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Case Report

A 45 Year Old Woman with Biloptysis – A Case Report of Broncho-billiary Fistula

Uma Dhar[1], Smaprity Islam[1]

Abstract

A bronchobiliary fistula or a biliobronchial fistula is a uncommon condition which occurs from an abnormal connection between the bronchial trees and biliary channel. It presents with pathognomonic bilious sputum or biloptysis with suspicious pneumonia. Broncho-biliary fistula is associated with Hydatid infection but recently found in patients with tumours. This case report presents a patient who had bronchobiliary fistula who had history of Adenocarcinoma of gall bladder with multiple space occupying lesions in liver and she underwent cholecystectomy

Keywords: *Biloptysis, Bronchobilliary Fistula, Hydatid infection, Adenocarcinoma, Cholecystectomy.*

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Introduction

A bronchobiliary fistula (BBF), also called biliobronchial fistula, is a rare condition caused by an abnormal connection between the biliary and bronchial trees. It is characterized by pathognomonic yellow bilious sputum, known as biliootysis exhibiting pneumonic infiltration. BBF first reported and described Peacock in 1850, a rare complication after hepatectomy, which consists of an abnormal intercommunication between the biliary tract and bronchial tree.[1,3] In the past, infectious diseases, such as a hydatid infection, have been known to cause BBF, but recent reports have suggested its association with an obstruction of the biliary tract by tumour , trauma or hepatic abscess. Especially in endemic regions, hydatid or amebic disease of the liver are common causes of BB tumors.[2] Bilious sputum aids in the diagnosis of patients with BBF but a definitive treatment is unclear and controversial. BBF is associated with high morbidity and mortality rates. The management of this rare entity is challenging with limited current evidence to date on how to treat this condition. Therefore, case-specific procedures and treatments, such as invasive surgical procedures and simple conservative treatment, differ between centers.[2] We report one case case of BBF who had history of Adenocarcinoma of gall bladder with multiple space occupying lesions in liver.

Case report

A 46-years-old Bangladeshi house wife got herself admitted into Bangabandhu Sheikh Mujib Medical University (BSMMU) on 13th December, 2022 with the complaints of cough with productive yellowish sputum and right sided chest pain for 1 month. She has history of adenocarcinoma of gallbladder for which she underwent cholecystectomy 6 months back. Her sputum culture shows Enterococcus sp. Which was sensitive to Gentamycin and vancomycin.Her CTscan of chest with contrast shows consolidation and cavitationa in middle lobe of right lung. CT guided core biopsy from right lung lesion showed inflammatory lesion. Her ultrasound of abdomen suggested multiple Space occupying lesions in liver.Ultrasonography guided FNA from liver SOL suggests metastatic adenocarcinoma.Her sputum was analysed for billurubin and it was 7.6 mg/dl.Her MRCP was suggestive of metastatic lesion at right anterior subphrenic region invaded the liver having communication with biliary tree also invaded right dome of diaphragm resulting broncho-billiary fistula.

1. Department of Respiratory Medicine, BSMMU, Dhaka
2. Department of Respiratory Medicine, BSMMU, Dhaka.

Corresponding author: Dr. Smaprity Islam, Assistant Professor, Department of Respiratory Medicine, BSMMU, Dhaka, Bangladesh. E-mail: umauma789@yahoo.com

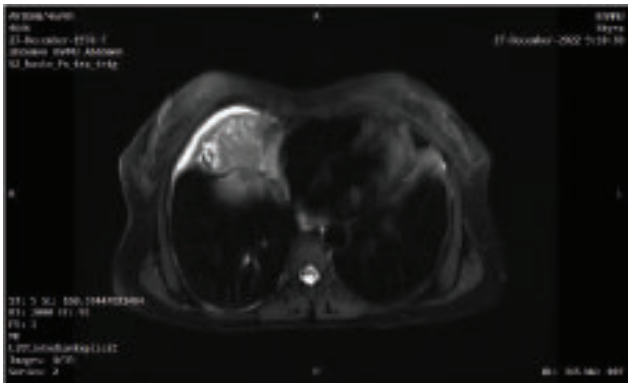


Figure 1.

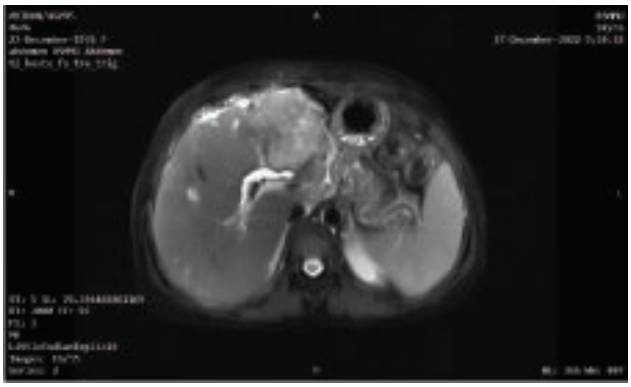
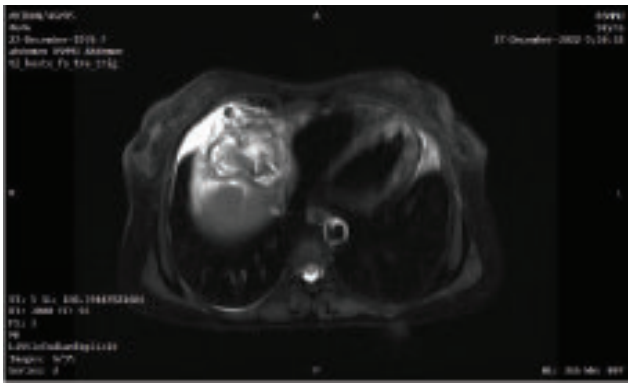


Figure 2.



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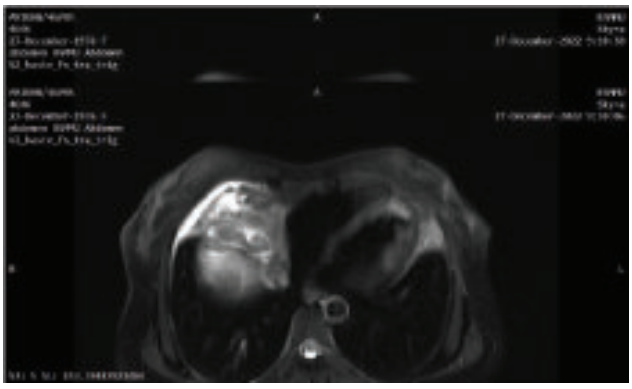

Figure 4.

Discussion

BBP is an rare 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, Carcinoma gall bladder followed by cholecystectomy with liver metastasis 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 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 diagnostic purpose, 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]

recently, there are no guidelines on how to treat this rare complication with both conservative and surgical approaches 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]

Conclusion

As pathogenesis of BBF is still unclear, BBF should be suspected in patients who have undergone liver resection and have fluid collection near the diaphragm, with bilious sputum exhibiting pneumonic infiltration. 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. Either ERCP or PTBD can be used for the primary management of BBF.Surgeons should have low suspicion of diagnosing and managing this complication after biliary tract operations.

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