CASE REPORT

Bilothorax—An Unusual Presentation of Biliary Tract Obstruction: A Rare Case

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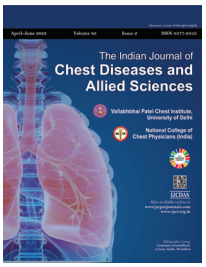

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Abstract

Introduction: Bilothorax is a rare condition defined by the presence of bile in pleural space. Notable causes are traumatic injury followed by hepatobiliary procedures. However, secondary to subphrenic/hepatic abscess, biliary obstruction is rare.

Case description: A 60-year-old female presented with abdominal pain and discomfort, fever, right-sided chest pain, dyspnea, and dry cough for 10 days. On examination, absent breath sounds on the right side of the chest with right hypochondrial tenderness. Ultrasound-guided diagnostic thoracocentesis showed exudative bilious pleural effusion with pleural fluid to serum bilirubin ratio above 1. Contrast-enhanced computed tomography (CECT) chest and abdomen reported right hydropneumothorax with empyema gallbladder, and dilated common bile duct (CBD) due to distal CBD stricture. The patient was managed with intravenous (IV) fluids, antibiotics, tube thoracostomy, and supportives. The patient was referred to the Department of Gastroenterology for which they advised surgical intervention.

Discussion: There are several possibilities of how bile travels into the pleural space, which includes the passive movement of bile through the diaphragm/lymphatic channels, traumatic or congenital defects in the diaphragm, and bilious fistulas. Other possible etiologies include the extension of biliary peritonitis, blunt trauma causing biliopleural fistula, or complication of open or percutaneous hepatobiliary procedures. The most specific test for diagnosis of bilothorax is the pleural fluid to serum bilirubin ratio.

Conclusion: Bilothorax is a rare manifestation. Unlike common etiologies of bilothorax such as trauma, and rupture of hepatic or subphrenic abscess, exclusively biliary tract obstruction due to CBD stricture is one rare possibility of bilothorax to be considered. Early diagnosis and treatment should be given to prevent the development of empyema as bile in enclosed areas is a good medium for the growth of bacteria. The linchpin management of bilothorax includes complete pleural drainage and correction of the root cause.

Keywords: Biliary obstruction, Biliopleural fistula, Bilothorax, Case report, Chest and abdomen tube thoracostomy, Contrast-enhanced computed tomography empyema, Endoscopic retrograde cholangiopancreaticography, Transhepatic drainage.

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Abbreviations used in this Article

ARDS = Acute respiratory distress syndrome; BP = Blood pressure; BPF = Biliopleural fistula; CBD = Common bile duct; CBP = Complete blood picture; CECT = Contrast-enhanced computed tomography; ERCP = Endoscopic retrograde cholangiopancreaticography; GHCCD = Government Hospital for Chest and Communicable Diseases; GI = Gastrointestinal; HBD = Hepatobiliary duct; HbsAg = Hepatitis B surface antigen; HCC = Hepatocellular carcinoma; HCV = Hepatitis C; ICD = Intercostal chest drain; IHBRD = Intrahepatic biliary radical dilatation; IV = Intravenous; LFTs = Liver function tests; MRCP = Magnetic resonance cholangiopancreaticography; MTBC = *Mycobacterium tuberculosis* complex; PBF = Pleurobiliary fistula; PEEVO = Pleural effusion of extravascular origin; PTBD = Percutaneous transhepatic biliary drainage; RIF = Resistance to rifampin; SGOT = Serum glutamic-oxaloacetic transaminase; SGPT = Serum glutamic-pyruvic transaminase; USG = Ultrasonography; VATS = Video-assisted thoracic surgery ; WBC = White blood cell; Xpert MTB/RIF = Xpert *Mycobacterium tuberculosis* and resistance to rifampin.

Introduction

Bilothorax is a rare condition defined by the presence of bile in pleural space. Notable causes of bilothorax are traumatic injury followed by hepatobiliary procedures or a complication of hepatic trauma, hepatic tumor, and percutaneous transhepatic biliary drainage (PTBD).[1–3] However, bilothorax secondary to subphrenic or hepatic abscess and biliary obstruction is rare.[4] Biliary tract obstruction usually presents with jaundice, abdominal pain, itching, nausea, and vomiting, but presenting with bilious pleural effusion is rare. Here we present a rare case report of bilothorax which is rare itself presenting as a result of biliary tract obstruction due to common bile duct (CBD) stricture.

Case Description

A 60-year-old female presented to our outpatient department (OPD) of the Department of Pulmonology in Government Hospital for Chest and Communicable Diseases (GHCCD), Guntur, Andhra Pradesh, India with shortness of breath, non-productive cough,

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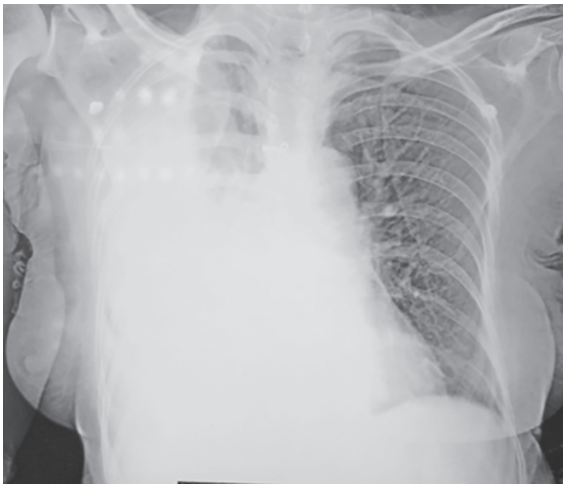

Fig. 1: Chest radiograph showing right-sided loculated pleural effusion

Fig. 2: Greenish color pleural effusion (bilothorax) collected in intercostal chest drain (ICD) bag

right-sided chest pain along with abdominal pain and discomfort and dyspepsia of 10 days duration. She was in tachycardia with low blood pressure (BP) recordings, temperature of 99°F, and with oxygen saturation of 90% on room air. On examination of the patient, a dull note on percussion of the right side of the chest, and on auscultation absent breath sounds were heard on the right side of the chest with right hypochondrial tenderness on palpation of the abdomen. Chest radiograph unveiled considerable right-sided pleural effusion, as depicted in (Fig. 1). Subsequent to an ultrasound-guided diagnostic thoracocentesis, an analysis of the pleural fluid was conducted. Notably, the obtained fluid exhibited a distinctive greenish hue, as illustrated in Figure 2. In addition to the standard pleural fluid analysis, the specimen was also subjected to specialized examinations for pleural fluid bile salts, bile pigments, and pleural fluid bilirubin. Complete blood picture (CBP) and other investigations such as liver function tests (LFTs) and viral markers were sent and the reports are tabulated in Table 1. Pleural fluid analysis indicated an exudative nature, and the pleural fluid to serum bilirubin ratio, quantified at 4.7, surpasses the threshold of 1, signifying a bilious effusion. Table 2 represents pleural fluid analysis results. Pleural fluid acid-fast stain; gene Xpert

Table 1: Blood investigations of the patient

S. No.	*Investigations*	*Result*
1	CBP (white blood cell [WBC])	14,800 cells/mm^3
2	Serum glutamic-oxaloacetic transaminase (SGOT)	64 U/L
3	Serum glutamic-pyruvic transaminase (SGPT)	24 U/L
4	Serum bilirubin	2.3 mg/dL
5	Hepatitis B surface antigen (HbsAg)	Nonreactive
6	Hepatitis C (HCV)	Nonreactive

Table 2: Pleural fluid analysis of right-sided pleural effusion

S. No.	*Investigations*	*Result*
1	Pleural fluid bilirubin	10.8 mg/dL
2	Pleural fluid protein	4.0 gm/dL
3	Pleural fluid sugar	10 mg/dL
4	Pleural fluid bile salts	Positive
5	Pleural fluid bile pigments	Positive
6	Pleural fluid/serum bilirubin ratio	4.7

Mycobacterium tuberculosis and resistance to rifampin (Xpert MTB/RIF); and culture were negative.

Tube thoracostomy was done to drain the fluid and the patient was managed with intravenous (IV) fluids, IV antibiotics, and supportives. Ultrasonography (USG) abdomen reported hepatomegaly with dilated hepatobiliary duct (HBD), grossly enlarged and edematous gall bladder, and dilated CBD with small calculi in clump at the distal end. Contrast-enhanced computed tomography (CECT) chest and abdomen reported edematous gallbladder and dilated CBD with intrahepatic biliary radical dilatation (IHBRD) due to distal CBD stricture (Figs 3 and 4). After clinical and radiological improvement, the patient was referred to the Department of Gastroenterology for further management for which they advised surgical intervention.

Discussion

The presence of bile within the pleural cavity is referred to as bilothorax, alternatively known as cholethorax, bilothorax, pleurobilia, and thoracobilia.[5,6] This constitutes a rare etiology contributing to pleural effusion of extravascular origin (PEEVO).[7–12] Williams et al. was the first study to describe bilothorax where blunt hepatic trauma instigated the development of a biliopleural fistula (BPF).[13] To date, approximately 60 cases have been reported in literature since 1960. As elucidated in the medical literature, bilothorax has garnered attention as a consequential outcome of trauma-induced injuries, encompassing blunt trauma, gunshot wounds, and stab injuries, alongside iatrogenic occurrences stemming from procedures such as transhepatic biliary drainage. Alternative etiologies include subphrenic and hepatic abscesses, hydatid disease (echinococcosis), biliary obstructions, complications arising from cholangiography, percutaneous liver biopsy, as well as procedural sequelae like gastropleural and cholecystopleural fistulas.[4] However, in our case, unlike common etiologies of bilothorax-like trauma, rupture of hepatic or subphrenic abscess,

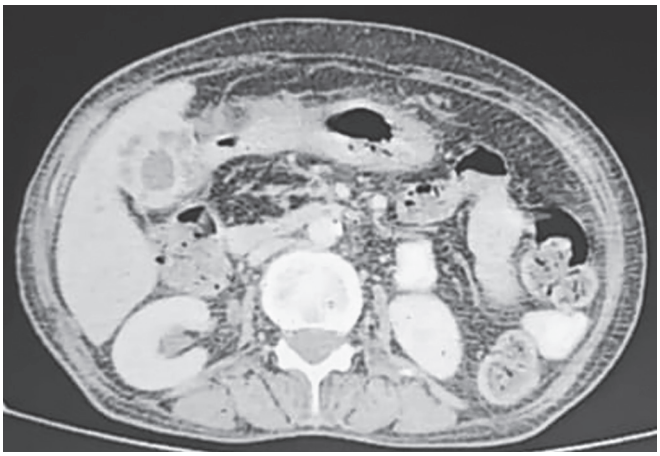

Fig. 3: The CECT abdomen showing edematous gall bladder with peri-gall bladder collection

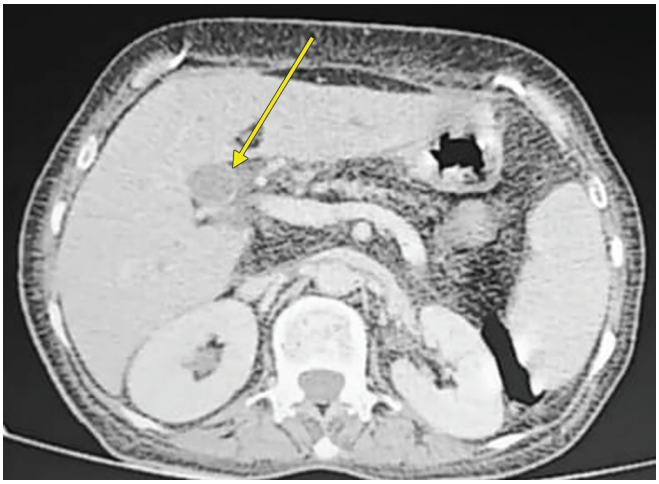

Fig. 4: The CECT abdomen showing dilated CBD

Table 3: Review of cases of bilothorax

Case study	*Etiology*	*Outcome*
Addas RA et al.[14]	Iatrogenic due to biliary draining catheter	Died due to advanced metastatic pancreatic cancer
Tchercanskya AN et al.[15]	Gallstone spillage due to laparoscopic cholecystectomy	Died on postoperative day 11
Reddy VD et al.[16]	No definitive cause of bilothorax was identified	Died due to a worsening condition
Sano and Yotsumoto[17]	PTBD	Resolved with thoracostomy
Owens and Sanders[18]	Acute pancreatitis	Resolved with thoracostomy
Kim and Zangan[19]	PTBD and biliary obstruction	Died due to a worsening condition
Bilal et al.[20]	PTBD	Died due to bowel perforation
Bulus et al.[21]	Hydatid cyst and PBF	Resolved after endoscopic retrograde cholangiopancreaticography (ERCP) and thoracostomy
Turong and Hauringa[22]	PTBD with biliary obstruction	Resolved after decortication with video-assisted thoracic surgery (VATS)
Cooper et al.[23]	Blunt trauma and PBF	Resolved with thoracostomy
Motika[24]	Hepatocellular carcinoma (HCC) and biliary obstruction	Resolved after biliary decompression with ERCP

exclusively biliary tract obstruction due to CBD stricture led to bilothorax.

So far, this is the first case of bilothorax due to obstruction of the biliary tract as a result of CBD stricture. Frampton et al. reported a case of bilothorax as a result of calculus in the distal CBD with pleurobiliary fistula (PBF) confirmed by magnetic resonance cholangiopancreaticography (MRCP).[12] Table 3 represents a review of a few cases of bilothorax published in *Pleura* in 2017,[4] and other publications.

Bile salts are inherently alkaline and caustic to pleural and peritoneal tissue and cause an intense inflammatory response which can eventually end with an acute respiratory distress syndrome (ARDS).[14] None of the cases documented in the medical literature were characterized as transudative effusions.[17] Frequently, bilothorax is complicated by infection due to biliary obstruction, as bile confinement provides an optimal environment for bacterial proliferation. Microorganisms commonly originate from the gastrointestinal (GI) tract, such as *Escherichia coli*, Enterobacteriaceae, and *Staphylococcus aureus*, are often implicated in the development of emphysematous bilious effusions.[25] Delayed diagnosis of bilothorax poses life-threatening risks and is associated with numerous sternly complications, including loculated effusion, empyema, and sepsis.[5,6] Although there is no established protocol for bilothorax management, the typical approach involves immediate chest tube insertion to achieve complete drainage of the bilothorax.[17] This intervention is crucial, as bilothorax has a high propensity to progress to empyema, and addressing the underlying cause is essential. Most reported cases endorse a conservative management approach, involving pleural drainage with the insertion of an ICD, which has proven to be an effective and appropriate method for resolving this condition.[26]

In this case, the patient presented with right-sided massive pleural effusion, and abdominal pain with jaundice. Thoracentesis on the right side yielded greenish color fluid with a raised pleural fluid bilirubin to serum bilirubin ratio of 4.7, which is more than 1 signifying bilothorax. Upon investigating further, CECT chest and abdomen reported biliary tract obstruction due to CBD stricture and did not demonstrate any fistula into the pleural space. Investigations such as MRCP or ERCP were not pursued in this case

due to limitations in the available resources at our hospital with cost-effective approach.

Therefore, after stabilizing the patient and complete drainage of fluid by tube thoracostomy, we referred the patient to the Department of Gastroenterology for further evaluation and management.

There are various prospects regarding how bile reaches the pleural space. Of them, few are by the laid-back movement of bile either through lymphatic channels or transdiaphragmatic pores and congenital defects in the diaphragm. Others include injury of the biliary tract either due to blunt trauma or procedural complications and due to extension of biliary peritonitis.[5,17,23,27,28] The most specific test for diagnosis of bilothorax is pleural fluid bilirubin to serum bilirubin ratio above 1.[4]

In our case, as there is no definite fistulous connection and concurrent ascites, we assert that etiological underpinning resides in the obstruction of the biliary tract secondary to distal CBD stricture. The heightened intrabiliary and intrahepatic ductal pressures are postulated to have precipitated the expansion of submicroscopic interconnections traversing the diaphragmatic barrier. Consequently, served as a conduit facilitating the retrograde transit of bile into the pleural space, thereby culminating in the accumulation of bile within the thoracic cavity characterized by its intrinsically negative pressure environment.

Conclusion

Bilothorax is a rare manifestation. Unlike common etiologies of bilothorax-like trauma, rupture of hepatic or subphrenic abscess, exclusively biliary tract obstruction due to CBD stricture is one rare possibility of bilothorax and no such case exists in the literature so far. Early diagnosis and treatment should be given to prevent the development of empyema as bile in a contained area is an optimal environment for bacterial proliferation. The linchpin management of bilothorax includes complete pleural drainage and correction of the root cause.

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