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Case Report

Bronchobiliary fistula causing gallstone formation in lung parenchyma: a rare case report

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ABSTRACT

Bronchobiliary fistula (BBF) is a rare condition characterized by an abnormal communication between the biliary tract and the bronchial tree. In developing countries, it most commonly occurs as a complication of hepatic hydatid cysts or amebic liver abscesses, whereas in developed countries, it is more frequently associated with trauma or previous biliary surgery. Bilioptysis, defined as the expectoration of bile-stained sputum, is a pathognomonic symptom observed in most patients. The diagnostic and therapeutic management of BBF requires a multidisciplinary and patient-specific approach, beginning with noninvasive or minimally invasive procedures and sometimes requiring major surgical intervention. This case is remarkable because, despite clinical findings suggestive of BBF, preoperative imaging failed to demonstrate the fistulous tract. Moreover, the intraoperative discovery of gallstones within a destructed lung parenchyma makes this case exceptionally rare.

Keywords: bronchobiliary fistula, gallstones, bilioptysis, lung diseases, thoracic surgery

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Introduction

Bronchobiliary fistula (BBF) is a rare condition characterized by an abnormal communication between the biliary tract and the bronchial system. It was first described by Thomas Bevill Peacock in 1950 [1]. BBF may develop as a complication following surgical procedures such as hepatic hydatid cyst surgery, cholelithiasis, choledocholithiasis, malignancy, or peptic ulcer surgery, and it may also occur secondary to congenital malformations [2]. Patients most commonly present with the pathognomonic symptom of coughing up bile-stained sputum (bilioptysis). Other nonspecific symptoms such as abdominal pain, fever, and dyspnea may also accompany this presentation [3,4].

A variety of imaging modalities can be utilized for diagnosis, including computed tomography (CT), bronchoscopy, magnetic resonance cholangiopancreatography (MRCP), endoscopic retrograde cholangiopancreatography (ERCP), hepatobiliary iminodiacetic acid (HIDA) scintigraphy, and endoscopic ultrasonography [5,6]. CT is valuable for assessing the lung and hepatic parenchyma, while bronchoscopy allows direct visualization of bile-stained secretions in the bronchial tree. MRCP is superior to CT in delineating the biliary system and identifying the fistulous tract. ERCP, though invasive, enables direct visualization of the biliary tree and allows stent placement for simultaneous diagnosis and treatment [7].

Management should be tailored to the etiology, fistula size, patient's overall condition, and associated disease. Endoscopic biliary stenting via ERCP is usually the first-line treatment to divert bile away from the fistulous tract. However, in cases of persistent bile leakage due to obstruction or hypersecretion, surgical intervention may be necessary. The main surgical principle is to identify and close the fistulous tract [6]. Herein, we present the diagnostic and therapeutic approach in a 54-year-old male patient operated for BBF, highlighting the clinical challenges encountered in management.

Case Report

The patient was first operated on in 2015 at an external center for a perforated hepatic hydatid cyst. In 2021, he developed bilioptysis and underwent ERCP, during which a stent was placed in the common bile duct, resulting in symptomatic improvement. Two years later, gallstones were detected, and cholecystectomy was performed, with simultaneous removal of the biliary stent. One year later, the recurrence of bilioptysis prompted repeat ERCP and re-stenting. As the symptoms persisted, the patient was referred to our clinic.

Retrospective evaluation of imaging studies did not reveal a distinct fistulous tract. Thoracoabdominal CT showed destructive changes in the right lower lobe, without clear visualization of the fistula, and the report stated: "Destructive parenchymal areas in the right lower lobe; no definite fistula observed; bronchobiliary fistula cannot be excluded" (Figure 1).

Bronchoscopy was performed after obtaining informed consent. Findings included mucosal hyperemia in the right intermediate bronchus and yellow-green secretions from the right lower lobe bronchi (Figure 2). Biochemical analysis of bronchoalveolar lavage fluid confirmed bilirubin, while cytology excluded malignancy.

The multidisciplinary team (general surgery, gastroenterology, thoracic surgery) recommended MRCP to evaluate the biliary system. MRCP did not clearly show a fistulous tract; however, the presence of contrast at the diaphragmatic level made exclusion of BBF impossible (Figure 3).

The patient was re-evaluated by the multidisciplinary council. Considering the possibility of dense adhesions between the lung parenchyma and diaphragm, the feasibility of direct evaluation of the fistulous tract via open thoracotomy, and the absence of any suspicion of malignancy, a parenchyma-sparing surgical procedure was planned. Written informed consent was obtained from the patient and his relatives prior to the operation.

During right thoracotomy and exploration, dense adhesions were observed between the right lower lobe and the diaphragm at the level of the fistulous tract; however, these adhesions could not be completely released. A localized area of lung injury was detected in the distal portion of the right lower lobe, while the remaining parts of the lobe appeared intact. Therefore, the region containing the fistulous tract and the surrounding destructed lung tissue was resected from the parenchyma by wedge resection using two separate cutting and stapling devices. A small portion of the diaphragm adjacent to the fistulous site was also excised and sent for histopathological examination (Figure 4). The dia-

phragmatic defect was repaired primarily. Macroscopic examination of the resected specimen revealed the presence of a gallstone within the lung parenchyma (Figure 5). After hemostasis and air-leak control were achieved, two chest tubes were placed in the thoracic cavity, and the thoracotomy incision was closed in layers according to anatomical planes.

Postoperatively, thoracic drains were removed on days 2 and 5. On day 7, yellowish serous discharge appeared at the incision. Upon reopening under local anesthesia, a large amount of yellow-green fluid resembling bile was drained. Vacuum-assisted closure (VAC) therapy and tube thoracostomy were applied. Biochemical analysis confirmed high bilirubin concentration. By day 5 of follow-up, drainage had ceased, and the tube was removed.

Repeat ERCP revealed an obstructed stent, which was removed. The patient underwent laparotomy for exploration of subdiaphragmatic tracts. Two tracts containing gallstones were identified in hepatic segments VII–VIII, and the diaphragmatic openings were closed.

Postoperatively, bilioptysis resolved completely. The thoracotomy wound was managed with VAC changes for 14 days under local anesthesia and closed anatomically. The patient achieved full recovery and was discharged. During outpatient follow-up, no recurrence of symptoms was observed.

Written informed consent was obtained from the patient for publication of this case report and any accompanying images.

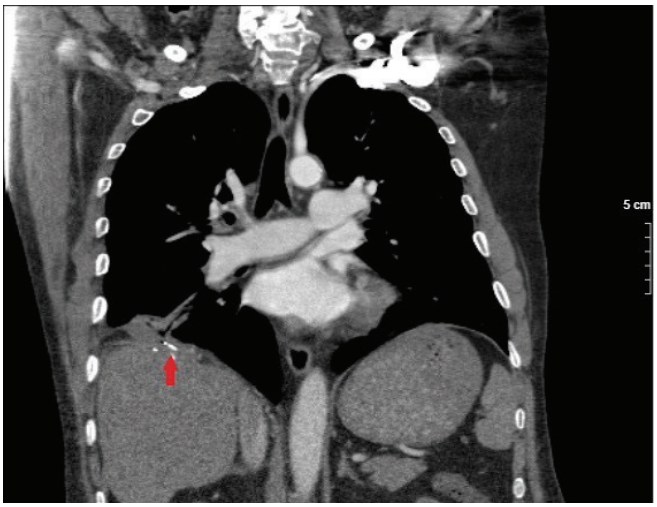

Figure 1. Thoracic CT demonstrating contrast material image at the level of the right diaphragm (area indicated by the arrow).

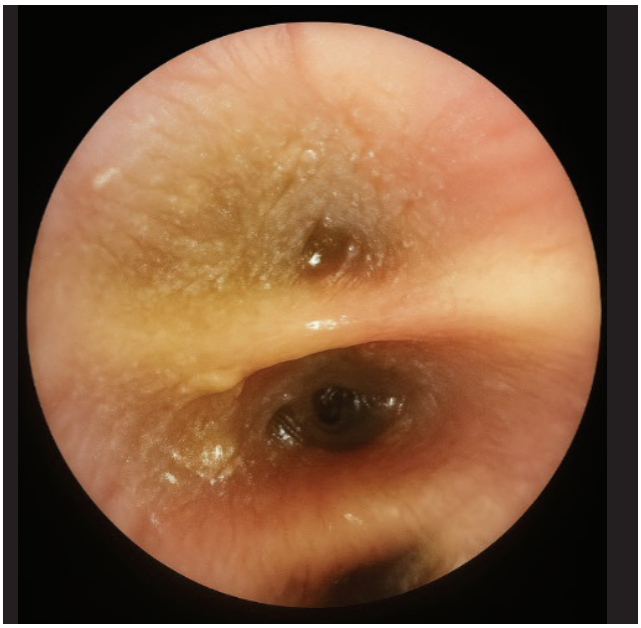

Figure 2. Visualization of bile-stained secretions during bronchoscopy.

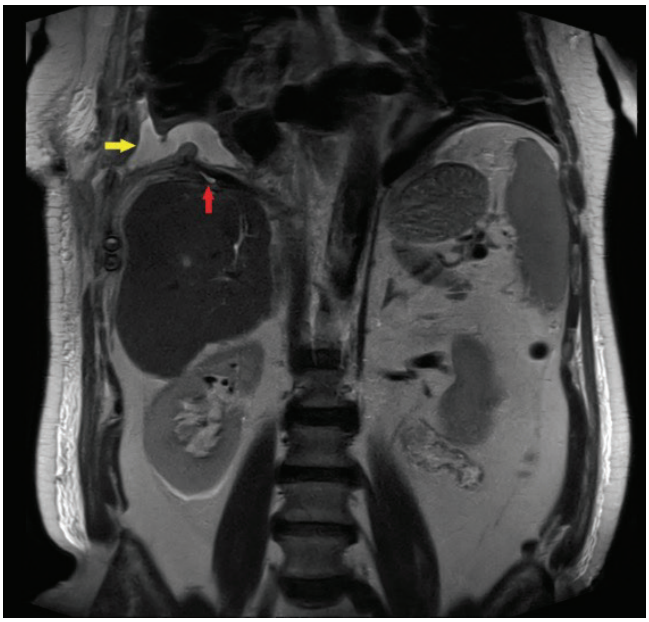

Figure 3. MRCP showing contrast material image at the level of the right diaphragm (red arrow) and right pleural effusion (yellow arrow).

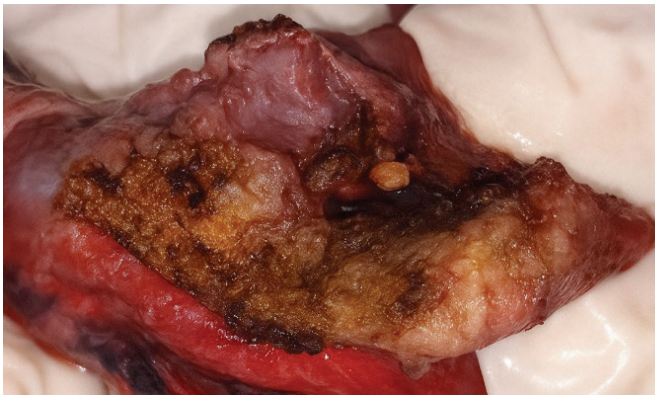

Figure 4. Resected destructive lung parenchyma specimen obtained by wedge resection.

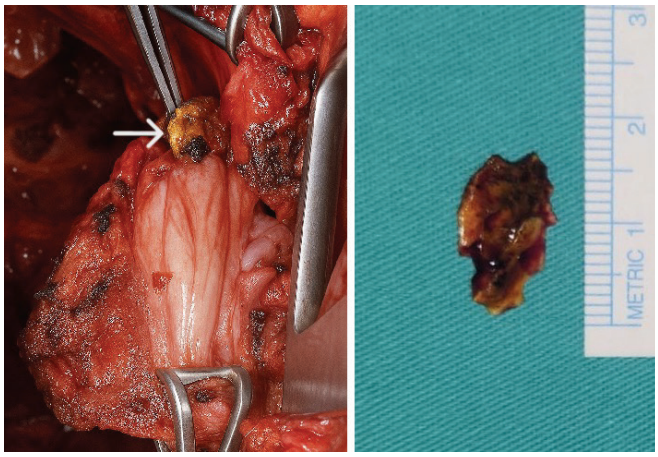

Figure 5. Intraoperative image showing a gallstone within the lung parenchyma (left) and the excised gallstone specimen (right).

Discussion

BBF is a rare pathological condition characterized by an abnormal communication between the biliary system and the bronchial tree [1]. Several mechanisms have been proposed to explain the development of BBF. The most widely accepted theory suggests that an inflammatory process arising in the subdiaphragmatic region or within the hepatic parenchyma causes erosion of the diaphragm, thereby establishing an abnormal connection between the biliary ducts and the bronchial system [5].

Historically, infectious etiologies particularly hydatid disease and pyogenic abscesses have been reported as the most common causes of BBF. In recent years, however, a marked increase has been observed in secondary cases associated with surgical interventions and their complications [4]. Among these, minimally invasive procedures such as transarterial chemoembolization (TACE) and radiofrequency ablation (RFA), which are frequently used in the treatment of hepatic tumors, have emerged as new etiological factors [6]. In our case, BBF developed following liver surgery, demonstrating characteristics similar to those reported in the literature for postoperative cases.

When bile leaks outside the biliary system or gastrointestinal tract, it exerts a strong irritant effect on surrounding tissues. Consequently, irritative cough and bilioptysis are considered pathognomonic findings of BBF [5]. The presence of bilioptysis and elevated bilirubin levels in sputum are valuable biochemical indicators for early diagnosis [8]. In our patient, cough, bilioptysis, and significantly elevated bilirubin levels in bronchoalveolar lavage samples were consistent with the diagnosis of BBF and in agreement with the findings in the literature.

Due to its rarity, there is no universally accepted diagnostic or therapeutic protocol for BBF. At present, curative treatment strategies are generally divided into two main categories: minimally invasive techniques and surgical interventions [9]. The most frequently used minimally invasive approach is ERCP with biliary stent placement. In addition, newer techniques such as ERCP-guided coil embolization and bronchoscopic histoacryl injection have been reported to yield favorable outcomes [6,8].

Physiologically, the basal pressure of the sphincter of Oddi is higher than that of the common bile duct, while intrathoracic pressure is lower than intra-abdominal pressure. These pressure differences facilitate the movement of bile toward the bronchial system in the presence of a fistulous tract [6]. By placing a stent in the common bile duct via ERCP, bile flow is redirected toward the duodenum, effectively eliminating bile drainage through the fistulous tract. This results in symptom regression and may allow for spontaneous fistula closure. However, stent obstruction or excessive bile production can lead to inadequate drainage, resulting in symptom recurrence and the need for surgical intervention [5].

Surgical treatment options include laparatomy, thoracotomy, thoracoabdominal approach. When planning surgery, careful assessment of the etiology, location, size of the fistula, and the extent of tissue injury caused by bile is essential. A multidisciplinary approach is crucial for accurate decision-making and improved treatment outcomes [10].

In this case, despite adequate biliary drainage, persistent symptoms and the development of parenchymal destruction in the lung necessitated surgical intervention. At this stage, the specific surgical approach was determined following a multidisciplinary council evaluation. The major challenge encountered during surgical planning was the inability to clearly delineate the fistulous tract using imaging modalities. Since the parenchymal damage in the right lower lobe was confined to a limited, localized area and the remaining portion of the lobe appeared intact, a parenchyma-sparing wedge resection was performed instead of lobectomy.

In conclusion, the management of bronchobiliary fistula represents a complex challenge that necessitates a tailored, multidisciplinary strategy ranging from minimally invasive interventions to major surgery. As demonstrated in this exceptionally rare case compli-

cated by intrapulmonary gallstone formation, treatment decisions must be guided by the specific anatomical pathology and the patient's overall physiological reserve. This report underscores that when endoscopic measures prove insufficient, individualized surgical planning remains the definitive solution for successful outcomes.

Declaration of conflicting interests

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Authors' contribution

MK: organized the article and wrote the paper TUK: FBE: contributed to the data collection MOK: revised the article. All authors revised the manuscript. The authors read and approved the final manuscript.

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