

Pancreatico-Pleural Fistula – from Diagnosis to Management. A Case Report

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ABSTRACT

Pancreatic pseudocysts are frequent complications of both acute and chronic pancreatitis. By contrast, pancreatico-pleural fistula is rare. Here we report a case of massive pleural effusion secondary to a fistula in the left hemi-diaphragm, between a pancreatic pseudocyst and the left pleura, in a patient with a right kidney tumor and bilateral massive pulmonary thromboembolism. This fistula developed after several episodes of un-investigated acute pancreatitis. The pleural effusion was treated by three thoracocenteses, without recurrence.

Key words: pancreatico-pleural fistula – pancreatic pseudocyst – pleural effusion.

Abbreviations: CT: computer tomography; TTE: transthoracic echocardiography.

INTRODUCTION

The first case of pleural effusion in pancreatitis was reported by Phillips (1901), followed by others until 1942, when Werner described the unusual nature of these effusions and the significance of pleural fluid amylase [1].

The pancreatic pseudocyst is a frequent complication of both acute and chronic pancreatitis. It is defined as a localized liquid collection with a non-epithelial wall (fibrous and granular tissue) containing high levels of pancreatic enzymes. A posterior rupture could cause a communication in the left diaphragm with the formation of a fistula and pleural effusion. Patient's symptoms may range from asymptomatic to progressively installed severe dyspnea, requiring thoracocentesis. Pancreatico-pleural fistula is an extremely

rare complication of acute or chronic pancreatitis, usually being treated by surgery [2].

The simultaneous finding of a pancreatic pseudocyst, a massive left pleural effusion, bilateral massive pulmonary thromboembolism and a malignant tumor could complicate the diagnosis route, making it necessary to accurately determine the etiology of the pleural effusion: neoplastic or pancreatic.

CASE REPORT

A 66-year-old male patient was hospitalized in the Emergency Department with transient severe dyspnea. The patient had presented several episodes of major abdominal pain 6 months before, for which he did not seek medical attention. Upon admission, the patient had a mildly influenced general condition, sinus tachycardia, without hemodynamic instability. He was underweight with a body mass index of 17.6 kg/m². No abnormal pulmonary sounds on auscultation were detected. Blood tests were within normal range. Thoracic computed tomography (CT) showed bilateral pulmonary thromboembolism (Fig. 1). The electrocardiogram showed sinus tachycardia; transthoracic echocardiography (TTE) was without significant structural or functional changes.

The patient's evolution was uneventful under anticoagulation therapy with unfractionated heparin during the first week. After a transient aggravation of dyspnea, repeated TTE showed no significant new cardiac structural abnormality, with a good

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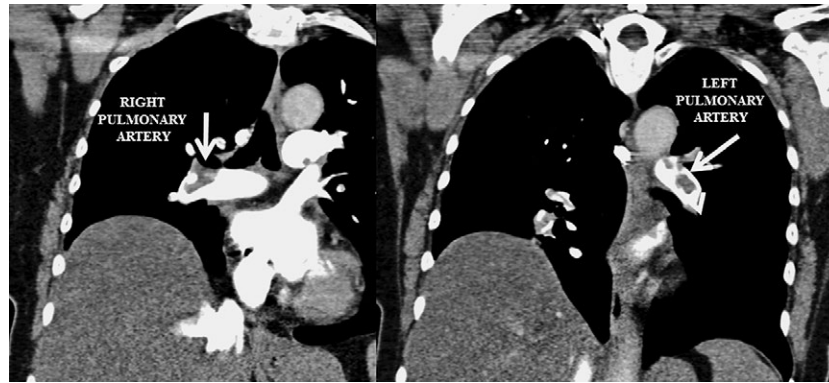


Fig. 1. Bilateral pulmonary embolism on thoracic computer tomography (arrows).

systolic left ventricular function and a large left pleural effusion, confirmed by chest X-ray (Fig. 2). A thoraco-abdominal-pelvic CT showed that the pulmonary arteries thrombi had disappeared, and revealed the large left pleural effusion with a maximum thickness of 110 mm and passive left lower lung lobe atelectasis. A milimetric discontinuity of the left posterior diaphragm suggested a fistula between the intra-abdominal collection and the pleural cavity (Fig. 3). The abdominal scan images revealed different collections suggestive of repeated episodes of acute pancreatitis: a fluid collection located in the gastro-colic segment; an extraperitoneal pararenal left anterior

collection that extended into the left perirenal space, in contact with the lower face of the left hemi-diaphragm muscles, a voluminous pancreatic pseudo-cyst (40x59x85mm). A small-sized homogeneously structured pancreas was detected, without calcifications; the Wirsung duct had 3 mm caliber in the body-caudal segment. The CT also depicted a solid mid-kidney nodule with a diameter of 22 mm. The vascular pattern of enhancing in the arterial phase and wash-out in the venous phase was suggestive for renal carcinoma. No local extension or metastases were revealed (Fig. 4).

The analysis of the pleural liquid revealed a hemorrhagic exudate fluid with elevated levels of amylase (20,087 u/L) and lipase (34,138 u/L) and the absence of neoplastic cells. After three repeated thoracocenteses, there was no need for surgical closure of the pleuro-pancreatic fistula. Blood tests revealed elevated levels of amylase (767 u/L) and lipase (335 u/L) and a non-specific inflammatory syndrome.

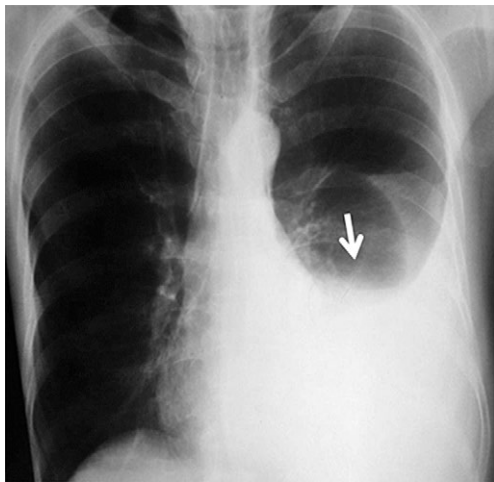


Fig. 2. Left pleural effusion on chest radiography (arrow).



Fig. 4. Right kidney tumour on computer tomography (arrow).

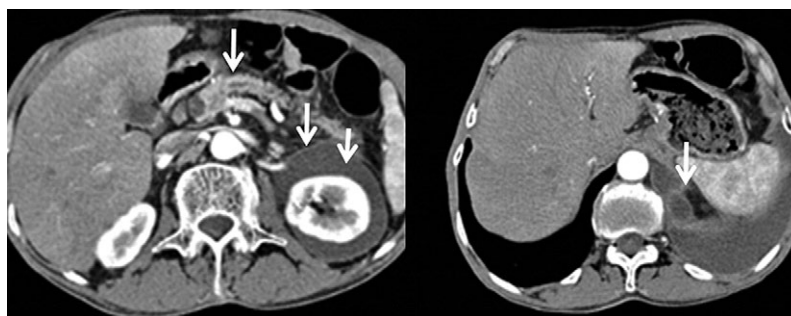


Fig. 3. Pancreatic pseudocyst and pararenal left anterior collection that extends into the left perirenal space surrounding the upper 2/3 of the kidney (left side) and fistula in the left hemi-diaphragm between the pancreatic pseudocyst and the left pleura (right side) on computer tomography.

After three weeks, the patient was referred to the Regional Institute of Oncology for surgical treatment of the right kidney carcinoma (T1N1Mx stage). The postoperative period was uneventful. The histology revealed type 2 papillary carcinoma. But one week after discharge, the patient had a fulminant stroke and died at home. Unfortunately, an autopsy was not performed.

DISCUSSION

Pancreatico-pleural fistula occurs in 0.4 and 4.5% of patients with chronic pancreatitis and is more rare than pancreatic ascites [1-3]. Pancreatico-pleural fistula allows the pancreatic juice to enter into the pleural cavity following post-traumatic or post-surgical rupture of the pancreatic duct [4-8]. Another cause can be the presence of a pancreatic pseudocyst communicating with the pleural cavity through a fistula [6, 7]. Pleural effusion is large, recurrent and refractory to repeated thoracentesis [4, 7]. It is frequently unilateral, predominantly on the left side and rarely on the right side (20%). It is unusual to be bilateral (15%).

Amylase-rich pleural effusion can be produced by many causes, including: neoplasia (either primary as mesothelioma or metastatic especially adenocarcinoma), esophageal rupture, pulmonary embolism, tuberculosis, lymphoma, leukemia, liver cirrhosis, hydronephrosis etc. [9].

Our patient was diagnosed with renal carcinoma and repeated episodes of acute pancreatitis. To establish the etiology, it was important to analyse the pleural effusion. The pleural fluid revealed the presence of an exudate fluid with very high levels of amylase, and no tumor cells. In addition, the patient had very high levels of serum amylase and serum lipase. Usually, a level of amylase >10,000 u/L in the pleural fluid can be explained only by the rupture of pancreatic pseudocyst into pleural cavity or the presence of a pancreatico-pleural fistula [9], as in this case.

In pancreatico-pleural fistulas, the cardiovascular and respiratory symptoms are more frequent than the gastrointestinal symptoms [7]. This patient presented respiratory symptoms (transient dyspnea), without any abdominal symptoms during hospitalisation.

A variety of imaging methods can be used to visualize a fistula: magnetic resonance cholangiopancreatography, computer-tomography [4, 7, 8]. In our patient, the diagnosis was established by a CT scan.

The available therapeutic options include: conservative management, endoscopic treatment (via endoscopic retrograde cholangiopancreatography) and surgery. The medical management includes administration of octreotide to inhibit the pancreatic exocrine secretions, and thoracentesis [4, 10]. Endoscopic treatment options are: sphincterotomy, stone extraction from the pancreatic duct or dilation of the Wirsung duct stricture. The surgical intervention is used when the medical treatment fails or in cases requiring immediate intervention [4].

In this case, the patient had a milimetric pancreatico-pleural fistula with spontaneous closure after thoracentesis. It is estimated that up to 70% of external pancreatic fistulas may heal with conservative management [6]. Somatostatin

analogues administration could promote the closure of a pancreatic fistula by decreasing the volume of the fistulous tract output [7].

Type 2 papillary renal cell carcinoma is a high-grade tumor, associated in some cases with venous invasion and thromboembolism [11]. Therefore pulmonary thromboembolism is not unusual in these patients. In our patient, kidney carcinoma was unmasked by bilateral thromboembolism. Papillary renal cell carcinoma is also associated with malignant pleural effusion [12]. However, in this case pleural effusion was not related with the kidney carcinoma.

CONCLUSIONS

This clinical case presented a complex pathology: repeated episodes of acute pancreatitis, complicated with a pancreatico-pleural fistula, and a massive pulmonary thromboembolism. Examinations also detected a previously unknown papillary renal cell carcinoma. Evaluation of the etiology of a pleural effusion must be very careful if there are associated pathologies.

Conflicts of interest: No conflict to declare.

Authors' contributions: F.M., L.N. and R.V. performed the acquisition of data and wrote the paper; C.M., C.C.P. and V.S. contributed to the acquisition and interpretation of data; V.S. and V.L.D. made critical revisions of the manuscript for important intellectual content.

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