

Haemosuccus pancreaticus.

How difficult is the diagnosis and how dangerous is the status?

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Haemosuccus pancreaticus (wirsungorrhagia, sanctorinorrhagia, haemoductal pancreatitis, pseudo-haemobilia) is a rare cause of gastrointestinal bleeding. However, the diagnosis could be difficult and the haemorrhage life threatening as demonstrated by Al-Tashi et al. (2) in this issue of the Journal.

The first reference of haemorrhage from the pancreatic duct was made in 1931 by Lower & Farell (cited from 3). The term "haemosuccus pancreaticus" was first used by Sandblom in 1970 (cited from 14). Haemosuccus pancreaticus is most often due to rupture of pseudoaneurysm of the splenic artery in chronic pancreatitis, pancreatic pseudocysts, and pancreatic tumours (5,12,14). Bleeding occurs when a pseudocyst or tumour erodes into a vessel, forming a direct communication between the pancreatic duct and blood vessel. It may also be seen after therapeutic endoscopy on the pancreas or pancreatic duct, including pancreatic stone removal, pancreatic duct sphincterotomy, pseudocyst drainage or pancreatic duct stenting (12). Haemosuccus pancreaticus in a heterotopic jejunal pancreas was reported as an exceptional case report (15). Other rare causes of haemosuccus pancreaticus include trauma and rupture of primary splenic aneurysm (4,14). These primary aneurysms occur more commonly in women, generally presented as a massive intraperitoneal bleeding, most often during pregnancy or in elderly multiparous women (6). Less common sites of rupture of pseudoaneurysms include the peritoneal cavity, retroperitoneum, duodenum, common bile duct, and colon (6,9). Penetration of the pancreatic pseudocyst, perforation into the stomach and massive bleeding is quite exceptional. Such cases were published by Kubo et al. (13) and Shahani et al. (18). Al-Tashi et al. (2) report another case in this issue of the Journal.

Pseudoaneurysms would form when enzyme-rich peripancreatic fluid (often within pseudocysts) leads to autodigestion and weakening of the walls of

adjacent arteries. These arteries then undergo aneurysmal dilatation. At this point, the dilated region is correctly termed an aneurysm rather than pseudoaneurysm, as the blood is still contained within the vessel although with a thinned arterial wall. In some instances, rupture of the aneurysm into the pseudocyst can occur, with conversion of the pseudocyst into a pseudoaneurysm, defined as an extravascular haematoma communicating with the intravascular space. Despite this distinction, both forms are generally grouped together as pseudoaneurysms (6).

Any vessel can be involved in pseudoaneurysm formation but the splenic artery is affected most frequently, followed by the gastroduodenal, superior mesenteric, dorsal pancreatic, hepatic and gastric arteries (6,8,9).

Bleeding from a pseudoaneurysm may be slow and intermittent or acute and massive. The most common course is a rupture of the pseudoaneurysm into the pseudocyst presented with intermittent bleeding into the gastrointestinal tract via the pancreatic duct, associated with abdominal pain (6). In many cases an initial self-limited haemorrhage occurs, followed hours or days later with a massive exsanguinating bleed. The initial self-limited haemorrhage may be due to transient tamponade of the bleeding vessel because of increased pressure within the enlarged pseudocyst (8). If the pseudocyst does not communicate with pancreatic duct, blood may be confined to the pseudocyst, manifested by rapid pseudocyst enlargement, pain and sudden fall of haemoglobin (6).

Diagnosis of haemosuccus pancreaticus can be difficult as bleeding can be intermittent. Thus establishing proper diagnosis may take even several months (10,14,16). Timely duodenoscopy may reveal active oozing haemorrhage from the major or minor papilla (1,11,20), such a finding is suggestive of haemosuccus pancreaticus. The diagnosis can be confirmed by abdominal CT scan, ERCP, angiography,

or intra-operative exploration. If pseudoaneurysm is suspected, CT scan should be performed first because it is least invasive followed by angiography (to define and embolize the pseudoaneurysm).

The literature on haemosuccus pancreaticus has been mostly limited to case reports. Only a few papers published larger sets of patients. Shahani et al. (18) reported six male patients with chronic alcoholic pancreatitis massively bleeding from pseudoaneurysms of the splenic artery. Etienne et al. (7) published a series of 9 cases (from period 1981 – 2003). Seven patients presented with overt bleeding, anaemia was the initial sign in another one and abdominal pain in the last patient. Embolization was effective in 3 cases, surgery was performed on 5 persons (after embolization failure in one). One female patient did not require either surgery or arterial embolization. Nobody from this series died (7). Elton et al. (6) treated three patients with bleeding pancreatic pseudoaneurysm with a combination of embolization and endoscopic drainage of pancreatic pseudocyst (thus avoiding the need for subsequent surgical intervention). Suter et al. (19) gathered 4 cases in a university hospital from 1972 to 1993. Three patients were treated surgically, successful radiological embolization was performed in the fourth case. Woods et al. (21) published four cases of chronic alcoholic pancreatitis, two were operated and two were treated with successful embolization of the pseudoaneurysm. Risti et al. (17) reported three cases of haemosuccus pancreaticus treated surgically.

Upon diagnosis, treatment follows either by means of interventional radiological clotting of the artery (by

coil embolization mostly), surgical resection or ligation (7,8,11). Splenic infarction may follow ligation or clotting of the splenic artery but reduced blood flow to the spleen is usually compensated by sufficient collateralization (3). Benz et al. (3) reported implantation of a metallic stent into the splenic artery as a novel successful treatment of haemosuccus pancreaticus. Unless arterial embolization is performed first, a pseudoaneurysm represents an absolute contraindication to endoscopic intervention (endoscopic drainage of pancreatic pseudocyst).

Haemosuccus pancreaticus is a life threatening condition. A mortality rate of 12.5 % was reported in treated patients and more than 90 % in those untreated. Although reported as a rare complication of chronic pancreatitis, a pseudoaneurysm is encountered in 5 – 10 % of patients with chronic pancreatitis and as many as 20 % of all cases operated on for chronic pancreatitis (8,21). Thus timely diagnosis and treatment seems to result in markedly reduced mortality. Once a pseudoaneurysm has been identified, it should be treated whether or not it has caused bleeding (8).

The differential diagnosis of gastrointestinal bleeding should include haemosuccus pancreaticus, especially when chronic pancreatitis is present, other sources of haemorrhage seem unlikely or have been excluded and a bleeding episode is accompanied by an attack of severe abdominal pain. Mesenteric arteriography with coil embolization can control acute haemorrhage. If bleeding persists or is massive, pancreaticoduodenectomy or pseudocyst resection and ligation of the bleeding vessel definitively prevents rebleeding.

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