the preoperative and postoperative evaluation of patients, this study was supported by PT. All authors contributed to the drafting and critical revision of the paper and gave conclusions approving the version to be released.

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Availability of data and materials

The datasets used and analyzed during the current study are available from the corresponding author on reasonable request.

Ethics approval and consent to participate

This study strictly followed the basic principles of medical ethics and

was approved by the medical ethics committee of the first affiliated hospital of Guangxi medical university.

Consent for publication

Written informed consent was obtained from the patient to publish this case report and the accompanying images.

Declaration of competing interest

The authors declare that they have no competing interests.

Acknowledgments

Not applicable.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.rmcr.2020.101075>.

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