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CASE REPORT

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Pancreaticopleural fistulas of different origin: Report of two cases and a review of literature

Katarzyna Wypych[1], Zbigniew Serafin[1,2], Przemysław Gałązka[3], Piotr Strześniewski[1,2], Włodzimierz Matuszczak[4], Katarzyna Nierzwicka[3], Władysław Lasek[1], Andrzej I. Prokurat[3], Marek Bąk[4]

[1] Department of Radiology and Diagnostic Imaging, Nicolaus Copernicus University, Collegium Medicum, Bydgoszcz, Poland
[2] Department of Radiology, MCD Voxel, Bydgoszcz, Poland
[3] Department of Pediatric Surgery, Nicolaus Copernicus University, Collegium Medicum, Bydgoszcz, Poland
[4] Department of Surgery, MSWiA Hospital, Bydgoszcz, Poland

Author's address: Zbigniew Serafin, Department of Radiology and Diagnostic Imaging, Nicolaus Copernicus University, Collegium Medicum, M. Skłodowskiej-Curie 9 Str., 85-094 Bydgoszcz, Poland, e-mail: serafin@cm.umk.pl

Summary

Background: Pancreaticopleural fistula (PPF), a form of internal pancreatic fistula, is a rare complication of acute or chronic pancreatitis or pancreatic trauma.

Case Report: We report two cases of PPF resulting in formation of pleural pancreatic pseudocysts. A 35-year-old male alcoholic patient with a history of recurrent episodes of acute pancreatitis was admitted due to a severe dyspnea. A CT scan showed a significant left pleural effusion with a total left lung atelectasis, compression of the mediastinum, and dislocation of the left diaphragm. A follow-up CT showed a fistula between the abdominal pancreatic pseudocyst and the left pleural cavity. The second case was a 13-year-old male patient, who was admitted for a splenic stump excision. Two weeks after the surgery the patient presented a massive pleural amylase-rich effusion. CT exam suggested a PPF, which was indirectly confirmed by a thoracoscopy.

Conclusions: PPF should be considered in cases of massive pleural effusion and encapsulated pleural fluid collections in patients with a history of acute pancreatitis and surgery involving pancreas.

Key words: acute pancreatitis • pleural effusion • pancreaticopleural fistula • splenectomy

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Background

Pancreaticopleural fistulas (PPF), a form of internal pancreatic fistulae, are rare complications of acute or chronic pancreatitis or pancreatic trauma [1,2]. PPF may enter the pleural cavity through the sternocostal triangle, the caval hiatus or directly through the diaphragm.

The presence of the PPF is rarely associated with an active acute pancreatitis. Patients with fistulae tend to have a history of chronic, relapsing inflammation of the pancreas. Other pathologies inducing formation of the fistulae include gallstones, trauma, and anatomic anomalies of pancreatic ducts [2]. The presence of the fistula can be demonstrated in magnetic resonance (MR) or computed tomography (CT) imaging as a pleural fluid collection, but the canal connecting the pleural cavity and the abdominal cavity is difficult to visualize. More definite may be the biochemical fluid analysis, demonstrating a markedly increased amylase activity.

Because of its low incidence, PPF is rarely considered as a cause of pleural effusion. We present two cases of this entity with a different clinical presentation and a different CT image.

Case Reports

Case 1

A 35-year-old male, who was addicted to alcohol and had a history of recurrent episodes of acute pancreatitis, was

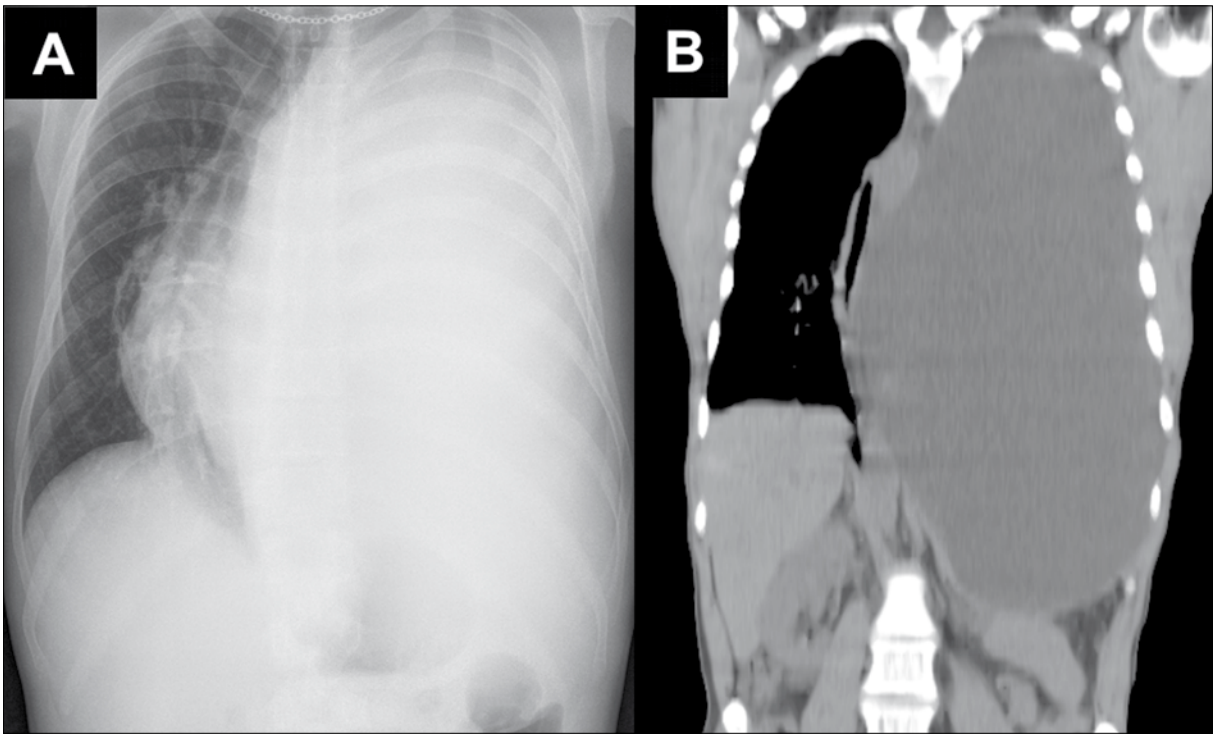

Figure 1. Case 1. Massive pleural effusion at the time of admission (**A**) on chest radiography, (**B**) on coronal CT scan reconstruction.

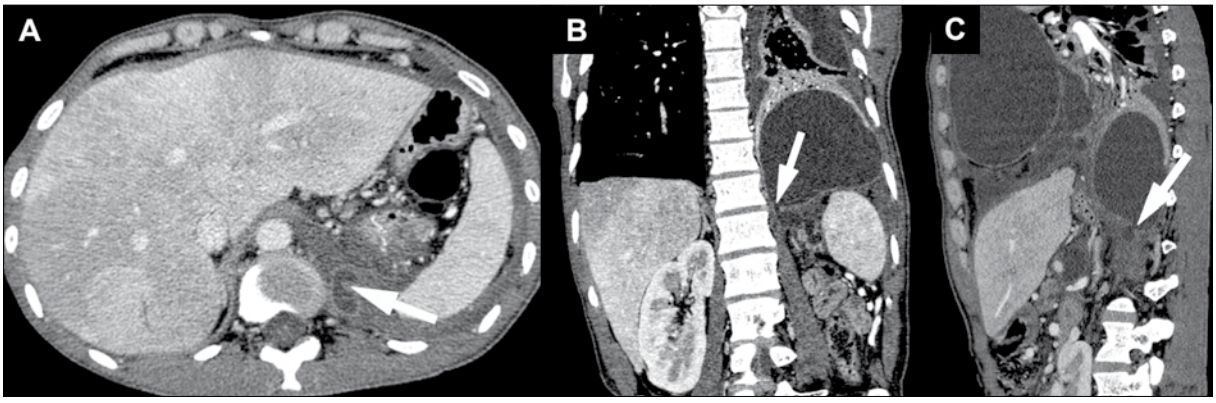

Figure 2. Case 1. Pancreaticopleural fistula (arrows) detected with the second CT examination, which was performed after the thoracocentesis: (**A**) axial image, (**B**) coronal reconstruction, (**C**) sagittal reconstruction.

admitted to the hospital for severe increasing dyspnea, cough, and general weakness. A chest X-ray revealed left-sided massive pleural effusion (Figure 1). The severe effusion was confirmed by CT examination, which also showed few abdominal fluid collections close to the pancreas. Based on those examinations, the patient was scheduled for a thoracocentesis. Laboratory analysis of the fluid revealed an elevated level of amylase (8.400 U/L). A second CT scan, which was performed after the thoracocentesis, revealed a fistula between one of the parapancreatic pseudocysts and the pleural cavity (Figure 2). The patient was scheduled for a conservative treatment but after a partial release of the symptoms he decided to discharge himself. His further history is unknown.

Case 2

A 13-year-old male with a history of hereditary spherocytosis, and a partial splenectomy carried out 4 years ago, was admitted to the hospital due to regrowth of the splenic stump and recurrent symptoms of hypersplenism. During an elective laparotomy and splenic stump excision, considerable adhesions of the stump to the left diaphragm were noted, which required a sharp dissection of the splenic tissue from the diaphragm. An early postoperative course was uneventful, and results of all routine laboratory tests were normal. On 11th postoperative day an unexpected elevation C-reactive protein level (90 mg/L) was noted without fever or any complaints.

Thoracic and abdominal sonography revealed a significant left pleural effusion without any abnormalities within the abdomen (Figure 3A). The pleural fluid was evacuated, and a laboratory analysis showed a slightly increased concentration of amylase (430 U/L). Within the next two days of observation the amount of pleural fluid was increasing. The level of amylase in fluid that was evacuated during a

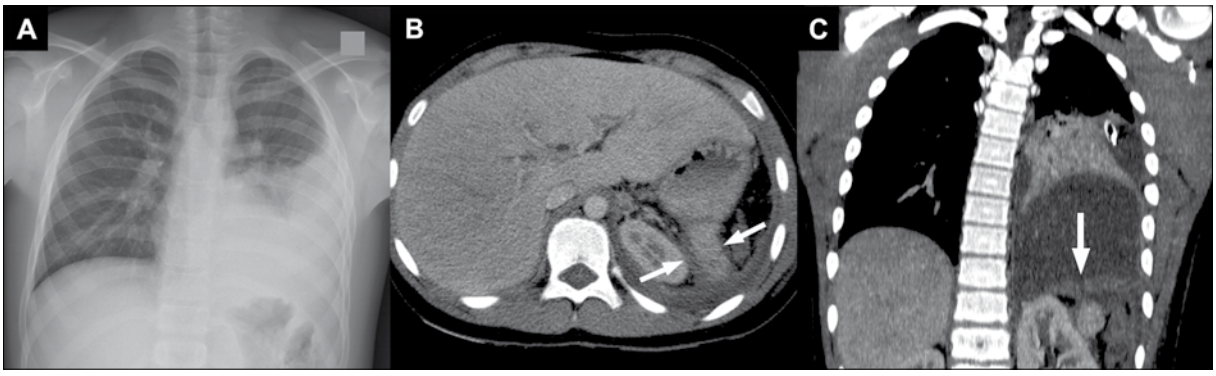

Figure 3. Case 2. (**A**) left pleural effusion, (**B**) the tail of the pancreas (arrow) located close to the diaphragm, surrounded by a limited fluid collection, (**C**) pancreaticopleural fistula (arrow).

second thoracocentesis was significantly increased (6.000 U/L), while serum and urine amylase levels remained unchanged. CT examination revealed a pleural fluid collection, and a left pulmonary atelectasis. The tail of the pancreas was seen close to the diaphragm, surrounded by a limited fluid collection (Figure 3B). Based on CT multiplanar reconstructions a radiologist raised a suspicion of PPF, which was to be located in the central part of the left diaphragm leaflet, directly above the tail of the pancreas (Figure 3C).

Due to the atypical course, and the ambiguous result of the CT examination, a left-sided thoracoscopy was undertaken, which revealed significant pulmonary-pleural adhesions, and significant inflammatory changes of the visceral and parietal pleura. The pancreaticopleural fistula was not directly seen, but the most probable locations were the region the aortic hiatus, and the central part of the left diaphragm leaflet. The patient was left on *nothing per os* regimen and TPN for 12 days, and the pleural drainage was maintained for 3 weeks. A follow-up chest CT-scan performed 2 months after the surgery showed only small pleurodiaphragmatic adhesions in the area of the costo-diaphragmatic recess.

Discussion

Pancreatic pathologies can be complicated by two types of pleural effusion. The first one is usually small, located in the left pleural cavity. The effusion is characterized by a normal amylase activity (below 100 U/L), and a low protein concentration (below 3 g/dL) [2,3]. This type is associated with acute pancreatitis, and is absorbed during recovery. The second kind of effusion is related to the presence of PPF in the course of chronic or recurrent pancreatitis, which can penetrate to the pleura, bronchi, mediastinum or pericardium due to the gradients of pressure between the abdomen and the thorax. The effusion is usually large, single-sided, recurrent, and contains a high level of amylase (usually over 1000 U/L) and proteins (over 3 g/dL) [4–6]. In the published cases of PPF the pleural fluid amylase activity ranged between 400 and 446.600 U/L [7]. A connection between the fistula and a pseudocyst, or an injured pancreatic duct can frequently be documented. These two forms of pleural effusion should be clinically recognized, in view of their different complication rate, prognosis, and treatment [2,5].

PPF is considered a rare pathology [2,3,5]. Its incidence is estimated at 0.4% in patients with pancreatitis and 4.5% in those presenting with a pancreatic pseudocyst [8]. A systematic review of the English language literature from years 1960 to 2007 by Ali et al. [7] allowed for identification of fifty-two documented cases of pancreaticopleural fistulae in the English literature. Using the same methodology for years 2008–2009, one can find additional nineteen reported cases [9–15]. Tajima et al. suggested, that the rate of the diagnosed or reported cases of pancreaticopleural fistulae is underestimated and will increase due to continual improvement in imaging technology [16].

Patients diagnosed with PPF are middle-aged (40–50 years), predominately male (83%), with chronic pancreatitis related mainly to alcohol abuse (67%) [7]. Clinical presentation of PPF varies between patients due to different level of coexisting pancreatic injury and the amount of fluid collected in the pleural cavity. According to Ali et al. [7], the most common symptoms are related to the pleural effusion. These include dyspnea in 65% of patients, cough in 27%, and chest pain in 23% of them. Abdominal pain was reported in 29% of cases. Our first patient presented a typical clinical PPF profile. The second one was a rare case of iatrogenic injury, which was not frequently reported [10]. Moreover, PPF in pediatric patients is unusual pathology as well [17].

Clinically, the massive pleural effusion with strongly increased activity of amylase and increased protein concentration in a patient with pancreatitis suggests a diagnosis of PPF [3,6,7]. However, a direct demonstration of the fistula may be difficult in a number of cases. Ultrasonography, which is an examination commonly performed in pancreatitis, was reported to visualize PPF in a limited number of patients, because of bowel loops decreasing the image quality in upper abdomen [18,19]. There was also a report of PPF diagnosis based on endoscopic ultrasound [20]. CT was performed in most studies reporting PPFs, but it was able to present the fistula only in 33–47% of cases [2,7]. The reason may limit contrast resolution of CT in the case of narrow and tortuous canal with poorly enhancing walls, which is frequently hidden within pancreatic pseudocysts [2,7,16,21].

Magnetic resonance cholangiopancreatography (MRCP) is considered a method of choice for suspected PPF [2,7,16, 22–24]. The advantages of MRCP include noninvasiveness

and the possibility to detect the fistula even in case of severe strictures of the pancreatic duct [7,24]. It allows for mapping of the ductal anatomy along with pathologic changes in adjacent structures that may provide with important information to understand the localanatomy and to plant an optimal treatment [16,25]. The sensitivity of MRCP is estimated at 80%. False negative results may be caused by multiple pseudocysts or ascites, which decrease image quality [16]. Endoscopic retrograde cholangiopancreatography (ERCP) is the second effective modality to diagnose PPF, with sensitivity of 46–78% [2,7]. Despite its invasiveness, ERCP is widely used due to the possibility for simultaneous endoscopic treatment. However, this method may be ineffective in cases of anatomic anomalies of pancreatic ducts and severe ductal strictures or obstructions [7]. The value of ERCP may be further improved by performing a CT scan after the endoscopy do determine the course of the fistula filled with contrast medium [26,27]. Such approach enables for three-dimensional analysis of the lesion and its relation to adjacent anatomic structures, which may help to plan surgical treatment [1,6,7]. It should be noted, that ERCP, either diagnostic or therapeutic, is an invasive procedure and may lead to some complications. Sut et al. reported a patient with PPF that developed in the course of MRCP-induced pancreatitis, who had to be treated surgically [10].

Recently, Tajima et al. proposed an algorithm for imaging PPF starting with CT and MRCP in all patients suspected of the fistula, with ERCP reserved only for patients with inconclusive MR or scheduled for an endoscopic treatment [16]. In our first patient the fistula could be visualized only after thoracocentesis which reduced the pleural pressure and allowed for recanalization of the fistula. Before the pleural fluid evacuation, the pressure was high enough to close the fistula in a valve-like mechanism. Therefore, we consider that in cases with such a massive pleural effusion, complicated by atelectasis and displacement of the mediastinum, the pleural fluid evacuation may help to find the PPF. In the second patient, despite the use of 64-MDCT and the known high level of pleural fluid amylase, the diagnosis was not possible with CT imaging and was established on the basis of thoracoscopy. To our knowledge, this was an unusual approach, not reported in the literature previously. Alternatively, an MR examination would be of value in this patient.

Therapeutic options for PPF include conservative treatment, endoscopic management, and surgery. Medical treatment is usually attempted for 2–3 weeks using analogs of somatostatin to reduce pancreatic secretion and pressure in pancreatic ducts [2,24]. The success rate of this approach yields 30–60% [7,28]. ERCP allows for simultaneous assessment of the fistula and for decompression of ducts. Endoscopic procedures include sphincterotomy, stone removal, balloon stricture dilatation, and stenting [2,7]. Surgery is necessary in cases of noninvasive treatment failure, which is frequent in patients with severe pancreatic duct strictures and multiple polymorphic pseudocysts [2,7,16]. Depending on the local anatomy, distal pancreatectomy, pancreatojejunostomy of fistula closure via transthoracic approach is performed [2,7,10], which was the case in our second patient. However, all the surgical methods require a previous careful imaging of both the pancreatic ducts and the surrounding structures [2,7,16].

Conclusions

In conclusion, pancreaticopleural fistula should be taken into account in case of massive pleural effusion and encapsulated pleural fluid collections in patients with a history of acute pancreatitis and surgery involving pancreas. Advanced cross-sectional imaging enables visualization of the fistulous tract, but pleural fluid amylase determination is still the most important method to diagnose this pathology.

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