Bronchobiliary fistula after hepatectomy: A case report and review of the literature

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Abstract. Bronchobiliary fistula (BBF) is a rare complication following hepatectomy, and consists of an abnormal intercommunication between the biliary tract and bronchial tree. The management of this rare entity is challenging with limited current evidence to date on how to treat this condition. Herein, we present a case of BBF following a central hepatectomy and the successive steps of the management. In addition, we sought to conduct a review the literature to summarize all the available evidence regarding post-hepatectomy BBFs.

Case presentation

A 64-year-old Caucasian male was diagnosed with adenocarcinoma of the rectum in 2012 for which he underwent low anterior resection with adjuvant chemotherapy (FOLFOX). The histological report of the specimen revealed a moderate to poorly differentiated adenocarcinoma of colon, and the patient was referred to the oncology department.

Three months later, the patient was admitted under the care of the medical oncology team with a history of fatigue for two days, weight loss and fever up to 39°C. A CT scan demonstrated a postoperative intrahepatic collection (Fig. 2A) and a 12F drain was inserted by a radiologist with CT guidance. An endoscopic retrograde cholangiopancreatography (ERCP) was performed which demonstrated a bile leak, and a 9 cm 10 F stent was placed (Fig. 2B). The patient was discharged 20 days...
later with the drain in situ and was examined regularly in our outpatient clinic.

However, 14 months postoperatively, the patient presented to the oncology department complaining of new onset fever and expectoration. A chest x-ray was performed demonstrating atelectasis of the lower right lobe and was treated as suspected pneumonia. Given the patient previous history, an inflammatory tumor was included in the differential diagnosis. Although afebrile, the patient reported episodes of a productive cough with dark-yellow sputum, raising concerns for bronchobiliary fistula. The patient underwent a CT scan which confirmed the clinical suspicion and he was subsequently referred to our department for further treatment. Since the patient had a biliary stent in situ, the surgical team decided that a further operation was required. Using a transabdominal approach, a communication between the initial intrahepatic collection and one inferior lobular bronchus was demonstrated. Transhiatal removal of fistula was performed with closure of the defect through the abdomen (Fig. 3). There was no bile leak through the remaining liver parenchyma.

The postoperative course was uneventful with no further bilioptysis or other pulmonary symptoms. At radiological follow up one month postoperatively, a CT showed a postoperative biloma for which the patient underwent CT guided drainage with a 12F catheter. A further ERCP revealed leakage and communication of the bile duct with the biloma which was drained and a stent was inserted. The patient was discharged 2 months after surgery and was examined regularly in the outpatient clinic. He was readmitted on a schedule to remove the plastic stent by ERCP. Currently, there are no signs of further bile leakage, BBF or pulmonary symptoms.

**Discussion**

BBP is an uncommon condition whose etiology can differ by geographical region; in developing countries echinococcal and amebic disease are the most common causes, whereas in developed countries trauma and biliary obstruction predominate (6). According to a recent systematic review, tumor is the most frequent cause (32.3%) of BBF formation, followed by bile duct obstruction (30.8%) (7). Other causes have also been identified, such as chronic pancreatitis, trauma and congenital malformation. The pathogenesis of fistula formation has not been completely understood yet, although increased pressure within the biliary tree and local inflammation appeared to be the two major factors that contribute to the development of BBF. In our case, intrahepatic abscess triggered the formation of adhesions between the diaphragm and the lung and was thought to be the cause of the BBF formation (4,8-10).

The clinical presentation is variable, and may include fever, irritating cough, jaundice, and abdominal pain. The pathognomonic feature is a productive cough of bile stained sputum (7,11). Chest pain or episodes of dyspnea occur only in a minority of patients, while nausea, vomiting, portal hypertension and liver dysfunction are rarely found. In terms of diagnosis, CT imaging is usually the predominant modality or biliary scintigraphy with hepatic iminoacetic acid (HIDA) which demonstrates initial tracer activity in the liver which migrates into the chest cavity (12). BBF can also be diagnosed by interventional techniques, such as ERCP, percutaneous transhepatic cholangiography, bronchoscopy or fistulography (4,13,14).

Currently, there are no guidelines on how to treat this rare complication with both conservative and surgical approaches.
Table I. Review of reported cases with bronchobiliary fistula after hepatectomy.

<table>
<thead>
<tr>
<th>Author, Year</th>
<th>Age/sex</th>
<th>Type of disease</th>
<th>Primary disease</th>
<th>Treatment</th>
<th>Bilioptysis</th>
<th>Fever</th>
<th>Jaundice</th>
<th>Approach</th>
<th>Outcome</th>
<th>(Refs.)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Lucero et al, 2005</td>
<td>63/F</td>
<td>Benign</td>
<td>Hydatid cyst</td>
<td>Right hepatectomy</td>
<td>Yes</td>
<td>N/A</td>
<td>N/A</td>
<td>Surgery</td>
<td>Cure (29)</td>
<td></td>
</tr>
<tr>
<td>Memis et al, 2000</td>
<td>40/M</td>
<td>Benign</td>
<td>Hepatic Alveolar disease</td>
<td>Partial hepatectomy</td>
<td>Yes</td>
<td>N/A</td>
<td>Yes</td>
<td>Histoacryl injection through microcatheter and ERBD</td>
<td>Cure (26)</td>
<td></td>
</tr>
<tr>
<td>Senturk et al, 1998</td>
<td>26/M</td>
<td>Benign</td>
<td>Hydatid cyst</td>
<td>Partial right lobectomy</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>ERBD</td>
<td>Cure (21)</td>
<td></td>
</tr>
<tr>
<td>Schwartz et al, 1988</td>
<td>18/M</td>
<td>Unknown</td>
<td>Unknown</td>
<td>Right heptectomy</td>
<td>Yes</td>
<td>N/A</td>
<td>N/A</td>
<td>PTCD and balloon dilation cholangioplasty</td>
<td>Cure (24)</td>
<td></td>
</tr>
<tr>
<td>Chua et al, 2000</td>
<td>61/M</td>
<td>Benign and Malignant</td>
<td>Gangrenous Cholecystitis Carcinosarcoma</td>
<td>Right hepatectomy</td>
<td>Yes</td>
<td>Yes</td>
<td>N/A</td>
<td>Surgery</td>
<td>Cure (9)</td>
<td></td>
</tr>
<tr>
<td>Ong et al, 2004</td>
<td>71/F</td>
<td>Malignant</td>
<td>Metastatic colon Ca</td>
<td>Right hepatectomy</td>
<td>Yes</td>
<td>Yes</td>
<td>No</td>
<td>Octreotide injection surgery</td>
<td>Cure (30)</td>
<td></td>
</tr>
<tr>
<td>Kaido et al, 2006</td>
<td>76/M</td>
<td>Malignant</td>
<td>Hepatocellular Ca</td>
<td>Right hepatectomy</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Failure</td>
<td>Cure (28)</td>
<td></td>
</tr>
<tr>
<td>Jung et al, 2003</td>
<td>65/M</td>
<td>Malignant</td>
<td>Metastatic colon Ca</td>
<td>Right hepatectomy</td>
<td>Yes</td>
<td>Yes</td>
<td>N/A</td>
<td>Multiple ERBD then surgery</td>
<td>Cure (27)</td>
<td></td>
</tr>
<tr>
<td>Oettl et al, 1999</td>
<td>56/M</td>
<td>Malignant</td>
<td>Metastatic colon Ca</td>
<td>Right hepatectomy</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>ERBD</td>
<td>Cure (19)</td>
<td></td>
</tr>
<tr>
<td>Rose et al, 1998</td>
<td>61/M</td>
<td>Malignant</td>
<td>Metastatic colon Ca</td>
<td>Wedge liver resection</td>
<td>Yes</td>
<td>N/A</td>
<td>Yes</td>
<td>ERBD</td>
<td>No BBF died 5 months later (20)</td>
<td></td>
</tr>
<tr>
<td>Velchik et al, 1991</td>
<td>38/F</td>
<td>Malignant</td>
<td>Mucinous adenocarcinoma</td>
<td>Right hepatectomy</td>
<td>Yes</td>
<td>N/A</td>
<td>N/A</td>
<td>PTCD</td>
<td>Cure (23)</td>
<td></td>
</tr>
<tr>
<td>George et al, 1988</td>
<td>58/M</td>
<td>Malignant</td>
<td>Metastatic colon Ca</td>
<td>Hepatic resection</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Surgery</td>
<td>Failure (22)</td>
<td></td>
</tr>
<tr>
<td>Hai et al, 2016</td>
<td>70/M</td>
<td>Malignant</td>
<td>Hepatocellular Ca</td>
<td>Partial resection</td>
<td>Yes</td>
<td>Yes</td>
<td>N/A</td>
<td>ERBD then surgery</td>
<td>Cure (11)</td>
<td></td>
</tr>
<tr>
<td>Kuo et al, 2014</td>
<td>68/M</td>
<td>Malignant</td>
<td>Hepatocellular Ca</td>
<td>Central lobectomy</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>Surgery</td>
<td>Cure (25)</td>
<td></td>
</tr>
<tr>
<td>Current case, 2019</td>
<td>64/M</td>
<td>Malignant</td>
<td>Metastatic colon Ca</td>
<td>Central hepatectomy</td>
<td>Yes</td>
<td>Yes</td>
<td>Yes</td>
<td>ERBD then surgery</td>
<td>Cure (Present study)</td>
<td></td>
</tr>
</tbody>
</table>

Ca, carcinoma; ERBD, endoscopic retrograded biliary drainage; ENBD, endoscopic nasobiliary drainage; PTCD, percutaneous transhepatic cholangiography and biliary drainage; N/A, not applicable.
considered acceptable to date. Some authors suggest that BBF associated with benign causes should be treated conservatively; endoscopic retrograded biliary drainage (ERBD), endoscopic nasobiliary drainage (ENBD), percutaneous transhepatic cholangiography and biliary drainage (PTCD), and abscess drainage (15-17). Adjunct use of octreotide might have a role in some cases due to reports of successful resolution or reduction of symptoms with its use (18). On the other hand, surgical management should be considered in case BBF has developed in a background of tumor, trauma or obstruction and conservative therapy has previously failed (7). Operative exploration and repair of the initial injury is usually performed, whereas resection of the involved pulmonary area and removal of the fistula is advised in the form of thoracobiliary fistula decortication (18).

Table I summarizes a brief review of the literature on bronchobiliary fistulas after liver surgery for both benign and malignant diseases (9,11,19-30).

Due to low incidence of BBF, there is no clear consensus on the treatment of this uncommon complication to date. Multidisciplinary management of such patients should be considered taking into consideration the underlying pathology leading to this rare complication. Conservative treatment should be considered first, while surgical resection of the BBF remains an option when other therapies have failed. Surgeons should have low suspicion of diagnosing and managing this complication after biliary tract operations.

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Availability of data and materials
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Authors’ contributions
VL, DM and APa were involved in the study conception and design. VL and APa contributed to the acquisition of data. VL, DM, DIT and GSK were involved in data analysis and interpretation. DM, DIT and GSK drafted the manuscript. APa and APe critically revised the manuscript.

Ethics approval and consent to participate
The study was approved by the Ethics Committee of Nicosia Hospital.

Patient consent for publication
Informed consent was obtained.

Competing interests
The authors declare that they have no competing interests.

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