Pancreatico-pleural fistula: A rare cause of massive right-sided pleural effusion

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Abstract Pancreatico-pleural fistula is a rare entity and it accounts for a small percentage of cases of pleural effusion. We present a case of massive right sided pleural effusion that was attributable to pancreatico-pleural fistula in a 46 year old male alcoholic patient who had dyspnea, terepopnea and dull aching right sided chest pain. ERCP showed a fistulous track between the main pancreatic duct and right hemithorax. This case is very rare as the pleural effusion due to pancreatico-pleural fistula is usually left-sided in up to two thirds of cases.

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Introduction

Pancreatic pleural effusion is a rare condition and is defined as significant fluid accumulation in the pleural space with high amylase content resulting from the leakage of fluid from the pancreatic duct after its disruption [1].

Pancreatic fistula is a complication of acute or chronic pancreatitis. It may follow surgical pancreatic resections, percutaneous drainage for a pseudocyst or after abdominal trauma [2]. This pancreatic fistula may communicate with the peritoneal cavity causing pancreatic ascites or with the pleural cavity causing pancreatico-pleural fistula. Both of them have the same pathogenesis which, as mentioned, is the disruption of the main pancreatic duct, leading to the leakage of pancreatic fluid [3,4].

Pancreatico-pleural fistula was first described in the late 1960s and early 1970s after publishing case reports [5]. It has been predominantly seen in alcoholic middle-aged men who usually present with dyspnea and a left-sided pleural effusion [6]. Rarely, it can present with right-sided or bilateral pleural effusions [7].

Case report

Our patient was a 46 year old male who works as a mechanic. He is a heavy smoker with a smoking index of thirty pack-years. He drinks alcohol regularly for the last 25 years.

The patient was referred to our hospital because he had dyspnea associated with dry cough for the last month that increased when lying on his left side. He also had right sided dull aching chest pain. The patient had no wheeze, no expectoration, and no hemoptysis. He had no fever, no weight loss and no appetite change.

Past medical history showed that the patient was healthy and athletic with no diabetes, hypertension, or chest troubles. He had no history of allergy or TB. He used to receive oral antacids with some antispasmodic and analgesic drugs from his family physician for the last 2 years because of epigastric pain and dyspepsia.
General examination was unremarkable apart from abdominal distension with resonance on percussion and no organomegaly. Chest examination showed breath rate of 24 breaths per minute, right sided stony dullness with overlying nearly absent breath sounds. Trachea was shifted to the left side.

Chest X ray showed total opacification of right hemithorax with massive right-sided pleural effusion and mediastinal shift to the left side. CT chest showed massive right sided pleural effusion with collapsed right lung (Fig. 1A).

Tuberculin test was negative, CBC was normal, and ESR was 35 mm/h. Rheumatoid factor, ANA, ANCA, anti double strand DNA and other collagen disease markers were all negative.

Pleural fluid was serous and analysis showed that it was exudate with normal pH and normal glucose content. Adenosine deaminase (ADA) was 10 U/L. Pathologically it was negative for malignancy.

Pleural fluid was rapidly re-accumulating after repeated aspirations so, chest tube was inserted. After that, the patient became less dyspneic but pleural fluid drainage through the tube was as much as 400–600 ml per day for more than 2 weeks. The patient was prepared for thoracoscopy to visualize the pleura and take biopsies if applicable. At that time abdominal ultrasound showed fatty normal sized liver, normal spleen, colonic distension, right sided pleural effusion, and no ascites. The radiologist commented that there was an abnormal echogenicity in the area of pancreas with no evidence of cyst formation. So, revision of the upper abdominal cuts of CT chest was done and showed evidence of chronic pancreatitis.

Serum amylase was high (750 U/L) and pleural fluid amylase was 1350 U/L.

Thoracoscopy was postponed and the patient was sent to do abdominal MRI that showed evidence of chronic pancreatitis, and Magnetic resonance cholangiopancreatography (MRCP) showed mildly dilated main pancreatic duct so the patient was referred to gastroenterologist to do Endoscopic retrograde cholangiopancreatography (ERCP).

ERCP showed dilatation of main pancreatic duct and a fistulous track extending from the pancreatic duct to right hemithorax (Fig. 1B). On the same setting a stent has been placed in the main pancreatic duct at the site of leakage. Fig. 1C shows the stent in place with abdominal CT done later.

The pleural fluid drainage from the tube decreased markedly after ERCP and stent placement and the chest tube was removed on the 15th day after ERCP.

Discussion

Pleural effusion is a diagnostic dilemma and has a very big list of differential diagnosis. Pleural effusion due to pancreaticopleural fistula is very rare as it accounts for less than 1% of cases, while pleural effusion can be seen in 3–7% of patients with pancreatitis [8,9].

Usually the diagnosis of pancreatico-pleural fistula is delayed unless there is a high index of suspicion especially in alcoholic patients or known cases of chronic pancreatitis along with pleural effusion. The delay in diagnosis usually ranged from 12 to 49 days [10]. In our case the presence of massive right sided effusion added to the difficulty of diagnosis because pancreatico-pleural fistula usually comes with left sided effusion. Diagnosis of our case was also difficult due to the predominance of pleuro-pulmonary symptoms more than abdominal symptoms.

We could not diagnose the case by using Magnetic resonance cholangiopancreatography (MRCP) although it is reported to be particularly useful in demonstrating the pancreatic pathology and the fistula [9]. In our patient, only the MRI and MRCP showed evidence of chronic pancreatitis with mild dilatation of main pancreatic duct.

Endoscopic retrograde cholangiopancreatography (ERCP) is essential in both the diagnosis and management of pancreatico-pleural fistula. It not only shows the site of leak but also reveals the ductal morphology [11]. In our patient, the diagnosis was confirmed with ERCP and in the same setting treatment was done by placing a stent in the main pancreatic duct at the site of leakage which was very effective and the patient improved dramatically.

Conservative treatment with pancreatic duct stenting and/or octreotide has been successful in 31–45% of cases, while surgery is curative in 80–90% of cases but with a high rate of mortality (up to 10%) [3,11]. Our case has been treated successfully with stent in the main pancreatic duct that inhibited the leakage.

Amylase-rich pleural effusion can be detected in cases of malignancy either primary (mesothelioma) or metastatic (especially adenocarcinoma), esophageal rupture and pancreatic-related conditions including, acute and chronic pancreatitis and pancreatico-pleural fistula [12]. Lymphoma, leukemia, liver cirrhosis, hydronephrosis, and pulmonary tuberculosis can also account for elevated pleural fluid levels of amylase.

Figure 1 CT chest showing massive right sided pleural effusion with collapsed right lung (A). ERCP showing mild dilated pancreatic duct and a fistulous tract from it to right hemithorax (B). CT abdomen showing the stent in the main pancreatic duct after ERCP (C).
however, very high levels of amylase in pleural fluid can be explained only by the rupture of pancreatic pseudocyst into pleural cavity or the presence of pancreatico-pleural fistula [13]. In our patient, the pleural fluid amylase was very high and even exceeded the serum amylase which was also markedly elevated.

Conclusion

Although uncommon, pancreatico-pleural fistula can present with massive right-sided pleural effusion. Also, pancreatico-pleural fistula should be put in the differential diagnosis of pleural effusion especially in middle aged men with a history of alcohol consumption.

Conflicts of interest

None.

References