Cholecystopleural Fistula in a 71-Year-Old Man with Symptoms of Pneumonia and Pleural Effusion

Puja Gaur, MD
Alexandra L. B. Webb, MD

Cholecystopleural fistula (CPF) is a rare condition in which a tract forms between the gallbladder and the pleura. It may be caused by instrumentation or can be spontaneous due to inflammation of the gallbladder. CPF is a relatively benign condition that can be successfully treated with antibiotics and surgery if recognized early in its pathologic course; otherwise, it can lead to systemic inflammatory response, multiorgan failure, and death. This article presents the case of an elderly patient who presented with symptoms of pneumonia and isolated right-sided pleural effusion and was noted to have air in the gallbladder lumen on sequential imaging. After an appropriate diagnosis and initiation of antibiotic therapy, the patient underwent successful laparoscopic removal of the gallbladder and repair of the fistula tract. This case illustrates the importance of including CPF in the differential diagnosis for patients with isolated right-sided pleural effusion, particularly if it is refractory to medical therapy.

Cholecystopleural fistula (CPF), the formation of a tract between the gallbladder and the pleura, is a rare clinical entity that is often overlooked by clinicians, resulting in a delayed diagnosis. CPF typically presents with symptoms of pneumonia, sometimes including a right-sided pleural effusion. On imaging, a thickened, shrunken gallbladder with or without air in the lumen is often seen. Timely diagnosis followed by prompt medical and surgical intervention can significantly reduce the morbidity and mortality associated with CPF.[1]

CASE PRESENTATION

Initial Presentation and History

A 71-year-old man residing at a nursing home presented to the emergency department with labored, shallow breathing. The patient reported having low-grade fever and nonproductive cough for 1 week prior to presentation. He complained of dyspnea and indicated that he had no pleuritic pain, chest pain, or abdominal pain. His past medical history was significant for coronary artery disease, bipolar disorder, dementia, and vitamin B_{12} deficiency. Findings on additional review of systems were unremarkable.

Physical Examination

On admission, the patient was afebrile and had stable vital signs and oxygen saturation of 91% while breathing room air. He was noted to be disoriented to person, place, time, and situation. He appeared confused and had an ataxic gait. Lung examination revealed right-sided coarse crackles with poor inspiratory effort and decreased breath sounds bilaterally. No wheezes, rhonchi, splinting, egophony, fremitus, hyperresonance, or dullness to percussion were appreciated. The remainder of the examination was unremarkable. The abdominal examination was normal, and neither right upper quadrant pain nor Murphy's sign was elicited.

Laboratory Examination and Radiologic Studies

Initial laboratory testing revealed an elevated white blood cell count of 18,800 cells/μL with a left shift and C-reactive protein level of 75.1 mg/L. Levels of electrolytes, liver enzymes, and cardiac enzymes were normal, as was an electrocardiogram. Chest radiography revealed a right lower lobe infiltrate and opacification, consistent with consolidation. A small right pleural effusion was also noted, suggesting an abscess (**Figure 1**). On chest computed tomography (CT), consolidation of the right middle and lower lobes with air

Dr. Gaur is a resident, Department of General Surgery, Emory University Hospital, Atlanta, GA; Dr. Webb is an assistant professor, Department of General Surgery, Emory University School of Medicine, and Chief, General Surgery Section, Atlanta Veterans Affairs Medical Center, Decatur, GA.

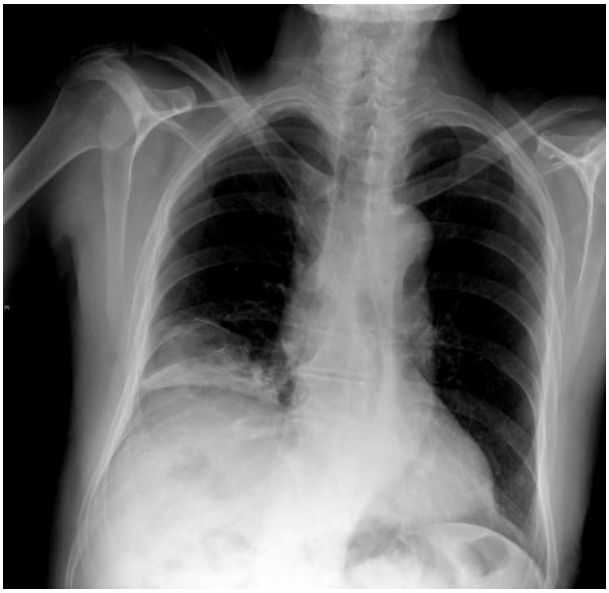

Figure 1. Chest radiograph obtained at admission revealing right-sided consolidation and a small pleural effusion.

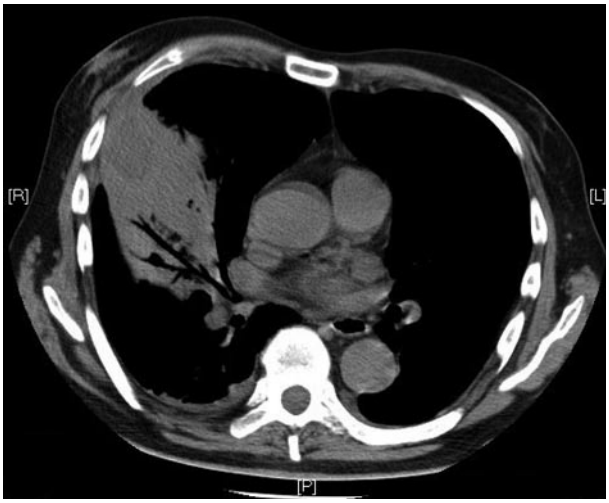

Figure 2. Chest computed tomography scan obtained at admission demonstrating a right-sided pleural effusion with air bronchograms.

bronchograms was evident (**Figure 2**). A moderate-size loculated right pleural effusion was noted to extend into the major and minor fissures. Several pockets containing air-fluid levels with enhanced margins were also seen within the consolidation, suggesting parenchymal abscess and empyema formation. Notably, the left lung was clear. Multiple scattered, small (< 1 cm) mediastinal lymph nodes as well as several benign liver cysts were seen. A 1.6-cm gallstone was noted, but there was no evidence of gallbladder-wall thickening, pericholecystic fluid, or common bile duct dilatation.

Treatment, Clinical Course, and Outcome

On admission, blood, sputum, and urine cultures were sent for analysis, and the patient was started on empiric therapy with intravenous ceftriaxone and azithromycin for community-acquired pneumonia. Despite treatment with intravenous antibiotics, the patient remained febrile, with body temperature up to 100.8°F. On the sixth hospital day, because there had been minimal improvement in the patient's mental status and no growth in cultures, piperacillin/tazobactam was added to the patient's antibiotic regimen. Ceftriaxone and azithromycin were both discontinued the next day. Nutrition was initiated via a Dobhoff tube on hospital day 7. Over the next 3 days, the patient's body temperature returned to normal, and the white blood cell count and C-reactive protein level normalized. By hospital day 10, the patient had high oxygen saturation while breathing room air and was converted from tube-feeding to an oral diet. Over the next 2 weeks, the patient continued to receive intravenous antibiotics and physical therapy. Due to discharge and placement issues, the patient had a prolonged hospital stay. In order to determine the duration of intravenous antibiotics and assess for resolution of pleural effusion, a repeat chest CT scan was obtained on hospital day 31. CT demonstrated resolution of the pleural fluid collection (**Figure 3**), but a 2-cm gallstone was also noted in a thickened, contracted gallbladder without any pericholecystic fluid adjacent to the diaphragm. In addition, air was noted in the lumen of the gallbladder (Figure 3B), suggesting that a CPF was the cause of the patient's pneumonia and empyema. These findings were not appreciated on the initial CT scan, where cholelithiasis was noted without any evidence of cholecystitis or air in the gallbladder lumen.

A surgical consultation was requested, and on hospital day 36 the patient was taken to the operating room for laparoscopic cholecystectomy and perineorrhaphy. A laparoscopic approach was used to minimize operative morbidity. A shrunken, scarred gallbladder was found to be adherent to the diaphragm, thus suggesting the presence of a CPF. Multiple adhesions were lysed, and starting at the apex the gallbladder was carefully dissected away from the diaphragm and its liver bed. After removal of the gallbladder, the fistula tract was identified, which indeed confirmed the presence of CPF. The fistula tract was repaired with two 2-0 Prolene figure-of-eight sutures placed laparoscopically. To our surprise, the patient did not suffer from

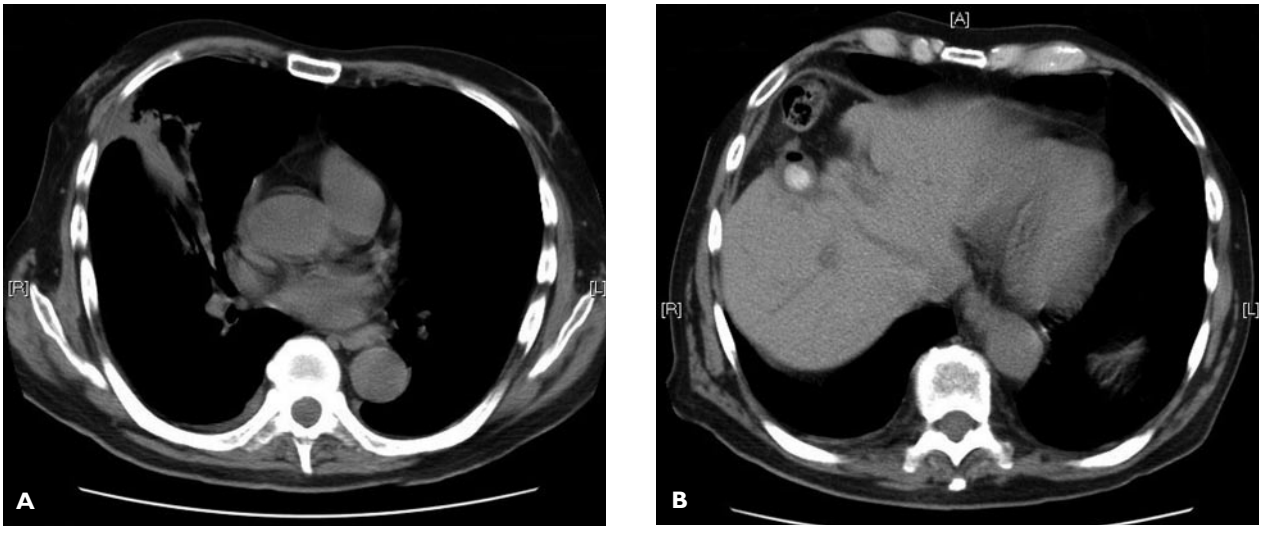

Figure 3. Repeat computed tomography scan on hospital day 31 revealing (**A**) interval decrease in the size of the pleural effusion and organization of the subacute process and (**B**) pneumobilia, suggesting the presence of a cholecystopleural fistula.

pulmonary collapse secondary to pneumoperitoneum. Pathologic examination of the specimen demonstrated suppurative, acute inflammation with fibrosis and perforation, consistent with a fistula tract.

After surgery, the patient was transferred to the intensive care unit because of his comorbidities, and 2 days later he was transferred to a regular hospital room. He resumed an oral diet on postoperative day 2. On postoperative day 14, he was discharged to a skilled nursing facility with good oral intake, significantly improved mental status, and the ability to ambulate with assistance.

CASE DISCUSSION

Community-acquired pneumonia is usually diagnosed in patients who present with fever, cough, and dyspnea with characteristic radiologic findings and laboratory abnormalities.[2] In some cases, however, the symptoms of pneumonia can mask an underlying process that usually is not suspected until after treatment failure.[2] As this case demonstrates, CPF is one such entity that may present with clinical, radiologic, and laboratory characteristics of community-acquired pneumonia. It is a rare condition that may go undiagnosed initially but should be considered in the differential diagnosis of isolated right-sided pleural effusion of unknown source that is refractory to antibiotics. Early diagnosis, management, and surgical intervention in patients with CPF can significantly reduce the associated morbidity and mortality.[1]

CHOLECYSTOPLEURAL FISTULA

Spontaneous CPF usually results from chronic inflammation of the gallbladder wall due to gallstones. Calculous disease of the gallbladder affects approximately 10% to 20% of the adult population in the United States; however, many of these patients remain asymptomatic.[3] Cholecystectomy is indicated for patients who develop symptoms or complications as a result of cholelithiasis.[3] Two of the most common presentations of gallstones are biliary colic and acute cholecystitis. Gallstone pancreatitis, Mirizzi syndrome, choledocholithiasis with or without ascending cholangitis, biliary cirrhosis, hepatic abscess, and gallbladder cancer are less common complications of gallstones but are associated with higher morbidity and mortality. In addition, various fistulae may form between the gallbladder and the abdominal wall, lung, stomach, small or large bowel, or biliary ductal system (cholecystocholedochal fistula), predisposing patients to conditions such as pneumonia, pleural effusion, spontaneous biliothorax, recurrent pancreatitis, cirrhosis, small bowel obstruction (gallstone ileus), and choleretic diarrhea.[3–6]

Etiology

Echinococcal and amebic diseases of the liver account for most cases of CPF in developing countries, while in the Western world trauma and biliary obstruction due to previous biliary surgery are the most common causes of CPF.[1,7] Biliary fistulae are reported to occur in 2% to 4% of all patients with hepatic trauma,

regardless of the mechanism of injury.[1] A search of the literature over the last 20 years revealed reports of CPF in patients who had undergone hepatic surgery, manipulation of the liver with surgical instruments, percutaneous transhepatic procedures, liver biopsy, or radiofrequency ablation and in patients with complete biliary obstruction because of malignancy or benign strictures and subsequent cholangitis.[8–12]

Diagnosis

Suspicion of CPF should be especially high in patients with right-sided pneumonia and pleural effusions who have not had their gallbladder removed and who are refractory to medical therapy. We identified 2 previous case reports in the literature of this type of clinical presentation of spontaneous CPF.[5,6] One was an autopsy report of a CPF that caused sepsis and death in an elderly patient who had presented with a massive right pleural effusion and septic shock.[5] At autopsy, multiple gallstones were recovered from the right pleural space. The second case report described a patient who presented with acute respiratory insufficiency, tachypnea, fever, right-sided pleural effusion, and lung consolidation. CT-guided thoracocentesis yielded a greenish fluid with elevated bilirubin levels, and the presence of a CPF was confirmed at the time of cholecystectomy.[6] Initial evaluation of a patient with suspected CPF should include blood, sputum, and pleural fluid cultures, chest radiography, and gallbladder sonography. CT of the chest and abdomen should be performed to elucidate underlying pathology and identify any pleural or intra-abdominal fluid collections that need percutaneous drainage.

Treatment

Initial treatment should include aggressive hydration, supplemental oxygen, bowel rest, and broad-spectrum antibiotics with coverage of common biliary flora; antibiotic therapy can be tailored once the causative pathogen is identified. Eventually, surgical intervention is warranted for treatment of a CPF if the patient is able to tolerate the procedure. Placement of chest tubes or CT-guided placement of drainage catheters may be necessary to stabilize the patient before surgery. Patients with CPF who are not diagnosed early and treated promptly usually become seriously ill and die of comorbidities associated with this condition. These patients manifest different features of systemic inflammatory response, including acute renal failure, pulmonary insufficiency, cardiac stress, adrenal insufficiency, hypoxia-induced neurologic changes, and sepsis. Ideally, these patients are treated with supportive physiologic measures, such as vasopressors, dialysis, and ventilation, while an attempt is made to control the inciting stimulus with either open or laparoscopic surgery if the patient is able to tolerate the procedure.[7,8]

CONCLUSION

This case report serves to make clinicians aware of CPF, a rare clinical entity that usually presents with symptoms of pneumonia and is therefore often missed, resulting in a delayed diagnosis. CPF should be considered in the differential diagnosis for patients with isolated right-sided pleural effusion, sometimes associated with right upper quadrant pain, who have not had their gallbladder removed. In such patients, the morphology of the gallbladder and whether air is present in the gallbladder lumen should be investigated. Prompt diagnosis of CPF permits timely surgical intervention, which can significantly reduce the morbidity and mortality associated with this condition. **HP**

Corresponding author: Puja Gaur, MD, Emory University Hospital, H120, 1364 Clifton Road, NE, Atlanta, GA 30322. pujagaur@hotmail.com.

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