Atypical pancreatic pseudocysts

By K. Oleszczuk-Raszke, Z. Domanski and K. Niezabitowski

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The authors present three cases of atypical pancreatic pseudocysts, occurring in the course of chronic pancreatitis: intrasplenic pseudocyst, pseudocyst in the hilus of spleen and bleeding into the pseudocyst of the tail of pancreas. Such cysts are extremely difficult, or even impossible to be recognized by conventional clinical and radiological methods. Located in the parenchyma or in the hilus of spleen, they are formed by means of fistulization along the lienorenal ligament. In every case when the chronic pancreatitis coexists with the splenic cyst, the intrasplenic localization of the pancreatic pseudocyst should be taken into consideration. The cyst in the hilus of spleen may imitate intraperitoneal mass and its relation with the pancreas may be recorded angiographically only. Angiography is also the basic diagnostic method in the haemorrhage into the pseudocyst. Bleeding into the pseudocyst has a form of vascular blush appearing during arteriography. The blush increases its intensity and persists after an examination. Early diagnosis in atypical pseudocyst is vital for the patient's fate.

Until recently, pancreatic pseudocysts occurring in about 10% of chronic pancreatitis (9) were radiologically diagnosed on the basis of indirect symptoms exclusively. The introduction of angiographic diagnostics into the studies of the pancreas facilitated both a direct insight into the organ and a much more precise diagnosis of the pseudocysts. However, the indirect symptoms as well as the directangiographic picture may lead to misdiagnosis when atypical pseudocysts or their complications are not taken into consideration.

Although sporadical, complications in the form of cyst fistulous extension to the adjacent organs, even to peritoneal cavity (3), rupture of the spleen (1, 6) or internal haemorrhages (2) in the course of chronic pancreatitis, have been known to surgeons for many years.

Also cases of dissection into the retroperitoneum (9), mediastinum (4), thorax (8) and scrotum (12) have been reported. Although rare and not known enough, fistulization into the spleen, with subsequent development of intrasplenic or perisplenic pseudocyst, has been described (9, 12).

As such cases are clinically and radiologically nonspecific and uncharacteristic, they may be recognized arteriographically only. Complication in the form of bleeding into the pseudocyst is also difficult to be diagnosed. This accounts for a small number of reports on correct preoperative diagnoses of pseudocysts either atypically located or complicated (5, 7). The majority of the cases published so far have been recognized surgically or anatomopathologically only. In our paper we present three cases of atypical pancreatic pseudocyst, diagnosed on the basis of arteriography: intrasplenic pseudocyst, perisplenic pseudocyst in the hilus of the spleen and bleeding into the pseudocyst.

Report of cases

Case I. A 45-year-old woman was admitted to the hospital with the symptoms of chronic pancreatitis. An oral barium examination revealed no abnormality. Splenomegaly appeared in the course of the disease. Selective celiac and superior mesenteric arteriography revealed the chronic pancreatitis and avascular mass (15 × 8 cm), in the external part of the spleen. The normal splenic tissue, as well as the main and intrasplenic branches of splenic artery were displaced medially (Fig. 1a and b). Radiologically, intrasplenic pseudocyst of the pancreas was recognized. At operation, a large, intrasplenic cyst, chronic pancreatitis, and dense fibrotic adhesions, enveloping both the tail of the pancreas and the spleen were found. A splenectomy and distal pancreactectomy were performed. The cyst was filled with yielded fluid, containing pancreatic enzymes (diastase determination was 8888 Wohlgemuth's units, lipase determination was — 0,6

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Case II. A 23-year-old woman admitted to the hospital due to a palpable mass in the left upper quadrant. An upper gastrointestinal examination revealed distinct displacement of the stomach posteriorly, which suggested intraperitoneal mass (Fig. 2a). Selective splenic arteriography revealed avascular mass (8 cm in diameter) in the region of the hilus of spleen, as well as straightening and displacement of pancreatic branches in the region of the tail of pancreas (Fig. 2b). Moreover, large splenic collateral veins were opacified indicating splenic vein obstruction (Fig. 2c). Pancreatic pseudocyst in the hilus of spleen was recognized. A surgical procedure was performed and the cyst with the distal part of pancreas and the spleen was removed. At operation, the radiological diagnosis was confirmed.

Case III. A 20-year-old man admitted to the hospital because of acute pancreatitis. Some weeks after the acute symptoms regressed, selective splenic arteriography and hepatic arteriography were carried out. Chronic pancreatitis and obstruction of the splenic vein were diagnosed. As soon as the improvement was achieved, the patient was discharged. After four months, the patient was readmitted to the hospital on the ground of palpable mass in the left upper quadrant. An upper gastrointestinal examination demonstrated retrogastric mass, displacing the body of stomach anteriorly. Since a pancreatic tumor was suspected another arteriography was performed. Selective splenic, hepatic, and upper mesenteric arteriography revealed again findings of chronic pancreatitis and obstruction of the splenic vein. Furthermore, during the arterial phase of selective splenic arteriography there appeared a well-circumscribed, regular, oval blush (7 × 9 cm) in the region of the tail of pancreas, (Fig. 3a) increasing its intensity in the late phase of arteriography. This blush was seen in the whole series of selective hepatic arteriography, carried out after the interval of 20 minutes (Fig. 3b). A haemorrhage to the pseudocyst of the tail of pancreas was diagnosed and later confirmed surgically.

Discussion

The presented cases indicate the possibility and the value of roentgenographic studies in the diagnosis of atypical pancreatic pseudocyst. Taking into account the fact that such cysts occur quite rarely and are uncharacteristic, sometimes their indirect symptoms being even misleading, it is essential for the radiologist to get acquainted with the etiopathogenesis of that disease and with criteria facilitating correct diagnosis.
Pancreatic pseudocysts, filled with proteolitic enzymes, have the tendency to extend along the soft tissues, and they may form fistulisations even with remote organs. The anatomical vicinity of the tail of the pancreas and the spleen provides conditions for the cyst to develop in the hilus of the spleen and even in the splenic parenchyma. A natural pathway for extension of pancreatic secretions from the tail of the pancreas into the spleen, containing both the tail of the pancreas and the splenic vessels, entering the hilus. Correct diagnosis of atypically localized pancreatic pseudocysts is extremely difficult, even when the cases are clinically typical, because the roentgenograms of gastrointestinal tract lack the characteristic displacement of the neighboring organs. The only symptom may be splenomegaly, as it was observed in case 1.

Ramer suggests that in differential diagnosis of all unclear masses in the upper left quadrant, the atypical pancreatic pseudocyst should be taken into consideration, particularly in every case of pancreatitis, coexisting with the splenic cyst (9). In case 1 the correct diagnosis was facilitated by the vascular findings of chronic pancreatitis (Fig. 1a), coexisting with the cyst in the splenic parenchyma (Fig. 1b). It may seem strange that there is a relatively small number of reports on intrasplenic cyst of the pancreas, despite the anatomical neighborhood of these organs. It seems correct to accept Warshaw's hypothesis that undiagnosed intrasplenic pseudocyst of the pancreas is lying at the base of, very often described, spleen rupture in the course of pancreatitis (12). Such a cyst is, however, possible to be diagnosed only by means of arteriography. Pancreatic pseudocyst in the hilus of spleen also fails to be diagnosed on the basis of the indirect findings provided by the examination of gastrointestinal tract, as well as by other
conventional, clinical and radiological investigations since it imitates the intraperitoneal masses. Only the arteriography enables a relation of the cyst with the pancreas to be determined. In our II case, the cyst of the tail of pancreas, resting in the hilus of spleen, clinically manifested by palpable mass in the upper left quadrant, caused a displacement of stomach posteriorly (Fig. 2a). Thus, the barium gastrointestinal examination could not lead to establishing a diagnosis of cyst originating from the pancreas. The superselective lenal arteriography revealed a cyst in the hilus of the spleen, however in the area being supplied by the pancreatic branches (Fig. 2b). Obstruction of the splenic vein was demonstrated, too (Fig. 2e). It is emphasized in the literature that the obliteration, resulting from compression or thrombosis, of the lenal vein appears to be one of the first symptoms of pancreatic pseudocyst in the hilus of the spleen (10). Such a cyst, if unrecognized and treated incorrectly may lead to serious complications, including the dissection and perforation to the peritoneal cavity (3).

Every perforation and drainage of the cyst can, moreover, be followed by the danger of internal haemorrhage (2). The proteolitic action of enzymes filling the pancreatic pseudocyst, may be responsible for the erosions in the wall of adjacent artery and subsequently, for the bleeding into the cyst. This is a rare complication and before the arteriographic study era, it was unrecognizable without a surgical procedure. Despite the development of angiographic diagnostics it still remains a poorly known entity, although the angiographic picture is rather characteristic: the bleeding to the cyst becomes evident in the form of regular blush appearing approximately 3 seconds after the administration of contrast medium, and increasing its intensity in the following seconds (6). As it is apparent from individual reports, the characteristic focal, vascular blush may occasionally represent other common or rarely-seen entities, and it must be differentiated primarily from islet-cell tumor of the pancreas, accessory spleen, hyperplastic lymph node, focal nodular hyperplasia of the liver and aneurysms as well (4, 6, 10).

It is obvious that such a complication of the cyst cannot be diagnosed without resorting to angiographic study and in order to locate the source of bleeding it is necessary to use superselective method.

In Warshaw's opinion, the most frequent source of bleeding to the pseudocyst is the splenic artery (12). The French authors (7) collected 6 cases of this unusual complication, in which the source of bleeding were the erosions in the splenic or gastroduodenal artery. However, the bleeding in the described cases was much smaller than that in our investigation. The source of bleeding can rarely be located unequivocally, preoperatively even if the superselective method of arteriography is applied. The source of bleeding has failed to be determined both in our cases and in the majority of other so far described.

The recognition of bleeding into the pseudocyst is of vital importance as it necessitates the surgical procedure. Conservative treatment or drainage of the cyst similarly as in spontaneous perforation, may lead to exsanguination. Violent drainage of the cyst reduces the pressure on the wall of the altered vessel thus provoking the bleeding (2). The radiologist is almost totally responsible for the diagnosis since only the angiographic study provides data for the recognition of atypical pancreatic pseudocysts. Relatively early diagnosis and the appropriate treatment may prevent from serious complications and all the consequences.

Conclusions
1. In all the cases of chronic pancreatitis, the possibility of pseudocyst atypically located or complicated by bleeding should be considered.
2. Pancreatic pseudocyst in the splenic parenchyma or in the hilus of the spleen, and bleeding to the pseudocyst as well, may be diagnosed radiologically, even if the clinical picture is nonspecific.
3. Indirect symptoms, observed in the barium examination of the gastrointestinal tract, are useless for the diagnosis of this type of alterations leading thus to false conclusions.
4. Selective celiac, splenic, hepatic and superior mesenteric arteriography is the basic method for the diagnosis of the atypical pancreatic pseudocyst.

Fig. 2e. Indirect splenoportography in the same case. Large, tortuous venous collaterals from the spleen indicating splenic vein obstruction. Vena porta is opacified by collateral circulation.
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Fig. 3a. Case III. Selective splenic arteriography in the case of bleeding into the pseudocyst of the tail of pancreas in course of chronic pancreatitis. Arterial phase. Regular, oval blush (7 - 9 cm) in the tail of pancreas.

Fig. 3b. Selective hepatic arteriography in the same case. Irregularity of vessels in the head of pancreas indicating chronic pancreatitis (arrows), and a vascular blush which persists after splenic arteriography (about 20 min).

Literature


K. Oleszczuk-Raszke,
I. Dep. of Roentgenological Diagnostics of Radiology Institute,
Pomeranian Medical Academy in Szczecin,
Szczecin/Poland