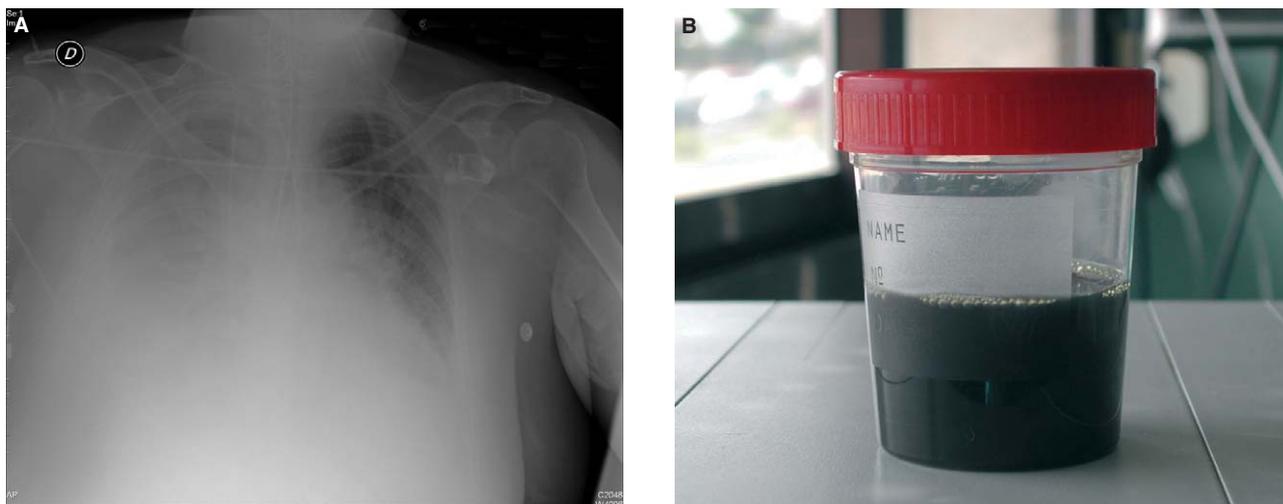


### **Biliothorax Unrelated to Fistula: A Rare Complication in Gallbladder Disease**

**To the Editor:** Biliothorax, or pleural effusion composed of bile, is a rare complication of diseases originating in the biliary tree, secondary to a limited number of clinical entities. We describe the case of a patient with biliothorax not related to pleurobiliary fistula.

A 74-year-old woman with a history of hypertension came to our hospital for pain in the right hypochondrium and mild jaundice. She was diagnosed with acute biliary pancreatitis and admitted to the hospital ward. After her condition deteriorated and she went into shock, we performed an emergency laparotomy, which revealed around 300 mL of free bile in the peritoneal cavity. Cholecystectomy and lithotripsy of the biliary tract performed using a Fogarty balloon catheter obtained a scant amount of bile and a very small gallstone. The diaphragm was not approached during the procedure. Due to postoperative septic shock, the patient was admitted to our unit for inotropic support. Blood gas analysis showed an initial PaO<sub>2</sub> of 70 mm Hg (inspired oxygen fraction, 0.8 + 10) and persistent metabolic acidosis (serum lactate levels as high as 15 mg/dL). Empirical antibiotic therapy with piperacillin-tazobactam was started per protocol and invasive hemodynamic follow-up was performed using a PiCCO catheter. The hemodynamic values were indicative of septic shock, although the preload values were adequate. Chest radiography revealed proper positioning of the right jugular catheter and endotracheal tube, as well as bilateral infiltration. The right hemithorax also showed diffuse opacification indicative of right pleural effusion (Figure A). A sample of 50 mL of greenish fluid (Figure B) obtained by needle thoracentesis was sent for biochemical and microbiologic analysis, which showed a white cell count of 180/μL (40% mononuclear and 60% polymorphonuclear); glucose, 119 mg/dL; proteins, 2.6 g/dL; lactate dehydrogenase, 5134 U/L; amylase, 6136 U/L; urea, 92 mg/dL; creatinine, 2.7 mg/dL; total bilirubin, 5.35 mg/dL; direct bilirubin, 3.6 mg/dL; and positive test for cholesterol. Gram staining showed no microorganisms. A drain was inserted and a total of 1750 mL was collected; the radiologic study confirmed that the effusion had disappeared. Despite the treatment and the measures taken, the patient showed a progressive pattern of sepsis and worsened, with death occurring 36 hours after admission to our unit, due to multiorgan failure.



**Figure. A:** chest radiography at admission to our unit shows diffuse opacification in the right thorax, indicative of right pleural effusion. **B:** macroscopic characteristics of the fluid taken from the pleural cavity.

Biliothorax is rare in both humans and animals. In fact, a MEDLINE search not limited in time that combined the medical subject headings *bile* and *pleural effusion* located only 32 documents, with most in the context of veterinary medicine or biliopleural fistulae secondary to perforated cholecystitis or complications of choledocholithiasis.<sup>1</sup> Gallbladder perforation is known to be present in approximately 5% of patients with acute cholecystitis and is associated with a mortality of 27%.<sup>2</sup> In recent years, the number of cases has increased with the development of techniques involving the biliary tract. McAllister et al<sup>3</sup> reported right pleural effusion in 33% of patients following uncomplicated laparoscopic cholecystectomy. Biliary fistulae have also been described in up to 0.6% of cases after cholecystectomy.<sup>4</sup>

If no fistulae are present, then peritoneal fluid is passing toward the thorax by some other mechanism. Large quantities of air, ascites, or blood in the intra-abdominal region are known to be capable of crossing the diaphragm and penetrating the pleural space, mostly by directly passing through diaphragmatic defects (found on autopsy or during video-assisted thoracoscopy) which could be secondary to muscle stretching in the cases of tension ascites. In these patients, the hydrostatic pressure of the ascites causes the collagen fibers to separate, with evagination and communication with the pleural space.<sup>5</sup> In some cases, the fluid may be crossing the diaphragm via the lymphatics, although this is an apparently secondary mechanism and may

be exacerbated by a pressure change in the thorax.

The diagnosis of biliothorax is based on a pleural-fluid-to-serum bilirubin ratio greater than 1.<sup>6</sup> Pleural bilirubin is being “cleared” rapidly, even during the first 12 hours,<sup>4</sup> and, therefore, early thoracentesis may fail to show bilirubin in pleural fluid or repeat thoracentesis may yield a pleural-fluid-to-serum bilirubin ratio below 1. Biliuous pleural effusion should only be diagnosed after repeated laboratory measurements.

In our case, a key differential aspect was the unusual presentation, in which surgery revealed only a small amount of bile (barely 300 mL) in the abdomen, but an abundant amount in the pleura (almost 2 L). No congenital diaphragmatic defects or biliary fistulae were found and, therefore, bile most probably crossed the diaphragm from the abdomen to the thorax via the lymphatics; otherwise, the absence of bile peritonitis could be due to adhesions of the greater omentum as a result of gallbladder surgery.

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