



Surgical Treatment of Bronchobiliary Fistula with Pulmonary Resection and Omentopexy

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Bronchobiliary fistula is a rare disease defined as an abnormal connection between the biliary tract and the bronchial tree. We report the successful surgical repair of bronchobiliary fistula. A 78-year-old man underwent surgery and several rounds of transcatheter arterial chemoembolization and radiofrequency ablation as treatment for hepatocellular carcinoma. He presented with greenish sputum and chronic cough for several months, and his symptoms did not resolve after endoscopic treatment. We performed lobectomy of the right lower lobe and omentopexy for bronchobiliary fistula under laparotomy and thoracotomy. The bronchobiliary fistula was successfully closed, and the bilious sputum disappeared after surgery.

Keywords: Bronchial fistula, Omentopexy, Respiratory tract fistula

Case report

A 78-year-old man presented with bilious sputum after several rounds of transcatheter arterial chemoembolization (TACE) and radiofrequency ablation (RFA) as treatment for hepatocellular carcinoma (HCC). He was diagnosed with HCC 13 years ago, underwent central lobectomy of the liver, and received 3 rounds of TACE and 5 rounds of RFA, which extended until 8 years ago. Four years ago, the patient was hospitalized for a liver abscess, which was treated with percutaneous abscess drainage. Upon presentation, his symptoms included greenish sputum, as well as an irritating cough, which he had experienced for the past 3 months and which decreased immediately after insertion of a percutaneous transhepatic bile drainage (PTBD) catheter. Bronchobiliary fistula was diagnosed upon visualization of a fistula tract between the biliary tract and the right lower lobe bronchus by cholangiography through the PTBD catheter (Fig. 1), and fiberoptic bronchoscopy also showed biliary secretion from the RB8 bronchus in response to a saline injection through the PTBD catheter (Fig. 2). Although biliary secretion decreased after PTBD catheter insertion, the symptom recurred after 2 months, when we attempted to remove the catheter after clamping. Considering the surgical risk and the poor overall condition of

the patient, non-surgical treatment was initially proposed; however, drainage via endoscopic retrograde cholangiopancreatography failed, and embolization through the PTBD catheter was not possible.

Therefore, a surgical approach to treat the bronchobiliary fistula was necessary, but fistulectomy through an abdominal approach was impossible due to severe adhesions near the fistula tract and the area of the liver dome. We therefore planned fistula closure by repair of the diaphragm via a thoracic approach and reinforcement with omentopexy. First, a laparotomy incision was made and sufficient omental tissue was mobilized, after which the omentum was pulled up through the substernal tunnel. Then, a right posterolateral thoracotomy incision was made along the sixth intercostal space following adhesiolysis of the pleural adhesion, particularly on the diaphragm side. Because the majority of the right lower lobe exhibited severe inflammation and damage, lobectomy of the right lower lobe was performed. There was no definite pore or fistula tract on the diaphragm side, but injection of saline through the PTBD catheter led us to suspect the location of the lesion. The fistula was closed primarily with silk sutures, and omental tissue was fixed between the suspected fistula tract and the bronchial stump to reinforce the fistula closure and to prevent fistula recurrence around the bronchial stump (Fig. 3).



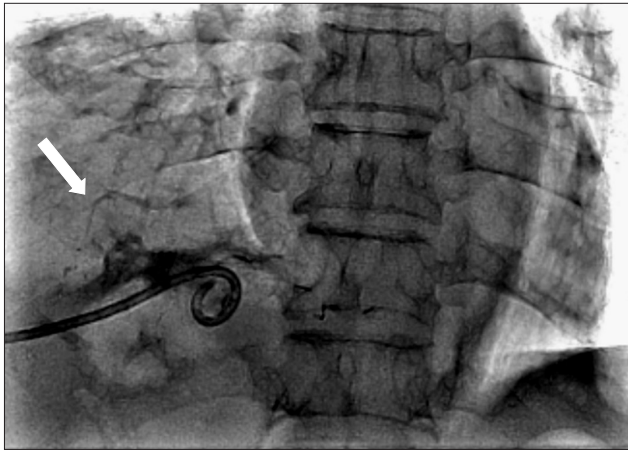


Fig. 1. Cholangiography through a percutaneous transhepatic bile drainage catheter shows a fistula tract extending from the biliary tract to the bronchial tree (white arrow).

After fistula closure, the substernal tunnel was closed. The operation was completed after the insertion of 3 indwelling chest tube drains in the thoracic cavity and 1 Hemovac drain in the abdomen.

After the operation, the patient's biliptysis disappeared, and the patient did not present any other sign or symptom of bronchobiliary fistula. Postoperatively, we assessed bilirubin levels in the chest tube drainage, and we removed the last chest tube on postoperative day 13. No remnant bronchobiliary fistula was evident on cholangiography through the PTBD catheter, and no biliary tract dilatation was visible on dynamic liver computed tomography (CT). Follow-up concluded on postoperative day 68 with no recurrence of bronchobiliary fistula.

We got a verbal informed content from a protector of the patient for publication of clinical details and images.

Discussion

Bronchobiliary fistula is a rare disease defined as an abnormal connection between the biliary tract and the bronchial tree. It may occur congenitally or secondarily to other conditions, such as liver malignancy, infection, trauma, or biliary obstructive disease, or as a complication of liver surgery, TACE, or RFA. The diagnosis is made based on the clinical symptom of biliptysis or sputum analysis. Imaging studies, such as cholangiography, chest/upper abdominal CT scans, contrast-enhanced magnetic resonance cholangiography, and hepatobiliary iminodiacetic acid scanning, can also be utilized for diagnosis [1]. Bronchobiliary fistula can have a mortality rate of over 50% without invasive

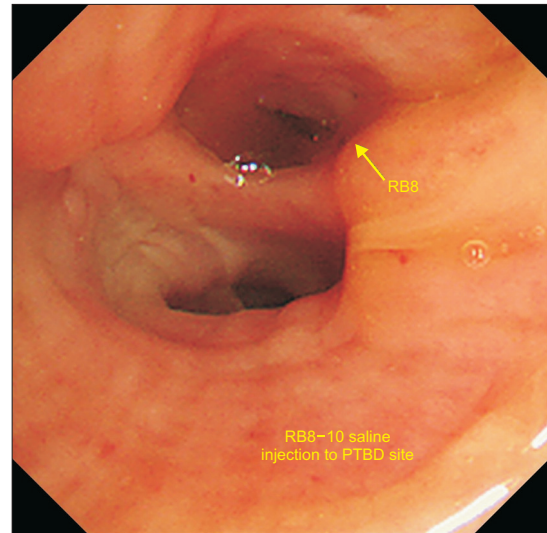


Fig. 2. Fiberoptic bronchoscopy image of the patient. The top hole is the RB8 bronchus, and bile secretion increased when saline was injected through the PTBD catheter. PTBD, percutaneous transhepatic bile drainage.

treatment. Despite its high mortality and morbidity rates, there is no definite consensus regarding the treatment of bronchobiliary fistula [1-3].

Various treatments have been utilized for bronchobiliary fistula, including medical treatment, non-surgical treatment, and surgical treatment. Medical treatment includes conservative management with antibiotic therapy, chest physiotherapy, and nutritional support, the last of which is used because a massive amount of bile loss may cause malnutrition and electrolyte imbalance. Endoscopic techniques have been the most frequent type of non-surgical treatment performed in recent years; these procedures include retrograde biliary drainage, endoscopic nasobiliary drainage, and sphincterotomy. Percutaneous drainage is another choice for main or supplementary treatment [1,2]. Histoacryl embolization under bronchoscopy is a recently-introduced option for the treatment of bronchobiliary fistula [4]. Surgical treatment also varies, from abscess drainage alone to a thoracoabdominal approach for pulmonary resection and subdiaphragmatic exploration for complete exposure of the fistula tract [5]. Usually, non-surgical treatment is preferred over surgery due to its lower invasiveness and high success rate. However, surgical treatment may be indicated in some patients for whom other treatments fail or who are unsuited for non-surgical treatment [6].

Because most patients with bronchobiliary fistula have been exposed to chronic malnutrition and underlying liver disease, it is important to minimize the time and extent of

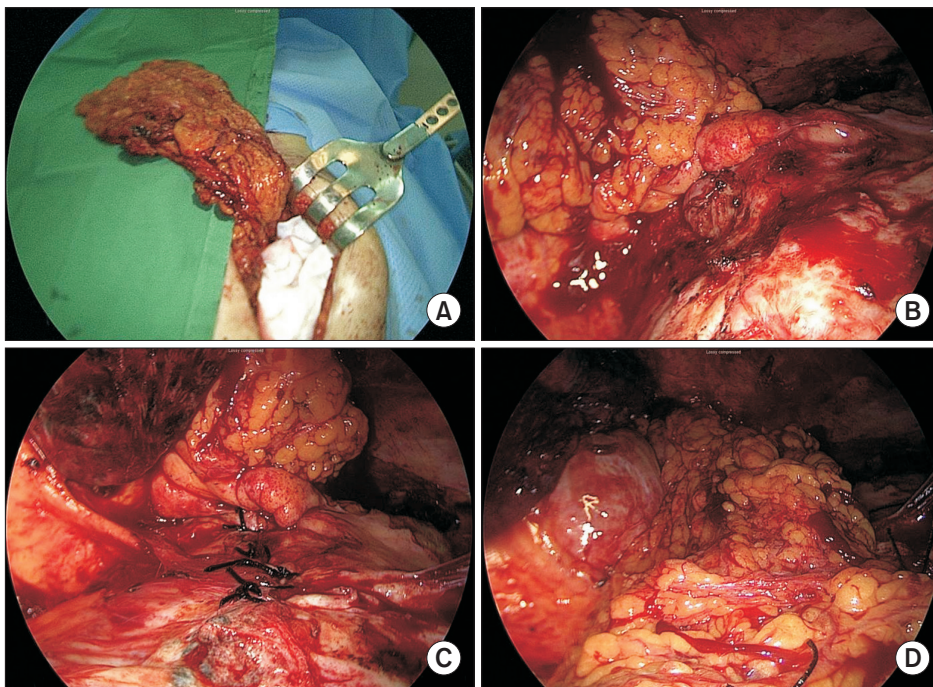


Fig. 3. (A) Mobilized omental tissue under the laparotomy incision. (B) The suspected fistula at the diaphragm surface is visible inside the thoracic cavity. (C) Primary closure of the diaphragm with silk suture. (D) Omentopexy between the fistula tract and the bronchial stump.

surgery, while minimizing the risk of fistula recurrence. Previous reports have insisted that the complete resection of the fistula tract is important, making subdiaphragmatic exploration through phrenotomy necessary [2,3]. However, in this case report, the patient underwent surgery and several rounds of TACE and RFA starting more than 10 years previously, so we chose to utilize an omental flap at the supradiaphragmatic fistula site instead of phrenotomy and dissection around the liver dome. Using this approach, we successfully treated the bronchobiliary fistula. The management of bronchobiliary fistula remains challenging, and each treatment plan should be determined prudently. Surgical treatment may be indicated in some patients, in which case exploratory thoracotomy and omentopexy could be an option for treatment.

Conflict of interest

No potential conflict of interest relevant to this article was reported.

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