CASE REPORT

Primary presentation of chronic calcific pancreatitis with massive unilateral pleural effusion

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SUMMARY

We described a 45-year-old previously healthy man presenting with progressively worsening breathlessness for 10 days. Physical examination was suggestive of a left-sided pleural effusion. A chest X-ray was confirmatory. Analysis of aspirated fluid showed a lymphocytic exudate with grossly elevated amylase and lipase levels. CT revealed chronic calcific pancreatitis as the underlying cause of effusion. Retrospective questioning failed to identify classical symptoms of chronic pancreatitis including abdominal pain and steatorrhoea. The patient was managed with intercostal drainage and supportive care. Although unusual, chronic pancreatitis should be kept as a differential diagnosis in patients with unilateral exudative pleural effusion. Elevated fluid levels of amylase and lipase are useful clues to this uncommon diagnosis.

BACKGROUND

Left-sided pleural effusion is a recognised complication of acute pancreatitis, occurring in 4–17% of cases.¹ Typically, such effusions are clinically silent and incidentally detected in patients presenting with abdominal pain. Although acute pancreatitis may occasionally present with primarily respiratory symptoms arising from massive pleural effusion, such presentations are rare for chronic pancreatitis. Moreover, a review of sporadic reports of massive pleural effusion in the chronic pancreatitis reveals a history of recurrent abdominal pain and a history of alcoholism compatible with chronic pancreatitis in most patients.

Contrastingly, our patient never experienced abdominal symptoms and possessed no risk factors for developing chronic pancreatitis. As this case amply illustrates, absence of risk factors and symptomatology of chronic pancreatitis cannot be reliably employed to rule out this disorder as the aetiology of pleural effusion. Elevated levels of amylase and lipase in pleural fluid constitute important diagnostic clues; consequently, these enzymes should be estimated in all patients with otherwise unexplained unilateral exudative pleural effusion, even in the absence of clinical evidence of chronic pancreatitis.

CASE PRESENTATION

A 40-year-old Asian-Indian man presented with progressively worsening breathlessness for 10 days. There were no associated symptoms of cough, pleuritic chest pain or fever. His medical history was uneventful. He denied any history of alcohol consumption or other forms of substance misuse. There was no history of abdominal trauma in the

past, and no history of pancreatitis in his family members.

General physical examination did not reveal any signs of malnutrition. He was haemodynamically stable. Respiratory examination revealed tracheal deviation to the right side, a stony dull note to percussion over the entire left hemithorax, and absent breath sounds on auscultation over the same areas. Respiratory movements were diminished on the left side. There were no adventitious sounds. An abdominal examination ruled out epigastric tenderness or the presence of ascites. A cardiac examination showed medial shifting of the apical impulse, with slight muffling of the heart sounds.

INVESTIGATIONS

Routine laboratory tests revealed hyperglycaemia (random blood glucose 227 mg/dL; glycated haemoglobin 7.6%). Relevant haematological investigations were within the normal limits. A chest X-ray confirmed a massive left-sided pleural effusion with mediastinal shift, with a clear right lung field (figure 1). Thoracentesis yielded straw-coloured fluid; analysis revealed an exudate (fluid protein 3.9 g/dL; serum protein 7.6 g/dL; fluid glucose 69 mg/dL; fluid lactate dehydrogenase 973 U/L; serum lactate dehydrogenase 191 U/L; fluid adenosine deaminase 1.4 U/L). Cytology and Gram stains of the fluid were negative for malignancy and microorganisms respectively. Staining for

Figure 1 Chest X-ray showing massive left-sided pleural effusion with mediastinal shift to the right.

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Figure 2 Contrast-enhanced abdominal CT showing pancreatic calcification (coronal and axial sections, arrow) and pleural effusion (coronal section, arrow).

acid-fast bacilli was negative. A conventional aerobic culture was sterile. Nested PCR for *Mycobacterium tuberculosis* complex (IS6110) was negative. Amylase and lipase levels were assessed when the effusion re-accumulated rapidly after aspiration, and were grossly elevated (28 644 and 82 398 U/L, respectively). Serum amylase and lipase were sent thereafter and were mildly elevated (107 and 77 U/L, respectively).

CT of the abdomen showed atrophy of the head and body of the pancreas with an irregularly dilated main duct and multiple parenchymal and intraductal calcifications, and no evidence of peripancreatic fat stranding or fluid (figure 2). Pancreaticopleural fistulae could not be delineated. A thoracic CT performed after insertion of an intercostal drainage tube (see below), revealed gross left-sided pleural effusion with partial collapse of the underlying lung (figure 3). There was no discernible pathology within the lung parenchyma, mediastinum or pericardial compartment.

An endoscopic retrograde cholangiopancreatography and magnetic cholangiopancreatography were offered for identifying possible pancreaticopleural fistulae but refused on financial grounds.

In the absence of alcohol consumption, chronic pancreatitis was presumed to be idiopathic. The abdominal CT ruled out a congenital anomaly of the pancreas. The absence of family history made hereditary pancreatitis and cystic fibrosis less likely. Further evaluation including genotypic analysis for serine protease inhibitor Kazal-type 1 (SPINK-1) and cystic fibrosis transmembrane conductance regulator (CFTR) were not performed due to financial constraints. Sweat chloride determination was not conducted due

to unavailability of the test at our hospital and relative rarity of cystic fibrosis in Southern India. PET was similarly not attempted due to its relatively prohibitive cost.

DIFFERENTIAL DIAGNOSIS

- ▶ Massive unilateral pleural effusion secondary to chronic calcific pancreatitis
- ▶ Malignant pleural effusion with unknown primary
- ▶ Lymphoma-related pleural effusion
- ▶ Tubercular pleural effusion

TREATMENT

The patient was initially managed with emergent thoracocentesis and aspiration of 600 mL of fluid, with partial relief of symptoms. Rapid fluid re-accumulation necessitated repeated aspiration on the second and third day of hospitalisation. An intercostal drainage tube was then inserted to allow continuous drainage.

The chronic pancreatitis was managed conservatively with pancreatic enzyme replacement, antioxidants and intravenous octreotide therapy (50 µg subcutaneous every 8h).

The secondary diagnoses listed above were carefully considered. Persistently negative microbiological and cytological analyses of pleural fluid rendered these possibilities unlikely, and empiric treatment was therefore not attempted.

OUTCOME AND FOLLOW-UP

Although significant relief was gained after insertion of the intercostal chest tube, large volumes of fluid (upto 500 mL/day) continued to drain 1 week after the procedure. Alternative strategies including chemical pleurodesis were planned once drainage came down. Owing to financial constraints, however, the patient refused further hospital stay, electing instead to follow-up at a local healthcare facility. Surgical decompression/endoscopic stenting were suggested as means to decelerate pleural fluid production but refused again on financial grounds. The risks and benefits of retaining the chest drain, that is, infection and sepsis against a reduced chance of breathlessness, were explained and the patient elected for retention. He was discharged with the chest drain in situ and was subsequently lost for follow-up.

DISCUSSION

Chronic pancreatitis is uncommonly associated with massive symptomatic pleural effusion, and reports of such presentation are limited to sporadic case reports.^{2–5} All these cases except one, however, were marked by obvious clinical clues to a

Figure 3 Contrast-enhanced thoracic CT showing pleural effusion (axial section, arrow).

possibility of chronic pancreatitis including a history of alcoholism, chronic abdominal pain or previous attacks of acute pancreatitis.² In one instance, the patient had a prior diagnosis of chronic pancreatitis and subsequently developed pleural effusion.³ In another, bilateral haemorrhagic pleural effusion developed in a patient with previously undiagnosed chronic pancreatitis, and no history of alcoholism.⁴ Careful questioning revealed a history of vague abdominal pain; in our own patient such symptoms could not be elicited despite repeated questioning.

Formation of pancreaticopleural fistulae has been suggested as the primary mechanism of development of pleural effusion in patients with chronic pancreatitis.² Although such fistulae were directly demonstrated by MR and/or endoscopic retrograde cholangiopancreatography in several case reports,^{1–3} they could not be demonstrated in our patient. Rapid re-accumulation of pleural fluid and the absence of associated ascites, served as indirect evidence of an underlying fistulous communication to the pancreas.

Interestingly, rapid re-accumulation of pleural effusion following pleurocentesis appears to be a characteristic feature of pancreatic effusion,⁶ and was a hallmark of our case. A combination of recurrent large unilateral exudative effusion and presence of risk factors such as chronic alcoholism appears to be pathognomic of a pancreatic pleural effusion. Moreover, chronic pancreatitis can be easily, reliably and non-invasively differentiated from other causes of amylase-rich exudative effusion⁷ including malignancy, oesophageal rupture and tuberculosis by means of CT. Nevertheless, diagnosis is frequently delayed with a time to diagnosis ranging from 12 to 49 days,⁸ possibly reflecting a relative lack of awareness of this entity among medical professionals.

Once diagnosed, a conservative management with intercostal drainage is indicated along with somatostatin analogues. Failure to respond is an indication for endoscopic or surgical decompression of the obstructed pancreatic duct. Endoscopic stenting is now accepted as the primary modality of intervention, while a surgical approach is reserved for patients failing to respond to endoscopic stenting or those with an unsuitable pancreatic anatomy.⁶

In summary, routine estimation of fluid amylase and lipase in patients with rapidly accumulating pleural effusion, followed by a thoracoabdominal CT in cases with elevated enzyme levels appears to be the best diagnostic approach to this disorder. With the advent of specific therapy in the form of endoscopic and surgical intervention, increased awareness should be generated about this unusual complication of chronic pancreatitis.

Learning points

- ▶ Chronic pancreatitis can be complicated by massive pleural effusion.
- ▶ Pleural effusion in such instances can occur unaccompanied by classical symptoms of chronic pancreatitis including abdominal pain and steatorrhoea.
- ▶ Elevated levels of amylase and lipase in pleural fluid are important diagnostic clues; consequently, these enzymes should be estimated in all patients with otherwise unexplained unilateral exudative pleural effusion.

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Competing interests None.

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