Recurrent Pleural Effusions: An Unusual Presentation of Chronic Pancreatitis

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Abstract
Chronic pancreatitis is a rare cause of recurrent pleural effusion. Here is a case of recurrent massive left pleural effusions due to pancreaticopleural fistula (PPF) secondary to asymptomatic chronic pancreatitis. Pleural fluid analysis was inconclusive. Diagnosis was made by CT chest and abdomen and confirmed by MRCP and MRI. He required surgical intervention although medical management with pancreatic ductal stenting is the first line of treatment.

Introduction
Small, reactive, self-limiting left sided pleural effusion commonly occurs in acute pancreatitis. But recurrent, massive pleural effusion due to pancreaticopleural fistula (PPF) following chronic pancreatitis are rare. Though case reports were published as early as the 1960s, it is extremely unusual.

Case Report
A 49 year old man, a timber merchant, known smoker and alcoholic presented with left sided chest pain and worsening dyspnoea, associated with weight loss of ten to twelve kilograms since two months. One year ago, he had an event of acute left femoropopliteal and tibial thrombosis for which he had undergone grafting of great saphenous vein, following which he was on Acitrom 4 mg with INR at admission of 1.39. There was no other significant past illness except for mild vague abdominal pain for a brief period, eight months back. During these two months of illness, he was evaluated and found to have recurrent left pleural effusions. He had undergone therapeutic thoracocentesis five times without a proper diagnosis. CT chest done elsewhere showed massive left pleural effusion which was drained resulting in two liters of exudative fluid with lymphocyte predominance. Pleural fluid amylase and lipase was marginally elevated, 305IU/L and 685IU/L respectively. Polymerase chain reaction was negative for tuberculosis. At our institute, based on these initial reports, a provisional diagnosis of pulmonary tuberculosis/malignancy was made. Left side ICD was inserted and 2.1 liters of chocolate brown fluid was drained. Pleural fluid analysis was unremarkable except for marginally raised amylase, 320 IU/L. A video assisted thoracoscopy with pleural biopsy after contrast enhanced CT chest and abdomen was planned. CT abdomen was done to rule out any abdominal malignancy as the cause. The CT features were suggestive of subacute pancreatitis with multiple hypodense fluid pockets throughout the parenchyma. There was a pseudocyst in the tail region extending along the diaphragmatic hiatus into the thorax with adjacent small pockets along the lower esophagus, aorta, mediastinal pleura and pericardium. On the left side there was moderate pleural effusion with left lower lobe and lingular consolidation with atelectatic changes. MRCP and MRI Abdomen (Figure 1) were done which were more superior in delineation of structure of PPF and corroborated with CT findings.

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Pancreatogram showed an incomplete pancreatic divisum with disruption of the pancreatic duct just distal to the genu. With contrast, the pancreatic duct in the body and tail was not opacified indicating that the ductal leak was proximal to it (Figure 2). As a therapeutic measure pancreatic duct stenting was attempted through the minor papilla which was unsuccessful. As a definitive measure he underwent distal pancreatectomy with drainage of pseudo-pancreatic cyst. Splenectomy also was done in order to release dense inflammatory adhesions involving the tail of pancreas, spleen, splenic flexure, and distal stomach. Doppler study of his left lower limb done as a preoperative workup showed chronically occluded, fibrotic band in place of bypass graft. His ankle-brachial index was 0.62.

Discussion

PPF presenting as pleural effusion is uncommon, with incidence being 0.4% -1% of cases.\textsuperscript{5,7} Pleural effusions are usually the result of leak from an incompletely formed or ruptured pseudocyst and occasionally due to direct pancreatic duct leak.\textsuperscript{4,5} If the pancreatic duct disruption occurs anteriorly, a pancreaticoperitoneal fistula will develop manifesting as ascites.\textsuperscript{6,7} Pancreatic secretions flow into retroperitoneum if the duct disrupts posteriorly which may dissect into the mediastinum through aortic or oesophageal hiatus. It may either form a direct pleural fistula or present as mediastinal pseudocyst which in turn ruptures into the pleural cavity resulting in pleural fistula.\textsuperscript{6,7} Men aged between 40 and 50 years who have history of chronic alcoholism and develop pancreatitis are susceptible to develop PPF.\textsuperscript{5,7} About half of the patients do not have history of pancreatitis. Trauma contributes for 0.5% of the cases.\textsuperscript{7} The clinical manifestations are often misleading as pulmonary symptoms predominate. Symptoms are usually associated with significant pleural effusion with dyspnea being the most common.\textsuperscript{7,8} The average duration of symptoms is 5.6 weeks.\textsuperscript{5,7} Pleural effusion is predominately left sided; are reported to account for 76% of cases.\textsuperscript{5,7} However, right-sided and bilateral effusion occurs in 19% and 14% of patient’s, respectively.\textsuperscript{1} Pleural effusion of this nature tends to be large and recurrent despite repeated thoracentesis.\textsuperscript{5,7} Superinfection is a major complication and contributes to significant morbidity and mortality in these patients. The time to diagnosis is frequently delayed and is reported to range from 12 to 49 days.\textsuperscript{5} A suspicion of PPF should arise if the clinical picture is suggestive and the pleural fluid analysis shows an extremely elevated amylase level (normal <150 IU/L), lipase, and albumin content (>3 g/dL)\textsuperscript{1,6} (see algorithm). In our case, on the contrary, serial dilutions of the pleural fluid did not show any significant rise in amylase. The serum amylase is usually mildly elevated but not invariably and is thought to be partly secondary to reabsorption of amylase from pleural surfaces.\textsuperscript{5,7} Chest radiography is the first line of investigation, but a CT scan of the chest and abdomen is valuable in the diagnosis as it proved in our case.\textsuperscript{6,7} ERCP leads to diagnosis in 80% of cases and demonstrates the fistulous tract in 59% to 74% of the cases.\textsuperscript{5,9} Magnetic resonance cholangiopancreatography is an important aid to demonstrate the pancreatic pathology and the fistula.\textsuperscript{6,8} It is a noninvasive alternative to ERCP, visualizes the duct beyond the strictures where ERCP fails which was true in our patient.

Current treatment methods include medical management with octreotide and thoracentesis, ERCP with or without endoscopic pancreatic stent placement, and surgery.\textsuperscript{9,10} Medical treatment involves the administration of somatostatin analogues to reduce stimulation of pancreatic exocrine secretions,
Conclusions

PPF is difficult to diagnose and at times difficult to treat. They require a high index of clinical suspicion to diagnose, particularly in the setting of recurrent pleural effusions with coexisting history of pancreatitis or alcohol abuse. Pleural fluid amylase testing will avoid delayed diagnosis. The first line of treatment should be chest drain, octreotide therapy, and ERCP with an attempt at pancreatic stent insertion which is shown to shorten the duration of hospital stay. Surgical intervention is the second line of treatment with an appreciable morbidity and mortality.

References