Journal of Gastroenterology and Hepatology (1999) **14**, 172–175

ACUTE PANCREATIC LESIONS

CASE REPORT: Haemosuccus pancreaticus: A clinical challenge

GHULAM NABI YATTOO, MOHAMMAD SULTAN KHUROO, NAZIR AHMAD WANI,* KHURSHID ALAM WANI* AND FAYAZ AHMAD BHAT

*Departments of Gastroenterology and *General Surgery, Sher-i-Kashmir Institute of Medical Sciences, Soura, Srinagar 190 011, Kashmir, India*

Abstract

***Background*:** Haemosuccus pancreaticus is a rare complication of pancreatitis. It is a diagnostic problem for even the most astute clinician and a challenge for the expert endoscopist. We report a 25-year-old male patient who had all the features usually seen in haemosuccus pancreaticus patients: recurrent obscure upper gastrointestinal bleeding, pancreatitis, pseudocyst formation, ductal disruption, fistula and pancreatic ascites. The patient was treated by subtotal pancreatectomy, splenectomy and drainage of the pseudocyst. Although pancreatic duct communication with the surrounding vasculature could not be ascertained, we strongly believe the patient had haemosuccus pancreaticus because, over a follow-up period of 3 years, the patient was not only ascites free, but did not experience any further upper gastrointestinal bleeding. We believe that in evaluating patients with recurrent obscure gastrointestinal bleeding, one should always remember that the pancreas is a part of the gastrointestinal tract and, like other organs, is prone to blood loss.

Key words: aneurysm, chronic pancreatitis, fistula, haemobilia, pancreatic ascites, pseudoaneurysm, upper gastrointestinal bleeding.

INTRODUCTION

Haemosuccus pancreaticus (HSP) describes the syndrome of gastrointestinal (GI) bleeding into the pancreatic duct (PD) manifested by blood loss through the ampulla of Vater.[1] A rare cause of GI haemorrhage, it is usually the result of rupture of pseudoaneurysms of the splenic artery or its branches into the PD.[2,3] Less commonly, pseudoaneurysms of the hepatic or gastroduodenal arteries are involved.[4] Most cases occur in acute or chronic pancreatitis, either of which may be a contributory factor in the pathogenesis of pseudoaneurysms.[5] Communication between the PD and surrounding vasculature is hardly ever demonstrated, although rarely, selective coeliac angiography may localize extravasation through the PD or aneurysmal vessel.[6] Distal pancreatectomy and splenectomy is the treatment of choice in most cases.[2,7]

CASE REPORT

A 25-year-old patient was referred to our Gastroenterology Unit with a 1 month history of rapidly accumulating ascites demanding frequent therapeutic paracentesis at his local hospital. The patient had a past history of recurrent upper GI bleeding (haematemesis and/or melaena) with normal upper GI endoscopy at each bleed. For the first 2 years each bleed was preceded by sudden upper abdominal pain relieved immediately after haematemesis or passing a melaenous stool. More recently, the bleeding episodes have become painless. The origin of bleeding remained obscure with investigations showing a normal coagulation profile, normal lower intestinal tract at colonoscopy, normal barium follow through and normal coeliac angiography. Over a period of 5 years, the patient had been transfused with 24 units of group-specific blood and was

Correspondence: Ghulam Nabi Yattoo, Assistant Professor, Department of Gastroenterology, Sher-i-Kashmir Institute of Medical Sciences, Soura, Post Bag No. 27, Srinagar 190 011, Kashmir, India.

Accepted for publication 26 August 1998.

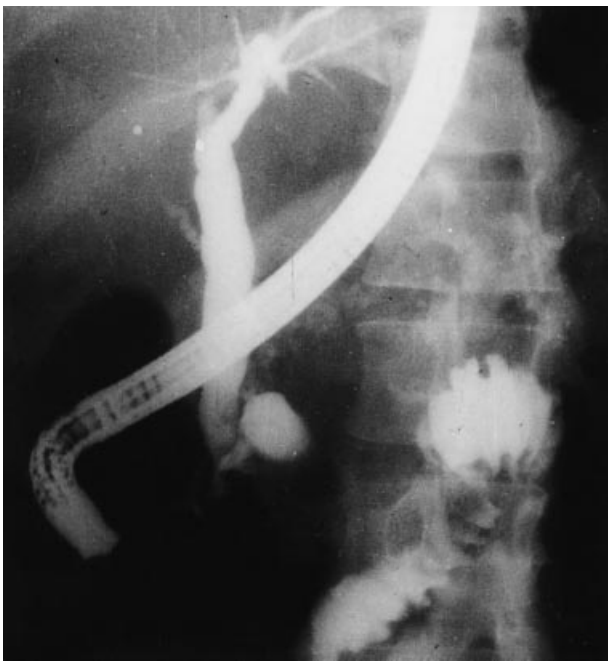

Figure 1 Endoscopic retrograde cholangiopancreatography demonstrates a pseudocyst (cystic cavity opacified by contrast) in the head of the pancreas. Marked changes of chronic pancreatitis are present in the pancreatic duct including tortuosity, sacular dilations and strictures. Contrast is retained poorly in the pancreatic duct because of leakage from a fistulous opening. The bile duct is normal.

facing long-term iron therapy with his haemoglobin concentration never exceeding 8 g/dL (range 4.5–8.0 g/dL). On initial clinical examination he was emaciated, tachypnoeic, anaemic and with orthostatic hypotension. Chest examination revealed bilateral basal dullness and a haemic cardiac murmur. Abdominal examination revealed tense ascites with fluid thrill. There was no abdominal visceromegaly when examined after the ascites were drained. Except for a low haemoglobin (5.4 g/dL), complete blood counts, liver and renal function tests were normal. Ascitic fluid analysis revealed a very high amylase (1200 IU/L) level, suggesting pancreatic ascites. Ultrasonography (US) and computerized axial tomography (CAT) scanning showed fluid in the peritoneal cavity, a normal hepatobiliary tree, an echogenic pancreas with a dilated (1.1 cm) PD and a cystic mass near the head and neck of the pancreas consistent with a pancreatic pseudocyst. An upper GI endoscopy showed a normal oesophagus, stomach and duodenal bulb. In the second portion of the duodenum near the major papilla, a puddling of pink-coloured fluid was noted: it was positive for occult blood, suggesting the diagnosis of HSP. Endoscopic retrograde cholangiopancreatography (ERCP; Fig. 1) revealed a normal biliary tract, a large cystic cavity in the head region of the pancreas, marked changes of chronic pancreatitis in the PD, including dilation, strictures and tortuosity. During ERCP, contrast was seen leaking out of the PD suggesting a PD fistula. In view

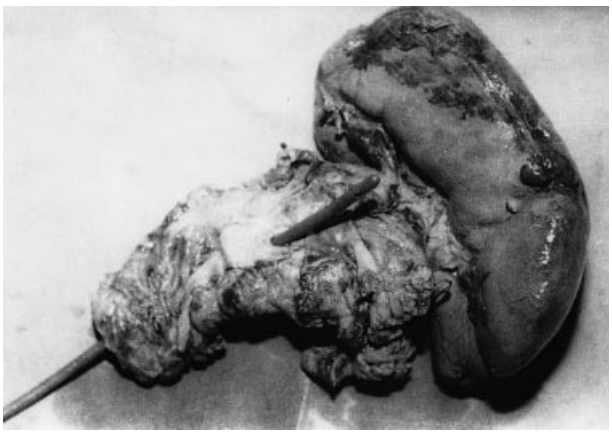

Figure 2 Resected specimen of the pancreas and spleen. A pancreatic duct fistula is demonstrated by passing a rubber catheter through the pancreatic duct. The other end of the catheter is seen coming out through the fistula. The pancreas shows much scarring and is adherent to the spleen.

of these findings it was decided to operate and subtotal pancreatectomy, splenectomy and drainage of the pseudocyst were performed. The PD fistula and extensive scarring of pancreas with the spleen adherent to it were readily demonstrated on the resected specimen (Fig. 2). On histopathological examination of the specimen, changes of chronic pancreatitis were seen. The patient had an uneventful postoperative period. Six months after surgery, the patient developed diabetes mellitus that was controlled with insulin therapy. Over a follow-up period of 3 years, there has been no recurrence of GI bleeding or ascites. The haemoglobin at his last follow-up visit was 14.5 g/dL. The clinical response to surgery performed on this patient supports the diagnosis of HSP, although communication between the PD and surrounding vasculature was not documented.

DISCUSSION

Haemosuccus pancreaticus is a syndrome of blood loss through the pancreatic duct and is usually accompanied by pancreatitis. In most cases, a severe bout of pancreatitis precedes, sometimes by years, the first episode of bleeding. Haemosuccus pancreaticus should be suspected in any patient with pancreatitis in whom GI haemorrhage develops without an obvious source. Bleeding can be vigorous with rapid development of shock, requiring urgent operative intervention to identify the source of bleeding. In other instances, the urgency of the condition becomes apparent only if the bleeding continues or occurs repeatedly in patients who have exhibited only mild clinical symptoms. Thus, the diagnosis may be overlooked at initial evaluation, only becoming apparent after repeated hospital admissions and unsuccessful operative procedures.[6] Although bleeding into a pancreatic pseudocyst is somewhat more common, the term HSP defined by Sandblom[1] is properly applied only to blood loss that occurs into the GI

tract through the ampulla of Vater. Other than a report by Longmire and Rose of a young girl with HSP caused by a gastric and duodenal duplication that ulcerated and bled through the ampulla of Vater,[8] the cause of this disease has always been identified as an aneurysm or pseudoaneurysm of a visceral artery. The splenic artery is most frequently involved. Blood issuing forth from the ampulla of Vater at upper GI endoscopy has been reported to be highly specific for HSP. In fact, all three cases reported to date with this endoscopic finding were confirmed to have HSP at angiography.[2,6,7] A variety of procedures has been reportedly successful in treating this problem, ranging from transarterial electrocoagulation, embolization or balloon occlusion to pancreatic resection.[9–12] In most cases, the procedure of choice has been distal pancreatectomy with splenectomy.[2,3,7] Some authors have advocated proximal and distal ligation of the aneurysmal vessel, with either oversewing of the ductal communication or drainage of a pseudocyst, if present.[9,12]

To date, some 23 cases of HSP have been reported, most commonly in the setting of chronic pancreatitis with rupture of pseudoaneurysms of the splenic, gastroduodenal or hepatic artery into the PD. In most of these cases, pseudoaneurysms with or without extravasation into the PD, were outlined by selective coeliac angiography, abdominal US and CAT scanning.[2,7,9] However, in some cases, PD communication with surrounding vasculature was appreciated only at surgery or after histopathological examination of the resected specimen. Reviews of bleeding associated with HSP note several common clinical presentations: sudden abdominal pain associated with the appearance or enlargement of an abdominal mass considered to be a pseudocyst; a bruit heard over a pancreatic mass; sudden unexplained blood loss in a patient with a known pseudocyst; GI bleeding in a patient with a known or suspected pseudocyst; or abdominal pain and evidence of bile duct obstruction as blood passes through the common channel of the pancreatic and bile ducts.[7,13] The pain produced by this bleeding is secondary to rapid ductal distention, which also causes the leak to seal as pressure in the duct rises. As the duct is decompressed through the ampulla of Vater, clinical GI blood loss is seen and pain subsides. The primary role of upper GI endoscopy in patients with associated pancreatic disease is to eliminate other potential bleeding sites. A negative upper endoscopy, with the exception of blood in the second portion of the duodenum, is suggestive but not diagnostic for HSP. In contrast, blood seen issuing from the papilla is a much more specific finding.[2,6,7]

In patients with recurrent, obscure upper GI bleeding who have suspected but not proven pancreatic disease, a reasonable next step is to image the pancreas with abdominal US, CAT scanning and ERCP. A rapidly enlarging cystic mass demonstrated by US in a patient with pancreatitis, recurrent pain, and GI bleeding should alert the clinician to the possibility of HSP and lead to further evaluation. Pulsatile flow in a cystic mass has been described in some patients with pseudoaneurysms using pulsed Doppler ultrasound.[14,15] Technetium-99m-labelled red blood cell scanning has also been used successfully to localize bleeding into the GI tract.[16] The combination of an enlarging cystic pancreatic mass seen on US and radionuclide pooling in the same area is probably sufficient evidence to confirm the diagnosis of HSP. The rapidity, safety and ease with which these procedures are performed makes them very useful as early screening procedures in these patients, who are often quite ill and tolerate more lengthy, complex procedures poorly.

Computerized axial tomography scanning is a valuable procedure for diagnosing abnormalities of the pancreas, particularly masses and pseudocysts. Findings in patients with pseudoaneurysm include high-density fluid (blood) in the pseudocyst, a central transiently enhancing circular vessel within a cystic pancreatic mass or blood in the PD.[7,17] Any of these findings should prompt urgent further evaluation by means of angiography. Bolus dynamic CAT scanning appears to have the highest sensitivity. Endoscopic retrograde cholangiopancreatography is also an excellent diagnostic modality in suspected, but not proven, chronic pancreatitis complicated by a pseudocyst and fistula as shown in the present case. Bowers *et al*. demonstrated multiple clots in the PD of a patient with haemorrhage from a ruptured aneurysm of the splenic artery.[18] The procedure also helped demonstrate a filling defect representing a clot in the duct in a location corresponding to that of the pseudoaneurysm found on angiography.[2]

Of all the techniques available, angiography is the gold standard for the demonstration of pseudoaneurysm. Angiographical findings include encasement and distortion of vessels and extravasation of contrast material without obvious pseudoaneurysm.[2,3,19] To demonstrate extravasation through the PD, angiography should be performed during the patient's attack of pain, because this is when bleeding is actually taking place. If angiography is delayed until blood is seen, only the aneurysmal vessel will be found; this discovery leads to clinical suspicion but not to definitive diagnosis. Angiography also allows for precise operative planning if surgery is anticipated, or for angiographic therapy, if needed, at the time of diagnostic study.

In summary, HSP may be a potentially life-threatening event during the course of chronic pancreatitis. The key to timely and appropriate evaluation and therapy requires a high suspicion of this vascular complication of pancreatitis. Only early recognition and treatment will allow a timely application of the therapeutic options which are presently available. Non-invasive radiological evaluation can assist in suggesting the diagnosis, but angiography is the cornerstone of both evaluation and planning of therapy. Nevertheless, a highly suggestive clinical history and endoscopic visualization of blood from the ampulla of Vater may suffice for the diagnosis of this rare clinical entity.

REFERENCES

1 Sandblom P. Gastrointestinal hemorrhage through the pancreatic duct. *Ann. Surg.* 1970; **171**: 61–6.

2 Clay RP, Farnel MB, Lancaster JR *et al*. Hemosuccus pancreaticus: An unusual cause of upper gastrointestinal bleeding. *Ann. Surg.* 1985; **202**: 75–9.
3 Starling JR, Crummy AB. Hemosuccus pancreaticus secondary to ruptured splenic artery aneurysm. *Dig. Dis. Sci.* 1979; **24**: 726–9.
4 Eckhauser FF, Stauley JC, Zelenock GB *et al.* Gastroduodenal and pancreaticoduodenal artery aneurysms: A complication of pancreatitis causing spontaneous gastrointestinal hemorrhage. *Surgery* 1980; **88**: 335–44.
5 Stroud WH, Culloun JW, Anderson MC. Hemorrhagic complications of severe pancreatitis. *Surgery* 1981; **90**: 657–63.
6 Cahow CE, Gurberg RI, Gottlieb LJ. Gastrointestinal hemorrhage from pseudoaneurysms in pancreatic pseudocysts. *Am. J. Surg.* 1983; **145**: 534–41.
7 Forsmark CE, Wilcox CM, Grendell JH. Endoscopy negative upper gastrointestinal bleeding in a patient with chronic pancreatitis. *Gastroenterology* 1992; **102**: 320–9.
8 Longmire WP, Rose AS. Hemoductal pancreatitis. *Surg. Gynecol. Obstet.* 1973; **136**: 246–50.
9 Hamel AE, Pare R, Adda G, Bouteloup PY, Huguet C, Malafosse M. Bleeding pseudocysts and pseudoaneurysms in chronic pancreatitis. *Br. J. Surg.* 1991; **78**: 1059–63.
10 Mandel SR, Jaques PF, Sanofsky S *et al.* Non-operative management of peripancreatic arterial aneurysms. *Ann. Surg.* 1987; **205**: 126–8.
11 Stechman MI, Dooley MC, Jaques PF *et al.* Major gastrointestinal hemorrhage from peripancreatic blood vessels in pancreatitis treatment by embolotherapy. *Dig. Dis. Sci.* 1984; **29**: 489–97.
12 McMahan MJ, Playforth MJ, Hill GL. The management of hemorrhagic complication of pseudocysts and abscesses of the pancreas. *Aust. NZ J. Surg.* 1980; **50**: 141–6.
13 Gadacz TR, Trunkey D, Kleffer RF. Visceral vessel erosion associated with pancreatitis. *Arch. Surg.* 1978; **113**: 1438–40.
14 Derchi LE, Biggi E, Cicio GR, Bertoglio C, Neumaier CE. Aneurysms of the splenic artery: Noninvasive diagnosis by pulse doppler sonography. *J. Ultrasound Med.* 1984; **3**: 41–4.
15 Falkoff GE, Taylor KJW, Morse S. Hepatic artery pseudoaneurysm: Diagnosis with real-time and pulsed doppler US. *Radiology* 1986; **158**: 55–6.
16 Bunker SR, Brown JM, McAuley RL *et al.* Detection of gastrointestinal bleeding sites. Use of *in vitro* technetium ^{99m}Tc labeled RBCs. *JAMA* 1982; **247**: 789–92.
17 Burke JW, Erickson SJ, Kellum CD, Tegtmeyer CJ, Williams BRJ, Hansen MF. Pseudoaneurysms complicating pancreatitis: Detection by CT. *Radiology* 1986; **161**: 447–50.
18 Bowers J, Koehler PR, Hammer SP, Nelson JA, Tolman KG. Rupture of a splenic artery aneurysm into the pancreatic duct. *Gastroenterology* 1976; **70**: 1152–5.
19 Vujic I. Vascular complications of pancreatitis. *Radiol. CI North Am.* 1989; **22**: 81–91.