Hemosuccus pancreaticus caused by rupture of a splenic artery pseudoaneurysm complicating chronic alcoholic pancreatitis : an uncommon cause of gastro-intestinal bleeding

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Abstract

We present a case of a 52-year old female patient with intermittent gastrointestinal bleeding and iron deficiency anaemia. Repeated endoscopic investigation revealed no diagnosis, but contrast-enhanced computed tomography showed a splenic artery pseudoaneurysm secondary to chronic alcoholic pancreatitis. A distal pancreatectomy and splenectomy was performed.

Hemosuccus pancreaticus is an uncommon cause of gastrointestinal bleeding, most frequently associated with chronic pancreatitis. Erosion of a peripancreatic artery by a pseudocyst can cause a pseudoaneurysm and rupture occurs in up to 10% of the cases. Bleeding from a pseudocyst wall or rupture of an atherosclerotic or traumatic aneurysm is rare. Angiography, contrast-enhanced computed tomography and endoscopic findings can be diagnostic in the majority of cases. Angiographic embolization or surgery are both therapeutic options depending on underlying nonvascular pancreas related indications requiring surgery. We discuss diagnostic pitfalls and current therapeutic strategies in the management of this disease. (Acta gastroenterol. belg., **2015**, 78, **427-430**).

Key words : hemosuccus pancreaticus, wirsungorrhage, gastrointestinal bleeding, chronic pancreatitis.

Introduction

Hemosuccus pancreaticus, gastrointestinal bleeding from the Ampulla of Vater via the pancreatic duct, is an uncommon cause of upper gastrointestinal bleeding, most frequently associated with chronic alcoholic pancreatitis. Chronic pancreatitis is reported as the cause of pancreatic disease in 75-90% (1). Lower and Farrell reported the first case in 1931. The term "hemosuccus pancreaticus" was first introduced by Sandblom in 1970. Literature on hemosuccus remains limited to individual cases and small series.

The reported incidence of hemosuccus pancreaticus as the cause of upper gastrointestinal bleeding is 1 per 1500 cases of upper gastrointestinal tract haemorrhage (1-3). There is a predominance for men with a reported 7:1 sex ratio, especially in relation to alcohol intake (1,3). Hemosuccus complicates underlying pancreatic disease in 80% of the cases, while 20% are related to vascular anomaly (1). The mean age at onset is about 50 years in patients with underlying pancreatic disease versus 60 years when the etiology is of primary vascular origin (4). Different causes of vascular anomalies are summarized (Table 1), and most commonly involve a primary aneurysm of a digestive tract artery : the splenic artery in 60-65%, followed by the gastroduodenal (20-25%), pancreaticoduodenal (10-15%), hepatic (5-10%) and left gastric arteries (2-5%) (1,6). Aneurysms are usually asymptomatic until they rupture leading to the release of red blood in the duodenum via the papilla (1).

Other rare causes of hemosuccus include ectopic pancreas and pancreas divisum, arteriovenous malformations, pancreatic carcinoma and villous adenomatosis. Hemosuccus pancreaticus can also occur during acute pancreatitis after necrosis of an arterial wall, or can occur as an iatrogenic complication of endoscopic retrograde cholangiopancreaticography (2).

Determination of the exact bleeding location can be a diagnostic challenge and may cause a delay in treatment. Hemosuccus is most commonly caused by rupture of a splenic artery pseudoaneurysm associated with acute or chronic pancreatitis. Pseudoaneurysms of the hepatic, gastroduodenal and pancreaticoduodenal artery have also been reported as causes of bleeding (5). They result from either autodigestion of the peripancreatic artery or erosion of a pseudocyst into the arterial wall with conversion of its cavity into a pseudocyst (6). Arterial aneurysms and pseudoaneurysms have a known prevalence up to 10% in chronic pancreatitis (1,2,7), but no causal relationship between chronic pancreatitis and aneurysm has been clearly established. Pancreatic pseudocysts and pancreatolithiasis are uncommon causes of hemosuccus pancreaticus. Several mechanisms might be involved. A pseudocyst can be haemorrhagic or can erode pericystic arteries. Intraductal stones and a dilated cystic main duct can cause vascular ulceration.

We present a case of hemosuccus pancreaticus and discuss diagnostic pitfalls and current therapeutic strategies in the management of this disease.

Case Report

A 52-year old female patient presented with cramping epigastric pain and recurrent melena. Two years previously, she was diagnosed with chronic alcoholic

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Table 1. — **Causes of vascular anomalies**

Atherosclerotic aneurysm
Traumatic aneurysm
Hereditary dystrophy of elastic tissue
Fibromuscular dysplasia of the arterial wall
Portal hypertension
Vasculitis
Syphilis
Alpha-1 antitrypsin deficiency

Table 2. — **Chronological overview of admissions and diagnosis**

Chronic alcohol abuse
03-2003 : Alcoholic hepatitis
03-2007 : Episode of epigastric pain, chronic pancreatitis
04-2007 : Acute on chronic pancreatitis
07-2007 : Acute on chronic pancreatitis, hyperplastic gastropathy
08-2008 : Acute on chronic pancreatitis
03-2009 : Gastric variceal bleeding
05-2009 : Acute on chronic pancreatitis
07-2009 : Diverticular bleeding
09-2009 : Haemorrhoidal bleeding

pancreatitis after a first episode of epigastric pain radiating to the back. She was admitted several times since then, because of multiple episodes of acute on chronic pancreatitis, epigastric pain, gastrointestinal bleeding and anaemia (Table 2), without diagnosis of a distinct bleeding focus.

Clinical examination on admission showed a hemodynamically stable, mildly anaemic patient. Biochemical analysis revealed iron-deficiency anaemia with serum haemoglobin of 8.4 g/dL (reference range : 14-18 g/dL). Liver, kidney and pancreatic function tests were in the normal range. Gastroduodenoscopy was performed, but no distinct bleeding focus could be identified. Because of the recurrent symptoms with a biochemical diagnosis of chronic anaemia, further diagnostic work-up was planned. Contrast-enhanced computed tomography (Fig. 1) with 3D angiographic reconstructions (Fig. 2) revealed a not previously described contrast capturing splenic artery pseudoaneurysm, presumably secondary to chronic alcoholic pancreatitis. The patient developed acute abdominal pain and became hemodynamically unstable with a significant drop of haemoglobin until 4.7 g/dL. Urgent distal hemipancreatectomy and splenectomy was performed and post-operative recovery was uneventful.

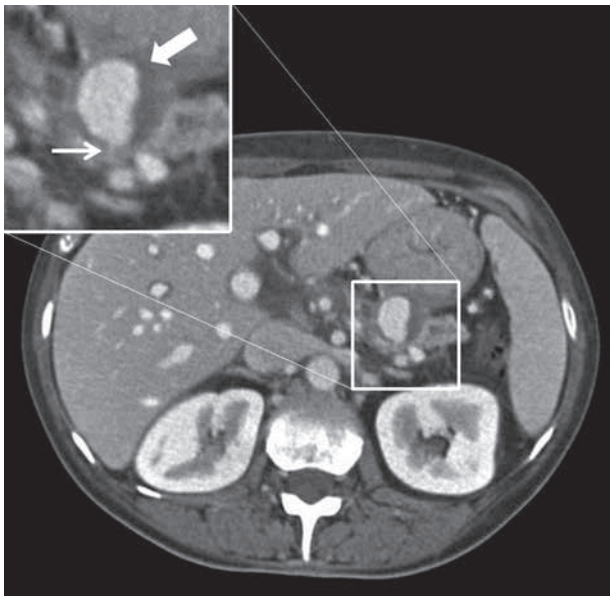

Fig. 1. — Contrast-enhanced computed tomography cross section showing a splenic artery pseudoaneurysm (broad arrow). In the inlet, the neck of the aneurysm can be appreciated (small arrow).

Discussion

We presented a typical case of hemosuccus pancreaticus as a cause of gastro-intestinal bleeding. An history of chronic alcoholic pancreatitis is present in the majority of cases and the onset of symptoms during the 5th decade is typical. Difficulty to confirm the diagnosis lead to prolong the clinical course and can cause a potentially life-threatening treatment delay.

Patients can report different symptoms, including melaena, episodic abdominal pain, hematemesis, hematochezia and combinations of these symptoms. Even though potentially life-threatening, clinical presentation may be similar to chronic pancreatitis (8). Rupture of a bleeding pseudoaneurysm is associated with a mortality rate of 12-57% (2). Iron-deficiency anaemia can be the only symptom (1). Prevalence of iron deficiency in one series of 17 patients was reported to be 58% (9). Elevated pancreatic enzymes might be present, caused by rapid dilatation of the pancreatic duct due to bleeding. Increased pressure within the duct leads to abdominal pain irradiating posteriorly and a tamponade of the aneurysm. The clot is then lysed and the cycle repeats itself (1,2,10). Digestive bleeding can occur up to 48 hours later, relieving the pressure and the pain. None of these symptoms however are pathognomonic. The type of pain and time sequence, elevated pancreatic enzymes and gastrointestinal bleeding are a triad that should create suspicion of a hemosuccus pancreaticus (1,9).

Diagnosis of the exact location of bleeding can be a diagnostic challenge since the site of intermittent bleeding is not readily accessible by endoscopy. Upper gastrointestinal endoscopy demonstrates active bleeding from the Ampulla of Vater in 30% of patients, and allows exclusion of other forms of upper gastrointestinal bleeding (1). In case of intermittent bleeding and normal findings on endoscopy, the procedure should be repeated.

Endoscopic retrograde cholangiopancreaticography may either confirm the diagnosis or raise suspicion. Filling defects within the pancreatic duct suggest the

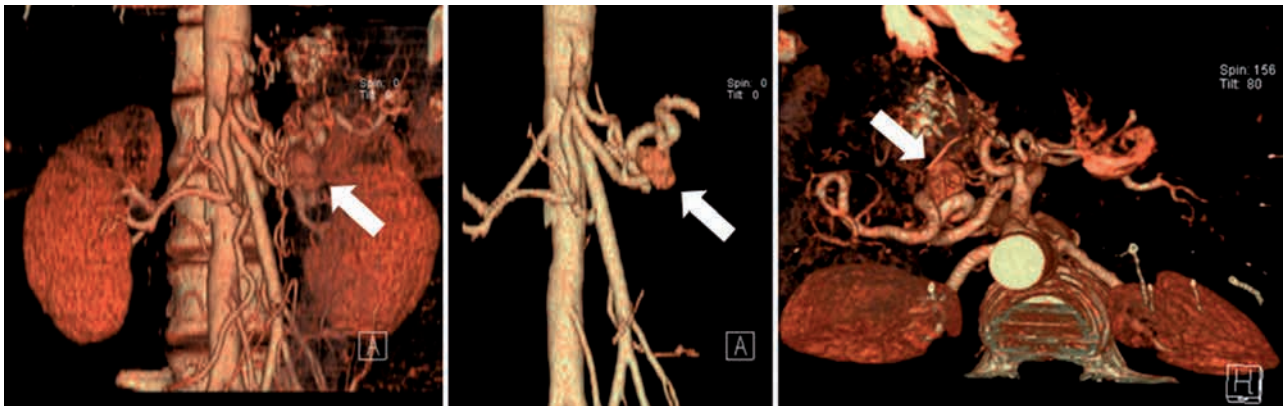

Fig. 2. — 3D-angiographic reconstructions of contrast-enhanced computed tomography clearly demonstrates a splenic artery pseudoaneurysm (arrow), allowing evaluation of its relation to neighbouring structures.

presence of blood clots, or rarely, of a pseudoaneurysm impinging on the lumen (9). Angiography remains the gold standard for diagnosis with a 96% sensitivity (1,6). In hemodynamically stable patients, high resolution multidetector CT with the possibility of 3-dimensional angiographic reconstructions increases diagnostic accuracy, even in periods without active bleeding (7). Contrast enhanced CT has a sensitivity of 96% and remains an excellent modality for characterizing pancreatic pathology, features of chronic pancreatitis, pseudocysts and pseudoaneurysms (3). MRI and MR angiography have added advantages over a CT scan including radiation reduction and angiography-like image reconstruction (4). Hemosuccus pancreaticus should be included in the differential diagnosis in patients with unexplained upper gastrointestinal bleeding and repetitive negative investigations. Underlying chronic pancreatitis should augment suspicion. In our patient, prior upper gastrointestinal endoscopy showed no bleeding focus or alterations of the duodenum. Further investigation with CT angiography and 3D reconstructions allowed visualisation of the pseudoaneurysm and evaluation of its relation to neighbouring structures.

Management of hemosuccus pancreaticus aims to eradicate the source of bleeding. Two therapeutic strategies are generally accepted.

Endovascular treatment is suggested as the preferred first line of therapy during an active episode of pain, when there is active bleeding (10). Endovascular treatment includes coiling, gel foam embolization, detachable balloon occlusion and stent grafting of the source of bleeding (4). The success rate of angio-embolization approaches 67-100% in hemodynamically stable patients (1,3,6). Coil embolization techniques provoke thrombus formation and can cause ischemia if collateral circulation is insufficient. Implantation of arterial covered and uncovered stents has been reported as a successful therapy in selected cases (11). Recurrence rates of 30% at six months after endovascular treatment have been observed (1). Endovascular techniques employed as either definitive therapy or as temporizing measure might be effective in selected patients, particularly in the elderly and those at high operative risk (2).

Surgical therapy is indicated in hemodynamically unstable patients, when angiography shows abnormal findings not suitable for endovascular treatment, after failure of endovascular treatment or when underlying nonvascular complications of pancreatitis require surgery. Reported procedures include transcystic ligation of the bleeding vessel with internal or external drainage of the pseudocyst, external ligation of the feeding vessel, hemipancreatectomy with or without splenectomy, pancreatectomy or rarely pancreaticoduodenectomy (12). Surgery is required in 17-37% of patients with recurrent bleeding following embolization (6). Most surgical series have a documented success rate of 70-80% with mortality rates of 20-25% and rebleeding rates of 0-5% (1,3,6). In patients with chronic pancreatitis, pancreaticoduodenectomy or hemipancreaticosplenectomy should be preferred, as leaving behind a diseased gland in close proximity to the previously injured artery is leaving the patient with an organ prone to recurrent pancreatitis and possible recurrence of arterial injury and bleeding (10). Although endovascular therapy is suggested as the preferred first approach during active bleeding, our patient was managed surgically. First of all, limited availability and expertise in endovascular treatment made this approach less attractive in a hemodynamically unstable patient. Secondly, there was a known history of chronic alcoholic pancreatitis and the recently revealed splenic artery pseudoaneurysm was highly suspected as the origin of hemorrhage. This could guide surgical exploration to diagnose the origin of bleeding and facilitate treatment. Furthermore, at the acute event, surgical therapy was readily available and was therefore preferred in this hemodynamically unstable patient. Hemipancreatectomy with splenectomy was successfully performed without complication.

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