BLEEDING FROM THE SPLENIC ARTERY INTO A PANCREATIC PSEUDOCYST WITH A COEXISTING MICROFISTULA TO THE TRANSVERSE COLON AS ATYPICAL RECURRENT MASSIVE GASTROINTESTINAL BLEEDING

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Here we present a case of a 49-year old male patient who was hospitalized at our Clinic from 2 to 16 December 2008 due to recurrent massive gastrointestinal bleeding. It was a patient with a history of recurrent pancreatitis. He had a history of surgical treatment for postinflammatory pancreatic cyst (Jurasz cystogastrostomy). From 28.01.2007 to 16.12.2008 he was hospitalized five times. During the last hospitalization he received a total of 12 units of packed red blood cells. Neither gastroscopy nor colonoscopy did demonstrate the site of bleeding. AngioCT of the abdominal cavity demonstrated clearly enlarged spleen and a well delineated region, 30x35 mm, reaching spleen hilum, filled with dense fluid suggesting a vascular fistula, in the projection of the body and tail of the pancreas. The patient was qualified for laparotomy. Intraoperatively, bleeding from the splenic artery into the pancreatic pseudocyst with coexisting microperforation to the transverse colon was detected. The pancreatic cyst was opened and drained, the bleeding blood vessel as well as the splenic artery were underpinned. Splenectomy was performed and wall of the transverse colon was repaired. The patient underwent reoperation due to adhesion related small bowel obstruction on day 30 after the procedure. Currently the patient is in good general condition, without complaints, undergoes periodic follow up in the outpatient setting.

Key words: gastrointestinal bleeding, pancreatic cyst, splenic artery, computed tomography angiography

Gastrointestinal bleeding, upper gastrointestinal bleeding in particular, is a very common problem in the routine medical practice worldwide. It is one of the commonest clinical situation that require rapid management (1). It is considered as the most common urgent condition in gastroenterology. Its estimated incidence is 50-170/100 000 annually in the population (2). When the bleeding is located proximally to the ligament of Treitz, it is termed upper gastrointestinal bleeding, when distally – it is termed lower gastrointestinal bleeding (1). Signs and symptoms of gastrointestinal bleeding depend on its cause, location of its source as well as intensity. They include coffee-ground vomiting (melaenemesis), bloody vomiting (haematemesis), tarry stools (me-laena), stools with blood admixture (haemato-
Atypical recurrent massive gastrointestinal bleeding at the age of 40-60 years are diseases of the rectum and anus (hemorrhoids, fissures), diverticuli of the large intestine (17-40%), inflammation of the large intestine (infectious, nonspecific) and various types of angiodysplasia (2-30%) (1), while in children and young adults – Meckel’s diverticulum (3). Sometimes it is difficult to unequivocally determine the site of bleeding. This situation is estimates to apply to approximately 5% of all gastrointestinal bleedings. Such bleeding is termed a bleeding of unknown etiology.

Our patient reported passing large amounts of bloody stools in the short period of time, causing weakness and drop of arterial blood pressure. This could indicate both massive upper gastrointestinal bleeding as well as bleeding into a small or large intestine. Endoscopy (either gastroscopy or colonoscopy) did not reveal the potential site of bleeding.

A gastrointestinal bleeding of unknown etiology is a recurrent or persistent bleeding which source was not located using preliminary conventional endoscopy of the upper and lower gastrointestinal tract (gastrofiberoscopy, colonoscopy). Bleedings of unknown etiology are an important diagnostic and therapeutic problem. They could be clinically overt, with typical signs and symptoms, or latent, causing iron deficiency anemia. A typical sign of these bleeding is their recurrence while their source is usually located in the small intestine. In some cases, pathological lesions overlooked in a preliminary endoscopic assessment are the source of the bleeding; most commonly they include: Cameron’s ulcers accompanying hiatal hernia, peptic ulcer disease of the stomach or duodenum, vascular lesions (angiodysplasia, Dieulafoy’s lesions, GAVE (Gastrin antral vascular ectasias), esophageal varices, isolated varices of the gastrin fundus) while in the lower gastrointestinal tract: angiodysplasia and malignancies.

Atypical gastrointestinal bleedings also include: bleeding from Meckel’s diverticulum, described below (5), aorto-intestinal fistulas (11) or ruptured aneurysm of the splenic artery into the lumen of the large intestine (4).

**CASE REPORT**

A patient W.P., age 49, coexisting diseases (-). Hospitalized 6 times:

- 28.01.-14.02.2007 due to acute pancreatitis (confirmed by CT imaging of the abdominal cavity and by laboratory biochemical tests),
- 12.04.-16.04.2007 due to acute recurrent pancreatitis,
- 03.09.-18.09.2007 due to acute recurrent pancreatitis with pancreatic cyst,
- 14.11.-27.11.2008 due to massive gastrointestinal bleeding,
- 02.12.-16.12.2008 due to massive gastrointestinal bleeding,
- 31.12.-09.01.2009 due to adhesion-related obstruction of the small intestine.

During the first two hospitalizations the patient was treated medically. During the third, due presence of postinflammator pancreati cyst, the patient underwent successful Juraz operation. In November 2008, he was hospitalized due to massive gastrointestinal bleeding. Endoscopy did not reveal a clear bleeding site. After resolution of the symptoms of bleeding and after improvement of his general condition, the patient was discharged home. On 2 December 2008, he was admitted again to the hospital due to a massive, recurrent gastrointestinal bleeding. At admission his general condition was moderate; the patient was pale, sweating, BP 90/50, HR 110 bpm, per rectum examination demonstrated fresh blood. Laboratory abnormalities included: HGB – 9.79 g/dl, HCT – 31%, WBC – 22,5 K/µl. Gastroscopy and colonoscopy did not demonstrate the bleeding site. During the hospitalizations due to recurrent massive gastrointestinal bleeding episodes, the patient received a total of 12 units of packed red blood cells. AngioCT of the abdominal cavity demonstrated clearly enlarged spleen, with small amount of free fluid around and irregular hypodense parahilar area and well delineated area, 30x35 mm, reaching the plenic hilum, filled with dense fluid, suggesting a vascular fistula. A paramural hyperdense lesion, 13x9 mm, was found in the intestinal lumen, in the region of splenic flexure.

The patient was qualified for laparotomy. A presence of pseudocyst of the pancreatic tail in the region of splenic hilum, posterior gastric wall and transverse colon was found. After it has been opened, a large amount of blood clots from adjacent splenic artery was evacuated. The cyst was lined with an endothelium-like tissue. The bleeding vessel, branching from
DISCUSSION

In this paper we present a rare case of recurrent massive lower gastrointestinal bleeding, caused by the bleeding from splenic blood vessels into the pancreatic pseudocyst along with coexisting microperforation into the colon. A few similar cases have been reported in the available literature (e.g. Krwawienie z tętnicy śledzionowej do torbieli trzustki ze współistniejącą przetoką do okrężnicy – Acta Angiologica 2002). It is a perfect ex ample of atypical gastrointestinal bleeding. Sometimes an aneurysm of the splenic artery can be a potential cause of massive gastrointestinal bleeding. Prevalence of this anomaly ranges from 1.6% in general population to 7.1% in population of patients with portal hypertension caused by liver cirrhosis (4). Early diagnosis of these vascular lesions is important due to possible complications, including the most dangerous – aneurysm rupture (the risk of rupture is 3-9.6%) (4). Rarely the aneurysm ruptures into the lumen of the large intestine, which may be the cause of recurrent, atypical massive bleeding into the gastrointestinal tract.

Other source of atypical, recurrent bleeding include the above mentioned Meckel’s diverticulum. This is the most common anomaly of the small intestine, reported for the first time in 1598 by Fabricius Hildanusa, resulting from incomplete obliteration of the vitelline duct (omphalo-mesenteric duct). Embryonal origin of the diverticulum was established in 1808-1820 by Johann Friedrich Meckel. The Meckel’s diverticulum occurs in approximately 2% population. It usually can be found approximately 60 cm proximal to the ileocecal valve and usually is asymptomatic, being found accidentally, e.g. during appendectomy. Complications occur only in 2-4% of cases; intestinal obstruction (35%) and bleeding (32%) are the most common.

An important pathology occurring in the diverticulum and contributing to gastrointestinal bleeding is ectopic gastric mucosa. It is found in 80% of Meckel’s diverticuli (5). Bleeding from the Meckel’s diverticulum usually occurs suddenly, without any preceding symptoms, and usually is very massive. Most common diagnostic modality used to diagnose the diverticulum is scintigraphy with a labeled sodium pertechnetate $^{99m}$Tc, which exhibits high affinity towards gastric parietal cells. Sensitivity of this method is 85-90%. False positive results are found in 15% of cases and false negative – in approximately 25%. An estimated specificity of this examination is 80% (5). In vivo or in vitro $^{99m}$Tc labeled red blood cells or $^{99m}$Tc sulphide colloid are used to locate the site of bleeding in the gastrointestinal tract. A focus of increased radioactivity is obtained at the bleeding site in consecutive scintigraphic images. This method allows for localization of the bleeding site when blood loss is at least 0.2 ml blood/min (6).

Capsule endoscopy introduced in 2000 is a new technique, allowing for precise visualization of such potential sources of bleeding in the small intestine as Meckel’s diverticulum or Crohn’s disease, angiodysplasia, tumors, polyps. It is safe and exhibits low invasiveness for the patient. The complete system includes three main parts: an endoscopic capsule, an external recorder with an antenna and a computer system to read and interpret the recorded images. This examination can essentially be used in a patient at any age. An estimated sensitivity of this method in the diagnostics of gastrointestinal bleeding of unknown etiology in adults is 89% while specificity 95% (7). The principal failure of the examination is entrapment of the endoscopic capsule in the gastrointestinal tract (e.g. polyps, tumors, diverticuli). Some sites use so called trial capsule that is digested in the event of entrapment. Previous contrast enhanced examination is also recommended to
rule out stenosis. Other less common complication include lack of image, lack of transition of the capsule in the specified time window, mechanical damage (chewing). Obviously, the biggest drawback of this wonderful method is its high cost and very low availability.

X-ray contrast enhanced imaging studies (passage, enteroclisis), angiography, enteroscopy (push enteroscopy, double balloon or intraoperative enteroscopy) are also used to demonstrate a potential source of atypical gastrointestinal bleeding in the small intestine. Enteroscopy involves direct endoscopy of the small intestine. Its principal advantage apart from direct assessment of the bleeding site, is possibility to take specimens for histopathological examination and to use endoscopic methods of stopping the bleeding (thermal, injection-based or mechanical). Limitation of push enteroscopy is its relatively small range (assessment of approximately 100 cm of proximal small intestine) and poor tolerance of this examination by patients. So called double balloon enteroscopy is newer and more accurate endoscopic method. As with push enteroscopy, this method also allows for taking specimen and performing a proper treatment procedure. Advantage of double balloon enteroscopy over push enteroscopy is ability to assess the whole small intestine. The whole small intestine can be also assessed with X-ray examinations such as gastrointestinal passage or enteroclisis (an examination involving administration of methylcellulose and barium to the duodenum through a probe) (8). Both these methods are significantly cheaper than the previously described methods, and their availability is bigger. Unfortunately, sensitivity of these methods is limited and their utility in the diagnostics of gastrointestinal bleeding is low.

Apart from the above mentioned scintigraphy with technetium 99 labeled red blood cells, angiography of the superior mesenteric artery (that demonstrates extravasation of a contrast agent) can be performed in patients with active bleeding (>0.1 ml blood loss/min). It is less sensitive than scintigraphy, while more invasive and efficient only when the blood loss is at least 0.5 ml/min. However, it allows for concurrent stopping of the bleeding with embolization or intravascular drug administration (e.g. vasopressin). It is especially recommended in patients with active gastrointestinal bleeding with high perioperative risk (8).

Aorto-intestinal fistulas are another cause of atypical, massive gastrointestinal bleeding. They have various causes, however most commonly are a consequence of surgical aortic repair and damage of the intestinal wall by implanted vascular grafts. Primary aorto-intestinal fistula results when the disease process in one of these organs creates connection between the aorta and the intestinal lumen. Causes of primary aorto-intestinal fistulas include: abdominal aorta aneurysm, aortic inflammation (bacterial, syphilitic, tuberculous), penetrating peptic ulcer, malignant infiltration, radiation therapy, injury.

Secondary aorto-intestinal fistulas result from surgical aortic repair procedures, e.g. surgical treatment of an aneurysm or surgical treatment of Leriche syndrome (in 0.6 to 1.5% cases) (9). In majority of cases, the fistula occurs in the proximal anastomosis of the graft and aorta. It results from continuous adhesion of the implanted vascular graft to the intestinal wall, leading to necrosis of the intestinal wall due to compression and formation of aorto-intestinal connection. Most commonly a fistula is formed in the line of sutures, leading to massive bleeding. There are also fistulas with pseudoaneurysms and perigraft “fistulas”, accounting for 15 to 20% of cases. In the latter case there is no connection with the aortic lumen and damaged intestinal wall is the source of bleeding. Most commonly (in 78.5% of cases), the aorto-intestinal fistula occurs in the distal duodenum. Other locations, such as ileum (9%), jejunum (4%), large intestine (4%), stomach (3%), appendix vermiformis (1%), rectum (0.5%), are less common. The time between fistula formation and the gastrointestinal bleeding ranges from a few days to 14 years, 3 years on average (9).

We begin diagnostic procedures of bleeding from aorto-intestinal fistula from routine endoscopy which can provide diagnosis in less than half of cases. Normal endoscopic image does not rule out the fistula. This examination should be performed in conditions allowing for immediate surgical intervention since a significant risk of endoscopy is triggering of severe hemorrhage by removal of a clot that blocked the fistula lumen. Computed tomography imaging is very helpful when aorto-intestinal fistula is suspected. Ectopic mass, fluid and soft tissue in the region of the graft, local intestinal thickening and pseudoaneurysm are suggestive of
pancreatic fistula. CT only rarely demonstrates a patent connection between the aorta and intestinal lumen. Arteriography is effective in the event of active bleeding and demonstrates contrast extravasation into the intestinal lumen. Demonstration of a pseudoaneurysm in the graft region is suggestive of fistula. All the above mentioned diagnostic examinations can be performed in hemodynamically stable patients. Exploratory laparotomy is performed in patients with severe hemorrhage or when results of other examinations are normal.

Let us go back to our case. Routine endoscopy twice did not demonstrate a potential source of bleeding in the discussed patient. Due to recurrent bleeding episodes, we decided to perform computed tomography angiography (angioCT). This very useful diagnostic modality was made possible by development of spiral CT and its quality improves along with evolution of multi-slice technology. It is a method characterized by low invasiveness. During the examination the patient is exposed to risk related to use of contrast agents and ionizing radiation. To visualize a blood vessel and a potential bleeding site, we have to mix the circulating blood with a contrast agent. Since large amounts of contrast agent need to be administered (usually 2 ml/kg body weight or more), non-ionic contrast agents are used (lower risk of complications). Side effect of intravenous administration of contrast agents are rare (0.5-1%) and are usually not life threatening. An estimated mortality is 1:50 000-100 000 examinations (10).

AngioCT performed in the discussed case, demonstrated a clearly enlarged spleen with a small amount of free fluid around and irregular hypodense perihilar region and a well delineated region, 30x35 mm, reaching spleen hilum, filled with dense fluid suggesting a vascular fistula, in the projection of the body and tail of the pancreas. Due to lack of a potential bleeding site in endoscopy and bleeding episodes becoming more and more common and massive, the patient was qualified for laparotomy.

As we have already mentioned in Introduction, the most common site of gastrointestinal bleeding of unknown etiology is the small intestine. This was an additional significant diagnostic problem because an existing micro-fistula to the transverse colon was missed twice in colonoscopy. The “pancreatic” past of our patient – recurrent acute pancreatitis, status post Jurasz cystogastrostomy, proved an important fact in the diagnostic workup of our case. Pancreatic pseudocysts are the most common complication of acute and chronic pancreatitis as well as injuries of the pancreas. Pancreatitis (most often chronic) is the cause of 60-80% pancreatic pseudocysts. Other causes include surgical procedures of other abdominal organs adjacent to the pancreas (e.g. splenectomy, partial gastrectomy due to a penetrating duodenal ulcer) (11).

Pancreatic pseudocysts occur in 5-10% of patients after acute pancreatitis and in 20-40% of patients with chronic pancreatitis (12). Pseudoaneurysms of splenic artery form in approximately 10% of patients with acute pancreatitis. Clinical signs and symptoms of pancreatic pseudocysts include predominantly abdominal pain, nausea, vomiting, lack of appetite and loss of body weight. They depend on the pseudocyst location and size. These complaints often result from complications of the cyst, e.g. cyst rupture into the peritoneal cavity with pancreatic ascites, formation of pancreatic fistula to the peritoneal or pleural cavity. Damage of adjacent blood vessels can sometimes result in formation of an aneurysm of splenic artery and bleeding resulting from rupture of its wall. The final diagnosis is made on the basis of imaging studies, such as US, EUS, CT and MRI (13).

5-10% of patients with pancreatic pseudocyst develop bleeding into the cyst lumen, usually caused by injury of the splenic artery. Injury of blood vessels usually results from necrosis of their wall, secondary to compression of their wall by an enlarging cyst or lytic activity of pancreatic enzymes (14). Sometimes a pancreatic pseudocyst occurs that communicates with the lumen of the gastrointestinal tract. The most common site of perforation is colon, more accurately a region of splenic flexure (15). Obviously it is related to proximity of the colon, splenic vessels and pancreatic pseudocyst. When a blood vessel adjacent to the pancreatic cyst is injured with resulting hemorrhage into the cyst lumen, clinical signs and symptoms of gastrointestinal bleeding are rare because most commonly small blood vessels are injured and the cyst wall is thick enough to prevent its perforation.

Fortunately, injury of large arteries (e.g. splenic artery) is a rare cause of gastrointestinal
bleeding in the course of postinflammatory pancreatic cyst with coexisting fistula to the gastrointestinal tract. If we do encounter such case in our clinical practice, we must realize that finding of a potential bleeding site using routine endoscopy will be very difficult. This is because a fistula of the pancreatic pseudocyst to the intestine is usually very small and often intermittently spontaneously closes after the bleeding, manifesting as brief, massive gastrointestinal bleeding episodes. According to the literature, the most sensitive method to precisely localize a bleeding site is arteriography of the celiac trunk (16). Unfortunately, this procedure is unavailable in many sites.

In the reported case we used angioCT to localize a bleeding site quite accurately. Precise localization of the bleeding site before the surgical treatment markedly improves the prognosis. Therefore, if only possible, this diagnostic examination should be performed in the event of bleeding of unknown etiology, to more or less accurately localize the bleeding site. If possible, such patients should be transferred to reference centers, possessing adequate diagnostic equipment to precisely diagnose the patient and improve the chance of successful treatment.

Diagnostics, qualification and surgical treatment of patients with hemorrhagic complications of pancreatic pseudocysts according to many authors are the most difficult of all bleedings in the abdominal cavity (17). Due to high mortality of patients with pseudoaneurysms of splenic vessels, surgical treatment should be instituted as early as possible.

CONCLUSIONS

The reported case of recurrent massive lower gastrointestinal bleeding caused by bleeding from splenic vessels to the pancreatic pseudocyst along with coexisting microperforation to the colon is a rare case of gastrointestinal bleeding of unknown etiology. In the event of recurrent, massive gastrointestinal bleeding of unknown etiology, angiography of celiac arteries or abdominal angioCT, preferably during an active bleeding, should be considered after a routine endoscopy. In most of the cases it allows to precisely localize a bleeding site. Special attention should be paid to patients after an episode of acute pancreatitis, with recurrent massive gastrointestinal bleeding. In such cases, rare hemorrhagic complications of the pancreatic pseudocyst should be suspected, such as bleeding from the splenic vessels to the pancreatic cyst with coexisting microfistula to the transverse colon. It should be noted that adequate diagnostic workup, allowing precise localization of the source of bleeding before patient qualification for surgical treatment, significantly increases the chance of therapeutic success in such cases.

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Received: 27.08.2009 r.
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COMMENTARY

The paper discusses an interesting case of bleeding from the splenic artery to a pancreatic pseudocyst with coexisting microfistula to the transverse colon: a rare cause of gastrointestinal bleeding. The Authors presented the most common causes and signs and symptoms of gastrointestinal bleeding. Furthermore, the Authors comprehensively discussed various rare causes of gastrointestinal bleeding and diagnostic possibilities and difficulties related to the presented subject.

Pancreatic pseudocysts are one of the complications of acute and chronic pancreatitis, pancreatic injury and obstruction of pancreatic duct. It should be noted that bleeding complications occur in 6-30% of pancreatic pseudocysts. The cause of bleeding complications of these cysts can be directly related to the inflammatory process of the pancreas (pseudoaneurysm, necrosis of the cyst wall, esophageal and gastric varices caused by thrombosis of the splenic vein and portal hypertension, bleeding from the vessels of the cyst wall, rupture of the spleen) or coexisting disorder (peptic ulcer, hemorrhagic gastritis or duodenitis, esophageal varices secondary to portal hypertension caused by liver cirrhosis, Mallory-Weiss syndrome). Hemorrhagic pancreatic cyst may perforate into the stomach, duodenum and colon. In the event of perforation into the stomach, signs and symptoms of upper gastrointestinal bleeding occur. When a cyst penetrates to the colon, signs and symptoms of lower gastrointestinal bleeding are observed. In the reported case the authors detected lower gastrointestinal bleeding, caused by a microfistula to the transverse colon which was omitted twice during colonoscopy. Small diameter and intermittent closure of the fistula after the bleeding are the reasons for diagnostic problems in typical endoscopy (panendoscopy and colonoscopy). Therefore, most commonly it manifests as recurrent gastrointestinal bleeding episodes. It is important that when a pancreatic pathology coexist, as in the reported case, one must remember of possibility of such bleeding and undertake adequate angiographic diagnostic procedures to accurately detect its cause. Currently angi-CT is an recommended, noninvasive diagnostic procedure. The Authors used it to qualify the patient for the surgical procedure. Often precise diagnosis and finding of a bleeding site is possible only during a laparotomy, as in the re-
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Reported case. Adequate and rapid diagnostic workup allows for proper and effective treatment of the patient, which was emphasized by the Authors of the presented paper.

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