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

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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 pyocholethorax. We report a case of pyocholethorax 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 pyocholethorax. We report a case of pyocholethorax 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 followup. 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 greencolored pleural fluid was drained (Figure 2). A chest tube was then place, but there was minimal output. A clinical diagnosis of an organized empyema was made. The pleural fluid analysis was showed a bilirubin level of 22.9 mg/dL and pleural fluid cultures were positive for Klebsiella pneumoniae, making a diagnosis of pyocholethorax.

#### 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 xray 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 pyocholethorax. 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 pyocholethorax 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 (incluiding cholelithiasis) [17-18], iatrogenic perforation (e.g. PTC [3], PTBD [2, 4, 19], open cholecystectomy [5], liver biopsy [8], and bilioenteric 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 biliarypleural 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 9t and 10th ribs in the midaxillary line	h
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 pyocholethorax, 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 pyocholethorax 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 pyocholethorax is made when bilious pleural fluid grows out bacteria.

# Table 2

Most Common Organisms isoloated from infected bile by percentage.

Organism	%
Escherichiacoli	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), contrastenhanced 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 pyocholethorax leading to rapid clinical deterioration. Therefore, prompt diagnosis and treatment are essential.

#### References

1. Basu S, Bhadani S, Shukla VK. A dangerous pleural effusion. Ann R Coll Surg Engl. 2010;92(5):W53-4. 2. Al-qahtani HH. Biliopleural fistula with cholethorax. A rare complication of percutaneous transhepatic biliary drainage. Saudi Med J. 2011;32(11):1189-92. 3. Park CS, Lee SJ, Do GW, Oh SY, Cho H, Kim MS, et al. A case of cholethorax following percutaneous transhepatic cholangioscopy. Tuberc Respir Dis 2008;65:131-6. 4. Turkington RC, Leggett JJ, Hurwitz J, Eatock MM. Cholethorax following percutaneous transhepatic biliary drainage. Ulster Med J. 2007;76(2):112-3. 5. Prabhu R, Bavare C, Purandare H, Supe A. Pleuro-biliary fistula -- a delayed complication following open cholecystectomy. Indian J Gastroenterol. 2005; 24(1):28-9. 6. Hollands MJ, Little JM. Post-traumatic bile fistulae. J Trauma. 1991;31(1):117-20. 7. Sheik-gafoor MH, Singh B, Moodley J. Traumatic thoracobiliary fistula: report of a case successfully managed conservatively, with an overview of current diagnostic and therapeutic options. J Trauma. 1998;45(4):819-21. 8. Pisani RJ, Zeller FA. Bilious pleural effusion following liver biopsy. Chest. 1990;98(6):1535-7. 9. Rowe PH. Bilothorax--an unusual problem. J R Soc Med. 1989; 82(11): 687-688. 10. Ghritlaharey RK. Posttraumatic bilothorax in a child: A case report. Journal of Clinical and Diagnostic Research. 2008; 2:760-763. 11. Cooper AZ, Gupta A, Odom SR. Conservative management of a bilothorax resulting from blunt hepatic trauma. Ann Thorac Surg. 2012;93(6):2043-4. 12. Amir-jahed AK, Sadrieh M, Farpour A, Azar H, Namdaran F. Thoracobilia: a surgical complication of hepatic echinococcosis and amebiasis. Ann Thorac Surg. 1972;14(2):198-205. 13. Strange C, Allen ML, Freedland PN, Cunningham J, Sahn SA. Biliopleural fistula as a complication of percutaneous biliary drainage: experimental evidence for pleural inflammation. Am Rev Respir Dis. 1988;137(4):959-61. 14. Nichols DM, Cooperberg PL, Golding RH, Burhenne HJ. The safe intercostal approach? Pleural complications in abdominal interventional radiology. AJR Am J Roentgenol. 1984;142(5):1013-8. 15. Csendes A, Burdiles P, Maluenda F, Diaz JC, Csendes P, Mitru N. Simultaneous bacteriologic assessment of bile from gallbladder and common bile duct in control subjects and

patients with gallstones and common duct stones. Arch Surg. 1996;131(4):389-94.
16. Kolbakir F, Erk MK, Keceligil HT, Yilman M. Cholethorax. Journal of Experimental and Clinical Medicine. 1994; 11(1):45-46.

17. Córdoba lópez A, Monterrubio villar J, Bueno alvarezarenas I. [Biliothorax unrelated to fistula: a rare complication in gallbladder disease]. Arch Bronconeumol. 2008;44(7):396-7. (don't have)

18. Cunningham LW, Grobman M, Paz HL, Hanlon CA, Promisloff RA. Cholecystopleural fistula with cholelithiasis presenting as a right pleural effusion. Chest. 1990;97(3):751-2.

19. Thusgaard G, Johnsson BD, Fleckenstein P.

[Cholethorax. A complication of percutaneous transhepatic cholangiography]. Ugeskr Laeg. 1985;147(29):2304-5. (don't have)

20. Brunaud L, Sebbag H, Bresler L, Tortuyaux JM, Boissel P. Left hepatic duct injury and thoracobiliary fistula after abdominal blunt trauma. Hepatogastroenterology. 47(35):1227-9.

21. Cannon JW, Sebesta JA, Blackbourne LH, Smith DL,

and Holcomb JB. Thoracoabdominal war wounds
complicated by thoracobiliary fistulae: a report of cases
managed over the past eighteen years and a review of the
literature. J Surg Radiol. 2011 Jul 1;2(3).
22. Rothberg ML, Klingman RR, Peetz D, Ferraris VA,
Berry WR. Traumatic thoracobiliary fistula. Ann Thorac
Surg. 1994;57(2):472-5.

23. Frampton AE, Williams A, Wilkerson PM, Paterson IM. Thoracobilia: a rare complication of gallstone disease. Ann R Coll Surg Engl. 2010;92(5):W1-3.

24. Seong MH. A Case of Cholethorax Developed by Unknown Cause. Tuberculosis and Respiratory Diseases. 2011; 70 (3): 261-265

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