Case report

Massive pleural effusion from pancreaticopleural fistula: Successfully managed with endoscopic pancreatic duct stenting

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Abstract
We report a case of massive left sided hemorrhagic effusion secondary to pancreaticopleural fistula (PPF) in a young, previously healthy female of 17 years. High pleural fluid amylase levels prompted us for evaluation of this uncommon cause. Computed tomography (CT) of thorax and abdomen and magnetic resonance cholangio-pancreatography (MRCP) confirmed the diagnosis of PPF. Patient was managed with intercostal drainage of pleural effusion and therapeutic endoscopic retrograde cholangio-pancreatography (ERCP) with stenting of the pancreatic duct. Patient recovered completely and is doing well after one year of follow up without any recurrence of pleural effusion. Thus, high index of suspicion, and measurement of pleural fluid amylase, especially in large and or recurrent (left sided) pleural effusions, will help in early and definitive diagnosis of PPF. Above approach with therapeutic ERCP would cure this condition with less morbidity. Advances in the latter techniques dramatically reduced the need for surgical interventions and prolonged hospital stay with conservative treatment approach.

Key words: ERCP, MRCP, Pancreaticopleural fistula, Pancreatic duct stenting, Pleural effusion

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Pleural effusion from pancreaticopleural fistula (PPF) is a rare complication of chronic pancreatitis, accounting for less than 1 percent of patients1. It is mostly associated with chronic pancreatitis, even though it may occur from other secondary causes like acute pancreatitis, iatrogenic (ERCP-induced) or surgical pancreatectomy2,3. PPF is defined as the communication between pancreatic duct and pleural cavity leading to seepage of pancreatic secretions to form pleural effusion. This is one diagnosis that should be considered in a patient with a chronic undiagnosed pleural effusion. The diagnosis is virtually established if the amylase level is greater than 1000 U/L, avoiding the need for extensive pulmonary evaluation4.

Advances in endoscopic retrograde cholangio-pancreatography (ERCP) techniques have dramatically changed the treatment of PPF. It can help in diagnosis and stent placement. Thus, ERCP with stenting of the pancreatic duct may be suggested as the first line therapy in suitable patients which would enable early recovery and short duration of inpatient treatment without further recurrences3,5.
Case report

A 17 year old female who was having recurrent left sided chest pain and cough for over 3 months presented with complaints of gradually progressive shortness of breath and increased non-productive cough for the past 20 days. Prior to this consultation, patient received only symptomatic treatment elsewhere. She gave history of mild abdominal pain twice during the past three months. The patient has no known pre-morbidities, is a non-smoker and does not consume alcohol. There was no history of trauma or any relevant predisposing causes detected for her present condition during evaluation.

Physical examination revealed signs consistent with massive left pleural effusion. Chest x-ray revealed left sided pleural effusion with contralateral mediastinal shift.

Thoracocentesis revealed hemorrhagic pleural effusion. The fluid was exudative with 60% lymphocytes, plenty of red blood cells (RBCs) and had an Adenosine deaminase (ADA) of 26U/L. Pleural fluid and cell-block analysis was negative for malignant cells and revealed signs of chronic inflammation. Pleural fluid amylase levels were 1117 U/L. Serum amylase and lipase were 249 and 125 U/L respectively.

Contrast enhanced CT (CECT) of thorax and abdomen revealed left sided pleural effusion and signs of chronic pancreatitis. The main pancreatic duct was prominent in both head and body regions. On contrast administration, heterogenous enhancement is noted in the region of the tail with ill-defined fluid collection measuring 17x11 mm abutting the tail with suggestion of communication with pancreatic duct. A large non-enhancing hypodense lesion was also noted in the spleen superomedially suggestive of a pseudocyst (Fig 1).

An intercostal drain (ICD) was inserted into the left pleural cavity and the patient was managed with antibiotics and was given low fat diet, pending gastroenterologist’s advice.

MRCP (Fig 2 & 3) showed moderate to severe chronic pancreatitis with a PPF at the tail region associated with left pleural collection and 5.5 x 3.5cm intrasplenic pseudocyst. The patient underwent ERCP which showed pancreatic ductal leak from tail...
area. A pancreatic stent was placed. Patient tolerated the procedure well and was put under close observation. Octreotide and parenteral nutrition were not followed with after endoscopic therapy. The ICD was clamped and removed after 48 hours of observation. Patient was on antibiotics for two weeks. Repeat chest x-ray revealed significantly decreased pleural effusion. Patient was closely followed up and pancreatic stent was removed after four months. Pleural effusion had not recurred during the follow up of one year.

Discussion

Although most of the patients with pancreatic pleural effusion are alcoholics, only 50% of them have a clinical history and signs of previous pancreatitis.

Even though thoracic complications of chronic pancreatitis are very rare, they present with recurrent episodes of dyspnea and cough often not preceded by any abdominal symptoms. Pancreatic pleural effusion is an unusual complication of chronic pancreatitis and occurs only in 0.4% of patients with chronic pancreatitis and 4.5% of patients with pancreatic pseudocysts. It may occur through two mechanisms: directly via the fistulous track between main pancreatic duct and the pleural cavity or by direct extension of pancreatic pseudocyst through mediastinum.

Uchiyama et al in a review of 113 cases in Japan reported dyspnea, abdominal pain, cough and chest pain as the main symptoms in patients with pleural effusion from PPF. Pleural effusions with high amylase can also be found in other conditions such as pleural effusion resulting from acute pancreatitis, lymphomas, leukemias and tuberculosis.

There are two types of pleural effusion in pancreatic pathologies. The first is usually a small left-sided effusion and characterized by normal amylase activity (below 100 U/L) and low protein concentration (below 3 g/dL); this type is associated with acute pancreatitis and resolves during recovery. The second type of pleural effusion is related to the presence of PPF in the course of chronic or recurrent pancreatitis. These effusions are usually large, unilateral, recurrent and contains high level of amylase, with a range of pleural fluid amylase: 400-446,600 U/L, and protein above 3g/dl in one case series reported by Ali et al. Virtually no other disease entities produce such dramatic elevations of the pleural fluid amylase. These two forms of pleural effusion should be clinically recognized in view of their different complication rates, progress and approach to treatment.

Although some authors argue that despite having a low sensitivity of 47%, abdominal CT scans should be applied to diagnose features such as chronic pancreatitis, visualization of PPF and pancreatic pseudocysts, others argue that ERCP should be done earlier on in patients with high index of suspicion. Some series show confirmation of the diagnosis in 78% of cases with its use. CT scan should be the method of choice in cases where there is inability to demonstrate fistulas in locations where the breakpoint is in distal duct, as noted in our patient. MRCP which has 80% sensitivity was effective for the diagnosis of fistula in several studies and is a noninvasive imaging method to visualize PPF. Therefore, MRCP can be considered the initial modality of choice to diagnose PPF, when the facility is available. MRCP will also guide subsequent management plans for PPF which depends on the pancreatic duct morphology seen on this imaging modality. It will help decide which patient is likely to benefit from early endoscopic therapy.

In view of rarity of this condition, there are no standard guidelines as of now to evaluate the pros and cons of medical and surgical therapies. The main aim of medical treatment is to effectively reduce stimulation of pancreatic exocrine secretions. Measures like prohibition of oral intake, nasogastric tube insertion and total parenteral nutrition (TPN) which were used in the past are no longer recommended. Medical treatment now constitutes thoracocentesis and or tube thoracostomy and administration of somatostatin analogues such as octreotide.

Duration of conservative management varies from 2-4 weeks. In one study, chest tube drainage, which provides symptomatic relief was given for 6-24 days and octreotide was continued for 2.5-6 months. But, it should be noted that the reduction in pancreatic secretion is less important than the restoration of anatomic continuity of the pancreatic duct.

Advances in endoscopic therapies have dramatically changed the treatment approach of PPFs. ERCP can help identify the fistulous track and simultaneously enable stent placement in the pancreatic duct. ERCP with endoscopic pancreatic stenting has dramatically reduced the need for surgical intervention. Main aim of treatment with stent is to achieve drainage of ducts with fistulae (drainage into duodenum) in short term and drainage of stenosed pancreatic duct in long term. Owing to the rarity of PPF, the optimal duration for which the stent should be left in situ is unknown. Dhebri and Ferran in their review describe the time period for the ERCP therapy between 4-12 weeks. In our patient we
could successfully remove the stent in the fourth month. Success rates of ERCP with stent procedures were noted to be 96.4% in one study.

Indications for surgery are failure of medical and endoscopic therapy, large volume pseudocyst, persistent or recurrent effusions and multiple strictures or complete duct disruption.

It is said that the effectiveness of conservative treatment ranges from 30 to 60% in some series and 0% to 33% in others. Surgical treatment should be the second-line of option with attendant morbidity and mortality.

**Conclusions**

Pleural effusion with a PPF is a rare condition that is often elusive to early diagnosis. It is important to consider this entity since the condition is completely curable with appropriate and timely intervention. A high index of suspicion is required to clinch a diagnosis, especially in patients with history of large and or recurrent pleural effusion with co-existing history of pancreatitis or alcoholism. Pleural fluid amylase estimation, especially in left-sided effusions, even without any suggestive history of pancreatitis, will guide in further evaluation of PPF. Noninvasive imaging such as MRCP would help in better visualization of pancreatic duct anatomy. This will help in management decisions like conservative versus intervention therapy. ERCP with stenting of pancreatic duct is a very effective treatment modality. This approach may obviate the need for surgery or prolonged conservative medical treatment and hospitalization with attendant risk of failure.

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**Conflict of interest:** None

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