Spontaneous Left-Sided Cholethorax
A Case Report

Abstract

A 35-year-old man presented with a two day history of epigastric pain and was diagnosed with acute pancreatitis. During his workup, he was found to have a left-sided bilious pleural effusion. Further studies were unrevealing for an anatomic source. A bilious pleural effusion, almost always seen on the right side, is a rarity in clinical medicine and is usually secondary to traumatic injury. We report a case of an even more rare spontaneous, left-sided bilious pleural effusion.

Case

A 35-year-old man presented to the emergency department (ED) with a two day history of increasing epigastric pain. The pain had been present for six months and would wax and wane in intensity over hours to days. It was usually present at bedtime and would resolve by the following day. The pain seemed to worsen with food and improve with water intake. He has a history of hypertriglyceridemia, for which he was taking fenofibrate and ezetimibe. He was not taking any herbal medications or supplements. He has no other significant medical or surgical history, including no history of pancreatitis. He does not use tobacco and drinks alcohol occasionally, usually one or two drinks in social settings. He has no history of illicit drug abuse.

At the time of presentation, the epigastric pain was radiating to his back and was rated as a 10/10 on a pain scale. He had one episode of vomiting earlier in the day. Prior to coming to the ED, he went to his employee health office. He reported that while there, he was diaphoretic. He also complained of a one week history of cough but denied weight loss, anorexia, fever, diarrhea, chest pain or dyspnea.

Physical examination revealed the following: temperature, 98.80°F; pulse, 76 beats/min; respiratory rate, 24 breaths/min; BP, 128/85 mm Hg; and oxygen saturation on room air, 100%. He was in mild distress secondary to the pain. There were no masses or lesions noted on neck examination. His mucous membranes were moist. The heart exam did not reveal any murmurs or gallops. His lungs were clear to auscultation. The abdominal exam was significant for exquisite tenderness in the epigastrium. There was also tenderness in both flanks. Bowel sounds were diminished. Skin exam revealed no abnormalities. Neurologic exam revealed the patient to be lucid, speaking fluently and with no lateralizing motor or sensory deficits.

The WBC count was 13.1 x 103 cells/µL and the hemoglobin was 14.6 g/dL. The serum electrolytes were within normal limits. The serum glucose was elevated at 200 mg/dL. The BUN and serum creatinine were 17 mg/dL and 1.1 mg/dL, respectively. The serum albumin was 5.1 g/dL and his total bilirubin was 3.0 mg/dL. The aspartate aminotransferase was 617 units/L, alanine aminotransferase was 600 units/L and lipase was 7522 mg/dL. Serum cholesterol studies showed a total cholesterol of 233 mg/dL, an HDL of 26 mg/dL, a triglyceride level of 324 mg/dL and an LDL level of 153 mg/dL.

Abdominal radiographs showed no bowel obstruction or any other abnormality. A right upper quadrant abdominal ultrasound did not show evidence of gallstones or acute biliary tract disease. The pancreas was enlarged, as was the liver. There was diffuse fatty infiltration of the liver and a small amount of ascites was present. A computed tomographic scan (CT) of the abdomen suggested mild to moderate pancreatitis with ascites and diffuse fatty infiltration of the liver, and no evidence of biliary obstruction (Figure 1).
Surgical consultation was obtained on the day after admission for evaluation of his acute abdominal pain and elevated lipase and liver enzymes. The surgical team ordered an esophagogastroduodenoscopy, which was unrevealing except for mild erythema in the antrum of the stomach. Recommendations were to aggressively hydrate and provide analgesia. The presumptive diagnosis at this point was pancreatitis secondary to medications for hypertriglyceridemia. Several days into his hospital stay, the patient’s mental status declined and he was transferred to the medical intensive care unit (MICU). During the workup for altered mental status, a chest radiograph demonstrated a left pleural effusion (Figure 2).

Blood work was also repeated at the time of admission to the MICU. Serum electrolytes, lipase, and liver function tests had normalized, even though he continued to have left pleuritic pain, fever and leukocytosis. A diagnostic thoracentesis was performed to rule out a pancreaticopleural fistula or infected sympathetic effusion. The pleural fluid had a dark greenish-brown color. The pleural fluid analysis is outlined in Table 1.
After reviewing the pleural fluid analysis and taking into consideration the greenish-brown color of the fluid, a diagnosis of cholethorax was made after noting the ratio of pleural fluid to serum bilirubin was greater than one. Pleural fluid Gram stain and culture were negative. A magnetic resonance cholangiopancreatography (MRCP) scan was performed to rule out occult gallstone pancreatitis or another anatomic anomaly. No abnormalities were seen on the MRCP (Figure 3).

Fig. 3: Fluoroscopy of the abdomen.

Endoscopic cholangio-pancreatography (ERCP) showed no anatomic abnormality in the visualized biliary system.

A thoracostomy tube was inserted to drain the effusion. The patient improved over the next week and was able to be transferred back to the regular medical floor, where he continued to improve and was discharged home 15 days after admission. Chest X-ray obtained on the day of discharge demonstrated resolution of the cholethorax (Figure 4).
Discussion

Bilious pleural effusion, or cholethorax, is a rare disorder that occurs in only a few clinical situations. Thoracoabdominal trauma,\(^1\) biliary tract obstruction,\(^2\) complications from biliary tract surgery and percutaneous biliary drainage,\(^3\) parasitic infection,\(^4\) subphrenic abscess,\(^5\) and iatrogenic injury\(^6\) have all been associated with cholethorax. Cholethorax can occur spontaneously, but only two cases have been reported in the literature. In one, the patient had a ruptured gallbladder with spillage of gallstones into the pleural space.\(^2\) In the other, the patient had a necrotic gallbladder with a resultant fistulous tract into the right pleural space.\(^3\) To our knowledge, this is the first reported case of a spontaneous left-sided cholethorax in the setting of acute pancreatitis.

Our case is unique in several ways. This is a nontraumatic, nonsurgical cholethorax. Our patient had no biliary procedures, no evidence of liver or gallbladder infection, and no traumatic injury. Additionally, our patient’s effusion was on the left, which is distinctly unusual for a cholethorax. The case reported by Bini\(^6\) had a left pleural effusion, but it was secondary to trauma by a nasogastric tube. What makes this case even more unique is the fact that we were unable to locate a fistulous tract between his biliary system and the pleural space, even with an extensive workup. In the other reported occasions in the medical literature, the etiology of the cholethorax was either obvious or readily discovered. This was not the case with our patient, who developed a spontaneous cholethorax in the setting of acute pancreatitis.

References