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Case Report

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Favorable prognosis after pancreatico-pleural fistula and subsequent fungal empyema

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Abstract

Introduction: The diagnosis of pancreatico-pleural fistula is one that requires a high index of clinical suspicion. Furthermore, a subsequent fungal empyema is rare and life-threatening.

Case Presentation: A 48-year-old male presented with a physical exam, laboratory investigations, and imaging findings that were consistent with pancreatico-pleural fistula (PPF) and fungal empyema. The patient was adequately treated with cannulation, sphincterotomy, and stenting of the pancreatic duct via endoscopic retrograde cholangio-pancreatography along with sensitivity-directed antibiotics.

Conclusion: Although this patient presented with a life-threatening combination of PPF and fungal empyema, the patient had a favorable prognosis with the appropriate treatment.

***Keywords*:** pancreatico-pleural fistula; fungal empyema; acute pancreatitis; endoscopic retrograde cholangio-pancreatography.

Abbreviations: *CT: computed tomography; CXR: chest x-ray; ERCP: endoscopic retrograde cholangio-pancreatography; MRCP: magnetic retrograde cholangio-pancreatography; PPF: pancreatico-pleural fistula; VATS: video assisted thoracic surgery.*

Introduction

When a patient presents with respiratory distress, pneumonia, and amylase-rich pleural fluid the initial differential diagnosis is broad. It includes acute pancreatitis, pneumonia, parapneumonic effusion, pulmonary tuberculosis, numerous cancers, and various other conditions [1,2]. Therefore, a high index of suspicion and good clinical judgment are required to narrow down the list to the final diagnosis. Through the use of laboratory, imaging, and cultures our patient was found to have a pancreatico-pleural fistula (PPF) that produced bilateral pleural effusions and multilobar fungal pneumonia. This case report was prepared following the CARE guidelines [3].

Case narrative

A 48-year-old male has a history of recurrent acute alcoholic pancreatitis and five hospitalizations for cases of pneumonia with recurrent pleural effusions in one year prior to presenting. The patient presented to an outpatient pulmonology office for two weeks of persistent dyspnea. He admitted that his breathing gradually got worse and is currently having shortness of breath on rest and exertion. Moreover, he denied any cough, abdominal pain, fevers, chills, or recent triggers (including suspicious food). A lung ultrasound was done in the office that showed a moderate pleural effusion. After transferring to the hospital, he was afebrile and in no acute distress in the emergency room.

The physical exam revealed bibasilar wheezes and rales and the abdominal exam was insignificant. Initial laboratory investigations revealed anemia and elevated white count, lipase, and C-reactive protein. Chest x-ray (CXR) illustrated multilobar pneumonia and bilateral pleural effusions. Computed tomography (CT) angiography of the chest showed a multilobar pneumonia predominantly in the lower lobes and multiloculated moderate right and small left pleural effusions (Figures 1 and 2). A right-sided diagnostic thoracentesis removed 600 milliliters and pleural fluid studies indicated an amylase-rich exudative pleural fluid. Due to the history of recurrent acute pancreatitis and high clinical suspicion, a CT of the abdomen/pelvis with contrast was performed and it revealed findings consistent with mild acute pancreatitis and pancreatic cystic lesions in the head and uncinate process (Figure 2). We initiated broad antibiotic coverage. On day three of admission, a right chest tube was placed for lytic therapy, which consisted of alteplase 10 mg every 12 hours for six dosages.

Endoscopic retrograde cholangio-pancreatography (ERCP) was performed, but the pancreatic duct was unable to be cannulated despite pancreatic sphincterotomy. However, the common bile duct was able to be cannulated (Figure 3). Pleural fluid cultures grew Candida albicans and micafungin was added for initial coverage. After receiving sensitivities, we discontinued the current regimen and started fluconazole. The chest tube was removed on day 10 of admission. Upon stabilization, the patient was discharged home with instructions to continue fluconazole for six weeks, weekly lab monitoring, and repeat ERCP in three weeks to reattempt the cannulation of the pancreatic duct. On the repeat ERCP, the ampulla was cannulated with a 4.4 millimeter sphincterotome, pancreatic duct sphincterotomy was performed, and a four French, five-centimeter pigtail plastic stent was placed in the pancreatic duct (Figure 4). Two weeks later, an abdominal x-ray confirmed that the stent had migrated spontaneously. Since then, the patient has followed up with his primary care physician and has not had a recurrence of his pancreatitis or PPF.

Discussion

The true incidence of PPF is unknown, but it is estimated at 4.5% for patients with pseudocysts and 0.4% for those with acute pancreatitis. A pancreatic duct disruption causes persistent leakage of pancreatic secretions, which forms a fluid collection, known as a pseudocyst. Eventually, the secretions can

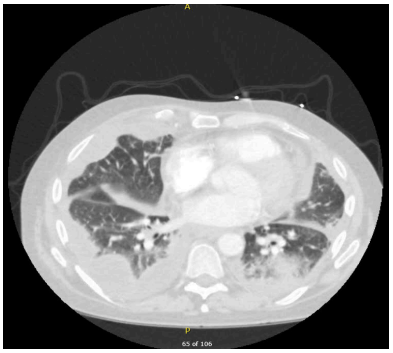

Figure 1: Admission CT angiogram of the chest - coronal view.
1. Multilobar pneumonia, predominantly in the lower lobes.
2. Mediastinal and hilar lymphadenopathy, likely reactive.

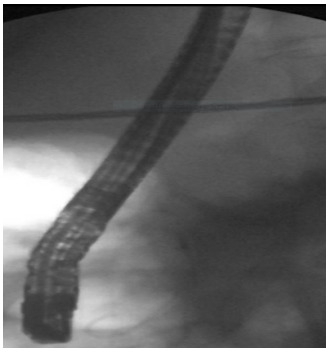

Figure 2: Admission CT angiogram of the chest - transverse view
1. Multiloculated moderate right and small left pleural effusions with pleural thickening and enhancement.
2. Atelectatic and emphysematous changes.

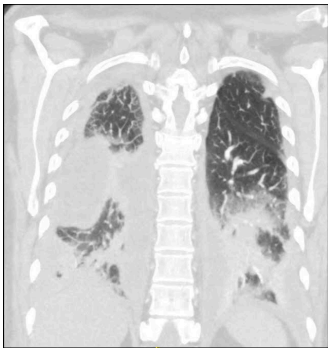

Figure 3: Admission CT Abdomen/Pelvis with contrast - Transverse view
1. Pancreatic edema, peripancreatic fat stranding, and free fluid in the pancreas, particularly the pancreatic body. Consistent with mild acute pancreatitis.
2. Stable pancreatic cystic lesions in the pancreatic head and uncinate process.
3. Possible chronic occlusion of the splenic vein with mild perigastric and perisplenic collaterals.
4. Complex loculated pleural effusions and bilateral airspace diseases.

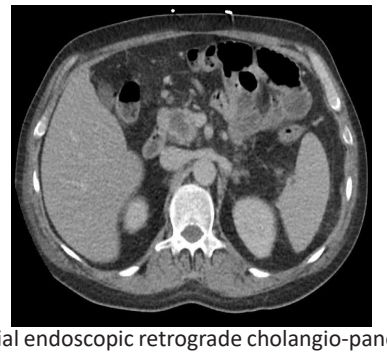

Figure 4: Initial endoscopic retrograde cholangio-pancreatography.
1. Common bile duct was cannulated and was shown to have an opacification.

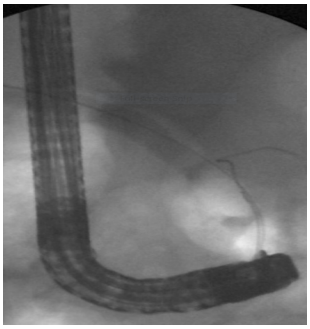

Figure 5: Repeat endoscopic retrograde cholangio-pancreatography
1. Pancreatic ampulla was successfully cannulated.

cause spontaneous erosion and subsequently leak into a neighboring organ or cavity. If the leak occurs anteriorly, it can form a pancreatico-peritoneal fistula, manifesting as ascites; however, a posterior leak causes PPF and pleural effusions [1,2]. Most PPF arises from the head or body of the pancreas [4,5].

Classical PPF patients are middle-aged, alcoholic males with a history of chronic pancreatitis and several acute flares. Other unlikely etiologies are after traumatic or surgical disruption of the pancreatic duct. Typical symptoms are dyspnea, cough, and chest pain; whereas, abdominal pain is less likely and may even be absent. Amongst laboratory findings, pleural fluid amylase is an important diagnostic feature of PPF. Currently, there is no threshold, but it is generally assumed that the amylase level will be over 1,000 U/L. Anecdotally, if the level is higher than 50,000 U/L, then it is invariably due to PPF. Additionally, the pleural fluid protein level is typically greater than 30 g/L. Lipase and albumin are also generally elevated [1,2,6]. On initial imaging with CXR and CT, pleural effusions and pseudocysts are common. Most often, PPF-induced pleural effusions are left-sided, at an incidence of 76%. However, right-sided and bilateral have been noted, at incidence rates of 19% and 14%, respectively. These effusions tend to be recurrent, as they rapidly accumulate and are refractory to repeated thoracentesis. Pseudocysts have been noted in up to 77% of PPF cases and linked as the most common cause of developing PPF, specifically posterior leakage from an incompletely formed or ruptured pseudocyst. [1,6]. Our patient did present with many classic PPF findings. However, some notable differences are that our patient had bilateral pleural effusions and larger on the right was found in this hospital admission or in his history.

Fungal empyema thoracis is a rare, fatal condition. Reports have linked its specific etiology to operations (such as thoracotomy, repeated thoracentesis, and previous tube thoracostomy), gastropleural fistula, and spontaneous esophageal rupture. It commonly develops as a nosocomial infection. Candida and Torulopsis species are often isolated from the pleural effusions. The crude mortality in a 67 patient study was 73% [7]. Additionally, an empyema arising from a PPF has been proposed to result in significant morbidity and mortality [4]. To the best of our knowledge, after researching three research articles engines, this is the first definitive report of a PPF with a subsequent fungal empyema. However, there have been three cases that have shown inconclusive findings of PPF and fungal empyema. One patient was a 43-year-old male who was diagnosed with distal esophago-nodal fistula and PPF. Cultures were negative, but the medical team assumed the patient had a fungal empyema and treated the patient appropriately [8]. A 78-year-old male presented with a left-sided pleural effusion, a left-sided subphrenic collection that was tethered to the pancreatic margin, and pleural fluid cultures growing Candida albicans and Enterococcus faecium. The patient was treated conservatively based on the clinical diagnosis of PPF without confirmation by ERCP and magnetic retrograde cholangio-pancreatography (MRCP) [9]. Finally, a 47-year-old male was discovered to have a small pseudocyst, pleural effusions, and Candida albicans positive pleural fluid. Despite not finding an inciting fistula on video-assisted thoracic surgery (VATS), a clinical diagnosis of PPF was made [10]. For our patient, the ERCP illustrated a PPF and the pleural fluid cultures grew Candida albicans. Additionally, our patient experienced a favorable prognosis with appropriate treatment, which is atypical with this life-threatening disease.

Multiple modalities have been used to diagnose PPF. Currently, CT abdomen, ERCP, and MRCP are most common. The sensitivities of these modalities for PPF diagnosis is 47%, 78%, and 80%, respectively. CT is useful in evaluating the pancreas, ducts, and pseudocysts, but has difficulty in accurately identifying the fistula. Through an ERCP, one can directly visualize the papilla and surroundings and perform endoscopic therapeutic maneuvers. However, it is operator-dependent, invasive, at risk for complications, and cannot evaluate a fistula if a ductal obstruction occurs more proximally than the site of ductal disruption. MRCP, on the other hand, is a non-invasive, non-contrast test that can visualize fistulas - even ones distal to ductal obstructions - pancreatic parenchymal and ductal structural changes, and small intra-/extra-pancreatic pseudocysts [2].

The different treatment options for PPF can be classified into 3 categories: conservative, endoscopic, and surgical. Conservative treatment includes symptomatic thoracentesis or tube thoracostomy. Octreotide, another option, is a long-acting somatostatin analog that inhibits pancreatic secretions and decreases the time for the fistula to close. Although pleural infection is not an approved indication for lytic therapy, a previous randomized controlled trial demonstrated that it improves fluid drainage and decreases the frequency of surgeries and hospital stay duration [11]. Conservative therapy failure is considered refractory pleural effusion or superinfection. Endoscopic techniques via ERCP include pancreatic sphincterotomy, stent placement, balloon dilatation, or stone extraction with or without extracorporeal lithotripsy [4]. The bridging stent is placed in a transampullary manner where it diverts pancreatic secretions away from the fistula and into the duodenum [5]. By creating a path of least resistance for the pancreatic secretions, the stent decreases intra-ductal pressures and eliminates pancreatic pressure gradients, which allows the fistula to rapidly close. Additionally, the stent mechanically blocks the fistula lumen to facilitate healing and restore anatomy [2]. An unknown concern with stent insertion is duration. Most reports cite a 4-12 week duration to allow for optimal healing, but long-term insertion can cause permanent ductal changes that persist even after stent remova [2]. Moreover, many stents occlude after three months of placement [4]. The surgical options include pancreatic resection, enteropancreatic anastomosis to the site of the pancreatic duct leakage, and VATS with pleural debridement and decortication [1,6,12].

Conclusion

This patient presented with a rare and life-threatening combination of PPF and fungal empyema. Moreover, the patient was found to have atypical findings of PPF, including bilateral

pleural effusions and no history of the previous pseudocyst. Despite the elevated mortality rate, the patient had a favorable prognosis after a therapeutic ERCP and sensitivity-directed antibiotics.

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