SCIENTIFIC LETTERS 105

administration with symptoms of dyspnoea, fever and cough.⁵ Most patients were treated with corticosteroids, sometimes given in combination with other immunosuppressants, and the mortality rate was 29%.5

In our case, the patient made a good recovery from the lung disorder with corticosteroid treatment, and is now asymptomatic.

In conclusion, despite the fact that the most common complications are infectious, inflammatory lung involvement related to anti-TNF- α drugs should be suspected in case of respiratory symptoms or impaired gas exchange. This complication has been described with the reference drug infliximab and can also occur with the infliximab biosimilar. In order to ensure early diagnosis and management of this complication, it is important to stress that patients receiving anti-TNF- α drugs can develop DILD, particularly if they have risk factors.

The authors notified the SEFV (*Sistema Español de Farmacovigilancia* [Spanish Pharmacovigilance System]) about this case.

References

 Harbord M, Annese V, Vavricka SR, Allez M, Barreiro-de Acosta M, Boberg KM, et al. The first european evidence-based consensus on extra-intestinal manifestations in inflammatory bowel disease. J Crohns Colitis. 2016;10:239–54.

- 2. Dixon WG, Hyrich KL, Watson KD, Lunt M, Symmons DP. Influence of anti-TNF therapy on mortality in patients with rheumatoid arthritis-associated interstitial lung disease: results from the British Society for Rheumatology Biologics Register. Ann Rheum Dis. 2010;69:1086–91.
- Curtis JR, Sarsour K, Napalkov P, Costa LA, Schulman KL. Incidence and complications of interstitial lung disease in users of tocilizumab, rituximab, abatacept and anti-tumor necrosis factor α agents, a retrospective cohort study. Arthritis Res Ther. 2015:17:319.
- 4. Sen S, Peltz C, Jordan K, Boes TJ. Infliximab-induced nonspecific interstitial pneumonia. Am J Med Sci. 2012;344:75–8.
- Perez-Alvarez R, Perez-de-Lis M, Diaz-Lagares C, Pego-Reigosa JM, Retamozo S, Bove A, et al. Interstitial lung disease induced or exacerbated by TNF-targeted therapies: analysis of 122 cases. Semin Arthritis Rheum. 2011;41:256-64.
- R. Ríos León^{a,*}, A. Jaureguizar Oriol^b, A. López-Sanromán^a, D. Jiménez Castro^b, R. Nieto Royo^b, A. Albillos Martínez^a
- ^a Servicio de Gastroenterología, Hospital Universitario Ramón y Cajal, Madrid, Spain
- ^b Servicio de Neumología, Hospital Universitario Ramón y Cajal, Madrid, Spain
- * Corresponding author.

E-mail address: raquelriosleon@gmail.com (R. Ríos León). 2444-3824/

© 2018 Elsevier España, S.L.U. All rights reserved.

Pancreaticopleural fistula associated with choledocholithiasis: An infrequent complication of acute pancreatitis*



Fístula pancreatopleural en relación con coledocolitiasis: una complicación infrecuente en la pancreatitis aguda

Pancreaticopleural fistula is an uncommon but potentially serious complication of pancreatitis, with an incidence of around 0.4–4.5% of patients with chronic pancreatitis. It is more common in the alcoholic aetiology and when pseudocysts appear. It is caused by disruption in the posterior aspect of the pancreatic duct, either directly or more often through the rupture of a pseudocyst or necrotic collection, with pancreatic secretions leaking into the pleural space through the aortic or oesophageal hiatus or more rarely through the diaphragm. The most common clinical presentation is dyspnoea secondary to pleural effusion, predominantly on the left side, which develops in 75% of cases.

We present the case of a 72-year-old male with a history of acute necrotising pancreatitis in April 2013 which required admission to the ICU due to sepsis secondary to infected necrosis. The patient developed spontaneous intestinal fistulisation of the collection, with thrombosis of the splenic vein and superior mesenteric vein, and development of left segmental portal hypertension with splenomegaly and collateral circulation. Subsequently, he had to be admitted in October 2015 and September 2016 for fever secondary to a necrotic collection treated conservatively with antibiotic therapy due to the presence of intestinal fistulisation. Clinically, he made good progress and did not require an invasive approach, so he was discharged to undergo follow-up as an outpatient. In January 2017 he was admitted for dyspnoea secondary to left pleural effusion, requiring pleural drainage, which showed fluid compatible with exudate. In light of elevated pleural amylase (9317 U/l), CT of the chest/abdomen was requested, showing the spread of the necrotic collection from the tail of the pancreas to the head towards the subphrenic region, with fistulisation through the diaphragm to the left pleural space. Juxtapapillary choledocholithiasis not seen in previous explorations was also observed, and confirmed by magnetic resonance (MR) cholangiography. Given the stability of the patient, treatment with octreotide (100 µg/8 h) was started, achieving clinical improvement, although the pleural effusion persisted. Because of the distal location of the pancreatic duct rupture associated with the necrotic collection, after considering an endoscopic approach to the fistula, the decision was finally made to use a combined approach with endoscopic retrograde cholangiography (ERCP) to extract

^{*} Please cite this article as: Carrión-Martín L, Lucendo-Jiménez L, Ordieres-Ortega L, Piqueras-Ruíz S, Cano-Ballesteros JC, Hernando-Alonso AI, et al. Fístula pancreatopleural en relación con coledocolitiasis: una complicación infrecuente en la pancreatitis aguda. Gastroenterol Hepatol. 2019;42:105–106.

106 SCIENTIFIC LETTERS

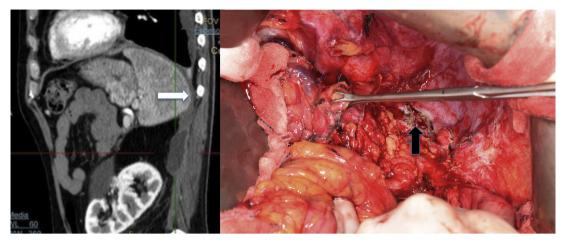


Figure 1 Lateral view of the abdominal CT showing the retroperitoneal collection which is fistulising into the pleural space (white arrow) and image of the surgical intervention (with fistulous orifice, black arrow).

the choledocholithiasis, followed by a surgical intervention with distal pancreatectomy and splenectomy in view of the presence of left segmental portal hypertension and the contact of the collection with the visceral aspect of the spleen (Fig. 1). The patient made satisfactory progress, the pleural effusion resolved and he is currently asymptomatic.

Pancreaticopleural fistula represents both a diagnostic and therapeutic challenge. Diagnosis is based on the analysis of pleural fluid and imaging tests. Pleural fluid is high in proteins (>3 g/dl) and has a high amylase concentration (>1000 U/l), although these findings can also be found in other aetiologies (infection, cancer, oesophageal perforation, cirrhosis, etc.), according to which the differential diagnosis must be made. It may be caused by the expansion or rupture of a pseudocyst or pancreatic collection or the direct disruption of the duct of Wirsung. The diagnostic imaging tests used are abdominal CT, ERCP and MR cholangiography, with a sensitivity of 47%, 78% and 80%, respectively, with MR cholangiography being the tool of choice due to its non-invasive nature and greater sensitivity.

First-line treatment consists of bowel rest and somatostatin analogues to reduce pancreatic secretions. If there is no response, a definitive approach should be considered, either endoscopic, with ERCP (with the aim of decompressing the duct of Wirsung, of greater benefit in partial, single disruptions at the proximal end and without large associated collections),⁴ or surgical. Delaying definitive treatment can increase the risk of complications.⁵ In our case, after the conservative treatment had failed, our approach involved the combination of endoscopic extraction of the choledocholithiasis followed by surgery for the definitive resolution of the collection located in the tail of the pancreas. Surgery also enabled us to resolve the segmental portal hypertension.

There were several unusual factors in this case: the fistulisation through the diaphragm (less common than through the aortic hiatus); the choledocholithiasis as a possible trigger of the fistula, which was not immediately apparent because of the increased intraductal pressure in the pancreatic excretory system; and the surgical approach that enabled both the fistula and the segmental portal hypertension to be resolved.

Pancreaticopleural fistula should be suspected in patients with a history of pancreatitis who present with pleural effusion with elevated amylase. The treatment approach is determined by the location of the duct of Wirsung disruption, the presence of pseudocysts, and the patient's comorbidity.

References

- Machado NO. Pancreaticopleural fistula: revisited. Diagn Ther Endosc. 2012;2012:815476.
- Aswani Y, Hira P. Pancreaticopleural fistula: a review. JOP. 2015;16:90-4.
- Wakefield S, Tutty B, Britton J. Pancreaticopleural fistula: a rare complication of chronic pancreatitis. Postgr Med J. 1996;72:115-6.
- 4. Ali T, Srinivasan N, Le V, Chimpiri AR, Tierney WM. Pancreaticopleural fistula. Pancreas. 2009;38:26–31.
- King JC, Reber HA, Shiraga S, Hines OJ. Pancreatic-pleural fistula is best managed by early operative intervention. Surgery. 2008;147:154-9, http://dx.doi.org/10.1016/j.surg. 2009.03.024.
- L. Carrión-Martín a,*, L. Lucendo-Jiménez a,
- L. Ordieres-Ortega^b, S. Piqueras-Ruíz^b,
- J.C. Cano-Ballesteros^b, A.I. Hernando-Alonso^a,
- R. Bañares a,c,d
- ^a Servicio de Aparato Digestivo, Hospital General Universitario Gregorio Marañón, Madrid, Spain
- ^b Servicio de Medicina Interna, Hospital General
- Universitario Gregorio Marañón, Madrid, Spain ^c Facultad de Medicina, Universidad Complutense de Madrid, Madrid, Spain
- ^d Centro de Investigación Biomédica en Red de Enfermedades Hepáticas y Digestivas (CIBEREHD), Spain
- * Corresponding author.

E-mail address: laura.carrion@salud.madrid.org

(L. Carrión-Martín).

2444-3824/

© 2018 Elsevier España, S.L.U. All rights reserved.