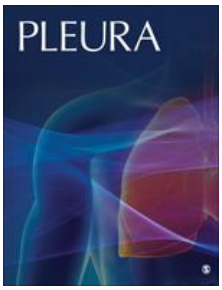

The Official Journal of the International Society of Pleural Diseases

Case Series & Review Article

The Green Pleural Effusion: A Comprehensive Review of the Bilothorax with Case Series

Adam Austin, MD[1], Nick Fox, MD[2], John Terrill Huggins, MD[2]*, Amit Chopra, MD[1]*

Abstract:
Bilothorax is a rare cause of an exudative pleural effusion and is characterized by the accumulation of bile into the pleural space. To date, there has been no systematic review of the clinical characteristics, pleural fluid analysis, pathophysiology and management of bilothorax. The most specific laboratory data to support the diagnosis of biliothorax is a total pleural fluid to serum bilirubin ratio greater than 1.0. Emergent diagnosis and drainage is essential to prevent the development of empyema from superimposed infection as bile fluid accumulates in the pleural space.

Keywords: Bile; Pleural Effusion; Thoracobilia; Cholethorax; Bilothorax

Institution:
[1]Department of Medicine, Division of Pulmonary and Critical Care Medicine, Albany Medical College, Albany, NY
[2]Department of Medicine, Division of Pulmonary, Critical Care and Sleep Medicine, Medical University of South Carolina, Charleston, SC
* Co-Senior Authors

Corresponding Author:
Adam Austin, MD
Department of Medicine
Division of Pulmonary and Critical Care
Albany Medical College
43 New Scotland Avenue
Albany, New York 12208
Email: Austina4@mail.amc.edu
Phone: 518-262-5377
Fax: 518-262-6472

Contact the Corresponding Author for reprints and permissions

---

PLEURA. 2017;4:21-31.

Received: 7/20/2017
Accepted: 10/1/2017
Published online: December/2017

Introduction:

Bilothorax is defined by the presence of bile in the pleural space. There are approximately sixty cases reported in the medical literature. The majority of the described cases are from bile tract injury resulting in a pleurobiliary fistulalization or from a sub-phrenic abscess formation. In previous reported cases, there has been a limited description of the pleural fluid criteria for the diagnosis of bilothorax and the pathophysiology of this condition.

In our case series, we describe five previous unpublished patients diagnosed with bilothoraces at the Albany Medical College and the Medical University of South Carolina (Table 1). In conjunction, we have additionally characterized the pleural fluid analysis (PFA), pathophysiology, etiology and management of this uncommon condition in the extant medical literature. We document fifty-two cases of bilothorax published in the English literature search via a Medline search from the years 1960 to 2016 using the search terms; *bilothorax*, *cholethorax*, *thoracobilia*, *pleurobilia*, *thoracobiliary fistula*, *bile* and *pleural effusion* (Table 2).

Case 1:

A 59 year-old man was admitted with obstructive jaundice from pancreatic head adenocarcinoma compressing the common bile duct. The patient developed ascending cholangitis and sepsis, which was treated with antibiotics and percutaneous biliary drain for source control. Subsequently, the patient developed pleuritic chest pain and was found to have right-sided, loculated pleural effusion. Bedside ultrasound demonstrated a complex, septated fluid collection. A small-bore chest tube was placed for management. The pleural fluid appearance was green and cloudy. The PFA was notable for a pH 7.02; pleural fluid to serum total protein ratio (PF/S protein) of 0.71, LDH 1170 IU/L, pleural fluid to upper limits of normal serum LDH ratio (PF/UNLS LDH) of 4.9 and total pleural to serum bilirubin ratio of 1.17. The total leukocyte count of pleural fluid was only 190 cells. Cytology was benign and bacterial cultures were negative. The patient was eventually discharged to hospice care.

Case 2:

A 46 year-old white man with cholangiocarcinoma with a recent biliary drain placement returned to emergency room with fever and new onset of shortness of breath. He was found to have sepsis secondary to Escherichia coli bacteremia with obstructive jaundice and a new right-sided effusion. An abdominal CT scan demonstrated a 7.2 x 9 cm sub-capsular fluid collection consistent with a biloma along with biliary obstruction (Figure 1). Thoracentesis revealed an exudative bilious pleural effusion with pleural bilirubin equal to serum bilirubin (7.9mg/dL). The fluid was negative for infection as well as malignant cells. Patient required surgical resection of the diseased part of the liver, subsequent to which pleural effusion resolved.

Case 3:

A 53 year-old white man with a history of non-alcoholic steatohepatitis related cirrhosis, underwent an orthotropic liver transplantation. Post-liver transplant course was complicated by biliary stricture and a post-transplant lymphoproliferative disorder. He presented with a new onset, right-sided pleural effusion, and underwent diagnostic thoracentesis that was consistent with exudative bilious pleural effusion with pleural bilirubin of 4.9 mg/dL (serum 4.8 mg/dL). Pleural fluid culture and cytology were negative. A percutaneous biliary drain was placed and patient was discharged home.

Figure-1: Contrast Tomography Scan of Abdomen demonstrating Biloma .

Case 4:
A 59-year-old white man with past medical history notable for poorly-differentiated gastric adenocarcinoma status-post distal esophagectomy and radical gastrectomy was admitted with cholecystitis and distal common bile duct stone. The patient underwent a placement of a percutaneous cholecystostomy tube, following which he became acutely dyspneic with chest radiograph demonstrating bilateral pleural effusions. A chest CT demonstrated a right greater than left pleural effusion with the internal-external biliary drain traversing the pleural space. Thoracentesis revealed an exudative, bilious pleural effusion with pleural bilirubin of 1.7 mg/dL (serum 1.3 mg/dL). Pleural fluid culture was negative for organisms. After removal of common bile duct stone, drainage from cholecystostomy tube decreased and pleural effusion resolved.

Case 5:
A 79 year-old white man with recent diagnosis of duodenal adenocarcinoma with duodenal stent placement developed new onset jaundice. He was found to have biliary obstruction and underwent percutaneous transhepatic cholangiogram with percutaneous drain placement. He later developed a large, loculated right-sided pleural effusion. Thoracentesis with pleural fluid analysis demonstrated a dual diagnosis of an exudative, bilious pleural effusion with pleural bilirubin 3.2 mg/dL (serum 2.3 mg/dL) along with pleural space infection. The effusion re-occurred necessitating chest tube placement with intrapleural tPA administration for complete pleural drainage.

Discussion:
Bilothorax is a rare cause of exudative effusion of an extra-vascular origin. Pleural effusions from extra-vascular origin can be classified as exudates and transudates. In addition to bilothorax, exudative pleural effusions of extravascular origin also include: (1) esophageal rupture, (2) extra-vascular migration of a central venous catheter with total parenteral nutrition infusion, (3) chylothorax, (4) enteral tube feeds leaking into pleural space, (5) gastropleural fistula and (6) pancreaticopleural fistula[1].

The term *bilothorax* was first coined by Williams[2] in 1971 in a case of blunt trauma resulting in a biliary fistulalization into the pleural space. In the extant English medical literature, bilious pleural effusions have several other monikers, including *cholethorax, bilithorax*, *pleurobilia* and *thoracobilia*. However, it is important to note that when Amir-Jahed[3] first used the phrase *thoracobilia*, it was originally defined as a syndrome of tender hepatomegaly and biloptysis with constitutional symptoms in setting of hepatic parasitic infections. Since this first use of the term in the surgical literature in 1972, thoracobilia has often been utilized as an all encompassing term to describe both pleurobiliary or bronchobiliary fistula and any pathology whereby bile enters the thoracic cavity regardless of the presence of biliary pleural effusion. The majority of the reported bilothoraces occur on the right-side of the thorax due to the anatomical location of liver and biliary system. In our review, there was one case of bilateral bilothorax in setting of bile spillage in the peritoneal cavity from a misadventure of a gastrojejunostomy placement resulting in perforation[4]. Only two pleural effusions were left-sided[5,6]; one case of acute pancreatitis[5] and another involving gastric perforation via nasogastric tube placement[6]. In our series, all of the cases of bilothorax were right-sided.

The most common etiologies of bilothorax include biliary obstruction [7, 8, 9, 10, 11], diaphragmatic trauma [2, 12, 13, 14, 15, 16, 17, 18], extension of hepatic infections [3, 16, 19], subphrenic abscess formation as a consequence of biliary obstruction[20, 21, 22] and iatrogenic complications of percutaneous hepatobiliary drain insertion[23, 24]. The exact mechanism is debatable, but postulated routes of bile entry into the pleural space include: 1) passive movement of bile through the diaphragm in a similar fashion as ascitic fluid leading to a hepatic hydrothorax whereby increased negative intrathoracic pressure facilitates biliary transit through diaphragmatic pores and 2) drainage of the collected sub-phrenic biliary fluid through lymphatics to pleural lymphatics via pleuro-peritoneal lymphatic connections.

Fistula formation can occur through multiple etiologies. Bile salts are inherently alkaline and caustic to pleural and peritoneal tissue, corroding the posterior recess of the diaphragm in cases with bile leakage even without iatrogenic perforation. Diaphragmatic injury, either through blunt trauma or iatrogenic causes, can form a connection from the peritoneal to pleural space. The presence of a percutaneous biliary drain potentiates the transit time of bile across the diaphragm into the pleural space by either direct or indirect mechanisms[24 25]. Direct

mechanism involved include perforation of pleural sulcus[26] via misplacement of percutaneous drain in the mid-axillary line between the 9th and 10th intercostal space[7]. Indirect mechanisms include bile accumulation in the sub-phrenic space due to either drain dysfunction, accidental displacement, or obstruction in drainage. Subsequently, biliary fluid can migrate from sub-phrenic space into the pleural space via above defined mechanisms. More common in the developing world, parasitic hepatic infections[3], especially Amebiasis[27, 28] and Echinococcosis[29, 19], can directly result in diaphragmatic extension into the pleural space as a result of the close proximity. In one retrospective analysis, 65% of amebic liver abscesses resulted in the extension into the thorax[27], and in another study of 123 patients developed thoracic rupture with hydatid hepatic cysts[29]. The development of chocolate-colored sputum, referred to as *anchovy-paste*[28], as opposed to biloptysis in the setting of bronchobiliary fistulalization, has been known to occur in setting of broncho-hepatic fistulas during the course of Entamoeba *histolytica* infections.

There is a strong association between the presence of a bilothorax and the development of an empyema. In a case series and review of thoracobiliary fistulalization by either gunshot, stab wound or blunt trauma by Oparah in 1978[15], it was demonstrated that of sixteen cases in the literature, twelve patients developed bilious empyemas. Bilious pleural effusions are mostly infected due to biliary obstruction. The most common organisms resulting in complicated and empyematous bilious effusions are gastrointestinal tract organisms like Escherichia *coli*[22, 24, 30], Enterobacter[15], Klebseilla *oxytoxa*[15, 22], Enterococcus *faecalis*[31] and Staphylococcus *aureus*[15]. In our review, there was one instance of an atypical biliary tract pathogen, a case of Stenotrophomonas (xantomonas) *maltophilia*, which occurred in the setting of a gastric perforation.

In the fifty-two published cases of bilothorax, the PFA was described in only eighteen (35%) of fifty-two cases. The pleural effusions were described as empyemas in seven (39%) cases, as an exudate in eight (44%), as complex fibrous effusions in two (11%) and as an effusion with lung entrapment in one case. None of the bilothoraces reported in the literature were described as a transudative effusion. In our series, five cases (83%) were associated with exudative effusions, and one was an empyema. In terms of the pleural fluid pH, it was reported in five (9.6%) of fifty-two cases. The mean pleural fluid pH reported was 7.16 (range 6.78 to 7.80). In our series, the mean pleural fluid pH was 7.42 (range 6.83 to 7.45). The PF/S bilirubin ratio was reported in only fourteen (27%) of fifty-two cases reported in the extant literature. The mean pleural fluid to serum bilirubin ratio reported was 8.4 (range 1 to 52.2). In our series, the mean was lower at 1.17 (range 1.00 to 1.39).

Pleural fluid to serum bilirubin ratios in all reported cases were greater than 1.0, with a range of 1.4 to 11.6. In five of the fifty-two cases only the total pleural fluid bilirubin level was reported, which ranged from 6.8 to 57.78. In terms of the remainder of the PFA on our review of the literature, only a limited number of cases detailed complete data of the pleural fluid and serum. The PF/S protein ratio was reported in two (3.8%) of fifty-two cases (3.8%) with values of 0.62 and 0.52. The mean PF/S protein ratio was 0.64 (range 0.52 to 0.80). The lack of PFA is also evident in pleural fluid LDH values, which ranged from 332 to 2251 IU/L and was reported in six (11.5%) of fifty-two cases. In our series, the mean PF/ULNS LDH was 2.10 with a pleural fluid LDH range of 131 to 1170 IU/L.

Diagnosis:

The diagnosis of a bilothorax requires a high level of suspicion. Conventional imaging modalities, including chest radiograph, ultrasound and CT can be used in establishing an accurate clinical diagnosis. The most specific test for the diagnosis of bilothorax is a total pleural fluid to serum bilirubin ratio greater than 1.0, with higher ratios increasing specificity for the diagnosis. A minimally invasive hepatobiliary scan can be useful in demonstrating the presence of a fistulalizing tract between the biliary tree and the thoracic cavity,[30] however it is not required for the diagnosis of bilothorax. Although a less common diagnostic tool, an exploratory laparotomy and thoracotomy can be both diagnostic and therapeutic by simultaneously identifying and repairing the communicating anatomical defect.

Management:

Successful management of a bilothorax requires a rapid and accurate diagnosis. Similar to hemothorax, Most reported cases described in the literature have a dual diagnosis of combined empyema and bilothorax. Therefore, we recommend early administration of broad-

spectrum antibiotics and immediate drainage of pleural space to prevent future infection. Early and aggressive surgical intervention is required in most of the cases. In the surgical management of thoracobiliary fistulas, it was even proposed by Ferguson and Burford[16] in 1967 after observations in combat wounds during World War II that diaphragmatic trauma and development of bile in the pleural space required steadfast thoracotomies, decortication and repair of the diaphragm. Conservative management unlikely will prevent re-accumulation of bile in the pleural space. With advancement in minimally invasive procedures, percutaneous drainage of the biliary tree and endoscopic retrograde cholangio-pancreatography (ERCP) alleviating biliary obstruction along with pleural drainage is the main stay of treatment.

Conclusion:

Bilothorax is a rare form of an exudative pleural effusion of extra-vascular in origin. The diagnosis of bilothorax requires a high degree of clinical suspicion and should be considered in cases of blunt abdominal trauma, severe hepatic infections and percutaneous transhepatobiliary interventions. The most specific finding is a pleural fluid to serum ratio of bilirubin greater than 1.0. Other PFA parameters may be variable depending on presence of concomitant infection. Complete emergent drainage of a bilothorax is essential to prevent empyema and fibropurulent effusions. Spontaneous closure of the bilopleural fistula typically never occurs and will require either direct surgical closure, percutaneous drainage of the biliary tree or ERCP to alleviate biliary obstruction.

Table-1: Bilothorax cases from AMC and MUSC

Pt.	Side	Type	Color	pH	P/S Bilirubin Ratio	P/S Protein Ratio	P/S LDH Ratio	Pleural LDH to Serum ULN (240 IU/L) LDH	Glucose	Nucleated Cells	WBC	PMN	Lymph	Etiology	Outcome
1	Right	Exudate	Green	7.02	8.8/7.5= 1.17	0.71	1170/225=5.2	1170/240= 4.88	85	295	190	70	7	Pancreatic adenocarcinoma with concomitant infection.	Patient choice palliative measures.
2	Right	Exudate	Yellow	7.42	7./7.9= 1.0	4.1/5.1=0.8	131/181 =0.72	131/240 = 0.55	99	1,323	1,125	79	6	Chronic biliary obstruction from malignancy.	Resolved after perihepatic drain placement.
3	Right	Exudate	Sero-sanguinous	7.41	4.9/4.8= 1.02	3.6/5.7 = 0.63	N/A	239/240 = 0.99	137	11,735	N/A	90	3	Biliary obstruction from biliary stricture post liver transplant.	Resolved after percutaneous biliary drain placed.
4	Right	Exudate	Yellow, cloudy	7.43	1.7/1.3= 1.31	3.3/5.3 = 0.62	N/A	474/240 =1.98	104	7,454	5,889	65	14	Bile leak post cholcystostomy tube placement for CBD stone.	Resolved after stone removal and drain removal.
5	Right	Exudate	Amber	6.83	3.2/2.3= 1.39	2.5/4.6 = 0.54	N/A	833/240 =3.33	43	N/A	N/A	N/A	N/A	Biliary obstruction from malignancy.	Patient decided on palliative care.

Legend:

N/A= Not Available
P/S= Pleural to Serum
LDH= Lactate Dehydrogenase
WBC= White Blood Cell
PMN= Polynuclear Cell

Table-2: Cases of Bilothorax in Literature Review

		Pleural Fluid Analysis						
	# Case	Exudate/Transudate	Total Bilirubin P/ S Ratio	Total Pleural Bilirubin (mg/dL)	pH	PFA	Etiology	Outcome
Sano[23] 2016	1	NR	NR	57.78	NR	NR	PTBD	Resolved with thoracostomy
Sano[23] 2016	1	Exudate	NR	NR	NR	NR	PTBD	Resolved with thorascopic curettage and drainage
Wu[32] 2016	1	NR	NR	NR	NR	NR	Bronchobiliary fistula	Resolved after choledoscopy
Begum[12] 2015	1	Exudate	NR	6.8	NR	Total Cell Count 4300/cumm w/ neutrophil predominance, Gram Stain demonstrated Gram Negative Bacilli, Culture Negative	Blunt Trauma	Resolved with conservative management
Owens[5] 2015	1	Exudate	4.6	NR	NR	LDH 1706, Protein Ratio 0.52	Acute Pancreatitis	Resolved with thoracostomy
Kim [7] 2015	1	NR	NR	NR	NR	NR	PTBD and Biliary Obstruction	Died due to worsening clinical course
Bilal[33] 2015	1	NR	NR	NR	NR	NR	PTBD	Died due to bowel perforation
Dahiya[13] 2015	1	NR	NR	NR	NR	NR	Pleurobiliary fistula due to gun shot wound	Resolved after subphrenic drainage and thoracostomy
Celis[34] 2015	1	Exudate	2.5	NR	NR	WBC 20,000 cells/cumm w/ neutrophil predominance, LDH 1850 mg/dL	PTGBD	Resolved after thoracoscopy with decortication
Bulus[19] 2015	1	NR	NR	NR	NR	NR	Hydatid cyst and pleurobiliary fistula	Resolved after ERCP and thoracostomy
Jenkinson[20] 2013	1	NR	5.52	NR	NR	NR	Hartmann's pouch perforation	Resolved after cholecystectomy
Hamers[31] 2013	1	NR	NR	20.8	NR	Pleural Fluid Culture positive for Enterococcus faecalis	Liver Mestastatsectomy	Resolved with thoracostomy
Aydogan[35] 2013	1	NR	NR	9.1	NR	NR	Laparoscopic cholecystectomy	Resolved with conservative treatment
Truong[36] 2013	1	Exudate	2.14	NR	NR	NR	PTBD with Biliary Obstruction	Resolved after decortication with VATS
Cooper[18] 2012	1	NR	6.125	NR	NR	NR	Blunt Trauma and Pleurobiliary fistula	Resolved with thoracostomy
Al-Qahtani[30] 2011	1	NR	>1	16.8	NR	Pleural Fluid Culture positive for E.coli	PTC complicated by Pleurobiliary fistula	Resolved after VATS decortication and thoracostomy
Frampton[8] 2010	1	NR	NR	10.53	NR	NR	Pleurobiliary Fistula and Obstruction	Resolved with ERCP and Relieving papilla stenosis
Basu[9] 2010	1	NR	NR	NR	NR	NR	Mirrizzi's syndrome with hepataticojejunostomy and cholecystectomy	Died on POD 21 due to ARDS and multi-organ failure
Ball[37] 2009	1	NR	NR	NR	NR	NR	Thoracoabdominal gunshot wound	Resolved after repair of diaphragmatic defect, chest and peritoneal tube placement and drainage, and ERCP
Motika[38] 2008	1	Exudate	10.6	NR	6.78	Total Cell Count 4221, WBC 2681 w/ neutrophil predominance, LDH 1195 U/L	HCC, Biliary Obstruction	Resolved after biliary decompression with ERCP
Turkington[10] 2007	1	NR	2.0	NR	NR	NR	PTBD and Biliary Obstruction	Resolved with PTC and biliary stent placement
Kaya[39] 2006	1	NR	NR	NR	NR	NR	Hemigoastrectomy and gastrojejunostomy	Abdominal exploration identified no diaphragmatic defect or

								fistula, negative fistulography. Resolved after 25th POD
Prabhu[40] 2005	1	NR	NR	NR	NR	NR	Pleuro-biliary fistula	Resolved with surgical Repair of Fistula
Bini[6] 2004	1	NR	NR	NR	NR	NR	Gastropleural fistula after NG tube stomach perforation	Resolved after repair of fistula via laparotomy
Navsaria[14] 2002	1	NR	NR	NR	NR	NR	Thoracobiliary fistula due to gunshot wound	Resolved after repair of fistula
Stockberger [41] 1999	1	NR	NR	NR	NR	NR	Chronic Klebsiella pneumoniae Subphrenic Abscess post-laparoscopic cholecystecomy	Resolved after Right Middle and Lower Lobectomies
Chopra[21] 1999	1	Exudate	NR	NR	NR	NR	Laparoscopic cholecystectomy complicated by subphrenic abscess and peritoneo-pleuro-bronchial fistula	Resolved with conservative treatment
Delco[25] 1994	1	Exudate	52.2	NR	7.38	WBC 250/cumm, Protein 30 g/L, LDH 2251 U/L	Cholecystopleural fistula	Resolved with cholecystectomy
Kolbakir[42] 1994	1	NR	NR	NR	NR	NR	Transdiaphragmatic perforation from echinococcal cysts	Resolved with surgical repair of diaphragm injury
Cunningham[22] 1990	1	NR	NR	NR	6.96	WBC 41,000 w/neutrophil predominance, Glucose 16 g/dL, Pleural Fluid Culture positive for Klebseilla oxytoxa and E.coli	Gangrenous Gallbladder and cholecystopleural fistula	Died of acute illness
Pisani[11] 1990	1	Exudate	2.6	NR	NR	LDH ratio 0.72, Protein ratio 0.62	Choledocholithiasis and ERCP	Died of multi-organ failure
Rowe[4] 1989	1	NR	NR	NR	NR	NR	Perforation of gastrojejunostomy	Resolved with Emergency Laporatomy
Strange[24] 1988	1	Exudate	11.6	41.7	6.90	WBC 1800/cumm w/ neutrophil predominance, LDH 1800 U/L, Protein 1.8 g/L, Glucose 13 mg/dL	Removal of PBD catheter and Biliary Obstruction	Resolved with subdiaphragmatic bilious drainage
Strange[24] 1988	1	Exudate	1.4	NR	7.80	WBC 9280/cumm w/neutrophil predominance, LDH 332 U/L, total protein 3.5 g/L, Pleural Fluid Culture positive for E.coli	Previous biliary trauma with PTC	Resolved after PBD catheter removal and thoracentesis
Strange[24] 1988	1	NR	NR	NR	NR	NR	Removal of PBD catheter and Biliary Obstruction	Resolved after replacement of PBD
Ivatury[43] 1984	1	Exudate	NR	NR	NR	NR	Gunshot Wound	Resolved after decortication and diaphragmatic repair
Ivatury[43] 1984	1	Exudate	NR	NR	NR	NR	Stab Wound	Resolved after thoracostomy, decortication and diaphragmatic repair
Ivatury[43] 1984	1	Exudate	NR	NR	NR	NR	Stab Wound	Resolved after thoracostomy, decortication and diaphragmatic, liver and lung repair
Oparah[15] 1978	1	Exudate	NR	NR	NR	Pleural Fluid Culture positive for Staph. Aureus and Enterobacter	Gunshot Wound	Resolved afer subphrenic abscess drainage and thoracostomy
Oparah[15] 1978	1	Exudate	NR	NR	NR	Pleural Fluid Culture positive for Klebsiella pneumonia and Enterobacter	Gunshot Wound	Resolved after antibiotics, thoracostomy, decortication and diaphragmatic repair
Oparah[15] 1978	1	Exudate	5.45	6	NR	Pleural Fluid Culture positive for Staph. Aureus	Stab Wound	Resolved after antibiotics and thoracostomy
Oparah[15] 1978	1	NR	10	1.0	NR	NR	Gunshot Wound	Resolved after thoracostomy
Dosik[17] 1975	1	NR	NR	NR	NR	NR	Percutaneous Liver Biopsy, Biliary Obstruction	Resolved after relieving biliary obstruction
Amir-Jahed[3] 1972	1	NR	NR	NR	NR	NR	E. histolytica liver abscess with bronchobiliary fistula	Resolved with amebic therapy
Williams[2] 1971	1	NR	NR	NR	NR	NR	Blunt trauma	Resolved after thoracostomy and choledochostomy

Ferguson[16] 1967	1	NR	NR	NR	NR	NR	Blunt trauma	Resolved after thoracotomy and subcostal drainage
Ferguson[16] 1967	1	NR	NR	NR	NR	Notes “infected bile”	Blunt trauma	Resolved after thoracotomy, decortication and subcostal drainage
Ferguson[16] 1967	1	NR	NR	NR	NR	NR	Blunt trauma	Resolved after thoracotomy and diaphragmatic repair
Ferguson [16] 1967	1	NR	NR	NR	NR	NR	Blunt trauma	Resolved after thoracotomy, diaphragmatic repair, laparotomy and roux-en-y choledochojejunostomy
Ferguson[16] 1967	1	NR	NR	NR	NR	NR	Pyogenic Hepatic Abscess with Staph. Aureus with bronchobiliary fistula	Resolved with antibiotic therapy
Ferguson[16] 1967	1	NR	NR	NR	NR	NR	Hepatic Abscess	Resolved with antibiotics, lobectomy, diaphragmatic repair and subcostal drainage
Ferguson[16] 1967	1	Exudate	NR	NR	NR	NR	Percutaneous Liver Biopsy	Resolved after thoracoplasty, diaphragmatic repair and subcostal drainage

References:

1. Sahn SA. Pleural effusions of extravascular origin. *Clinics in chest medicine* 2006; 27: 285-308.

2. Williams SW, Majewski PL, Norris JE, Cole BC, Doohen DJ. Biliary decompression in the treatment of bilothorax. *American journal of surgery* 1971; 122: 829-831.

3. Amir-Jahed AK, Sadrieh M, Farpour A, Azar H, Namdaran F. Thoracobilia: a surgical complication of hepatic echinococcosis and amebiasis. *The Annals of thoracic surgery* 1972; 14: 198-205.

4. Rowe PH. Bilothorax--an unusual problem. *Journal of the Royal Society of Medicine* 1989; 82: 687-688.

5. Owens WB, Sanders, M.B. . Spontaneous Left-Sided Cholethorax. *PulmCCM Journal* 2015; 1.

6. Bini A, Grazia M, Petrella F, Stella F, Bazzocchi R. Spontaneous biliopneumothorax (thoracobilia) following gastropleural fistula due to stomach perforation by nasogastric tube. *The Annals of thoracic surgery* 2004; 78: 339-341.

7. Kim SH, Zangan SM. Percutaneous Transhepatic Biliary Drainage Complicated by Bilothorax. *Seminars in interventional radiology* 2015; 32: 54-56.

8. Frampton AE, Williams A, Wilkerson PM, Paterson IM. Thoracobilia: a rare complication of gallstone disease. *Annals of the Royal College of Surgeons of England* 2010; 92: W1-3.

9. Basu S, Bhadani S, Shukla VK. A dangerous pleural effusion. *Annals of the Royal College of Surgeons of England* 2010; 92: W53-54.

10. Turkington RC, Leggett JJ, Hurwitz J, Eatock MM. Cholethorax following percutaneous transhepatic biliary drainage. *The Ulster medical journal* 2007; 76: 112-113.

11. Pisani RJ, Zeller FA. Bilious pleural effusion following liver biopsy. *Chest* 1990; 98: 1535-1537.

12. Begum S, Mukherjee S, Biswas D, Misra AK, Ghosh P, Bhanja P. A rare pleural effusion in a young male. *Lung India : official organ of Indian Chest Society* 2015; 32: 389-391.

13. Dahiya D, Kaman L, Behera A. Biliopleural fistula following gun shot injury in right axilla. *BMJ case reports* 2015; 2015.

14. Navsaria PH, Adams S, Nicol AJ. Traumatic thoracobiliary fistulae: a case report with a review of the current management options. *Injury* 2002; 33: 639-643.

15. Oparah SS, Mandal AK. Traumatic thoracobiliary (pleurobiliary and bronchobiliary) fistulas: clinical and review study. *The Journal of trauma* 1978; 18: 539-544.

16. Ferguson TB, Burford TH. Pleurobiliary and bronchobiliary fistulas. Surgical management. *Archives of surgery (Chicago,Ill: 1960)* 1967; 95: 380-386.

17. Dosik MH. Bil pleuritis: another complication of percutaneous liver biopsy. *The American journal of digestive diseases* 1975; 20: 91-93.

18. Cooper AZ, Gupta A, Odom SR. Conservative management of a bilothorax resulting from blunt hepatic trauma. *The Annals of thoracic surgery* 2012; 93: 2043-2044.

19. Bulus H, Aydin A, Koyuncu A, Yavuz A, Coskun A. Biliopleural fistula of a large retroperitoneal hydatid cyst. *The Turkish journal of gastroenterology : the official journal of Turkish Society of Gastroenterology* 2015; 26: 284-285.

20. Jenkinson MR, Campbell W, Taylor MA. Bilothorax as a rare sign of intra-abdominal bile leak in a patient without peritonitis. *Annals of the Royal College of Surgeons of England* 2013; 95: 118-119.

21. Chopra P, Killorn P, Mehran RJ. Cholelithoptysis and pleural empyema. *The Annals of thoracic surgery* 1999; 68: 254-255.

22. Cunningham LW, Grobman M, Paz HL, Hanlon CA, Promisloff RA. Cholecystopleural fistula with cholelithiasis presenting as a right pleural effusion. *Chest* 1990; 97: 751-752.

23. Sano A, Yotsumoto T. Bilothorax as a complication of percutaneous transhepatic biliary drainage. *Asian cardiovascular & thoracic annals* 2016; 24: 101-103.

24. Strange C, Allen ML, Freedland PN, Cunningham J, Sahn SA. Biliopleural fistula as a complication of percutaneous biliary drainage: experimental evidence for pleural inflammation. *The American review of respiratory disease* 1988; 137: 959-961.

25. Delco F, Domenighetti G, Kauzlaric D, Donati D, Mombelli G. Spontaneous biliothorax (thoracobilia) following cholecystopleural fistula presenting as an acute respiratory insufficiency. Successful removal of gallstones from the pleural space. *Chest* 1994; 106: 961-963.

26. Neff CC, Mueller PR, Ferrucci JT, Jr., Dawson SL, Wittenberg J, Simeone JF, Butch RJ, Papanicolaou N. Serious complications following transgression of the pleural space in drainage procedures. *Radiology* 1984; 152: 335-341.

27. Ibarra-Perez C. Thoracic complications of amebic abscess of the liver: report of 501 cases. *Chest* 1981; 79: 672-677.

28. Roy DC, Ravindran P, Padmanabhan R. Bronchobiliary fistula secondary to amebic liver abscess. *Chest* 1972; 62: 523-524.

29. Kabiri EH, El Maslout A, Benosman A. Thoracic rupture of hepatic hydatidosis (123 cases). *The Annals of thoracic surgery* 2001; 72: 1883-1886.

30. Al-Qahtani HH. Biliopleural fistula with cholethorax. A rare complication of percutaneous transhepatic biliary drainage. *Saudi medical journal* 2011; 32: 1189-1192.

31. Hamers LA, Bosscha K, van Leuken MH, Moviat MA, de Jager CP. Bilothorax: a bitter complication of liver surgery. *Case reports in surgery* 2013; 2013: 372827.

32. Wu Z, Wang L. A Man with "Abdominal" Pain. *Liver international : official journal of the International Association for the Study of the Liver* 2016.

33. Bilal M, Chong J, Lega M. Education and imaging. Hepatology: Bilothorax after percutaneous transhepatic biliary drainage. *Journal of gastroenterology and hepatology* 2015; 30: 802.

34. Celis CA, Tobon, M., Ramirez, R., Garcia, M., Suarez, J. Cholethorax Secondary to Percutaneous Transhepatic Gallbladder Drainage: A Case Report. *Am J Resp Crit Care Med* 2015; 191: A2978.

35. Aydogan A, Sukru Erden E, Akkucuk S, Davran R, Yetim I, Veli Ozkan O. Cholethorax (bilious effusion in the thorax): an unusual complication of laparoscopic

36. Truong V, Huaringa, A. Infected Biled In the Bellows: A Case of Pyocholethorax. *The Internet Journal of Pulmonary Medicine* 2013; 15.

37. Ball CG, Trexler S, Rajani RR, Vercruysse G, Feliciano DV, Nicholas JM. Importance of liver drainage in biliary-bronchopleural fistula resulting from thoracoabdominal gunshot injury. *Canadian journal of surgery Journal canadien de chirurgie* 2009; 52: E12-13.

38. Motika CA. A 66-Year-Old Man with Cryptogenic Cirrhosis and Hepatocellular Carcinoma with a Bilious Pleural Effusion [abstract]. *Chest* 2008; 134: c59001.

39. Kaya S, Hasanoglu, C., Demirbas, B., Aydin, R. . An Unusual Complication of the Surgery of Pyloric Stenosis: Bilothorax [abstract]. *Chest* 2006; 130: 302S.

40. Prabhu R, Bavare C, Purandare H, Supe A. Pleuro-biliary fistula--a delayed complication following open cholecystectomy. *Indian journal of gastroenterology : official journal of the Indian Society of Gastroenterology* 2005; 24: 28-29.

41. Stockberger SM, Jr., Kesler KA, Broderick LS, Howard TJ. Bronchoperitoneal fistula secondary to chronic Klebsiella pneumoniae subphrenic abscess. *The Annals of thoracic surgery* 1999; 68: 1058-1059; discussion 1059-1060.

42. Kolbakir F, Erk, M.K., Keceligil, H.T., Yilman, M. Cholethorax. *OMU Tip Dergisi* 1994; 11: 45-46.

43. Ivatury RR, O'Shea J, Rohman M. Post-traumatic thoracobiliary fistula. *The Journal of trauma* 1984; 24: 438-442.