EXTENSIVE PROPAGATION OF A PANCREATIC PSEUDOCYST ALONG THE LOWER LIMB THROUGH THE SCIATIC FORAMEN

B. Coulier¹, Ph. Maldague², I. Bueres-Dominguez², A. Ramboux³, F. Pierard³, B. Bienfait⁴

The extremely rare extensive propagation of a giant retroperitoneal pancreatic pseudocyst into the posterior compartment of the lower limb as far as the knee is reported. The extension was found producing through the sciatic foramen and the full diagnosis was made by MDCT. A complete healing was progressively obtained in the 78-year old female after a six months period of sequential multidisciplinary therapeutic approach comprising combined medical and surgical intra-abdominal and external drainage.

Key-word: Pancreas, cysts.

A pancreatic pseudocyst may rupture into the peritoneal cavity, an abdominal viscus, or the retroperitoneum with extension in either the supradiaphragmatic or subdiaphragmatic direction. Pseudocyst extension has been described and reported in the pelvis, mediastinum, pleural space and even in the neck (1-9).

We present a rare case in which a large retroperitoneal pseudocyst emerging from the tail of the pancreas migrated from the left anterior pararenal space to the infrarenal space, than to the pelvis to finally very unusually leave the abdomen through the sciatic foramen to extensively extend along the thigh up to the knee.

The report also emphasizes the results of a complex multidisciplinary therapeutic approach comprising the association of medical and intra-abdominal and external surgical drainage.

Case report

A 78-year old female was admitted in the department of gastroenterology with complaints of anorexia, alimentary vomiting, diarrhea, alteration of hepatic tests and a CRP level at 290 mg/l.

Contrast enhanced abdominal CT (not illustrated) demonstrated a major necrotic exudative pancreatitis. There were diffuse peripancreatic effusions but the predominant extensive necrotic effusion had migrated along the full length of the left paracolonic gutter to reach the pelvic floor. Extensions were also visible within a large preexisting hiatal hernia. A small amount of free ascitis was present.

After exclusion of other causes such as drugs or alcohol abuse, the origin of the pancreatitis was finally attributed to biliary stone migration. This hypothesis was reinforced by the ultrasound diagnosis of a multilitiasic gallbladder.

Retrograde wirsungography demonstrated a fracture of the caudal Wirsung with leakage of contrast. Sphincterotomy was first performed in order to reduce the pressure.

The clinical evolution was first favorable with reduction of clinical symptoms, decrease of the CRP level and progressive normalization of the hepatic and pancreatic tests. The patient was discharged at home with somatostatin.

Three weeks later, the patient was readdressed with complaints of massive progressive edema of the left leg and thigh suggesting first deep venous thrombosis. During Doppler ultrasound examination (not illustrated), the veins appeared free from thrombosis but numerous subcutaneous collections were found along the external side of the knee. These collections were prolonged by deep tubular intermuscular collections developed along the posterior side of the Additional deep collections were found in the buttock.

A new contrast enhanced continuous MDCT was performed from the top of the gastric hiatal hernia to below the knee. The multiple peripancreatic effusions had now formed large mature pseudocysts (Fig. 1). The dominant feature was a very long pseudocyst developing from the pericaudal pancreatic area to the left anterior pararenal area and then descending along the

infrarenal retroperitoneal spacer to reach the pelvis and largely expand in the retroperitoneal presacral area (Fig. 2). A digitiform extension of this pseudocyst trespassing the greater sciatic foramen was clearly visible. It was prolonged with gluteal collections - around the lesser trochanter and ischiatic tuberosities - and long perineural and perivascular tubular collections extending between the muscles of the posterior compartment of the thigh (Fig. Subcutaneous collections and effusions were found extending to the posterior and external side of the knee and were responsible of diffuse clinical swelling and edema.

A new retrograde wirsungography showed a persistent disruption of the distal caudal wirsung (Fig. 2C). A prosthesis was encased within the canal but failed to cover the site of the disruption. Thus no significant reduction of the main left abdominal pseudocyst was obtained observed after one week but during this time the collections of the thigh continued to expand drastically.

The high hydrostatic pressure exercised by the major volume and weight of the main left abdominal pseudocyst was considered responsible of the continuous pushing of fluid in the tight through the sciatic foramen. Active intra-abdominal drainage was thus proposed to reduce this pressure. Cysto-jejunostomy was performed at the lowest possible level of the pseudocyst. The pseudocyst was surgically maximally washed and drained before being anastomosed to a Roux-en-Y jejunal loop.

This internal drainage was effective on the abdominal component of the pseudocyst but remained without effect on the collections of the thigh. These continued to inflate with aggravation of the biologic inflammatory syndrome and development of a septic state (Fig. 3D). The collections of the thigh were thus directly

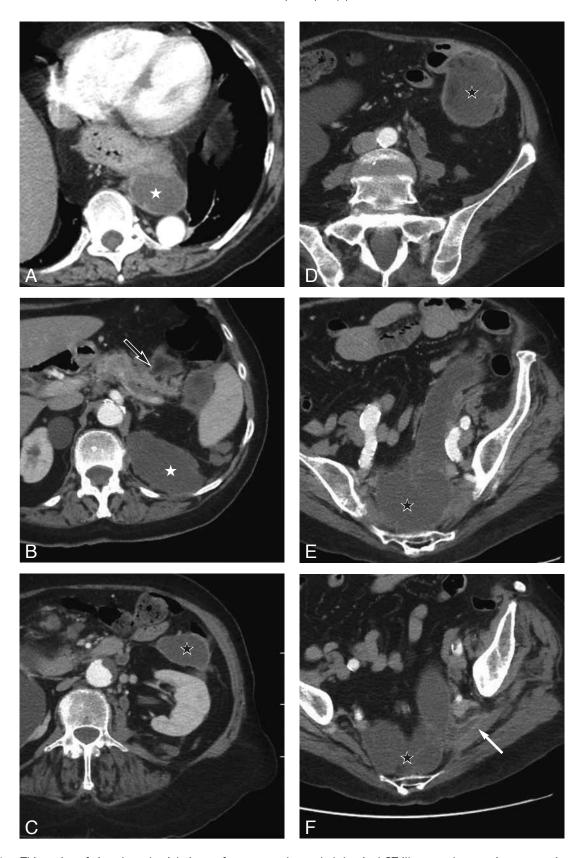
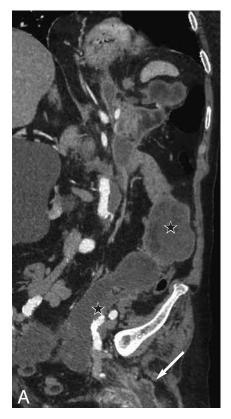


Fig. 1. — This series of six selected axial views of contrast enhanced abdominal CT illustrate the massive retroperitoneal extension of the long pseudocyst at the time of readmission of the patient three weeks after the initial onset of pancreatitis. The giant pseudocyst has developed from caudal pancreatitis (black arrow on B). A cranial extension is already visible within a large hiatal hernia (white star on A). Another extension reaches the left upper retrorenal space (white star on B). The most important extension develops in the left anterior pararenal space (black star on C), follows the anterior infrarenal space (black arrow on D) and distally extensively expands in the retroperitoneal presacral space (black star on E and F). A small digitiform expansion is already found through the greater sciatic foramen (white arrow on F).



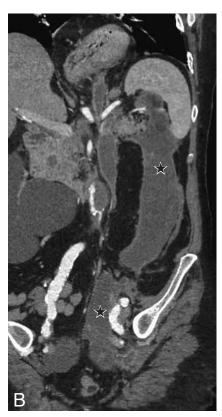




Fig. 2. — Curvilinear coronal oblique reformatted views (A&B) through the all length of the left abdomen illustrates the extensive craniocaudal retroperitoneal extension of the long pseudocyst (black stars) described in Fig. 1. The digitiform expansion through the greater sciatic foramen is pointed (white arrow on A). Retrograde wirsungography (C) illustrating a small disruption of the caudal Wirsung with leakage of contrast (black arrow).

surgically approached by multiple fasciotomies and debridements. High amylase and lipase levels were found in the aspired fluid. This direct drainage was effective but a large persistent subcutaneous open detachment failed to shut and secondarily appeared superinfected necessitating a new debridement after 2 months.

About nine months after the initial episode of pancreatitis, a control CT (not illustrated) showed a nearly complete decline of the abdominal pseudocysts and a complete resolu-

tion of the extensions in the lower limb.

Discussion

Pancreatic pseudocysts are local complications of acute or chronic pancreatitis (1-2). They are localized fluid collections which are rich in amylase and other pancreatic enzymes and are surrounded by a wall of fibrous tissue that is not lined by epithelium (2-3). Pseudocysts are connected with the pancreatic duct system either as a direct communi-

cation or indirect via the pancreatic parenchyma. They are cause by pancreatic ductal disruption following increased pancreatic ductal pressure, either due to stenosis, calculi or protein plugs obstructing the main pancreatic duct or as a result of pancreatic necrosis (2-5).

Pseudocyst formation is less common after acute (incidence of 7%) compared to chronic pancreatitis (incidence of 30 to 40%) and it is more common after alcohol-induced than after no-alcohol-related pancreatitis (6-7).

In chronic pancreatitis the pathogenesis of pseudocyst formation may result as a consequence of an acute exacerbation of the underlying disease and/or blockage of a major branch of the pancreatic duct by a protein plug, calculous of localized fibrosis.

After an episode of acute pancreatitis (7), the acute fluid collection must persist more than 4-6 weeks and be well-defined by a wall of fibrous or granulation tissue to be catalogued as an acute pseudocyst.

Most pseudocysts resolve with supportive medical care comprising intravenous fluids, analgesics and antiemetics, low fat diet, nasoenteral feeding or total parenteral nutrition (2). Choice is based on availability and local preferences. The rationale of using octreotide as a therapy for pseudocyst is that it will decrease pancreatic secretions and aid in pseudocyst resolution.

Larger cysts are more prone to become symptomatic or cause complications (8). However the size of the pseudocyst alone is not a sufficient indication for drainage.

Complications of pseudocysts (2-3) may be numerous comprising splenic complications (hemorrhage, infarction, sepsis and splenic vein thrombosis), hemorrhage due the erosion of a major vessel, pseudoaneurysms, infection, gastric outlet or biliary obstruction, biliary complications resulting in obstructive jaundice, portal hypertension and finally rupture which may produce spontaneously into the gastrointestinal tract (9), but also within the peritoneal cavity or along retroperitoneal spaces.

With rupture of a pancreatic cyst, pancreatic fluid classically dissects along the anatomic pathways and boundaries that provide the path of least resistance (10). The lesser sac and the anterior pararenal space are the most common sites of pancreatic fluid collection. There are reports of pseudocysts expanding into the

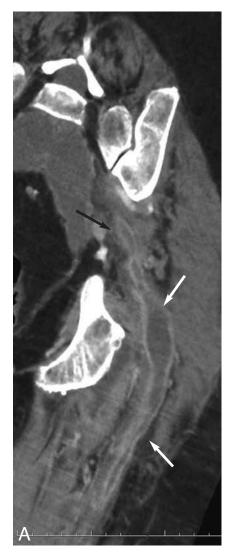






Fig. 3. — Contrast enhanced coronal MPR views (A to C) of the left buttock and thigh obtained just after the ultrasound examination of the left thigh and leg (not illustrated). Diffuse swelling and subcutaneous effusions extend to the posterior and external sides of the knee and are found being in relation with long perineural and perivascular tubular collections (white arrows on A & B) extending between the muscles of the posterior compartment of thigh. The extension has produced through the greater sciatic foramen (black arrow on A). These ramified collections will continue to inflate (white stars on C) during the hospitalization and will persist after efficacious large surgical drainage of the abdominal pseudocyst. This situation will impose specific surgical drainage of the thigh.

pelvis and involving the groin and scrotum (11-13) or involving the psoas and secondarily the thigh via the inguinal canal (10) or expanding along the fascia lata. Extensions of pancreas necrosis into the mediastinum (14-16) are also extremely rare and may produce via the aorta or the esophageal hiatus, through the diaphragmatic crura or through the erosion in diaphragm. Mediastinal extensions of pseudocysts may exceptionnaly be prolonged up to the neck (17) by different three routes.

To our knowledge our reported case illustrating an extensive propagation of a pseudocyst into the posterior compartment of the lower limb

to below the knee and producing through the sciatic foramen has not been described before.

Most authors recommend drainage for pseudocysts greater than 6 cm in size that persists for longer than 6 weeks (3).

Various options of drainage are possible. The use of percutaneous drainage progressively declines because of the discomfort of an external drain and the high risk of infections (18). Endoscopic transpapillary stenting is a useful procedure when a communication between the pancreatic duct and pseudocyst can be demonstrated.

Direct drainage of pseudocysts may be accomplished by providing a

communication between the cyst and the stomach or small bowel. Transpapillary or direct endoscopic drainage across the stomach or duodenum are now the preferred therapeutic approach because it is less invasive, avoids the need of an external drain and has a high long-term success rate (3).

Cystojejunostomy is felt by some authors to be associated with a low complication rate and a lower incidence of recurrence (3).

Endoscopic transpapillary stenting unfortunately failed in the reported case because the ductal leakage was too distally situated on the pancreatic tail. On the contrary, cystojejunostomy appeared effective to considerably reduce the volume of the abdominal retroperitoneal portion of the massive pseudocyst but revealed ineffective for the component extending in the lower limb. For this reason it appeared necessary to complete the drainage of the thigh by multiple fasciotomies and debridements.

References

- Xiao B., Zhang X.M., Tang W., et al.: Magnetic resonance imaging for local complications of acute pancreatitis: a pictorial review. World J Gastroenterol, 2010, 16: 2735-2742.
- Habashi S., Draganov P.V.: Pancreatic pseudocyst. World J Gastroenterol, 2009, 15: 38-47.
- Sanfey H., Aguilar M., Jones R.S.: Pseudocysts of the pancreas, a review of 97 cases. Am Surg, 1994, 60: 661-668.
- 4. Gumaste V.V., Pitchumoni C.S.: Pancreatic pseudocyst. *Gastro-enterologist*, 1996, 4: 33-43.
- Imrie C.W., Buist L.J., Shearer M.G.: Importance of cause in the outcome of pancreatic pseudocysts. Am J Surg, 1988, 156: 159-162.

- Parks R.W., Tzovaras G., Diamond T., Rowlands B.J.: Management of pancreatic pseudocysts. Ann R Coll Surg Engl, 2000, 82: 383-387.
- Boerma D., Obertop H., Gouma D.J.: Pancreatic pseudocysts in chronic pancreatitis. Surgical or interventional drainage? Ann Ital Chir, 2000, 71: 43-50.
- Yeo C.J., Bastidas J.A., Lynch-Nyhan A., et al.:The natural history of pancreatic pseudocysts documented by computed tomography. Surg Gynecol Obstet, 1990, 170: 411-417.
- Coulier B., Maldague P., Bueres-Dominguez I.: Spontaneous gastric drainage of a pancreatic pseudocysts. JBR-BTR, 2009, 92: 61.
- Lye D.J., Stark R.H., Cullen G.M., Wepfer J.F.: Ruptured pancreatic pseudocyst: extension into the thigh. *AJR*, 1987, 149: 937-938.
- Diego B.R., Tubet A.C., Baños G.J., et al.: Scrotal mass as result of the extension of a pancreatic pseudocyst. Actas Urol Esp, 2008, 32: 261-264.
- Gutiérrez Macías A., López Capel A., Eguía Larrea M., et al.: Inguinal tumor as the first manifestation of a pancreatic pseudocyst]. Gastroenterol Hepatol, 1998. 21: 486-488.

- Karantanas A.H., Sandris V., Tsikrika A., et al.: Extension of pancreatic pseudocysts into the neck: CT and MR imaging findings. AJR, 2003, 180: 843-845.
- Gupta R., Munoz J.C., Garg P., Masri G., et al.: Mediastinal pancreatic pseudocyst – a case report and review of the literature. MedGenMed, 2007, 9: 8.
- Sadat U., Jah A., Huguet E.: Mediastinal extension of a complicated pancreatic pseudocyst; a case report and literature review. J Med Case Reports, 2007, 1: 12.
- Wechalekar M., Falodia S., Gamanagatti S., Makharia G.K.: An extension of pancreatic pseudocysts in the neck. *Pancreas*, 2007, 34: 171-173.
- Farman J., Kutcher R., Dallemand S., et al.: Unusual pelvic complications of a pancreatic pseudocyst. Gastrointest Radiol, 1978, 3: 43-45
- Heider R., Meyer A.A., Galanko J.A., Behrns K.E.: Percutaneous drainage of pancreatic pseudocysts is associated with a higher failure rate than surgical treatment in unselected patients. *Ann Surg*, 1999, 229: 781-787.