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Case report

Resolution of pancreatico-pleural fistula with endoscopic ultrasound-guided therapy

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ABSTRACT

Pancreatico-pleural fistula is an uncommon cause of recurrent pleural effusion. Delayed diagnosis may occur if fluid amylase level is not obtained early in the clinical course. As most cases of pancreatico-pleural effusion are due to chronic pancreatitis, endoscopic therapy may be effective if pancreatic fluid secretion can be diverted to a more physiologic pathway. However, when severe pancreatitis leads to disconnection of the pancreatic duct, it precludes conventional endoscopic treatment via transpapillary stenting of the pancreatic duct. We describe a patient with a chronic, refractory pancreatico-pleural fistula arising from chronic pancreatitis with a disconnected pancreatic duct syndrome, which resolved following endoscopic ultrasound-guided therapy.

1. Introduction

Pancreatico-pleural fistula (PPF) is a rare cause of recurrent, large pleural effusions, usually resulting from chronic pancreatitis.[1–5] We report a unique case of successful resolution of PPF following endoscopic ultrasound (EUS)-guided therapy.

2. Case report

2.1. Patient presentation

A 58-year-old woman with a 30-pack-year smoking history and remote alcoholism presented with dyspnea due to large pleural effusions (Fig. 1).

She had a remote history of abdominal pain but no current pain or diarrhea. Over a period of four months, she underwent 4 large-volume thoracenteses, 2 chest tube placements and 2 thoracotomies for recurrent effusions; the etiology remained obscure until the 4th thoracentesis, when fluid amylase was measured and found to be markedly elevated (29,503 U/L).

2.2. Imaging studies

Chest radiography revealed pleural effusions. Magnetic resonance cholangio-pancreatography (MRCP) showed an irregular pancreatic duct with non-visualization of the tail portion, and two small adjacent fluid collections (Fig. 2).

2.3. Endoscopic management

A one-week trial of medical therapy with bowel rest and octreotide afforded no improvement in the pleural effusion(s). The patient was not considered a candidate for surgical therapy. Therefore, management with endoscopic retrograde cholangio-pancreatography (ERCP) and EUS was performed. ERCP confirmed the MRCP findings of ductal disruption with a disconnected pancreatic tail; the sub-diaphragmatic pancreatic fluid collection did not opacify with retrograde injection of contrast (Fig. 3). A transpapillary 7-French, single-pigtail stent (Hobbs Medical Co., Stafford Springs, CT, USA) was placed to facilitate drainage of the body and head of pancreas. EUS was performed using a linear-array echoendoscope (Olympus Corporation, Tokyo, Japan). The sub-diaphragmatic pancreatic fluid collection, attributable to the disconnected pancreatic tail, was targeted for EUS-guided therapy. Utilizing conventional techniques of EUS-guided pseudocyst drainage,[6] the collection was accessed via transgastric needle puncture (Echo-tip 19-gauge needle, Cook Medical, Bloomington,

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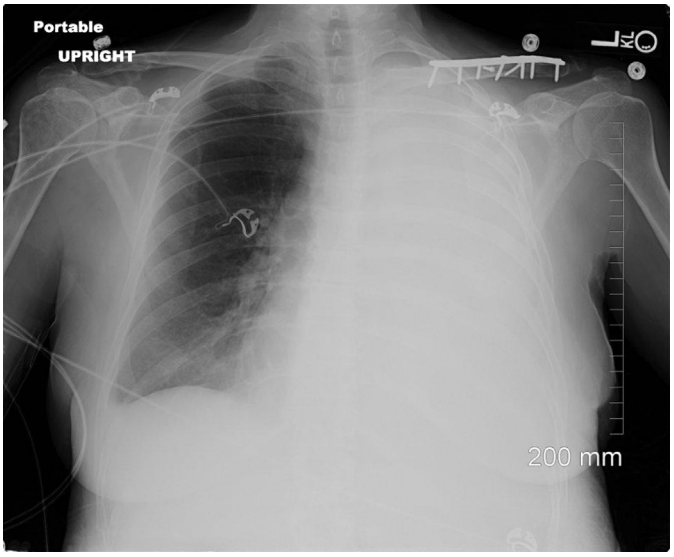

Fig. 1. Presenting anterior–posterior CXR showing large pleural effusion.

IN, USA) in the gastric fundus. Cyst access was confirmed by fluid aspiration followed by contrast injection under fluoroscopy (Fig. 4). A 0.035 guide-wire was passed through the needle and coiled in the cavity of the collection (Fig. 5). The transgastric tract was dilated to 8 mm using a balloon catheter. Two 5-cm double-pigtail Solus stents (Cook Medical) were placed, each with one pigtail within the collection and the other within the gastric lumen (Figs. 6).

2.4. Patient outcome

Plain X-rays obtained 12 days after the endoscopic procedures demonstrated spontaneous passage of the transpapillary pancreatic stent, stable position of the transgastric stents and resolution of pleural effusions. At 5-month follow-up the patient was asymptomatic without clinical or radiographic evidence of recurrent effusions; transgastric stents remained in stable position in the region of the gastric fundus (Fig. 7). Repeat MRCP showed resolution of peri-pancreatic fluid collections.

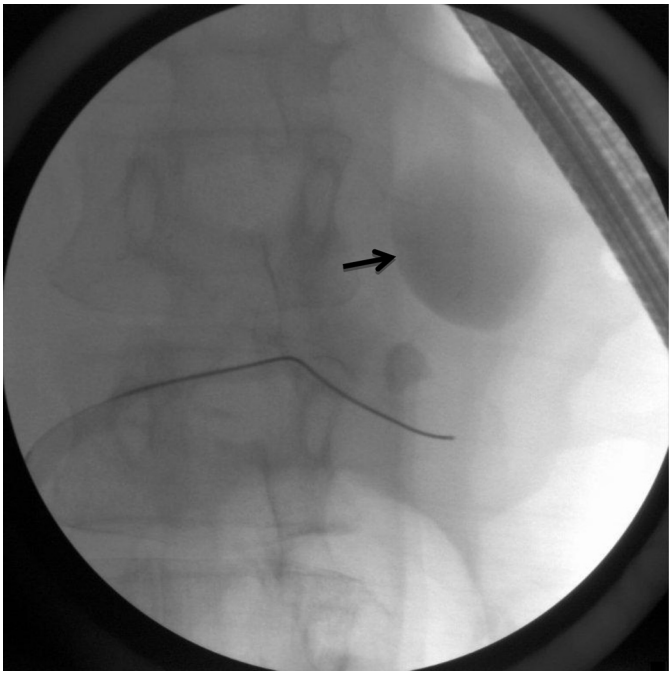

Fig. 3. ERCP with pancreatic wire cannulation demonstrating non-filling (disconnected) tail portion of pancreatic duct. There is opacification of the smaller fluid collection seen on MRCP (arrow), indicating communication with main pancreatic duct.

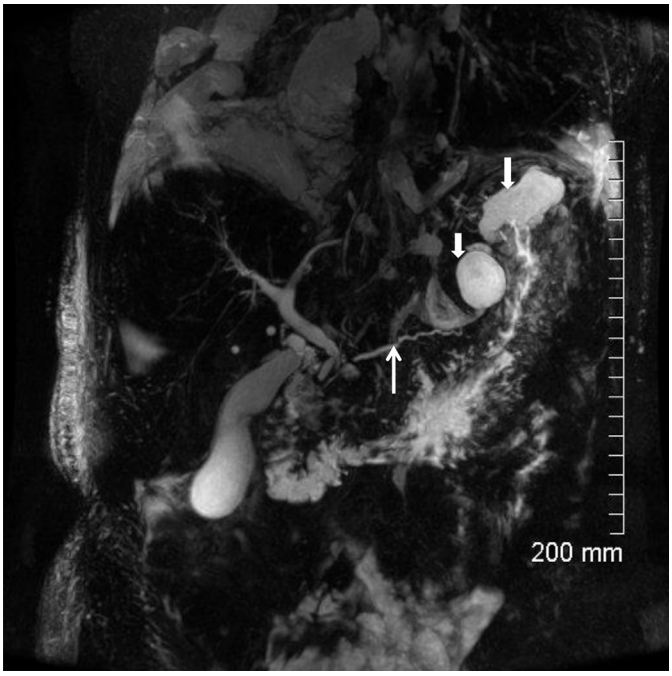

Fig. 2. MRCP demonstrating normal proximal pancreatic duct (arrow), non-filling of disconnected tail portion of pancreatic duct and adjacent pancreatic fluid collections (arrowheads).

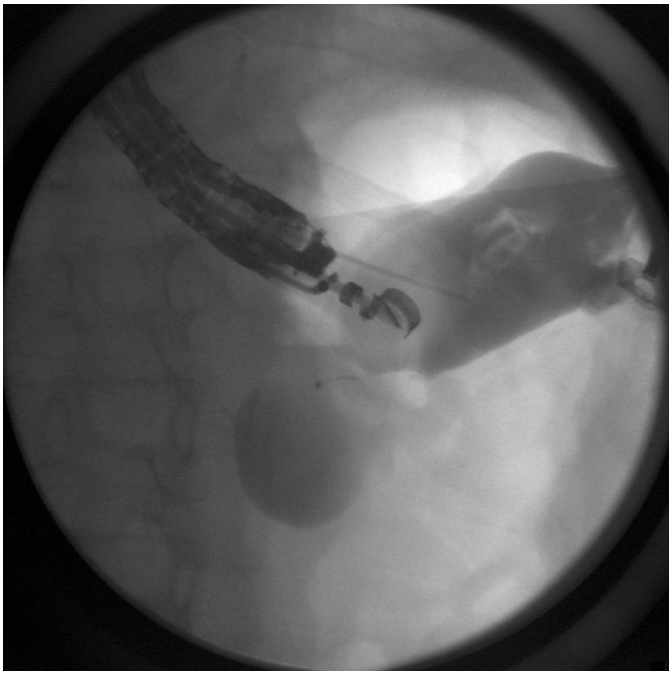

Fig. 4. EUS-guided wire cannulation of larger (disconnected) pancreatic fluid collection, which opacified only with EUS-guided transgastric contrast injection but not with transpapillary injection, consistent with ductal discommunication.

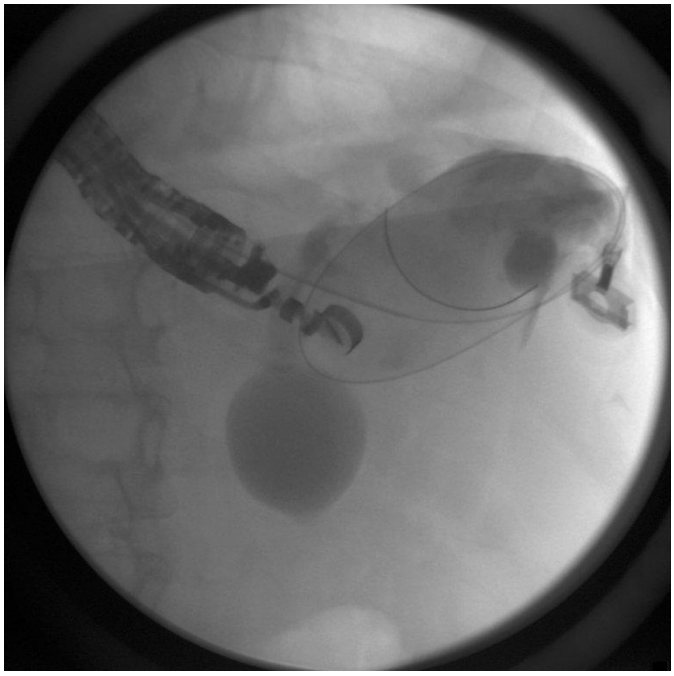

Fig. 5. EUS-guided wire cannulation of large pancreatic fluid collection.

3. Discussion

PPFs occur in 0.4%–4.5% of patients with pancreatitis.[1] PPF can arise from surgical procedures or trauma; more commonly it results from chronic pancreatitis with pancreatic duct disruption.[1–5] Transdiaphragmatic fistulous tracts allow communication between the peri-pancreatic retroperitoneal space and the pleural cavity, leading to large-volume pleural effusions.[7]

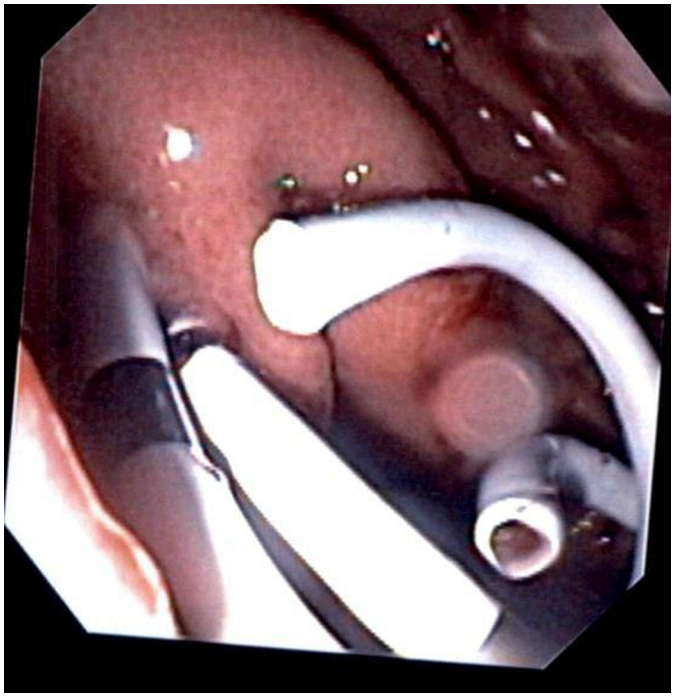

Fig. 6. Endoscopic view following EUS-guided placement of transgastric double-pigtail stents.

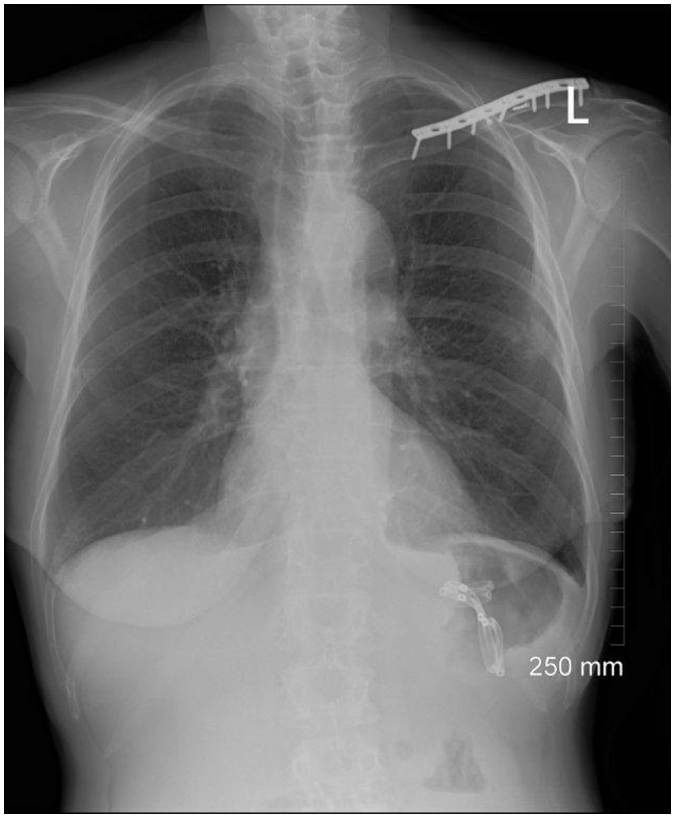

Fig. 7. Chest X-ray 5 months post endoscopic treatment of pancreatico-pleural fistula.

PPF may be diagnostically challenging because patients present with respiratory rather than abdominal symptoms.[1–5] Accordingly, chest radiography is typically the initial diagnostic study. Diagnosis of PPF can be secured by pleural fluid amylase elevation typically in the many thousands, and by radiographic imaging for a fistulous tract. The differential diagnosis of elevated pleural amylase include lung adenocarcinoma, female genital cancers, other solid organ cancers, esophageal rupture, and pancreatic disease.[3] MRCP is a useful imaging study since it allows complete imaging of the pancreatic duct, whereas ERCP cannot define ductal anatomy upstream of the disruption and associated pathology.[5,7,8]

Management of PPF includes thoracentesis, parenteral nutrition, and octreotide, which are effective in 40–60% of cases.[9–11] If conservative therapy fails, additional therapy is indicated. Options include percutaneous drainage, surgery, and endoscopic management. Endoscopic strategies strive to divert the anomalous pancreatic secretions toward the gastrointestinal lumen. When the pancreatic duct is intact, transpapillary stenting of the pancreatic duct has been employed successfully.[8,10] However, when a disconnected pancreatic duct leads to PPF as in our case, transpapillary stenting alone would only be expected to be efficacious if the disrupted duct can be bridged by the stent, a notoriously challenging undertaking.[12] In the present case EUS-guided therapy effectively addressed both the disconnected pancreatic duct and the PPF. Others have reported successful results with EUS-guided treatment for disconnected pancreatic duct syndrome.[12,13] However, to our knowledge, ours is the first reported case of EUS-guided therapy for PPF.

Surgical management of PPF entails pancreatectomy with splenectomy, pancreaticoduodenectomy, or pancreatic duct anastomosis to a loop of small intestine requiring an average hospital stay of 16 days.[14] Reported complications from surgical

management include wound dehiscence and intra-abdominal fluid collection requiring surgical drainage.[14] Successful EUS-guided treatment of PPF as described herein was accomplished with a 48-h hospital stay and avoided unnecessary loss of pancreatic or splenic function as well as the morbidity associated with laparotomy. Potential complications in the endoscopic management of PPF are empyema[15] and small bowel obstruction related to stent migration.[16] Empyema can be successfully managed with minimally invasive thoracotomy and laparotomy.[15] Small bowel obstruction due to stent migration is uncommon and often can be managed non-surgically.[16] The pigtail configuration of the stent acts as an anchor which resists migration. Material pliability, small caliber, and curvilinear poles are intrinsic properties of the double-pigtail stent which typically allow uneventful passage of migrated stents through the gastrointestinal tract.

Management of PPF requires a thoughtful multidisciplinary approach tailored to the individual patient and availability of endoscopic as well as surgical expertise. We believe our technique of EUS-guided therapy for PPF due to disconnected pancreatic duct syndrome is an appealing minimally invasive alternative to surgical therapy.

4. Conclusions

PPF is an unusual complication of chronic pancreatitis that should be recognized promptly if thoracentesis yields amylase-rich fluid. This case demonstrated effective EUS-guided therapy for a chronic PPF associated with chronic pancreatitis and disconnected pancreatic duct syndrome.

Conflict of interest

None declared.

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