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Case Report

A Case of Bronchobiliary Fistula Treated by Percutaneous Endobiliary Fistula Closure

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Abstract

Bronchobiliary fistula is a rare clinical condition with a characteristic clinical presentation of coughing bile-like sputum, which seriously affects the patient's quality of life. We report a case of a 60-year-old male patient with bronchobiliary fistula who was successfully treated with percutaneous hepatic puncture to closure the endobiliary fistula. The patient was followed up for 3 months without recurrence of bile-like sputum.

Keywords: Bronchobiliary fistula; Interventional therapy; Cholangiocarcinoma

Clinical Information

Male patient, 60 years old, was admitted with "cough and sputum for 1 week and fever for 2 days. The patient had a cough with no obvious cause and yellow bile-like sputum with bitter taste 1 week ago, and fever with a maximum of 39.6°C started 2 days ago. Past history: right hemicolectomy + hepatic caudatectomy + partial resection of hepatic segment 4b + cholecystectomy + left hepatic ductoplasty + left hepatic duct jejunostomy Roux-en-Y anastomosis on April 14, 2021; postoperative pathology: (liver) moderately differentiated intrahepatic cholangiocarcinoma with cancer visible in group 12b lymph nodes (1/1). Postoperative regularity to our hospital for chemotherapy treatment. The MRI examination suggested an encapsulated effusion in the right lobe of the liver with the effusion communicating with the right lower lung; abnormal signal in the head of the pancreas, involving the main pancreatic duct and portal vein, with truncation of the main pancreatic duct, distal pancreatic duct dilatation, and blurred and edematous peripancreatic soft tissue gap, considering tumor recurrence (Figure 1A-D). Fiberoptic bronchoscopy suggested a large amount of bile-like secretion in the right airway (Figure 1E). The secretion sent for examination suggested the presence of bilirubin component. The bronchobiliary fistula was considered to have formed; The symptoms did not improve after symptomatic treatment with internal medicine. After full communication with the patient and family and signing the informed consent form, percutaneous hepatic puncture was performed to closure the endobiliary fistula at an elective date.

The patient was taken to the interventional operating room. The operating area was disinfected, and after satisfactory basic + local anesthesia, the left intrahepatic bile duct was punctured under ultrasound guidance, and a 5F sheath was placed after successful bile duct puncture, and contrast was injected to reveal the left hepatic duct confluent with the intestine and a lamellar collection of contrast in the right hepatic region and visible communication with the bronchus, which was considered a endobiliary fistula (Figure 1F). Repeated attempts to access the proximal end of the fistula via the biliary anastomosis were unsuccessful. The fistula was then entered by direct puncture under fluoroscopy and the microcatheter was superselected to find the fistula. The fistula was first embolized with coils (five of 5mm and four of 4mm) and then embolized with approximately 3ml of a mixture of medical glue and super-liquidated iodine oil (1:2 mixture of medical glue: super-liquidated iodine oil). At the end of the operation, no fistula was visualized with the injection of contrast (Figure 1G), and the sheath was left in place and bandaged. The patient's coughing of yellow bile-like sputum was significantly reduced on the postoperative day. On the second postoperative day, there was no further coughing of yellow bile-like sputum. On the third postoperative day, the patient was sent to the interventional operating room for review, and the left sheath was injected with contrast and no fistula was visualized (Figure 1H), so the sheath was removed. The patient did not have any further coughing of bile-like sputum at the 3-month postoperative follow-up.

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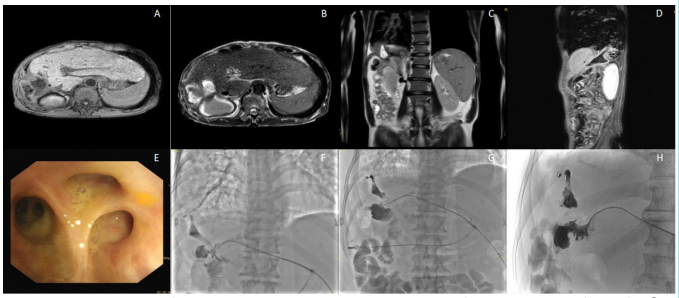

Figure 1A & B: The MRI examination suggested an encapsulated effusion in the right lobe of the liver with the effusion communicating with the right lower lung (Transverse plane); C Frontal plane; D Saggital plane; E Fiberoptic bronchoscopy suggested a large amount of bile-like secretion in the right airway; F DSA showed that the bile duct was connected to the bronchus; G No fistula was visualized after medical glue and coil embolization; H No fistula was visualized on postoperative day 3 review.

Discussion

Bronchobiliary Fistula (BBF) is a rare condition in which an abnormal traffic fistula is formed between the bile duct and bronchus by breaking through the diaphragm and bile can continuously flow into the bronchus through the fistula. The first case of BBF caused by hepatic hydatid disease was reported by Peacock in 1850 [1]. The mechanism of BBF formation remains unclear and can be classified as congenital and acquired. Congenital BBF is a developmental malformation of the biliary and respiratory systems, often starting in infancy and most of the fistulas are abnormal communications between the right bronchus and the bile ducts of the left lobe of the liver [2]. Acquired BBF often appears as a complication of diseases of the hepatobiliary system (liver tumors, biliary stones, inflammatory biliary strictures, hepatic hydatid disease, liver abscess, medical origin, etc.) [3]. The clinical symptoms of BBF are mainly manifested by cough, sputum, shortness of breath, chest pain, hemoptysis, fever, abdominal pain, and coughing bile-like sputum is a characteristic manifestation of the disease [4]. Relevant ancillary examinations can assist in the diagnosis and management of BBF; imaging examinations such as chest CT and airway reconstruction, MRI and MRCP show the presence of bile duct and bronchus abnormal communication; fiberoptic bronchoscopy shows bile-like sputum in the airway and bilirubin can be detected in the sputum; Percutaneous hepatic puncture cholangiography with bile duct injection showed contrast flow into the bronchus; Endoscopic Retrograde Cannulation of the Pancreatic (ERCP) showed that the bile duct communicated directly with the bronchus. the course of BBF is complex and difficult to heal, which seriously affects the quality of life of patients.

The main treatment methods for BBF are: 1) Surgical treatment: fistulectomy, fistula repair, lung segment or lobectomy can cure BBF radically [5]. However, surgical procedures are risky, traumatic, and require high physical condition of the patient; it is often difficult for patients with malignant and advanced tumors to tolerate surgical treatment. 2) Percutaneous hepatic puncture biliary drainage, nasobiliary drainage, and endobiliary stenting: by relieving biliary obstruction and reducing pressure in the bile duct, the bile output in the fistula is reduced and fistula healing is promoted [6]. However, there is no direct intervention of the fistula and there may be a non-healing of the fistula. 3) Selective bronchial occlusion: the output of the fistula is occluded under bronchoscopy by means of a bronchial silicone plug and/or a covered stent [7]. However, the output end of the fistula in bile duct bronchial fistula is often small, which makes it difficult to determine the location of the fistula and requires a high level of clinician experience. 4) ERCP: direct embolization of bile duct bronchial fistulas can be performed [8]. In patients with previous surgical procedures (e.g., biliary-intestinal anastomosis), the anatomy of the intestine and bile duct is altered, which may make the procedure more difficult. 5) Percutaneous hepatobiliary puncture fistula closure [9]: after percutaneous hepatic puncture of the bile duct, contrast is injected via the bile duct to clarify the location of the fistula, and the fistula is closed by embolization agent. The main embolization materials are coils and medical glue. The author suggests embolizing the fistula with a coil first and then combining it with medical glue to avoid incomplete embolization of the gap in the coil and the risk of misembolization and dislodgement of the medical glue. The embolization effect is enhanced by a structure similar to “steel and concrete” structure.

In bile duct bronchial fistula caused by malignancy, it is crucial to improve the quality of life of patients by sealing the fistula with minimally invasive treatment tolerated by patients. In this paper, the treatment of postoperative bile duct bronchial fistula after cholangiocarcinoma by percutaneous endobiliary fistula closure is safe, effective, and tolerated by patients.

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