CASE REPORT

Pancreaticopleural Fistula: A Rare Complication of Alcoholic Pancreatitis

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ABSTRACT

Pancreaticopleural fistula is a rare complication after pancreatitis, most commonly secondary to pseudocyst rupture with tracking of pancreatic fluid into the mediastinum. We describe a 45-year-old female with a long-standing history of chronic alcoholic pancreatitis with multiple recurrent episodes of acute pancreatitis who presented with dyspnea from a massive right pleural effusion, which was in communication with a large ruptured pseudocyst. Owing to failure of conservative therapy, she required endoscopic pancreatic duct stenting and surgical pulmonary decortication.

INTRODUCTION

Pancreaticopleural fistula (PPF) is a rare complication of pancreatitis, caused by communication of pancreatic ductal (PD) fluid with the mediastinum, most commonly due to a ruptured pancreatic pseudocyst. Chronic alcoholic pancreatitis is the most frequent culprit, with 0.4% of patients developing a PPF [1]. PPFs commonly manifest with dyspnea from massive pleural effusions, and thoracentesis along with abdominal imaging help establish the diagnosis. We present a case of PPF after chronic pancreatitis.

CASE PRESENTATION

A Forty-five-year-old-female with a 5-year history of chronic alcoholic pancreatitis and multiple recurrent episodes of acute pancreatitis complicated by pseudocysts presented with 3 days of epigastric pain radiating to the back associated with non-bloody non-bilious emesis and a new non-productive cough, with dyspnea at rest. Vital signs were notable for tachycardia of 128 bpm though she was normotensive and afebrile. On physical examination, she had decreased breath sounds, diffuse rales and dullness to percussion over the right hemithorax. Laboratory studies were significant for an elevated lipase level of 218 g/dL (reference range: <60 U/L), though no leukocytosis. Electrolytes and liver function tests were within normal limits. A chest radiograph demonstrated a moderate right pleural effusion and on computed tomography (CT) of the chest and abdomen, acute on chronic pancreatitis with a large pseudocyst, multiple peripancreatic fluid collections and mediastinal fluid were also seen **(Figure 1)**. A thoracentesis revealed fluid of an exudative pattern, with an elevated amylase level of 8773 U/L (reference range: <200 U/L), confirming the diagnosis of PPF. A magnetic resonance cholangiopancreatography (MRCP) revealed that the distal pancreatic pseudocyst communicated with the retroperitoneum, mediastinum and right pleural space by means of multiple fluid tracts **(Figure 2)**. The pancreatic duct (PD) was normal in size. The patient was managed conservatively for 6 weeks with a low-fat diet and octreotide. Despite initial symptom resolution, her dyspnea recurred and she required frequent thoracenteses. Given that the patient had failed 6 weeks of conservative management and was unable to remain asymptomatic without intervention, the decision was made to perform a right video-assisted thoracoscopic surgical (VATS) drainage with partial pleurectomy, pulmonary decortication and placement of a PleurX catheter to optimize her for future definitive management of the PPF.. Subsequently, after resolution of dyspnea, she underwent an endoscopic retrograde cholangiopancreatography (ERCP) at which time the pseudocyst was seen to be originating from a disruption

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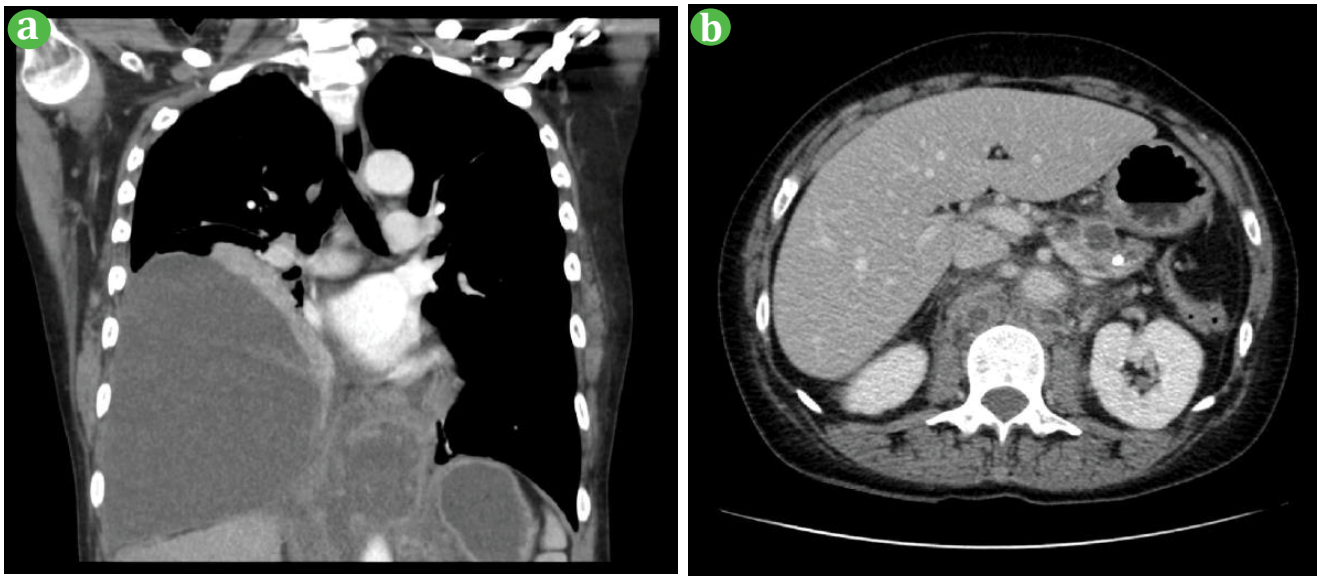

Figure 1. (a). Computed tomography of the chest revealed a massive right pleural effusion. **(b).** Computed tomography with intravenous contrast of the abdomen and pelvis revealed a lobulated pseudocyst of the pancreatic body and tail, a heterogeneous collection adjacent to the tail and calcifications from chronic pancreatitis.

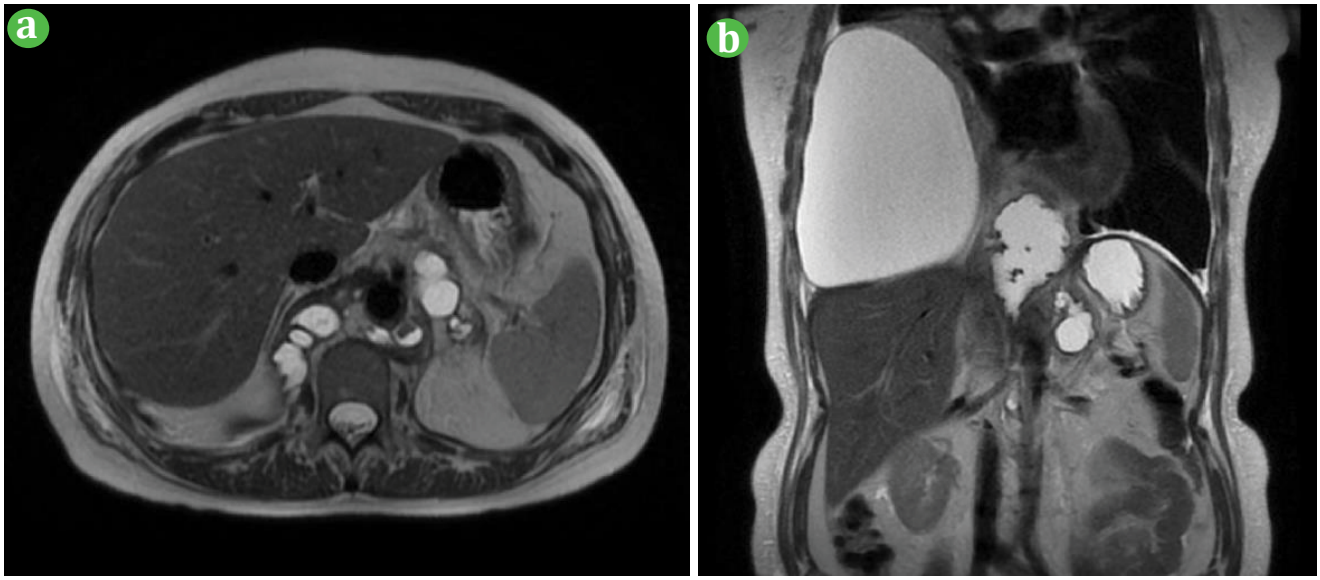

Figure 2. Magnetic resonance cholangiopancreatography demonstrated a large pseudocyst of the pancreatic body and tail extending into the retroperitoneum, mediastinum and right pleural space by means of multiple tracts. The pancreatic duct is of normal caliber. **(a).** Axial view. **(b).** Coronal view.

of the distal PD with associated stricture. A plastic PD stent was placed and a ventral pancreatic sphincterotomy performed **(Figure 3)**.

Patient Outcomes

Postoperatively, the patient had an uncomplicated course. The PD stent was subsequently removed 11 weeks afterwards, when a repeat ERCP demonstrated no further evidence of a fistulous tract from the distal PD. Octreotide was also discontinued at this time and the PleurX catheter removed. She has had no complications or symptom recurrence 7 months after the operation.

DISCUSSION

In the United States, the incidence of acute pancreatitis is 25/100,000 Annually, 10% will develop complications, most commonly chronic pancreatitis and pseudocysts [2] In very rare cases, when a pseudocyst wall ruptures, pancreatic fluid leaks from the PD via the path of least resistance and forms a PPF. The fluid typically tracks up the retroperitoneum and dissects through the aortic or esophageal hiatus into the mediastinum forming large pleural effusions, most frequently in the left hemithorax [3]. Patients classically present with dyspnea, however PPF may present with vague non-specific symptoms such as chest pain, palpitations, dysphagia, dyspnea, cough, or abdominal pain. Patients often undergo elaborate pulmonary work-up before a pancreatic source is identified, which can lead to significant delays in diagnosis of up to 49 days reported in the literature [4, 5].

Reactive pleural effusions from acute pancreatitis are much more common. However, PPF remains an important differential diagnosis in patients with an alcoholic etiology and persistent or worsening pleural effusions after resolution of acute episodes. Thoracentesis is helpful in

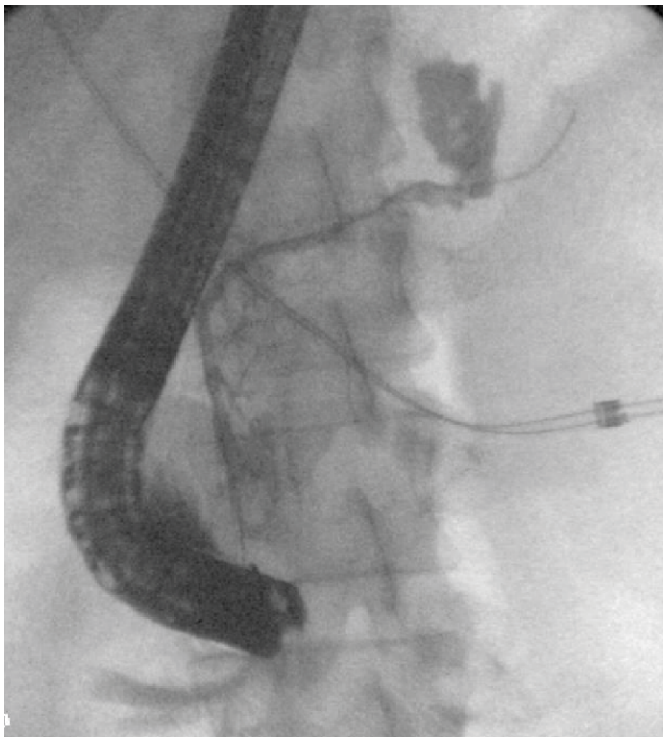

Figure 3. Endoscopic retrograde cholangiopancreatography demonstrated a pseudocyst of the pancreatic distal body and tail associated with a disrupted and strictured pancreatic duct.

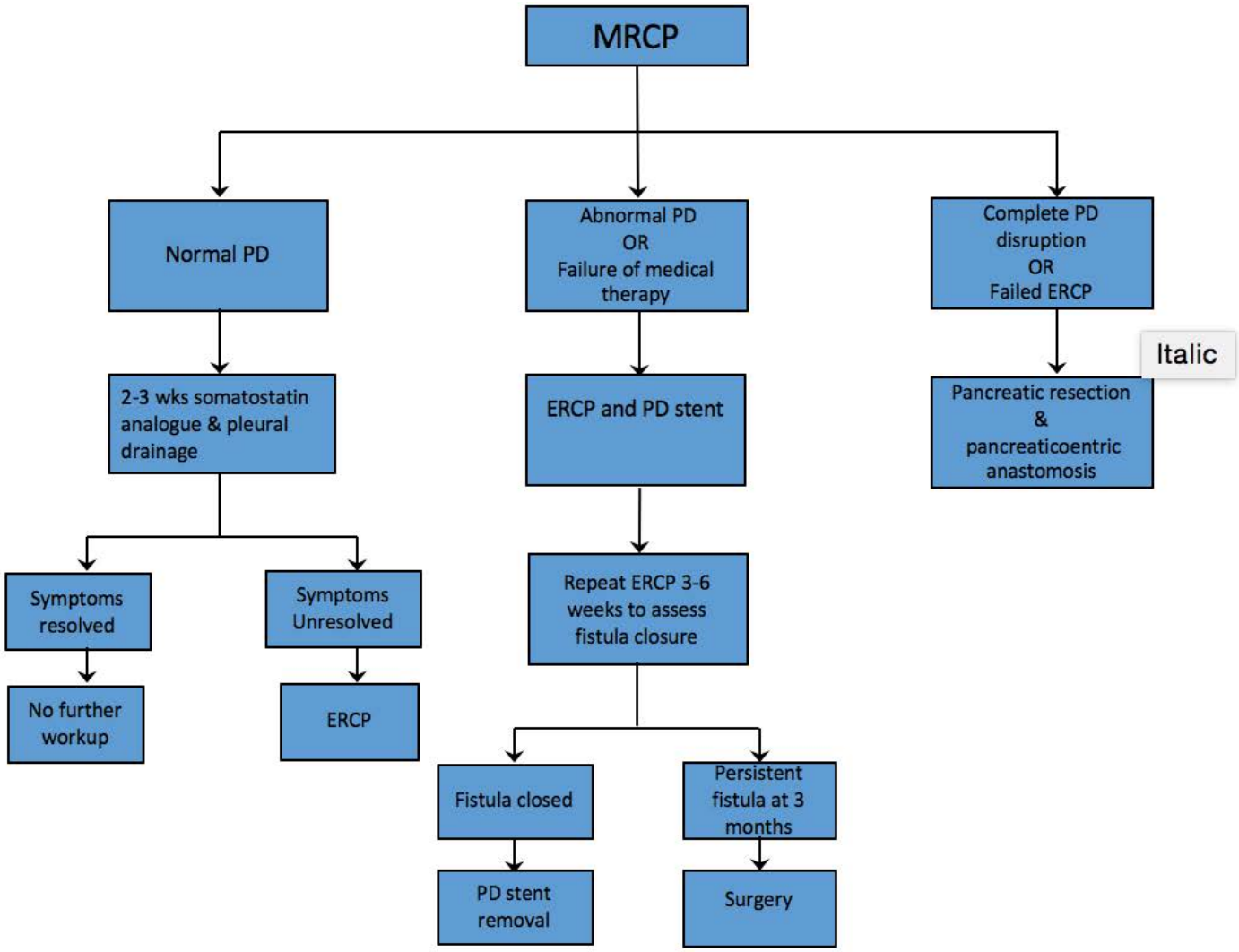

Figure 4. Treatment algorithm for the management of pancreaticopleural fistula.

ERCP endoscopic retrograde cholangiopancreatography; MRCP magnetic resonance cholangiopancreatography; PD pancreatic duct

establishing a diagnosis. High amylase levels in the pleural fluid and an exudative pattern are characteristic of PPF. Though pleural amylase may be detected in other disease processes (esophageal perforation, parapneumonic effusion and lung or pancreatic malignancy), the levels in PPF are usually much more elevated (>50,000 IU/L) [6]. Imaging is important both to visualize the fistulous tract as well as to define the underlying etiology and pancreatic anatomy. CT scans have the highest sensitivity (82-100%) for visualizing calcifications and other changes of chronic pancreatitis, and to assess for a pseudocyst [3]. MRCP is superior for evaluating PD anatomy and delineating the fistulous tract [7].

The treatment of PPF is largely dictated by patient symptomatology and PD anatomy **(Figure 4)** [8]. Medical approaches with pleural drainage and somatostatin analogues (to reduce fistula output) may be attempted provided the PD is normal in caliber and without stricture. Nil-per-os with total parenteral nutrition is no longer recommended [6]. A trial of conservative measures should be limited to 2-3 weeks, as more than 80% of patients develop complications beyond this time period. However, similar to our patient, 40-70% ultimately fail this treatment strategy [9, 10]. For patients who fail medical therapy or have MRCP findings of PD partial disruption or distal stricture, endoscopic or surgical measures are warranted. ERCP is a low-risk and less aggressive strategy with success rates up to 100% in some series, and should be considered first line in these patients over surgical intervention [11, 12, 13]. PD stenting covers any site of ductal disruption, and stricture dilation helps to restore a lower-pressure ductal flow towards the duodenum, aiding in fistula closure. Pancreatic sphincterotomy may also help in this regard [6]. At our institution, in line with other reported cases, plastic covered stents are used. Stent removal is important to prevent long-term ductal changes; however optimal duration is undefined owing to the rarity of PPF. Authors have suggested that up to 1-3 months may be appropriate; thus performing repeated ERCPs every few weeks to assess fistula closure is recommended [14]. Operative intervention is usually reserved for patients who fail medical and endoscopic therapy, however, it is an important first line therapy if a complete PD disruption or stricture proximal to the fistula is present [6, 8]. Though surgery offers thrice the success rate of medical therapy, it has much higher morbidity rates than does ERCP. Resection of the involved pancreas, pseudocyst and affected PD segment with an enteric anastomosis is most frequently performed [13]. Regardless of operative procedure, preservation of pancreatic parenchyma remains paramount, as most patients have diminished function from chronic pancreatitis. Should our patient fail endoscopic therapy in the future, consideration of a distal pancreatectomy with pancreaticojejunostomy would be appropriate.

CONCLUSION

Pancreaticopleural fistula is a rare entity, but should be considered especially in cases of alcoholic pancreatitis or pseudocyst with massive enlarging pleural effusions despite resolution of pancreatitis. Thoracentesis and MRCP are key in establishing the diagnosis and for treatment planning. The initial management strategy should be based upon pancreatic ductal anatomy.

Conflict of Interest

The author has no potential conflicts of interest.

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