

# Infected Bile In The Bellows: A Case Of Pycholethorax.

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## Abstract

Cholethorax is an unusual condition in which bile invades the pleural space. It may occur in any setting where the barriers between the pleural space and hepatobiliary system are broken. Of note, iatrogenic perforation has been increasingly reported cause in recent years [1-5]. The bilious pleural effusion may subsequently become infected becoming a pycholethorax. We report a case of pycholethorax in the setting of a percutaneous transhepatic biliary drainage (PTBD) in a patient with a biliary obstruction.

## INTRODUCTION

Cholethorax is an unusual condition in which bile invades the pleural space. It may occur in any setting where the barriers between the pleural space and hepatobiliary system are broken. Of note, iatrogenic perforation has been increasingly reported cause in recent years [1-5]. The bilious pleural effusion may subsequently become infected becoming a pycholethorax. We report a case of pycholethorax in the setting of a percutaneous transhepatic biliary drainage (PTBD) in a patient with a biliary obstruction.

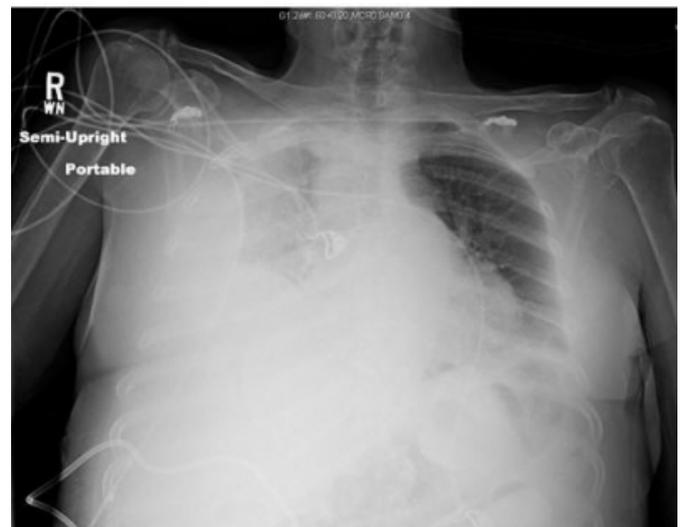
## CASE PRESENTATION

A 79-year-old woman with past medical history of coronary artery disease, hypertension, diabetes mellitus, and hypothyroidism presented with one month of progressively worsening pruritis and jaundice. A clinical diagnosis of biliary obstruction was made. Laboratory investigation revealed a bilirubin level at 10.7 mg/dL, AST 47 U/L, and ALT 29 U/L. A CT scan of the abdomen showed significant dilatation of the intrahepatic biliary tree. An endoscopic retrograde cholangiopancreatogram was attempted, however, due to marked stenosis of the duodenal bulb, the Ampulla of Vater could not be accessed by the endoscope. A PTBD procedure was subsequently performed. Next day, the patient's symptoms improved and the bilirubin level decreased. She was discharged home with outpatient follow-up. On post-operative day (POD) 4, the patient developed right sided pleuritic chest pain and dyspnea. On POD 7, the patient returned to the hospital with worsening dyspnea, tachycardia, and hypotension. Physical exam was significant

for decreased lung sounds and dullness to percussion on the right thorax. A chest x-ray revealed a large right sided pleural effusion (Figure 1). The patient was admitted to the ICU for further management.

## Figure 1

Total opacification of the right hemithorax.



A thoracentesis was performed and a small amount of green-colored pleural fluid was drained (Figure 2). A chest tube was then placed, but there was minimal output. A clinical diagnosis of an organized empyema was made. The pleural fluid analysis showed a bilirubin level of 22.9 mg/dL and pleural fluid cultures were positive for *Klebsiella pneumoniae*, making a diagnosis of pycholethorax.

**Figure 2**

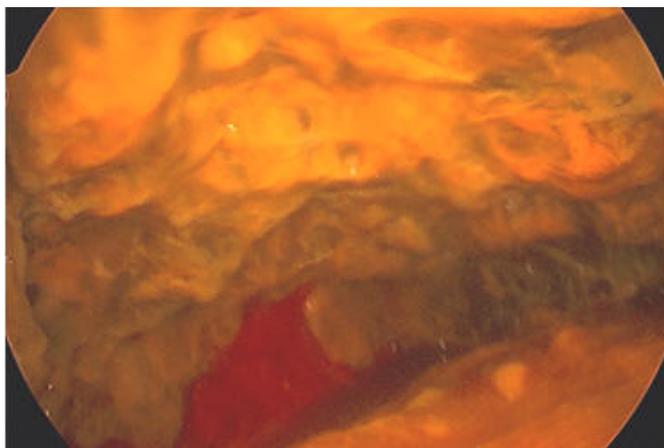
Fluid drained from chest tube with dark green appearance.



A video-assisted thoracoscopy surgery (VATS) decortication was performed. During this procedure, a large amount of green mucous-like material with thick consistency lining the interior thorax and lung parenchyma was removed (Figure 3). Three days later, the patient's dyspnea and her chest x-ray appearance improved. The patient was treated with appropriate antibiotics and transferred to a ward team for further work-up of her biliary obstruction.

**Figure 3**

Green mucous-like material with thick consistency lining the interior chest wall (top) and lung parenchyma (bottom).



## DISCUSSION

This case illustrates the clinical presentation of an iatrogenic pycholethorax. The PTBD in this patient was done via the

intercostal approach in which the biliary catheter was advanced through the pleural sulcus into the biliary tree. This approach has been widely used for diagnostic and therapeutic purposes. Pleural complications from the intercostal approach are uncommon as shown in a study of 230 interventional radiologic procedures over 3 years. In this study, complications (e.g. infection, hemothoraces, and pleural effusion) occurred in only 3% of patients and bilious pleural effusions only occurred 1 in one case [14]. In our case, the intercostal PTBD inadvertently led to a large amount of "Bile in the Bellows," and the development of pycholethorax which led to the patient's rapid clinical deterioration.

Cholethorax has been reported in a variety of settings including hepatic hydatidosis [16], amebiasis [16], liver abscesses [13], gallbladder disease (including cholelithiasis) [17-18], iatrogenic perforation (e.g. PTC [3], PTBD [2, 4, 19], open cholecystectomy [5], liver biopsy [8], and bilio-enteric bypass[1]), blunt hepatic trauma [10, 11, 20], penetrating thoracoabdominal trauma [7, 21, 22], and in one case the cause was unknown [24]. It may occur in any condition in which the barriers between the hepatobiliary system and pleural space are broken. In addition, cholethorax may occur with or without biliary-pleural fistula formation. Biliary fistulae occur in 2-4 % of all hepatic injuries and a subset of these cases will become biliary-pleural fistulae [6,7]. The risk factors of biliary-pleural fistula development has been described by Pistani and are described below in Table 1 [8]. The negative intrapleural pressure during inspiration aids in bile migration to the pleural space.

**Table 1**

Risk factors for fistula development:

Inadvertent removal of drainage tubes
Prolonged drainage
Tubes placed intercostally between the 9th and 10th ribs in the midaxillary line
Persistent biliary tract obstruction
Biliary tube dysfunction.

For clarification of terminology, bilious pleural effusions have been called cholethorax [2-4, 16, 19, 24] as well as bilothorax [9-11]. The term thoracobilia was coined in 1972 to describe any condition in which bile accumulates in the thorax not limited to the pleural space [12].

The prognosis of patients with cholethorax depends on the cause and development of complications. Generally, higher

rates of morbidity are associated with infectious etiologies of cholethorax (e.g. hydatidosis and amebiasis) and complications (e.g. progression to pycholethorax, fistula development)[12-13].

A cholethorax should be suspected in any patient with a recent PTBD who presents with dyspnea right sided pleuritic chest pain. A chest xray should be obtained to evaluate for a pleural effusion. A thoracentesis, which is both diagnostic and therapeutic, should be performed. Diagnosis of cholethorax is made with a thoracentesis aspiration showing bilious green fluid with elevated pleural fluid total bilirubin. The ration of pleural total bilirubin to serum total bilirubin > 1.0 has been used as a diagnostic parameter [8, 13]. In our case this ratio was 2.1. Cultures should be sent. Antibiotics should be started empirically if a pycholethorax is suspected. Of note, the most common organisms isolated from infected bile are Escherichia coli, Enterococcus, Klebsiella, and Enterobacter (Table 2)[15]. This is important when considering empiric antibiotic coverage. Diagnosis of pycholethorax is made when bilious pleural fluid grows out bacteria.

**Table 2**

Most Common Organisms isolated from infected bile by percentage.

Organism	%
Escherichia coli	41
Enterococcus	12
Klebsiella	11
Enterobacter	9

If pleural drainage is minimal a chest tube should be placed. If chest tube output is small, a more aggressive surgical approach should be considered. In our case, the patient a VATS decortication was required. If a fistula is present, imaging may be required. Imaging modalities include endoscopic retrograde cholangio-pancreatography (ERCP), percutaneous transhepatic cholangio (PTC), contrast-enhanced magnetic resonance cholangio (MRC) and hepatobiliary imino-diacetic acid (HIDA) scan. The latter two may be preferred for as they are less invasive. Treatment of biliary-pleural fistulas is surgical and carries a 97% success rate. Non operative management of biliary-pleural fistulas are successful in only 62% of cases [18]. Lastly, bile is a chemoirritant which causes extensive pleural inflammation and effective pleurodesis.

**CONCLUSION**

Cholethorax is a rare condition which has been increasingly

reported following PTBD. It has occur with or without biliary-pleural fistula development. Treatment consists of pleural fluid removal (e.g. thoracentesis, chest tube placement, and VATS decortication), fistula closure if present, and antibiotics. A cholethorax may become infected becoming a pycholethorax leading to rapid clinical deterioration. Therefore, prompt diagnosis and treatment are essential.

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