

SPLENIC ARTERY PSEUDOANEURYSM RUPTURE INTO A PANCREATIC PSEUDOCYST WITH ITS SUBSEQUENT PERFORATION AS THE CAUSE OF A MASSIVE INTRA-ABDOMINAL BLEEDING – CASE REPORT

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Pseudocysts account for approximately 70% of all cystic lesions of the pancreas. One of the most dangerous complications of pancreatic pseudocysts is bleeding into the cystic lumen; the most common cause of the bleeding is a splenic artery pseudoaneurysm rupture. This paper presents the case of a 37-year-old man treated surgically for a massive intra-abdominal haemorrhage caused by a splenic artery pseudoaneurysm rupture into the lumen of a tail of pancreas pseudocyst with its subsequent perforation into the abdominal cavity and retroperitoneal space. Peripheral resection of the pancreas together with the cyst and spleen resection was performed. There were no postoperative complications.

Key words: pancreatic pseudocyst, pseudoaneurysm, intra-abdominal haemorrhage

Pseudocysts account for approximately 70% of all cystic lesions of the pancreas. The term ‘pseudocyst’ refers to fluid reservoirs surrounded with a capsule devoid of epithelial lining, built of fibrous connective tissue and granulation tissue. They usually form in patients with alcohol dependence syndrome and are associated with acute or chronic pancreatitis (1).

According to Warshaw et al., in 8% to 85% of cases, pseudocysts undergo spontaneous resolution within 4–6 weeks. The majority of authors believe that after that period pancreatic pseudocysts with a diameter of over 6 cm should be treated; however, there is also an opinion that only symptomatic or complicated pseudocysts should be treated. At present there is a whole spectrum of management of

pancreatic pseudocysts, from minimally invasive drainage methods such as endoscopic gastrocystostomy, duodenocystostomy or transpapillary cystostomy to classic surgical procedures, including resections (1, 2).

One of the most dangerous complications of pancreatic pseudocysts is bleeding into the cystic lumen. This type of complication is reported in 6–10% of cysts and the most common source of bleeding is ruptured splenic artery pseudoaneurysm located in the cystic wall. Blood may extravasate into the peritoneal cavity or retroperitoneal space through a direct perforation of the cyst, into the duodenum through the duct of Wirsung communicating with the cystic lumen (haemoductal pancreatitis) or into the lumen of organs neighbouring the cyst through a fistula channel. According

to a number of authors surgical treatment of these complications is one of the most difficult surgical procedures for all cases of non-traumatic bleeding in the abdominal cavity and perioperative mortality ranges between 70% and 90% (3, 4).

This paper presents the case of a 37-year-old man treated for a massive intra-abdominal haemorrhage caused by a splenic artery pseudoaneurysm rupture into the lumen of a tail of pancreas pseudocyst with its subsequent perforation into the abdominal cavity and retroperitoneal space.

CASE REPORT

A patient K. C., aged 37, was admitted on 5 June 2015 to the Surgery Ward of the Cracow Remand Prison Hospital, Poland, due to severe girdle pain located in the upper abdomen with accompanying vomiting of food contents with traces of blood clots. The patient had been staying in a correctional facility for a few days. He had had an alcohol dependence syndrome for many years and a history of a few episodes of acute alcohol-related pancreatitis. He had been hospitalised for the last time at a gastroenterology ward of one of the Cracow hospitals shortly before being arrested. He was discharged on 21 May 2015 with the following diagnosis:

“Exacerbation of chronic pancreatitis. Thrombocytopaenia. Anaemia. Alcohol dependence syndrome”. An abdominal CT examination performed at the time revealed “(...) a thick-walled fluid lesion in the distal part of the tail of pancreas of 48 x 44 mm and an attenuation coefficient of 13 HU before and after administration of a contrast medium, with a significant enhancement of the walls following intravenous administration of a contrast medium. A splenic artery branch is pulled into the wall of the cystic lesion. The vessel is locally dilated and extends into the lumen of the cyst. CT angiography is recommended”.

Following surgical consultation which did not reveal any indications for surgical treatment, the patient was discharged with instructions on dietary and pharmacological treatment. CT angiography was not performed.

In clinical examination upon admission on 5 June the patient was cardiovascularly and respiratorily stable. Painful abdomen with no

resistance or peritoneal signs. Laboratory tests revealed: Hb 7.8 g/dl, HCT 24.1%, RBC $3.14 \times 10^6/\mu\text{l}$, amylase 137.3 U/l. The remaining parameters were within normal limits. Urgent gastroscopy excluded active bleeding into the gastrointestinal lumen. The hypothesis of another exacerbation of chronic alcohol-related pancreatitis was adopted and following the transfusion of 2 units of PRBC typical conservative treatment was pursued, with the perspective of performing an elective abdominal CT angiography.

On the third day of hospitalisation in the morning a severe pain episode occurred with a subsequent drop in the systolic arterial blood pressure to 60 mm Hg with a heart rate of 120/min. After a central line was set up fluid resuscitation using gelafundin and crystalloids was initiated. Blood was ordered. A large amount of free fluid was found in the abdominal cavity upon ultrasound examination. The tail of pancreas cyst reported previously was not visualised. In this situation the patient underwent an urgent operation at the General Surgery Ward of the Cracow Remand Prison Hospital.

The abdominal cavity was opened using a midline incision around the navel. Blood was found in the free peritoneum. 1500 ml of blood was suctioned out. In the omental bursa and retroperitoneal space extensive haematoma was found which was descending from the level of the pancreas downwards, at the left side of the aorta into the pelvis minor; the haematoma was pushing the stomach and the left part of the colon towards the layers of the anterior abdominal wall. In the colonic mes-



Fig. 1. Abdominal CT: splenic artery pulled into the cystic lesion of the tail of pancreas

entery near the splenic flexure a perforation opening communicating with the lumen of the tail of pancreas pseudocyst was identified. Active bleeding from the lumen of the perforated pseudocyst was present. The upper abdomen was packed. Following the stabilisation of the cardiovascular system using massive transfusions packing material was removed. The omental bursa was opened through the gastrosplenic ligament. Blood clots were evacuated. At the upper margin of the pancreas to the left of the aorta at the border between the body and tail of pancreas splenic vessels were identified and ligated. The stomach was separated along the side of the greater curvature up to the cardiac orifice through the gastrosplenic ligament. The left part of the colon was mobilised and blood clots were evacuated. The distal part of the body and tail of pancreas with the perforated cyst and the spleen were resected. The cross-section of the pancreas was coagulated and dressed using Prolene 0 mattress sutures. Irrigation drainage of the cross-section of the pancreas and the splenic bed using two-channelled Lavacuator tubes No 28 and 32 was applied. A draining tube was placed into the recto-vesical pouch.

After the procedure the patient, still under anaesthesia, was transferred to the ICU of the Ministry of the Interior and Administration Hospital in Cracow (there is no ICU within the Surgery Ward of the Cracow Remand Prison Hospital). Following the stabilisation of the cardiovascular and respiratory systems the patient returned to the ward on 10 June 2015 in a good general and local condition. There were no complications in the perioperative period. On the 4th day the draining tube was removed from the pouch. On the 6th day after the procedure the irrigation drains were removed following the exclusion of the presence of a pancreatic fistula as per the International Study Group of Pancreatic Surgery (ISGPS) standards. The patient was discharged from the ward on the 29th day after the procedure in a good general condition with the wound healed by primary intention. A histopathological examination of the resected specimen (No 1652) revealed an image typical of a pseudocyst. The spleen had signs of reactive lesions.

During the hospitalisation 10 units of packed red blood cells and 3 units of frozen plasma were transfused in total.

DISCUSSION

Splenic artery aneurysm was first described by Beaussier in 1770 (1). It accounts for 4% of all aneurysms and is the third most common abdominal cavity aneurysm after aortic and iliac artery aneurysms and the most common (60%) visceral artery aneurysm. Autopsy studies estimate the rate of splenic artery aneurysms to be 0.01 to 2%. It is four times more common in women than in men. According to Parks visceral artery pseudoaneurysms occur in approximately 10% of patients with a pancreatic pseudocyst; the risk of rupture is estimated to be 2 to 10% and it significantly increases in aneurysms with a diameter larger than 2 cm (5, 6).

Aneurysm rupture into the cystic lumen is the most dangerous complication of pancreatic pseudocysts; the most common source of bleeding is the splenic artery. Splenic artery pseudoaneurysms are located in its distal 1/3 in 74-87%. It needs to be stressed that in 22% of cases aneurysm rupture and massive, life-threatening bleeding into the gastrointestinal lumen or the free peritoneum is the first symptom of a pseudoaneurysm. The pathomechanism of vessel perforation and bleeding into the cystic lumen associated with chronic or acute pancreatitis is thought to involve vessel wall necrosis as a result of mechanical compression by the cyst and enzymatic lysis of the structures of the vessel wall by pancreatic proteases and elastases contained in the contents of the pseudocyst. It is also emphasised that bleeding into the lumen of the cyst may be provoked as a result of its decompression during puncture or drainage. It is particularly important in the light of the fact that EUS-guided internal drainage is currently the method of choice for the treatment of pseudocysts associated with chronic pancreatitis. Therefore, it is increasingly emphasised that before starting drainage treatment of a pancreatic pseudocyst the presence of an aneurysm in the cystic wall should be excluded using Doppler ultrasound, CT angiography, MRI angiography or selective visceral arteriography, which, as well as being a diagnostic tool, provides a whole range of endovascular therapeutic methods (1, 5, 6).

Visceral aneurysm rupture into the lumen of a pseudocyst is a rare complication of acute or chronic pancreatitis, but the most dangerous

one at the same time. It may occur in three clinical forms: bleeding into the gastrointestinal lumen (haemoductal pancreatitis, organ fistulas), peritoneal and retroperitoneal space bleeding and the combination of the two (4).

The first of these clinical forms belongs to the group of conditions which cause unexplained bleeding into the gastrointestinal lumen and account for 5% of all cases of bleeding into the gastrointestinal system together with diseases such as angiodysplasia, Dieulafoy's lesion, Cameron's ulcer, GAVE, Osler-Weber-Rendu disease and other conditions. The characteristic sequence of events is sudden pain in the upper abdomen with the frequently accompanying Kehr's sign, drop in arterial blood pressure, vomiting blood, melaena or blood in stool and gradual subsiding of pain with blood pressure normalisation. With FOBT being positive multiple urgent endoscopic examinations fail to determine the source of bleeding. After the event laboratory results are dominated by a significant decrease in blood count parameters. There is also a characteristic patient history: young age, alcohol dependence syndrome, multiple recurrences of pain associated with chronic pancreatitis, a pancreatic cyst found upon abdominal ultrasound, repeated hospitalisations due to bleeding recurrence, iron-deficiency anaemia or multiple deficiency anaemia.

There is a consensus that in this clinical form of visceral aneurysm rupture into the lumen of a pancreatic pseudocyst endovascular treatment is a method of choice, which is part

of interventional radiology. This type of minimally invasive procedures include coil embolisation and the use of stent grafts or cyanoacrylate glues with Lipiodol. These methods are sometimes supplemented with the administration of thrombin directly into the cystic lumen or their combinations are used. The effectiveness of these methods is consistently estimated to be 90% and mortality in elective embolisation procedures is not higher than 0.5%. In 2011 and 2012 coil embolisation was used at the ward headed by the author in two cases of such bleeding:

- in a gastroduodenal artery aneurysm ruptured directly into the descending part of the duodenum in a patient with alcohol-related, nodular pancreatitis (fig. 2);
- in a proper hepatic artery aneurysm ruptured into a head of pancreas pseudocyst (fig. 3).

The procedures were performed at the Department of Diagnostic Imaging of the University Hospital in Cracow. In both cases a full haemostatic effect was obtained without any complications. It needs to be emphasised that complementary use of embolisation as a procedure preceding surgery decreases the perioperative mortality in resection procedures from more than fifty down to a few percent (7, 8).

In the case of visceral aneurysm rupture into the peritoneal cavity or retroperitoneal space there is no time for radiological endovascular minimally invasive methods since time is of vital importance to the patient. Emer-

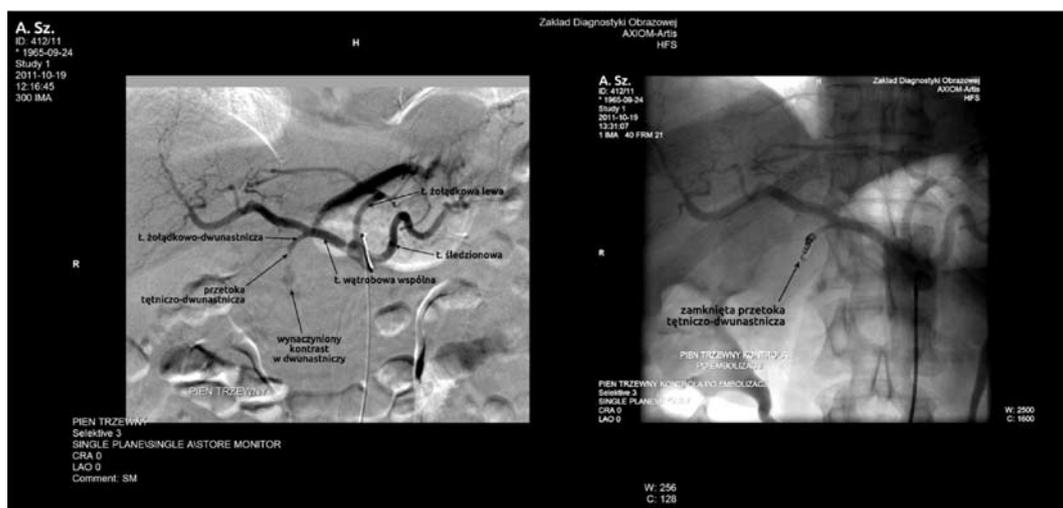


Fig. 2. Gastroduodenal artery aneurysm ruptured into the descending part of the duodenum. State before and after embolisation

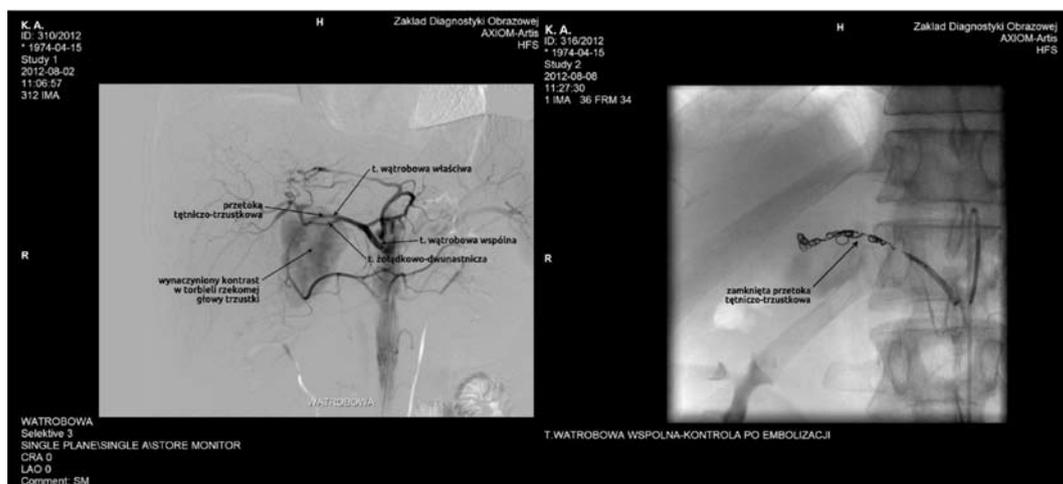


Fig. 3. Proper hepatic artery aneurysm ruptured into a head of pancreas pseudocyst. State before and after embolisation

gency laparotomy is the method of choice. It seems that in the case of aneurysm rupture into a cyst located in the proximal part of the pancreas transfixing of the aneurysm from the cystic lumen with a subsequent drainage is a life-saving procedure. In the case of aneurysm rupture into a cyst located in the distal part of the pancreas the optimal treatment is distal pancreatectomy with cyst removal and splenectomy. Perioperative mortality in this type of procedures is estimated to be 40 to 90% (4, 9, 10).

In 2015 there were approximately 80,000 inmates in Polish correctional facilities. This number is relatively constant with small fluctuations. Alcohol dependence syndrome is diagnosed in 30% of first time prisoners and in 70-90% of reoffenders, which means that 50% on average, i.e. 40,000 individuals serving prison sentences are alcoholics. Chronic pancreatitis is found in 10% of individuals with alcohol dependence syndrome; in this group

10% have a visceral aneurysm (7, 10). This means that in correctional facilities between three prison surgical wards (Bydgoszcz, Kraków, Łódź) there are 400 patients a year with visceral pseudoaneurysms having a risk of rupture with mortality as high as 90% (1, 5, 6). It is thus clear that prisoners are a special health care need group in relation to this condition. Therefore, physicians in correctional facilities should adopt the following patient management algorithm: an inmate with alcohol dependence syndrome, chronic pancreatitis, the presence of a pancreatic cyst upon abdominal ultrasound examination, periodic abdominal pain, unexplained drops in haematocrit, with a history of bleeding into the gastrointestinal lumen with an undetermined source requires abdominal CT angiography in order to exclude the presence of a visceral pseudoaneurysm. If such is found, the patient should undergo selective visceral arteriography and aneurysm embolisation.

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