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

ARKADIUSZ FABISZEWSKI, MAREK JACKOWSKI
Clinic of General Gastroenterological and Oncological Surgery, Collegium Medicum of Nicolaus Copernicus University in Toruń
Kierownik: dr hab. M. Jackowski, prof. UMK

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-

chezia), typical for the upper gastrointestinal bleeding (1). The most common symptom of lower gastrointestinal bleeding is stool with blood admixture. In majority of cases (3/4 of cases), the cause of gastrointestinal bleeding is found in the upper gastrointestinal tract. The most common cause of upper gastrointestinal bleeding is duodenal ulcer (26-40%) and gastric ulcer (10-20%), then hemorrhagic ulcerative gastritis (11-20%), varices of the esophagus and gastric fundus (10-16%) and Mallory-Weiss syndrome (10-12%). Neoplasm is a less common cause of bleeding (approximately 2%) (1).

Causes of lower gastrointestinal bleeding are somewhat different. They differ depending on the patient’s age. The most common cause of lower gastrointestinal bleeding in subjects
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 postinflammatory pancreatic 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 colon was found. The cyst was lined with an endothelium-like tissue. The bleeding vessel, branching from
the splenic artery, was underpinned. A site after the hematoma removal with microperforation, was found in the wall of the transverse colon. Single sutures were placed on the wall of the transverse colon. The splenic artery was underpinned. Splenectomy and drainage of the abdominal cavity were performed. The postoperative period was uncomplicated. On day 12 after the operation the patient was discharged home in good general condition. On 31.12.2008 the patient was again admitted to the Clinic with symptoms of gastrointestinal obstruction. He was operated on 2.01.2009 and intraoperatively an adhesion-related obstruction of the small intestine was found. The adhesions were freed along the whole small intestine. The patient was discharged home on 9.01.2009. Currently the patient is in good general condition, without any complaints, undergoes periodic follow up in the Outpatient Department of Surgery.

**DISCUSSION**

In this paper we present a rare case if 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-
rule out stenosis. Other less common complicati-
on include lack of image, lack of transition of
the capsule in the specified time window, me-
chanical 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, enteroclysis), angiography, enteroc-
scopy (push enteroscopy, double balloon or in-
traoperative enteroscopy) are also used to
demonstrate a potential source of atypical
gastrointestinal bleeding in the small intest-
tine. 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 histo-
pathological examination and to use endo-
scopic methods of stopping the bleeding (ther-
mal, injection-based or mechanical). Limita-
tion 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 enter-
scopy over push enteroscopy is ability to assess
the whole small intestine. The whole small
intestine can be also assessed with X-ray ex-
aminations such as gastrointestinal passage or
enteroclisis (an examination involving admin-
istration of methylcellulose and barium to the
duodenum through a probe) (8). Both these
methods are significantly cheaper than the
previously described methods, and their avail-
ability 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 scintigra-
phy 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 inva-
sive and efficient only when the blood loss is
at least 0.5 ml/min. However, it allows for
concurrent stopping of the bleeding with em-
bolization or intravascular drug administra-
tion (e.g. vasopressin). It is especially recom-
ended 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 com-
monly are a consequence of surgical aortic
repair and damage of the intestinal wall by
implanted vascular grafts. Primary aortointes-
tinal fistula results when the disease pro-
cess 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, tubercu-
losus), penetrating peptic ulcer, malignant in-
filtration, 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 oc-
curs in the proximal anastomosis of the graft
and aorta. It results from continuous adhesion
of the implanted vascular graft to the intesti-
nal 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 “fus-
tulas”, 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 intes-
tine (4%), stomach (3%), appendix vermiformis
(1%), rectum (0.5%), are less common. The time
between fistula formation and the gastrointes-
tinal 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 en-
doscopy 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 sig-
nificant risk of endoscopy is triggering of severe
hemorrhage by removal of a clot that blocked
the fistula lumen. Computed tomography imag-
ing is very helpful when aorto-intestinal fistula
is suspected. Ectopic mass, fluid and soft tissue
in the region of the graft, local intestinal thick-
ening and pseudoaneurysm are suggestive of
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. 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.

REFERENCES

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 (pseudoneurysm, 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 angio-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-
Atypical recurrent massive gastrointestinal bleeding

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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