Case Report

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Right hemithoracic pseudocyst with splenic artery aneurysm: two rare complications of an uncommon disease

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INTRODUCTION

Pleural involvement is an uncommon but well recognized complication of chronic pancreatitis [1], mainly in the form of pleural effusion affecting the left hemithorax. Pancreatic pseudocyst extending to the posterior mediastinum with or without communication with the pleural space is another rare form of this involvement.

The treatment of chronic pancreatic pleural effusions and pancreatic pseudocysts generally starts with a conservative approach including bowel rest, drainage of the pleural effusion by repeated thoracentesis or a chest tube, and total parenteral nutrition (TPN) for a period of time determined by the clinical course. Other treatment modalities including percutaneous drainage, endoscopic retrograde cholangiopancreatogram (ERCP) with stenting of the pancreatic duct and surgical drainage are used if conservative approaches fail.

We report a patient with a complicated pancreatic pseudocyst who showed an involvement of the posterior mediastinum and right pleural space, with conspectus sparing of the left hemithorax. The patient had a prolonged and

complicated course that included recurrence of the pseudocyst with oral feedings and the development of a splenic artery aneurysm. Some of the above findings have been reported separately as uncommon complications of chronic pancreatitis and pancreatic pseudcyst, but to our knowledge a single case with all these complications was not published in the English literature.

CASE REPORT

A 42-year-old male with history of alcohol abuse and pancreatitis, presented to Kino Community Hospital outpatient clinic with complaints of chest discomfort and shortness of breath over a two-week period. At presentation the patient was in no distress and had normal vital signs. The chest examination revealed decrease in air movement over the right hemithorax and the abdominal examination demonstrated mild epigastric tenderness.
A chest x-ray showed a large right pleural effusion and an epigastric shadow resembling a hiatal hernia (figure 1). A computed axial tomography scan (CAT scan) showed a right pleural effusion and a posterior mediastinal pseudo cyst (figure 2).

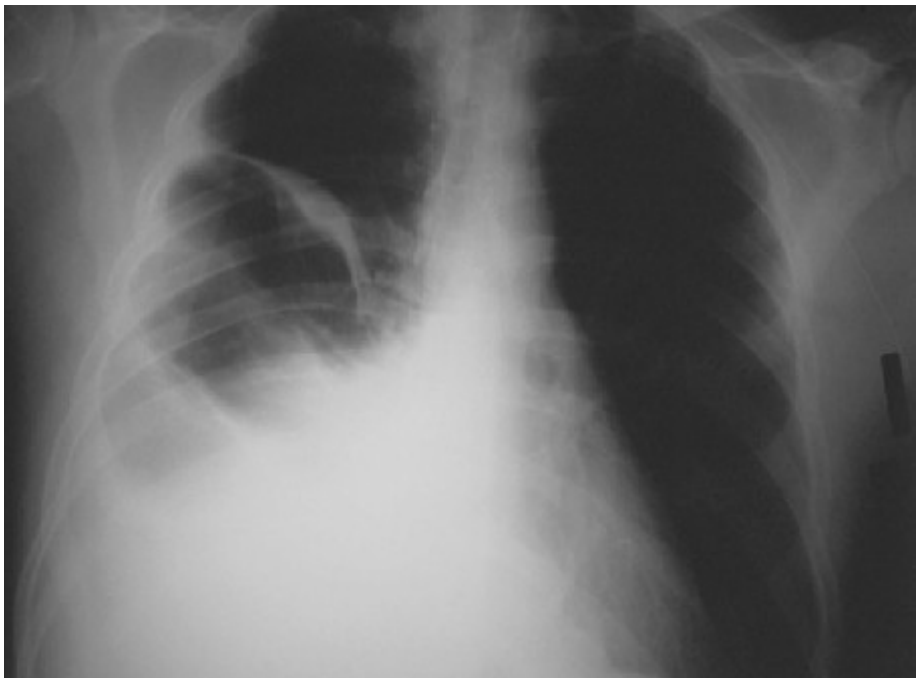
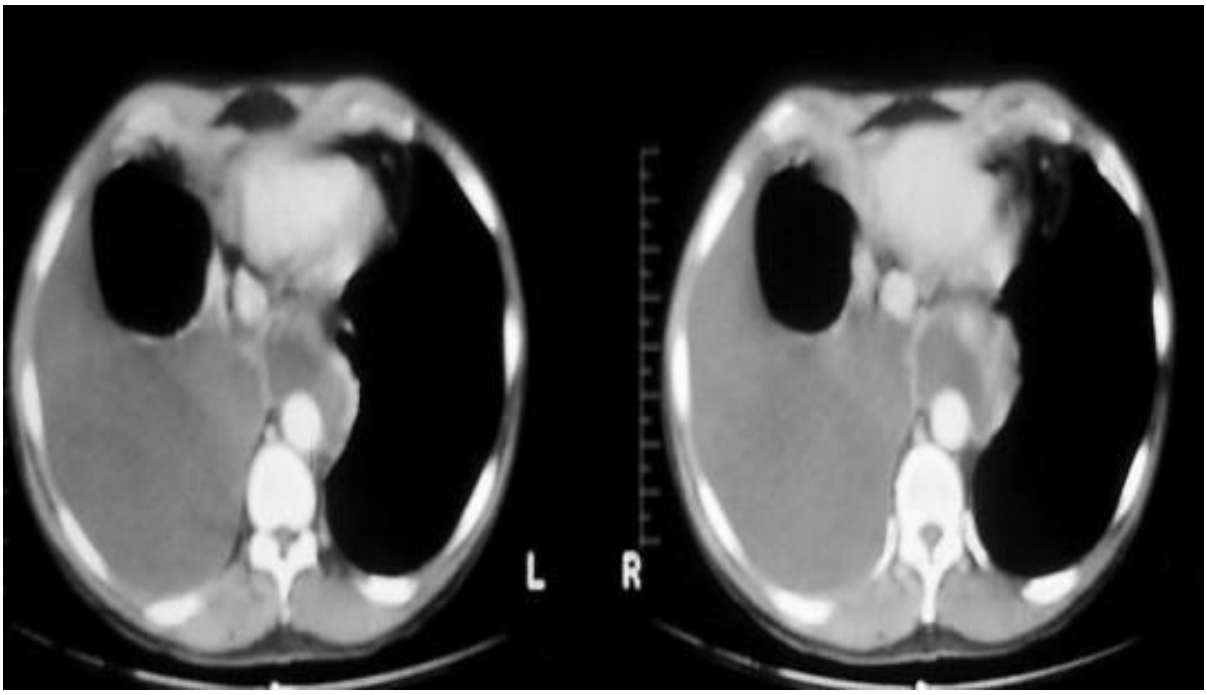


Figure 1: Chest X-ray of the right pleural effusion.

Laboratory investigations revealed the following; serum amylase 556 units, pancreatic isoenzyme 365 units, Lipase 143 units, total protein 5.9 mg/dL, total bilirubin 0.7mg/dL, albumin 2.4 mg/dL, alkaline phosphatase 75 IU/L, AST 68 IU/L, ALT 95 IU/L. Thoracentesis was performed and 1500cc of bloody effusion

was aspirated; WBC 2200 / cubic mm, RBC 16500 / cubic mm, LDH 1061 IU/L, protein 4300 mg/ dl, and amylase 22700, AFB smear and cultures were negative.

tion of the effusion and resolved pseudocyst. The chest tube output ceased, however, the serum amylase and lipase continued to be elevated.



The day after thoracentesis a chest x-ray revealed rapid re-accumulation of the pleural effusion. The patient was hospitalized; chest tube was inserted in the right pleural space and he was started on TPN. The following day the patient's symptoms resolved and the chest x-ray demonstrated complete resolution of the pleural effusion.

A week later the asymptomatic patient had a CAT scan, which demonstrated continued resolu-

***F**igure 2: CAT scan of the right pleural effusion and posterior mediastinal pseudo cyst.*

Brief oral feeding started on day ten followed by re-accumulation of the pleural fluid (figures 2&3). The patient continued on TPN and made non per os (NPO), Octeriotide acetate (Sandostatin, Novartis Pharmaceutical) was started to decrease pancreatic output. The chest tube was removed a few days later.

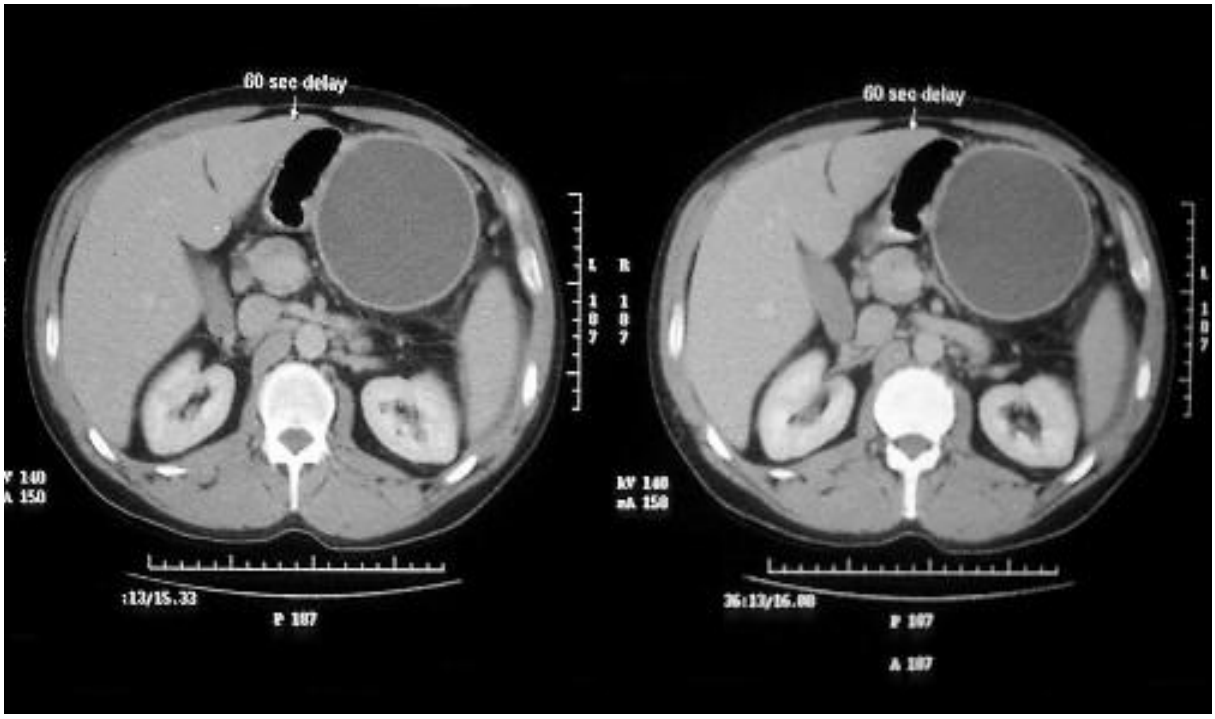


Figure 3: CAT scan of the abdominal view of the pancreatic pseudo cyst.

Three months later the serum amylase was still elevated 400 IU/L, but the patient was asymptomatic and a repeat CAT scan of the chest showed no effusion or pseudocyst. Oral feeding was attempted which again led to re-accumulation of the pleural effusion. The patient again was made NPO/TPN and was scheduled for surgical drainage. Prior to surgery the patient developed severe abdominal pain and was found to be acutely anemic (Hemoglobin 6.5g/dl). An emergent splenic artery angiogram revealed a bleeding splenic artery aneurysm (figure 4).

The patient underwent splenic artery embolization followed by splenectomy and cystoenteric drainage procedure in a later date. One year later the patient is asymptomatic with normal laboratory and radiological findings.

DISCUSSION

Pancreatic pleural effusion can complicate both acute and chronic pancreatitis. The former usually leads to a small sympathetic effusion, mainly involving the left side and is usually self-limiting [15]. Chronic pancreatitis may lead to an internal pancreatic fistula with leakage of pancreatic secretions

anteriorly to the peritoneal sac forming pancreatic ascites, or posteriorly to the retroperitoneal space forming a pseudocyst [2]. Rarely pseudocyst can dissect through the aortic or esophageal hiatus into the posterior mediastinum and present as a mediastinal pseudocyst.

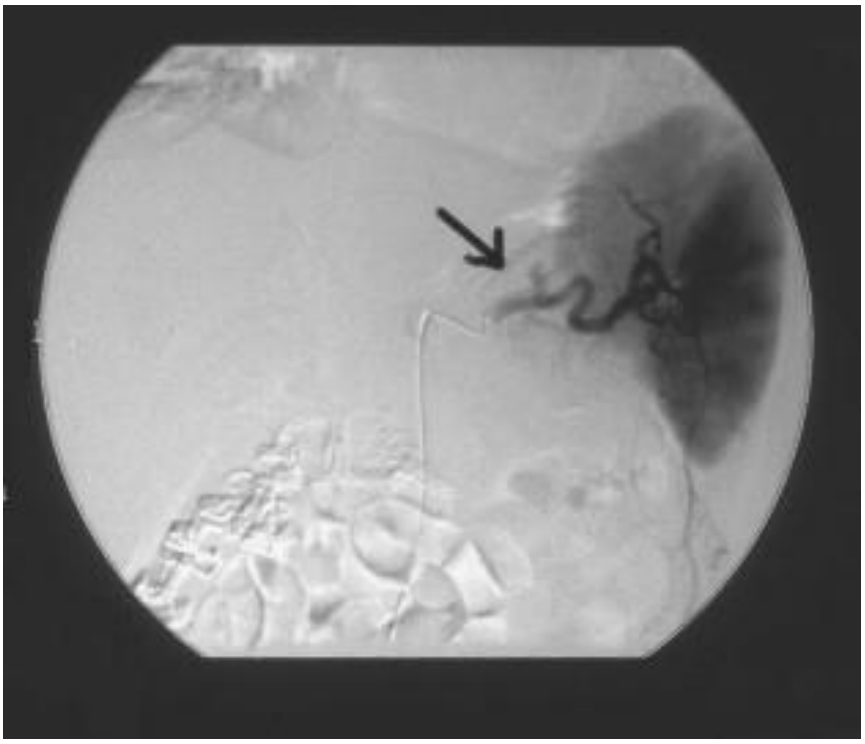


Figure 4: Angiogram of the splenic artery aneurysm.

Pancreatic pleural effusions are caused either by a pancreaticopleaural fistula passing through an opening in the diaphragm [2], or a mediastinal pseudocyst that has ruptured or extended into the pleura cavity [3]. Chronic pleural effusion is uncommon but a well recognized complication of chronic pancreatitis [1]. The majority of cases occur in men with a history of alcoholism [5]. The effusions are usually massive and left sided with high amylase levels, while serum amylase levels may only be mildly elevated [5, 6]. Clinical presentation is variable and often misleading, contributing to increased morbidity and mortality [4]. Pulmonary symptoms are more frequent than abdominal symptoms, with dyspnoea being the most common complaint.

The initial management of pancreatic pleural effusion is usually medical with bowel rest, drainage of the pleural space by repeated thoracentesis or a chest tube and TPN. Sandostatin, a long-acting somatostatin analogue, is occasionally added as part of the treatment for chronic pancreatic effusion [8, 9]. The favorable response to medical treatment is variable ranging between 40%-

60% in some reports [1, 5]. Failure of medical treatment is usually followed by several other modalities depending on the clinical situation and the radiological feature of the effusion and pseudocyst.

Åke Andren-Sandberg and Christos Devenis [15, 16], performed an exhaustive literature search in their review of the natural history and treatment of pancreatic pseudocyst. They pointed out the conflicting information regarding the natural history of pancreatic pseudocyst. The pathology of duct involvement is also considered a main factor in treatment selection; however, ERCP is not practical in all cases, and MRCP is not readily available. This makes evaluation of different treatment options difficult. Moreover, the selection of treatment is complicated by the high rate of spontaneous resolution of pseudocyst following the conservative approach.

Percutaneous drainage of the pseudocyst is an attractive approach that might circumvent an extensive surgical procedure if the pseudocyst is accessible [10]. ERCP with stent placement into the pancreatic duct to bridge the site of leakage can be used with success after the initial failure of medical management [11].

Surgical cystoenteric drainage is the ultimate procedure [12]; however, surgical drainage is technically difficult if done early in the disease course since the pseudocyst has no real boundaries and the pseudocyst wall in general is shaggy and frail [14].

Splenic artery aneurysm is a rare but frequently fatal complication of chronic pancreatitis. It is thought that the pancreatic secretions in contact with the splenic vessels may lead to the aneurysm formation. The treatment of such complication is usually surgical resection with or without initial arterial embolization in actively bleeding high-risk patients [13].

In our case the medical management was initially successful in resolving the effusion completely but the challenge was the persistent elevated amylase and the recurrence of effusion with trials of oral feeding. The pseudocyst was not accessible for percutaneous drainage, and it was felt that ERCP with placement of pancreatic duct stent if failed to resolve the pseudocyst carry the risk of introducing infection that may lead to urgent need for surgery, which is not advised early in the course of the pseudocyst formation.

The patient's reluctance to consent for a procedure that may lead to surgical intervention, the dilemma created by the course of his disease, and the literature reports of high incidence of spontaneous resolution, all influenced the relatively long period of conservative measures.

CONCLUDING REMARKS

Chronic pancreatitis and pancreatic pseudocyst may lead to pulmonary complications with significant morbidity and mortality. Early diagnosis and treatment with careful follow up is important to avoid unfavorable outcomes. Also, careful assessment of different treatment options and choosing the appropriate option on a case-to-case bases is paramount in the management of these complications.

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