A pancreatic pseudoaneurysm is a rare vascular malformation within the pancreas or in its immediate vicinity, resulting from vessel wall damage due to acute or chronic pancreatitis. Rupture of such malformation leads to a life-threatening hemorrhage. The treatment requires a precise and early diagnosis, indicating the need for surgical or conservative intervention.

In this paper, a rare case of a pseudoaneurysm of the pancreaticoduodenal artery causing recurrent gastrointestinal bleeding and portal hypertension is presented. The malformation was discovered by Doppler ultrasound and computed angiotomography of the abdomen, and the diagnosis was confirmed upon surgical intervention. The bleeding vessel was closed by a stick tie from within the pseudoaneurysm lumen. The portal hypertension was relieved by a partial resection of the pseudoaneurysm and the fibrosed pancreas. The postoperative course was uneventful.

A summary has been made of the available reports on pancreatitis complication presenting as fluid spaces and the causes of GI bleeding in the course of this disease have been reviewed.

**Key words:** pancreatic pseudoaneurysm, recurrent gastrointestinal bleeding, surgical treatment, casuistic case

Fluid collections form in 5-10% of patients after an episode of acute pancreatitis (AP) and in 20-40% of patients with chronic pancreatitis (CP). The underlying cause is usually a rupture of the main pancreatic duct or one of its larger tributaries or damage to the pancreatic parenchyma. At least 25% of these collections spontaneously resorb, while the remaining ones evolve into pancreatic pseudocysts through fibrosis, thickening and scarring of the adjoining tissues (1, 2).

A similar patomechanism, involving inflammation, tissue necrosis and secondary infections, leads to wall erosion of the vasculature of the pancreas and its surroundings. Arteries damaged this way may bleed into the peritoneal cavity, retroperitoneal space or the pseudocyst – resulting in the formation of a pseudoaneurysm. Such development is usually seen in AP, it can however accompany CP as well (3).

An estimated 10% of „acute” pseudocysts transforms into celiac pseudoaneurysms. The vessels most commonly involved are the splenic, gastroduodenal, superior and inferior pancreaticoduodenal arteries. The superior mesenteric, dorsal pancreatic, common hepatic and cystic artery are much less common locations (4). Other causes of peripancreatic pseudoaneurysm formation are anecdotal; cases of posttraumatic, iatrogenic (following pancreatoduodenectomy, pancreatojejunostomy, pancreas transplantation, necrosectomy, ERCP) pseudoaneurysms have been reported, as well as those observed in systemic lupus (5).

Portal hypertension is another severe complication of pancreatitis. It is evoked by compression or thrombosis of the portal, superior mesenteric or splenic vein (6). This can be caused by pancreatic edema, fluid collections and pancreatic fibrosis in the course of CP. The development of portal hypertension due to pressure by a pseudoaneurysm is extremely rare (7).
This report presents a case of a patient with numerous above-mentioned post-AP complications taking a dramatic course.

CASE REPORT

W.B., a 55-year-old male, ref. no. 1941/06, reported to the hospital with signs of active gastrointestinal bleeding (GIB). He had observed tarry and bloody stools for the previous few weeks. He had been experiencing abdominal pain for a few months prior to admission. The patient had a history of oral iron therapy for sideropenic anemia over the course of a few years. He had also been receiving oral pancreatin. Ten years previously the patient suffered from acute pancreatitis, which was initially treated conservatively and later by two laparotomies. Three years prior to admission, he had undergone an explorative laparotomy for a suspected tumor within the pancreatic head. Upon laparotomy the pancreatic head had been found to be enlarged and the whole gland slightly firmer than normally, with no infiltration of the neighboring tissues. A diagnosis of chronic inflammation and pancreatic fibrosis was made. Two years later, upon ultrasound, a “slightly enlarged spleen” was found (length: 130 mm) along with “an enlarged pancreatic head (54 mm thick) with mixed echogenicity, containing a polycyclic fluid collection measuring 39 by 43 mm with hyperechogenic areas inside (cyst?)”. Upon another laparotomy, none of these lesions were discovered within the pancreas, and the treatment was limited to the repair of an incisional hernia within the laparotomy scar.

Seven months after the second laparotomy the patient was once again hospitalized for lower gastrointestinal bleeding. Endoscopy showed 2nd degree hemorrhoids with no signs of active bleeding. Upon ultrasound, again an enlarged spleen (140 mm) was described, as well as an irregular pancreas with an anechogenic mass within its head, measuring 25 x 35 mm, within which hyperechogenic areas were seen. The patient was discharged in good condition after conservative treatment.

During the current hospitalization, upper abdominal tenderness was observed, with the absence of peritoneal signs and the liver and spleen not palpable. No dilation of superficial blood vessels was recorded. Upon digital rectal examination, tarry stool and fresh blood were found, as well as 3rd degree hemorrhoids. Laboratory findings included anemia (Hgb 6,1 g%, RBC 2,76 *10^7/ml) and increased amylase levels (serum – 375 U/l, urine – 875 U/l). Leukocytes, platelets and coagulation parameters were all normal. Upon colonoscopy 3rd degree hemorrhoids were confirmed; there was blood visible throughout the length of the colon and a small ulceration was found in the rectum. The blood was seen to come through the ileocecal valve in a continuous flow.

After conservative treatment a temporary cessation of bleeding was observed, with blood morphology returning to normal. However, after a few days the bleeding recurred, with blood hemoglobin falling to 8,2 g% and amylase concentrations rising (serum 615 U/l, urine 1971 U/l). Again, the bleeding stopped after conservative treatment.

Upon abdominal ultrasound a heteroechogenic lesion measuring 42 x 47 mm was observed within the pancreatic head and corpus, containing an anechogenic area of 26 x 22 mm, with the presence of a flow confirmed by Doppler. A partially thrombosed aneurysm of one of the arteries originating from the celiac

Fig. 1. Doppler abdominal ultrasound: A – within the head and partially the corpus of the pancreas, a heteroechogenic mass measuring 42 x 47 mm can be seen, containing an anechogenic area measuring 26 x 22 mm; B, C – upon Doppler imaging a flow is seen in the anechogenic area
trunk was suspected or a pancreatic lesion containing a pseudoaneurysm or hematoma. The length of the spleen was measured at 130 mm. Other organs were described as normal. According to the radiologist, the size of the fluid collection was similar to that measured a year before (Dept. and Chair of Radiology CM UMK, Head: prof. W. Lasek).

The diagnostics were extended to include endocrine assays; the levels of VMA, 5-HIAA, HVA, UNaV, UKV were all found to be normal. An abdominal CT scan with oral and intravenous contrast was performed, revealing a round area, diameter approx. 5-6 cm, containing fresh blood, with contrast extravasation in the arterial phase (pseudoaneurysm?). In the neighborhood, celiac trunk and superior mesenteric branches visible. The aforementioned mass compresses the portal vein. Within the head and corpus of the pancreas segmental dilation of the Wirsung duct up to 6 mm is visible, the distal portion of which including its duodenal terminus, cannot be visualized... In the mesogastrium, numerous dilated blood vessels were observed (possibly collateral pathways due to portal hy-
pertension), as well as an atypical course of an undilated splenic vein and an enlarged spleen (170 mm length) with a heterogenic structure in the arterial phase resulting possibly from peripheral hypoperfusion (forming focal infarctions?) According to the radiologist, the image could have resulted from portal hypertension. The gall-bladder was shrunken, with a thickened wall measuring up to 4 mm” (Dept. of Radiology, as above).

The patient was staged for surgery. Intraoperatively, the diagnosis of portal hypertension was confirmed (dilated veins with intensive bleeding, spleen enlarged). The pancreas was firmer than normal, and beyond its head and corpus a tumor the size of a man’s fist was found, having no connection to the aorta. The pancreas was transected at the isthmus and a compressed portal vein was uncovered. The corpus and tail of the pancreas were resected. A hard, nonpulsating tumor strictly adherent to the pancreatic head was uncovered. The anterior wall of the tumor was incised, and the diagnosis of a clot-filled pseudoaneurysm was confirmed. Following clot removal, an intense arterial bleeding was observed, which was managed by stick-tying the pancreaticoduodenal artery from within the pseudoaneurysm lumen. The walls of the aneurysm were partially resected, relieving the pressure on the portal system – the signs of portal hypertension receded and the bleeding diminished. The spleen – the size of 4 man’s fists – was also removed. A short film taken during the procedure is available at the following Web address: http://www.youtube.com/watch?v=GzZPlK9n95U

The postoperative course was uneventful. Proton pump inhibitors and LMWH were used. Persistent diarrhea and thrombocytopenia were observed postoperatively. Glucose profile was normal. The patient lost 8 kilograms during the hospital stay and was discharged 14 days after the procedure in good overall condition. During one year of follow-up the GI bleedings did not recur and the body mass returned to normal.

DISCUSSION

A rare case of recurrent upper gastrointestinal bleeding, caused by a pseudoaneurysm of the pancreaticoduodenal artery, was described in this paper. The diagnosis was based on radiological findings. Upon surgery, the aneurysm was reached through a resection of the tail and corpus of the pancreas. The aneurysm was partially resected, the bleeding artery stick-tied from within its lumen, which stopped the bleeding and relieved portal hypertension.

Pseudoaneurysm formation is caused by necrotic lesions of the pancreatic parenchyma and/or damage to the pancreatic ducts. This may lead to the formation of peripancreatic fluid collections, containing activated proteolytic enzymes. At this stage, a “real” pseudoaneurysm is formed – the blood remains within the distended artery. Once the aneurysm bursts, the blood extravasates into the cyst. If the cyst wall is weak, the pseudocyst ruptures and bleeding ensues into the peritoneal cavity or retroperitoneal space. If the cyst walls are fibrosed, fused with the surrounding tissues through an inflammatory infiltration, the blood fills the cyst and temporary pressure hemostasis ensues. Extravasation of blood into a pseudocyst converts it into a pseudoaneurysm (extravasal hematoma communicating with the vessel lumen).

Arterial walls can also be damaged through infection of the pseudocyst. Pseudoaneurysms most frequently form in patients after medium to severe AP, particularly if the disease leads to pseudocyst formation and/or peripancreatic abscesses.

A similar patomechanism may lead to pseudoaneurysm formation after pancreatic resections, when they are caused by anastomotic leaks or intraabdominal abscesses. Other factors inducing inflammation, even circumscribed, or purulent lesions may cause arterial wall damage. In consequence, a pseudoaneurysm forms, always bearing the threat of delayed rupture. A pseudoaneurysm may also result from pancreatic transplantation (8, 9).

A pancreatic pseudocyst must be differentiated with a primary aneurysm of the peri-pancreatic vessels. This pathology is found mostly in females; ruptures usually occur during pregnancy. The dominating presentation is that of an intraperitoneal hemorrhage with hemodynamic consequences (10).

One should suspect a pancreatic pseudoaneurysm in patients with a history of AP or active CP (alcoholics), in whom pseudocysts are found together with an epigastric vascular bruit. The clinical course is variable. In patients after pancreatitis one should be wary of: unexplained anemia, recurrent gastrointestinal bleeding
Pancreatic pseudoaneurysm presenting as recurrent gastrointestinal bleeding

and epigastric pain. Diagnosis is facilitated by radiological findings of an enlarging pancreatic pseudocyst or when a pulsating epigastric tumor forms, especially if accompanied by bruit and hyperamylasemia (6). Characteristic sequelae are associated with the pseudoaneurysm communicating with Wirsung’s duct: bleeding from the papilla of Vater with accompanying colicky pain and jaundice (hemosuccus, wirsungorrhagia) (11, 12).

Conservative treatment of a pancreatic pseudoaneurysm is risky, since there is no way to predict the development of the pathology. The most severe complication of a peripancreatic pseudoaneurysm is hemorrhage into the gastrointestinal tract, peritoneal cavity or retroperitoneal space. In a review of literature, 71 patients of 135 reported cases of a pancreatic pseudoaneurysm presented signs of bleeding, whereof 35 – to the intestinal tract. The bleeding may originate from the pancreatic duct, a fistula between the aneurysm and the stomach or intestine or from vascular malformations such as lesser gut varices. The mechanism of GI bleeding has been explained in detail in 9 cases only: in 5 of those the aneurysm communicated with the pancreatic duct, in 2 a fistula between the aneurysm and duodenum was found, in one case the fistula formed between the aneurysm and the duodenojejunal flexure and in the last case – between the aneurysm and the stomach.

Bleeding from a pancreatic pseudoaneurysm is usually violent and sudden, often with hypovolemic shock. Such a hemorrhage, if not stopped by emergency measures, is usually fatal. Even if proper management is performed, mortality remains high: from 13% (lesion within the corpus or tail) to 40% (head) (1, 13, 14).

If a GI bleeding is caused by a pancreatic pseudoaneurysm, only rarely can the source of bleeding be identified upon initial endoscopy. The most sensitive noninvasive diagnostic options are: Doppler ultrasound and angio-CT. The most specific tools are invasive – celiac and superior mesenteric arteriography. These methods offer therapeutic possibilities aside from pinpointing the origin of the hemorrhage (15, 16).

Non-surgical treatment is usually vessel embolisation with/without stenting. This can lead to definitive cure or provide additional time to prepare the patient for surgery. In recent years, the success rates for angioembolization have reached 67-100%. The best results can be achieved when the bleeding lesion lies within the head of the pancreas, previous surgical attempts have failed or the bleeding occurs in the postoperative period (17, 18).

The risks are typical for this type of procedure: vessel wall perforation by catheter tip, rupture of the aneurysm, thrombosis of an undesired vessel. Some authors indicate that bleeding recurs in 1/3 of the cases and the overall mortality for this type of treatment reaches 16% (8, 18).

Surgery is indicated particularly when angioembolization is ineffective or inaccessible. There is controversy regarding the way in which hemostasis should be achieved: the methods used include ties, stick ties and pancreatic parenchyma resections. The aneurysm itself may be partially resected or drained internally or externally (13, 15, 18).

Post inflammatory peripancreatic fluid collections are a common finding, yet reports of pancreatic pseudoaneurysms are relatively scarce – in 65 available reports a total of 135 cases have been presented. In Polish literature only one case of pancreatic pseudoaneurysm has been reported. In a 41-year-old-male a 12 cm cyst with a circulating fluid content has been revealed by abdominal ultrasound. Upon surgery, the source of bleeding was found to be the common hepatic artery. The lesion was repaired and the cyst anastomosed to the rear gastric wall. The postoperative course has been reported as uneventful (19).

The case reported here confirms the need for systematic follow-up of patients after medium to severe pancreatitis. Increasing post-inflammatory pseudocysts, anemia, hyperamylasemia or a tumor with a bruit audible in the epigastrium should alert the surgeon to initiate diagnostics for pancreatic pseudoaneurysm. Finding such a pathology requires vascular embolization or surgical management.

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COMMENTARY

Pseudoaneurysm of the arteries adjacent to the pancreas (hepatic artery, gastroduodenal artery, pancreatoduodenal artery) is a rare complication of pancreatitis. Its symptoms may include pain or intraperitoneal, extraperitoneal bleeding, common bile duct or pancreatic duct bleeding. In imaging studies it is revealed as a pseudocyst-like structure. Only Doppler US can demonstrate blood flow in the aneurysm. Angiography of celiac trunk can support the diagnosis and allows embolization of the aneurysm and thus it constitutes the best therapeutic option. Surgical treatment of bleeding pseudoaneurysms is difficult from the technical point of view and associated with high risk of complications and mortality.

Authors do not specify etiology of acute pancreatitis or surgery that the patient underwent. The following imaging studies were performed during the next hospitalization (its cause was gastrointestinal bleeding): abdominal US and triphase CT with an oral contrast agent. Unfortunately we have no information of possible use of celiac trunk angiography that would allow to establish diagnosis and initiate treatment (1). Interventional radiology that can perform embolization or percutaneous, US-guided injection of thrombin into the lumen of pseudoaneurysm is less invasive and safer alternative to surgery in the treatment of this disorder (2).

We should congratulate authors on the successful surgical procedure; gastrointestinal bleeding did not recur after the procedure. However, no imaging studies were performed that would assess the remaining parenchyma of the
pancreas. Precise US or CT monitoring could demonstrate possible neoplastic transformation in the pancreas. Thus both diagnostics and treatment of pancreatic pseudoaneurysms is a serious diagnostic and therapeutic problem.

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