

BILOTHORAX: Diagnostic and Therapeutic Approaches in a Rare Case - A Clinical Review

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ABSTRACT

Background: Bilothorax, or cholethorax, is a rare and serious condition where bile accumulates in the thoracic cavity, often due to complications from hepatobiliary procedures. This report examines a case managed at our institution and reviews the relevant literature to enhance understanding and management strategies.

Keywords: Bilothorax, Cholethorax, Thoracic Surgery, Bile Leakage, Case Report, Pancreatic Cancer, Pleural Effusion

Introduction

Bilothorax, also referred to as cholethorax, is an uncommon condition where bile accumulates in the thoracic cavity, requiring immediate medical attention. This report details a case of bilothorax treated at our facility and reviews related literature to improve understanding and management of this rare condition.

Case Presentation

A 62-year-old man with a history of pancreatic cancer diagnosed in early 2021 presented with obstructive jaundice. Imaging showed an enlarged pancreatic mass and increasing biliary dilatation. An attempt at endoscopic retrograde cholangiopancreatography (ERCP) for drainage was unsuccessful. An 8-French biliary drainage catheter was then inserted percutaneously, with the pigtail end in the biliary confluence.

Six weeks later, the patient arrived at the emergency department experiencing severe dyspnea, chest pain, and tachycardia. CT imaging revealed a large left hydropneumothorax compressing the lung and causing a mediastinal shift (Figure 1). The biliary drain was mispositioned, traversing the right pleural cavity and diaphragm into the liver (Figure 2), leading to bile accumulation in the thoracic cavity.



Figure 1: CT Angio was done to rule out pulmonary embolism. There were no embolism but it showed large right hydropneumothorax compressing the right lung and causing mediastinal shift to the left side.

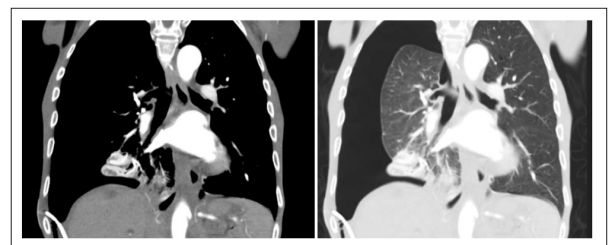


Figure 2: CT showed that the external biliary drain has an abnormal course through the right pleural cavity and the diaphragm into the liver.

A right-sided chest tube was immediately placed, draining about 450 mL of bile-colored fluid (Figure 3) and alleviating the

patient's symptoms. Pleural fluid analysis confirmed bilothorax with a pleural bilirubin to serum bilirubin ratio greater than 1.



Figure 3: Immediate drainage of around 450 mL of bile was evacuated and the drained fluid had a bile colour.

The patient was managed conservatively, with close monitoring, antibiotic therapy, and chest tube output surveillance. Interventional radiology replaced the biliary drainage catheter, positioning the new one in the right biliary duct under ultrasound guidance, and embolized the old tract to stop the bile leak. The patient's respiratory condition improved, and the chest tube was removed five days post-procedure.

Despite the successful management of bilothorax, the patient's condition worsened due to advanced metastatic pancreatic cancer, and he passed away three months later.

Discussion

Bilothorax, or bilious pleural effusion, is a rare condition often resulting from complications involving the gallbladder and biliary system. Causes include direct extension of bilomas, traumatic diaphragmatic perforations, percutaneous biliary procedures, spontaneous gallbladder perforations, and complications from surgeries such as cholecystectomy.

The formation of bilothorax typically involves small perforations in the diaphragm, allowing bile to flow from the peritoneal cavity into the pleural space. In percutaneous biliary interventions, the catheter may create an iatrogenic fistula. Increased pressure in the obstructed biliary system can also force bile into the pleural cavity, facilitated by negative inspiratory pressure.

Symptoms include dyspnea, cough, pleuritic chest pain, and signs of sepsis. Without treatment, respiratory deterioration can occur, potentially leading to acute respiratory distress syndrome (ARDS). Empyema, often involving gastrointestinal pathogens, is a common complication.

Diagnosis is usually straightforward but requires suspicion, particularly after hepatobiliary procedures. Chest radiographs

showing pleural effusion should prompt consideration of bilothorax. Characteristic bile-colored fluid and a pleural bilirubin to serum bilirubin ratio greater than 1 confirm the diagnosis. CT scans may show fistulas between the abdominal and chest cavities, and ERCP can demonstrate bile leakage.

Management typically involves conservative measures such as chest tube drainage, antibiotic therapy, and controlling the biliary source. Invasive surgical interventions may be necessary for persistent leaks or non-responsive empyema. Minimally invasive techniques like percutaneous drainage and ERCP to relieve biliary obstruction, combined with pleural drainage, have been effective. (Table.1)

Preventative measures include intraoperative checks for bile leaks and the use of intra-abdominal drains. Early broad-spectrum antibiotics can prevent infections. Close monitoring, adequate drainage, and control of the biliary source are crucial. Interventional radiology can be instrumental in stopping bile leaks, as seen in this case.

The prognosis of bilothorax depends on the underlying condition. In cases of advanced malignancies, like the metastatic pancreatic cancer in this report, prognosis remains poor despite successful management of bilothorax. Multidisciplinary care involving thoracic surgeons, interventional radiologists, oncologists, and palliative care specialists is essential for optimal patient outcomes.

Table 1: Overview Of Bilothorax - Causes, Symptoms, Diagnosis, and Management

Category	Details
Causes	- Iatrogenic complications (e.g., percutaneous biliary drainage)
	- Traumatic diaphragmatic perforations
	- Direct extension of bilomas through the diaphragm
	- Spontaneous gallbladder perforation
	- Open cholecystectomy
Symptoms	- Biliary peritonitis following jejunal, gastric, or bile duct perforations
	- Severe dyspnea
	- Chest pain
	- Tachycardia
	- Cough
Diagnosis	- Pleuritic chest pain
	- Signs of sepsis
	- Clinical suspicion following hepatobiliary procedures
	- Chest radiographs showing pleural effusion
	- Characteristic bile-colored pleural fluid
	- Pleural bilirubin to serum bilirubin ratio > 1
	- CT scans revealing fistulas between abdominal and chest cavities
	- ERCP demonstrating contrast spillage from the biliary tree into the pleural cavity

Management	Conservative Approach:
	- Chest tube insertion to drain pleural effusion
	- Antibiotic therapy to prevent empyema
	- Control of the biliary source
	- Monitoring vital signs, chest tube output, and regular chest imaging
	Interventional Radiology:
	- Removal and replacement of biliary drainage catheters
	- Embolization of leaking tracts
	Surgical Intervention:
	- Early and aggressive surgical intervention for persistent leaks or non-responsive empyema
Prevention	- Intraoperative checks for bile leaks using dyes or contrast agents
	- Intra-abdominal drains to divert bile externally
	- Early administration of broad-spectrum antibiotics to prevent infections
Prognosis Influencers	- Underlying condition severity (e.g., advanced malignancies)
	- Effective multidisciplinary care involving thoracic surgeons, interventional radiologists, oncologists, and palliative care specialists for comprehensive management

Conclusion

Bilothorax, an uncommon exudative pleural effusion, requires high suspicion for diagnosis. Common causes include iatrogenic hepatobiliary procedures, trauma, and infections. Management involves draining the pleural effusion, administering antibiotics, and addressing biliary obstructions. Early recognition and appropriate treatment are crucial to reduce morbidity and mortality associated with bilothorax.

Declarations: Ethical Approval

Human Subjects: Consent was obtained or waived by all participants in this study.

Consent to publish -also taken

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1. Mohammed Quader Naseer and Mohammed Afshar Alam wrote the main manuscript text.
2. Iqra Hoor collected figures and wrote the cover letter-All authors reviewed the manuscript.

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