Hemosuccus pancreaticus: a rare cause of gastrointestinal bleeding
A series of 9 cases

Sandrine ETIENNE, Patrick PESSAUX, Jean-Jacques TUECH, Paul LADA, Emilie LERMITE, Olivier BREHANT, Jean-Pierre ARNAUD

Service de Chirurgie Viscérale, CHU Angers, 4 rue Larrey, 49033 Angers Cedex 01.

SUMMARY

Aim — The purpose of this study was to analyze the diagnostic and therapeutic features of hemosuccus pancreaticus.

Methods — We conducted a retrospective study of nine patients (eight men and one woman, mean age 60 yrs) admitted to surgery or gastroenterology units for hemossucus p ancreaticus between 1981 and 2003. The following were studied: symptoms, contribution of established morphologic exams (upper digestive endoscopy, computed tomography (CT) and selective digestive angiography) and treatment.

Results — Hemosuccus pancreaticus occurred in chronic alcoholic pancreatitis (N=8) and chronic familial pancreatitis (N=1). Seven patients (77.8%) presented overt digestive bleeding (one melena, two hematochezia, two melena with hematochezia, one hematemesis, one hemorrhagic shock). The inaugural sign was anemia in one patient and epigastric pain in another. An upper digestive endoscopy, performed in eight patients (88.9%), revealed fresh red blood in the first or second duodenum in three and hemosuccus pancreaticus in three others. Arteriography was performed in eight patients (88.9%) and CT angiography in one. Surgery was performed in 5 patients (55.6%), after embolization in one. Embolization was effective in 3 patients. Therapeutic abstention proved successful in one patient. There were no deaths.

Conclusions — Hemosuccus pancreaticus is a rare cause of digestive bleeding. Upper digestive endoscopy and selective digestive angiography during active bleeding can provide the diagnosis. Endovascular treatment can control an unstable hemodynamic situation before elective surgery to prevent recurrence, which can be more severe than the first event.

Introduction

Acute digestive bleeding originating from the pancreas is the least frequent cause of upper gastrointestinal bleeding (1/500) [1, 2]. Hemosuccus pancreaticus is a rare cause of pancreatic bleeding, the prevalence being much lower than for segmentary portal hypertension or duodenal ulceration. About one hundred cases have been reported in the literature [1] since the first report by Lower in 1931 [3]. Blood is expelled into the duodenum via the main pancreatic duct. Several terms have been used to describe the phenomenon including wirsungorrhagia proposed by Vankemmel [4] in 1969 and currently used in France or the equivalent hemowirsungia proposed by others [2, 5]. Hemosuccus pancreaticus was proposed by Sandblom in 1970 [6] and hemoductal pancreatitis by Longmire and Rose in 1973 [7]. All these terms describe the emission of red blood along pancreatic ducts through the papilla.

The purpose of this study was to analyze the diagnostic and therapeutic features of hemosuccus pancreaticus.
Patients and methods

The files of nine patients with hemosuccus pancreaticus diagnosed between 1981 to 2003 were reviewed retrospectively. We noted demographic data, past history, diagnostic features (symptoms, results of the physical examination, time to diagnosis), results of complementary examinations, and therapeutic modalities as well as follow-up data.

Results

The main results are summarized in table I.

There were eight men and one woman, mean age 60 years (range: 35-70). The eight men had chronic alcoholic pancreatitis and five of them (55%) presented several episodes of acute pancreatitis. Four (44%) had a pseudocyst of the pancreatic head. One patient had histologically proven alcoholic cirrhosis (Child-Pugh A5). Two patients had a history of melena, associated with hematemesis in one. In the first patient with isolated melena, no cause could be identified. In the second with melena and hematemesis, upper digestive endoscopy demonstrated red blood in the second duodenum, probably issuing from a rupture of a pseudocyst of the pancreas. This patient had chronic familial pancreatitis. None of the patients were taking anticoagulant or anti-platelet drugs.

Five patients (55%) presented overt bleeding from the lower digestive tract: melena in one, hematochezia in two, melena and hematochezia in two. One patient presented with hematemesis. Two patients (22%) experienced pain in the epigastric region without digestive bleeding. The diagnosis was established in one other patient undergoing exploration for iron-deficiency anemia.

In one patient physical examination revealed a pulsating epigastric mass with a thrill at auscultation. Five patients (55%) had anaemia (serum hemoglobin 6.7-9.2 g/dL), associated with hemorrhagic shock in one. This latter patient required several packed red cell transfusions.

Upper digestive endoscopy was performed in eight patients (88.9%) and revealed bright red blood in the first or second duodenum in three cases (33.3%). Hemosuccus pancreaticus was visualized directly in three patients (33.3%). Two patients exhibited signs of portal hypertension (22.2%): cardial varices in one and grade II esophageal varices in the second. One patient with portal hypertension developed hemorrhagic shock during the endoscopic procedure. An attempt to drain a pancreatic pseudocyst lead to spontaneous fistulization of the pseudocyst directly into the duodenum with intracystic bleeding; drainage of the pseudocyst failed and the patient complained of violent pain in the right flank, followed by hemodynamic shock (with hemoglobin of 6.7 g/dL).

Abdominal computed tomography (CT) was performed in three patients (33.3%) and yielded information suggestive of the diagnosis (with iv contrast visualised in a pseudocyst located in the head of the pancreas in one patient (figure 1) and a large bleeding arterial aneurysm in another).

Selective arteriography of the celiac trunk and the superior mesenteric artery was performed in eight patients (89%) and enabled the etiological diagnosis in all cases. An angio-CT, performed in one patient, ruled out bleeding from a pseudocyst in the head of the pancreas and excluded the diagnosis of an aneurysm.

An esophagogastroduodenal barium study was performed in one patient and only demonstrated a widened duodenum. Abdominal ultrasonography was performed in two patients and enabled the diagnosis of aneurismal bleeding from the splenic artery in one case and a cyst fistulized into an artery in the head of the pancreas in another. Endoscopic ultrasonography, performed in one patient, revealed a pseudocyst in the head of the pancreas fistulized into the superior mesenteric artery.

An arterial abnormality was found to be at cause in five patients (55.6%): three aneurysms of the splenic artery (figure 2) including one eroded into a pseudocyst of the tail of the pancreas, one aneurismal dilatation of the duodenopancreatic artery and erosion of the gastroduodenal artery by a pancreatic pseudocyst, and a pseudo-aneurysm of the gastroepiploic artery directly fistulized into the duodenum. The four pancreatic pseudocysts were all located in the head of the pancreas and eroded a neighboring artery: the common hepatic artery and the gastroduodenal artery in one patient, arterioles arising from the superior mesenteric artery and the duodenopancreatic arcades in one, the gastroduodenal artery in one, and the superior mesenteric artery in one.

Surgery was undertaken in five patients (55.6%) to control the bleeding: after failure of arterial embolization in one and in an emergency setting in four. Procedures included distal pancreatectomy with splenectomy (N = 2), pancreatecoduodenectomy (N = 2), and total pancreatectomy associated with splenectomy (N = 1). There were no deaths. The patient who underwent total pancreatectomy had a biliary fistula treated medically and right-sided pneumonia. Mean follow-up of the operated patients was 54 months (range: 12-84). None developed recurrent bleeding. Three patients underwent arterial embolization (33.3%) which was successful and sufficient (after a second embolization in one patient). Two patients developed infectious complications: splenic necrosis with superinfection in one patient who required long-term antibiotic therapy and superinfection of a pancreatic pseudocyst drained endoscopically in one (a lost double pigtail stent and a naso-cystic drain); drainage itself was complicated by moderate stenosis of the pancreatic sphencter. These two patients were followed for 12 and 14 months and neither experienced recurrent bleeding. The other patient died at 64 months from lateral amyotrophic sclerosis with a frontal syndrome and swallowing disorders but had not experienced recurrent bleeding or complication. For the patient with chronic familial pancreatitis, active bleeding was not visualized on the angio-CT and no curative treatment was undertaken. This patient was followed for 42 months and remained symptom free; abdominal ultrasound demonstrated the presence of persistent calcifications and decreased size of the lesions in the head of the pancreas.

Discussion

Hemosuccus pancreaticus is a rare cause of acute digestive bleeding, observed predominantly in men (sex ratio 7:1), especially in relation to chronic alcohol intake. Mean age at onset is about 50 years when the pancreatic parenchyma is involved and about 60 years when purely of arterial origin [8]. The typical presentation associates abdominal pain and digestive bleeding. The epigastric pain, which irradiates posteriorly [9], is caused by increased intraductal pressure due to blood in the main duct [2, 10]. Digestive bleeding, generally melena or hematemesis, more rarely hematochezia, occurs 48 hours later, calming the pain. This type of pain and this time sequence are nearly pathognomonic signs of hemosuccus pancreaticus. The intermittent nature of the bleeding is also very specific [1, 5] and results from formation of a clot in the main pancreatic duct. Other clinical signs are more exceptional: jaundice by pancreatobiliary reflux secondary to clot formation [1, 2], vomiting, weight loss, palpable pulsating epigastric mass with a systolic thrill in the event of aneurysm. Iron-deficiency anaemia is frequent but liver blood tests are normal apart from increased serum bilirubin in the event of pancreatobilary reflux. Serum amylase is normal outside episodes of acute pancreatitis.
Upper digestive endoscopy can visualize active bleeding with red blood via the papilla in 30% of the patients [1], thus providing a certain diagnosis. It can also be normal in the event of intermittent bleeding and should be performed again later [1, 11, 12]. Other causes of upper digestive bleeding (esophagitis, gastritis, gastroduodenal ulcers, esophageal varices) can be ruled out [2] and evidence of point-ulcers in the gastroduodenal wall due to an aneurysm can also be identified. Combined with endoscopic retrograde cholangiopancreatography, endoscopy helps orient the etiological diagnosis by opacifying pancreatic pseudocysts or a communicating arterial aneurysm. Selective arteriography of the celiac trunk and the superior mesenteric artery

Table I. – Clinical and laboratory data in nine patients with hemosuccus pancreaticus.

<table>
<thead>
<tr>
<th>Patient, gender, age Year of diagnosis</th>
<th>Remarkable events in the patient’s history</th>
<th>Clinical presentation</th>
<th>Complementary explorations</th>
<th>Diagnosis</th>
<th>Treatment</th>
<th>Pathology</th>
</tr>
</thead>
<tbody>
<tr>
<td>1, male, 70 years 1981</td>
<td>07/1980: hemorrhagic shock, hematemesis and melena. Negative work-up</td>
<td>Melena</td>
<td>Arteriography</td>
<td>Aneurysm, splenic artery</td>
<td>Surgery: splenopancreatectomy</td>
<td>Chronic pancreatitis: focal hemorrhagic and necrotic pancreatitis</td>
</tr>
<tr>
<td>2, male, 62 years 1985</td>
<td>Episodes of acute pancreatitis for &gt; 20 years</td>
<td>Hematemesis</td>
<td>Pulsating mass</td>
<td>EGD: red blood in D2 Barium study CT: cephalic pseudocyst Arteriography</td>
<td>Aneurysm, duodenopancreatic arcade and gastroduodenal artery</td>
<td>Surgery: pancreaticoduodенectomy</td>
</tr>
<tr>
<td>3, male, 35 years 1989</td>
<td>Pseudocyst of the pancreatic tail, &gt; 3 cm, stable</td>
<td>Sideropenic anemia</td>
<td>Barium study: hemosuccus pancreaticus Arteriography</td>
<td>Aneurysm, splenic artery eroded into pseudocyst</td>
<td>Surgery: splenopancreatectomy</td>
<td>Chronic pancreatitis: focal hemorrhagic and necrotic pancreatitis</td>
</tr>
<tr>
<td>4, male, 64 years 1991</td>
<td>Hypertension, pseudocyst of the pancreatic head, insulin-dependent diabetes</td>
<td>Pancreatic pain, proctorrhagia, hemoglobin 9 g/dL, stable</td>
<td>Barium study: portal hypertension CT: contrast agent in the pseudocyst Arteriography</td>
<td>Gastroduodenal artery eroded by pseudocyst</td>
<td>Surgery: total pancreatectomy + splenectomy</td>
<td>Hemorrhagic pseudocyst of the pancreatic head with involvement of an arterial branch</td>
</tr>
<tr>
<td>5, male, 70 years 1993</td>
<td>Hypertension, non-insulin-dependent diabetes</td>
<td>Melena + hematochezia Hemoglobin 9.2 g/dL, stable</td>
<td>Barium study: red blood in D1 and D2 Ultrasound: cephalic pseudocyst fistulized into an artery Arteriography</td>
<td>Cephalic aneurysmal pseudocyst fistulized into common hepatic artery and gastroduodenal artery</td>
<td>Cephalic pseudocyst fistulized in the superior mesenteric artery</td>
<td>Embolization of the aneurysmal cyst: failure Second ultraselective embolization</td>
</tr>
<tr>
<td>6, male, 65 years 1998</td>
<td>Cirrhosis Bouvetot tachycardia</td>
<td>Melena + hematochezia Hemoglobin 7.5 g/dL</td>
<td>EGD: red blood in D2 Barium study CT: cephalic pseudocyst Arteriography</td>
<td>False aneurysm of the gastroepiploic artery</td>
<td>Cephalic pseudocyst fistulized in the superior mesenteric artery</td>
<td>Therapeutic abstention</td>
</tr>
<tr>
<td>8, male, 70 years 2003</td>
<td>11/2002: episode of acute pancreatitis Pseudocyst in the pancreatic head, 6 cm</td>
<td>Melena + hematochezia Hemoglobin 7.8 g/dL, stable</td>
<td>Barium study: portal hypertension Ultrasound and CT: intracystic aneurysmal bleeding</td>
<td>Cephalic pseudocyst fistulized in the superior mesenteric artery</td>
<td>Aneurysm of the splenic artery</td>
<td>Embolization of the splenic artery</td>
</tr>
<tr>
<td>9, male, 47 years 2003</td>
<td>Several episodes of acute pancreatitis Several operations for necrosectomy 10/1993 probable episode of hemosuccus pancreaticus Splanchnic alcoholization</td>
<td>Abdominal pain hematochezia Hemoglobin 8.2 g/dL</td>
<td>Barium study: portal hypertension Ultrasound and CT: intracystic aneurysmal bleeding</td>
<td>Aneurysm of the splenic artery</td>
<td>Embolization of the splenic artery</td>
<td></td>
</tr>
</tbody>
</table>

EGD: esogastroduodenoscopy
provides the formal proof of hemosuccus pancreaticus by opacifying the main pancreatic duct [4]. Its sensitivity is 96% [5]. It can localize aneurysms precisely [2] and provide information on the downstream or possible collateral circulation. An arterial aneurysm ruptured into a pancreatic pseudocyst, the digestive tract, or the biliary or pancreatic ducts can also be identified [13]. The procedure carries the risk of iatrogenic arterial dissection which is operator-dependent. Endovascular treatment by supraselective catheterization of the splenic or the gastroduodenal artery can also be performed. Ultrasonography can be used to visualize a pseudocyst in the pancreas or identify an aneurismal mass, and with Doppler duplex, can confirm the vascular nature of the lesions [2, 5, 12]. The same types of images are obtained with computed tomography (CT) and angio-CT which also give information on the type of pancreatic pathology. Hemosuccus pancreaticus is seen as a pool of contrast agent in the pancreatic region. Sometimes a clot can be directly visualized in the main pancreatic duct. 3D angio-CT reconstruction which demonstrated a fistula between an aneurysm and the splenic artery and the pancreatic ducts has been reported in one patient [14]; this finding was also confirmed by magnetic resonance imaging. 99m-Tc-labeled red cell scintigraphy can also visualize the bleeding zone if it is performed during a period of active bleeding [5, 10].

In 80% of the cases, hemosuccus pancreaticus complicates an underlying pancreatic disease; 20% of the cases correspond to a vascular anomaly [1]. In our series, as in others, chronic pancreatitis was the cause of 75-90% of the pancreatic diseases. Several mechanisms could be involved [1]: 1) the natural course of a pseudocyst could produce hemosuccus pancreaticus either because the pseudocyst is hemorrhagic or because it communicates and erodes a pericystic artery [15]; 2) vascular ulceration caused by an intraductal stone; 3) vascular ulceration by a dilated and cystic main pancreatic duct; 4) arterial aneurysm and pseudoaneurysm, known to have a higher frequency in chronic pancreatitis (10%) [12, 16]. Aneurysm and chronic pancreatitis are often associated but no causal relationship has been clearly established [9]. Other pancreatic causes of hemosuccus pancreaticus are rare: neuroendocrine tumor [1], ectopic pancreas [17], pancreas divisum [18].

During an episode of acute pancreatitis, hemosuccus pancreaticus can occur after necrosis of an arterial wall (duodenopancreatic arcade, gastroduodenal artery, splenic artery). Finally, hemosuccus pancreaticus can occur as a complication of endoscopic retrograde cholangiopancreatography.

In patients with an arterial anomaly, most of the cases involve an aneurysm of a digestive artery. There are several causes: atheroma, hereditary dystrophy of elastic tissue (Marfan disease, Ehler-Danlos syndrome), fibromuscular dysplasia of the arterial wall, portal hypertension, syphilis, vasculitis (particularly periarteritis nodosa), $\alpha_1$-anti-trypsin deficiency [19, 20]. By order of decreasing frequency, aneurysms are found in the splenic artery (60%) [21], the hepatic artery (20%), the superior mesenteric artery (5.5%) and its distal branches (3%), the celiac trunk (4%), the perigastric arterial circle (4%), the pancreaticoduodenal arteries (2%) and the inferior mesenteric and colonic arteries (1.5%) [19, 20]. The size of the aneurysm increases progressively until it ruptures into the pancreatic duct, leading to emission of red blood into the duodenum via the papilla. Aneurysms are usually asymptomatic [22]. One case of post-traumatic pseudoaneurysm has been reported [5].
Once the hemodynamic situation is under control, interventional radiographic methods are used for the initial treatment. Two techniques can be used: balloon occlusion and placement of prosthetic material. Inserting a balloon to obstruct the artery before open surgery can limit bleeding and shorten operative time, particularly in the event of portal hypertension secondary to thrombosis of the splenic vein or massive hemorrhage [1]. The balloon can be deflated and left in place for secondary use if needed. Coil embolization techniques provoke a thrombus in the aneurysm but also obliterate the artery [19, 23]. Ischemia can develop in the tissue supplied by the artery if the collateral circulation is insufficient, if the thrombosis is too extensive or if the embolization material migrates. Embolization of the celiac trunk, the common hepatic artery, or the superior mesenteric artery is thus contraindicated because of the important risk of ischemia and excessive thrombosis. Two other complications are aneurismal infection and splenic infarction which can be prevented by stent insertion: non-coated metallic stents favor the formation of a new arterial intima [11]. Smaller coated stents with a more flexible deployment system may become available in the future [14]. The overall success rate after endovascular is 80% (75% in our series) with a 70% recurrence-free rate at six months [24]. Conservative surgery by ligation of the pancreatic ducts can also be employed but results are unsatisfactory as the causal lesion remains intact. Arterial ligation is more effective but does not avoid the risk of recurrence. Drainage of the pancreatic pseudocysts, which can lead to serious recurrent bleeding, or arterial ligation associated with drainage is particularly effective, but there is a high rate of complications by infection, necrosis, or severe recurrence. More aggressive surgery with pancreatic resection enables treatment of both the pancreatic and arterial diseases. In patients with chronic pancreatitis, pancreaticoduodenectomy or splenopancreatectomy should be preferred. Selective embolization can be performed before surgery to favor hemodynamic control [1, 15, 19]. For patients with acute pancreatitis, arterial embolization followed by ligation or limited selective resection can be proposed. In all cases, the surgical procedure is quite difficult and mortality is high.

In conclusion, hemosuccus pancreaticus is a very rare cause of digestive bleeding. Diagnosis is based on two explorations which should be performed in all patients, and if possible, during a period of active bleeding: upper digestive endoscopy and selective arteriography of the celiac trunk and the superior mesenteric artery. Late diagnosis is however frequent due to the intermittent nature of the bleeding. Endovascular treatment by embolization is effective in most patients but there is no consensus concerning the need for associating surgery to achieve complete cure. As observed in our series, certain authors [25, 26] have reported that patients are free of recurrent bleeding after exclusive endovascular treatment. Other authors [27, 28] have had a recurrence rate to the order of 30%. Several conditions must be considered when deciding on whether or not surgery should be undertaken: the patient’s clinical status and the presence of co-morbid conditions, the type of pancreatic resection considered, the presence or not of a complication requiring surgery. Endovascular treatment can be sufficient to control an unstable hemodynamic situation, but ideally, surgical resection is the only way to prevent recurrent bleeding which can be more serious than the first episode.

REFERENCES


