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A Rare Case of Pancreaticopleural Fistula

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Abstract Pancreaticopleural fistulas are a rare complication of acute or chronic pancreatitis, pancreatic trauma, or complicated pancreatic pseudocyst rupture. It accounts for less than 1% of all cases resulting in pleural effusions and is seen in approximately 7% of patients with chronic pancreatitis and in 14% of patients with pseudocyst. Persistent pancreatic secretions result in erosion of neighboring tissue. Left sided effusions are more common than right sided effusion, accounting up to 76%. Pathophysiology incompletely formed or ruptured pseudocyst. The fistulous tract passes either through the sternocostal triangle, the caval hiatus or directly through the defects aortic or esophageal diaphragmatic orifice. If the pancreatic duct disruption occurs anteriorly, a pancreaticoperitoneal fistula will develop that will manifest as ascites. If the disruption develops posteriorly, pancreatic secretion will flow into retroperitoneum and may dissect through the diaphragm into mediastinum and form a pleural fistula or present as mediastinal pseudocyst. We present a case of a large bilateral pleural effusion secondary to a pancreaticopleural fistula in a patient with multiple risk factors.

Keywords: *Pancreas, Pleural Effusion, Fistula*

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1. Introduction

Pancreaticopleural fistulas are a rare complication of acute or chronic pancreatitis, pancreatic trauma, or complicated pancreatic pseudocyst rupture. It accounts for less than 1% of all cases resulting in pleural effusions and is seen in approximately 7% of patients with chronic pancreatitis and in 14% of patients with pseudocyst. Persistent pancreatic secretions result in erosion of neighboring tissue

Left sided effusions are more common than right sided effusion, accounting up to 76%. Pathophysiology incompletely formed or ruptured pseudocyst. The fistulous tract passes either through the sternocostal triangle, the caval hiatus or directly through the defects aortic or esophageal diaphragmatic orifice. If the pancreatic duct disruption occurs anteriorly, a pancreaticoperitoneal fistula will develop that will manifest as ascites. If the disruption develops posteriorly, pancreatic secretion will flow into retroperitoneum and may dissect through the diaphragm into mediastinum and form a pleural fistula or present as mediastinal pseudocyst. We present a case of a large bilateral pleural effusion secondary to a pancreaticopleural fistula in a patient with multiple risk factors.

2. Case Report

42 year old African American female with a history of Hypertension, Chronic pancreatitis, Pancreatic Pseudocyst, Hx of splenic rupture status post coil-embolization presenting with mild respiratory distress and complaints of progressive worsening dyspnea on exertion for the past month. Patient reported similar complaints 4 months ago when she was last hospitalized and had right sided pleural effusion and had a thoracostomy tube placed at an outside facility. CT Thorax was performed which revealed bilateral pleural effusion with left greater than right with contralateral mediastinal shift and abnormal paraesophageal fluid continuing through the esophageal hiatus into the gastrohepatic ligament. The patient subsequently underwent left sided thoracostomy tube placement with removal of 3.5 liters with improvement in the patient's dyspnea. Pleural fluid analysis revealed dark cola colored fluid consistent with an exudative pattern with high effusion amylase of 10377 U/L, effusion LDH 1560 U/L, and was started on empiric IV antibiotics. On day 3 of admission patient developed acute respiratory distress, interval imaging revealed left apical pneumothorax and re-expansion pulmonary edema. Patient was then taken to the OR where she underwent left thoracotomy with left lower lung lobe wedge resection and decortications for continued increase in left hydropneumothorax. Patient had bilateral thoracostomy tubes which was maintained to wall suction. Magnetic resonance cholangiopancreatography (MRCP) for possible pancreatic pseudocyst rupture as cause of pleural effusion, which was unremarkable. Hospital course was complicated with possible pneumomediastinum with recurrent right pleural effusion. CT thorax was performed for further evaluation, which showed posterior mediastinal fluid collection with air-fluid levels concerning for mediastinitis. Interventional radiology performed a CT guided pleural

effusion biopsy that removed approximately 35 cc of serous fluid that was non-purulent although pleural effusion analysis was consistent with exudative pattern. No surgical intervention was recommended, as there was no clinical evidence of mediastinitis. She was discharged on a 7 day course of antibiotics with outpatient follow up.

3. Discussion

In the above case presentation our patient presented with progressive worsening dyspnea attributed to the bilateral large pleural effusions seen on imaging. Pleural fluid analysis revealed dark cola colored fluid with a exudative pattern and a high effusion amylase of 10377 U/L. Black pleural effusions are extremely rare and have also been reported in association with Aspergillus niger infection, Rhizopus oryzae infection, metastatic melanoma, hemorrhage, charcoal containing empyema, and also seen in amylase rich pleural effusions. Cytology of pleural fluid showed no evidence of malignancy or infectious etiology and patient had a normal procalcitonin level and negative blood cultures. Computed tomography revealed an abnormal paraesophageal fluid continuing through the esophageal hiatus into the gastrohepatic ligament which showed a possible fistulous tract connecting the peritoneum and the pleura. Given the patient's prior history of chronic pancreatitis, history of pseudocyst, ETOH abuse, findings of elevated amylase level in pleural fluid and imaging showing a possible fistulous tract connecting the peritoneum and pleura the diagnosis of pancreaticopleural fistula was entertained. MRCP was performed to rule out pancreatic pseudocyst rupture as a underlying cause. Available medical treatment options include chest tube drainage, pancreatic replacement therapy, nasojejunal tube, use of a somatostatin analogue. endoscopic management such as ERCP with or without pancreatic stenting and IR guided therapy. When medical, endoscopic or IR guided therapy fail, surgical intervention is considered. Our patient had a complicated hospital course after she developed a left apical pneumothorax and re-expansion pulmonary edema for which she underwent left thoracotomy with left lower lung lobe wedge resection and decortications. Further complications during hospitalization included a posterior mediastinal fluid collection for which Interventional radiology performed a CT guided drainage with resolution of mediastinitis on subsequent imaging. Patient clinically improved with both medical and surgical management and patient was discharged with outpatient follow-up. Currently no uniform diagnostic and treatment standards exist for pancreaticopleural fistula, for which this makes the management of this condition a clinical challenge.

4. Conclusion

Pancreaticopleural Fistula is a known but rare complication of chronic pancreatitis. In our literature review, approximately 40% of PPF was diagnosed via MRCP/ERCP and 26% diagnosed via CT abdomen and pelvis imaging. To our knowledge, this is the first case report discussing PPF diagnosed via thoracocentesis showing very high amylase levels, with multiple imaging modalities mentioned above displaying unremarkable contour of the pancreas without abnormality. Further data and literature are limited attributing to the rarity of this disease. It is important to consider multiple differential diagnosis in a patient presenting with pleural effusion for better understanding of disease process which can ultimately affect treatment modalities and therefore outcome in patient.

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