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Case Report

Bilorrhea Secondary to Bronchobiliary Fistula

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Bronchobiliary fistula (BBF) is a rare condition which occurs most commonly as a complication of hydatid cyst liver disease. The following report describes a patient who presented with biliptysis 6 months following decortication of an empyema that had occurred following partial hepatectomy of a colon cancer metastasis. This is the only case to our knowledge that describes the presentation of a BBF in this context. The patient was diagnosed with BBF and successfully underwent open thoracotomy for fistulectomy and repair.

Key words: Fistula (bronchobiliary) – Bronchial disease – Surgery – Complications

In the United States, bronchobiliary fistula (BBF) most commonly occurs secondary to hydatid cyst liver disease or following surgical repair of trauma to the thoracic cavity. Less common causes include congenital formation of BBF, biliary lithiasis, hepatic or subphrenic abscess, cholecystitis, pancreatitis, a tumor involving the liver or biliary tree that obstructs the bile duct, and radiofrequency ablation.[1] Pulmonary inflammation and the destructive action of bile can erode the bronchus, leading to the formation of a fistula between the liver and lung. Persistence of this communication is favored by the pressure gradient formed by the positive pressure from the bile duct and the negative pressure in the thoracic space.[2] When bile escapes the biliary tree, it acts as an irritant that causes inflammation, leading to irritation and possibly necrosis of the diaphragm.[3] The classic presentation of a patient with BBF is chronic cough, biliptysis, persistent fever, and pain.[4] Mild to moderate jaundice may also be present. The history of such patients is typically positive for chronic illness or previous surgery following thoracic trauma.

Report of a Case

A well-nourished, well-developed 47-year-old woman was referred to the cardiothoracic surgical clinic for workup of pulmonary edema and a chronic cough accompanied by fever, chills, dyspnea, fatigue, night sweats, pleuritic chest pain, and expectoration of yellow-tinged fluid. Although she had been on IV antibiotics for several days, her condition remained unchanged. Approximately one

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year prior she had undergone a partial hepatectomy for metastasis, as well as colon resection. She developed an empyema after her initial hepatectomy, which was treated with video-assisted decortication. Although she did well after the procedure initially, her cough returned and gradually worsened, and she presented approximately 6 months after decortication of the empyema.

Based on her history and presentation, a fistula communicating between the biliary and bronchial systems was suspected. X-rays revealed what appeared to be an oval-shaped, ring-enhancing fluid collection posterior to the dome of the liver that possibly extended above and below the diaphragm. To better visualize the lesion identified on the chest X-ray, the patient was sent for a computed tomography (CT) pulmonary angiogram with IV contrast, with axial cuts from the thoracic inlet through the diaphragm with sagittal and coronal reconstructions. It was confirmed that the patient had a loculated cystic fluid collection of approximately 8.5 cm $\times$ 6 cm in the posterior aspect of the right lung base and a communication from the bronchus to the liver (Fig. 1).

The patient was scheduled for surgery for treatment of a suspected hepatobronchial fistula. Preoperatively, the patient was evaluated and cleared for bronchoscopy and right-sided thoracotomy, and the preoperative X-ray showed a right-sided effusion. In the operating room prior to surgery, a diagnostic bronchoscopy was performed using a single lumen endotracheal tube, which aspirated bilious-appearing fluid. The patient underwent a right thoracotomy with resection of the hepato-bronchial fistula, a right lower lobectomy, and decortication. The right lower lobe, which was densely adherent to the diaphragm, was freed up, revealing a thickened area that was consistent with a fistula. After this was removed, bile salt at the bronchial region was identified. The lower lobe was resected, as a significant portion was totally collapsed and nonfunctional. This finding precluded a limited pulmonary resection, such as a segmentectomy. The suprahepatic fluid collection was aspirated using a transdiaphragmatic approach. Bilious-appearing fluid (300 mL) was cultured and submitted for measurement of direct and total bilirubin. Examination of the resected right lower lobe revealed pathology consistent with a hepatopleural fistula (Fig. 2). A respiratory and squamous epithelial-lined fistula tract with mixed inflammation and hemorrhage was identified in the inferior aspect of the specimen. There was abundant yellow-green

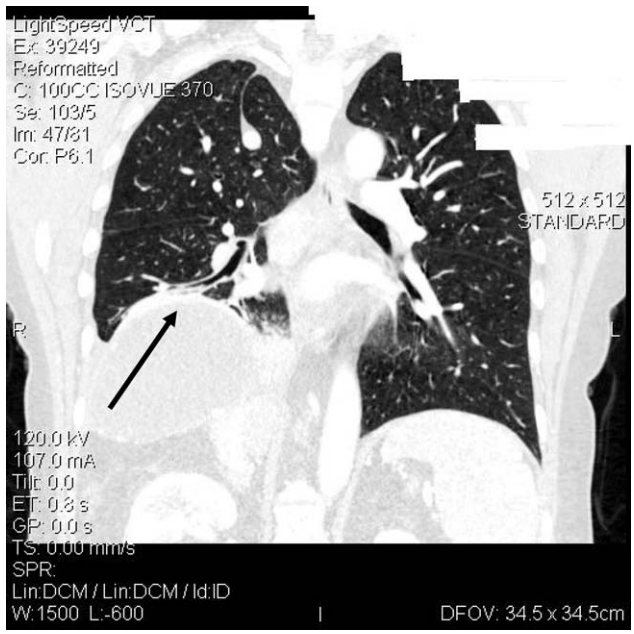

Fig. 1 CT scan of chest with hepato-biliary fistula (arrow).

pigmented material surrounding the tract and surrounding parenchyma (Fig. 3).

The use of a transdiaphragmatic approach for drainage and repair of the fistula prevented a secondary laparotomy. Postoperatively, the patient underwent esophagogastroduodenoscopy (EGD) with placement of a biliary stent to exclude the hepatic segment of the fistula. The patient tolerated the procedure well, recovered with complete resolution of symptoms, and was discharged on the fifth postoperative day. She continued to do well on appropriate postsurgical cardiothoracic surgical clinic visits, with complete resolution of the bilorrhea.

Discussion

Patients who have undergone surgical intervention of, trauma to, or who have had disease involving the lung, liver, biliary tract, or pancreas are at risk of developing BBF. Patients with BBF may present with symptoms including persistent cough, fever, and mild jaundice, and often have a history of liver pathology. Biliptysis, which is pathognomonic for BBF, is suggested by bitter-tasting or green-tinged sputum. Chest X-ray may reveal dense opacities in the right lower lobe. The best initial imaging evaluation is thoracic and upper abdominal CT to properly visualize the liver and lung. Endoscopic retrograde cholangiopancreatography (ERCP) can

Fig. 2 Resected right lower lobe with suture at fistula.

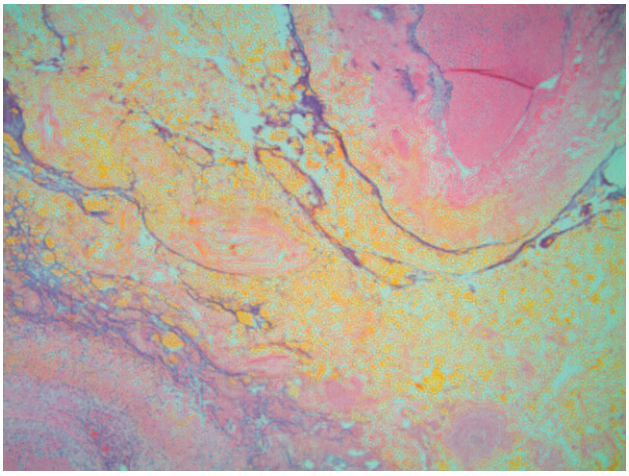

Fig. 3 Hematoxylin-eosin stain of respiratory and squamous epithelial-lined fistulous tract (10×, light microscopy).

confirm the presence of a fistula if the diagnosis remains in doubt and, indeed, is essential if an obstruction is suspected in the bile duct that would interfere with proper drainage into the duodenum.[1,5] Workup may include magnetic resonance cholangiopancreatography to visualize the communication between the bronchus and biliary tree and is fast, safe, and noninvasive. ERCP, although it has a higher failure rate and risk of complications, allows for biliary decompression in patients in whom biliary pathology is suspected.

Although minimally invasive and conservative management approaches have been used to treat BBF in select patients, direct, open repair is the definitively curative treatment.[5] The pleural, subdiaphragmatic, and perihepatic spaces must be completely drained to maintain closure of the fistula. Adequate drainage eliminates sources of inflammation and irritation as well as the pressure gradient

that favors continued bile drainage from the biliary tree to the bronchus. A patent bile duct with a BBF is an indication for thoracotomy, either via a thoracoabdominal or an inferior approach, depending on the degree of exposure needed to resect the entire fistulous tract. Subdiaphragmatic drainage of any remaining extrapleural fluid and closure of the diaphragm is required, and resection of part of the lung may be necessary in patients such as the one described in this case, in which there is a nonfunctional segment or lobe secondary to atelectatic collapse.[1] Open repair of BBF is preferred as the definitive treatment of BBF, and conservative management should be considered only in patients with short life expectancy or who are unable to tolerate open surgery. The emerging consensus is that access to the biliary tree to relieve obstruction, complete fistulectomy and closure of the diaphragm, and complete drainage of any fluid are key to achieving good outcomes for patients with BBF.

Conclusion

BBF presents a unique and interesting diagnostic challenge and highlights the importance of a thorough history and physical exam. This condition may arise in a patient with parasitic disease, in patients having undergone previous surgery, or in chronically ill patients with a long history of surgeries, abscesses, or radiofrequency ablation therapy. The prognosis in patients with BBF depends on their ability to tolerate surgery, their overall state of health prior to diagnosis (including their ability to undergo a lobectomy), and recognition of this condition early in its course. Direct open fistulectomy and repair is the most definitive approach to repairing BBF; with complete drainage and successful repair of the diaphragm, it provides curative treatment with excellent prognosis for patients with this condition.

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