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CASE REPORT

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Sudden death due to rupture of the arteria pancreatica magna: a complication of an immature pseudocyst in chronic pancreatitis

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Abstract Massive haemorrhage due to rupture of single pancreatic or peripancreatic vessels is a very rare but potentially lethal complication of acute and chronic pancreatitis. The splenic, gastroduodenal, and pancreatoduodenal arteries are the more commonly involved vessels, and rupture occurs mostly as a complication of large mature pseudocysts. We report a sudden death due to massive bleeding caused by rupture of the great pancreatic artery (arteria pancreatica magna), a complication of a small immature pseudocyst, in a 49-year-old male alcoholic with inactive chronic pancreatitis.

Keywords Forensic pathology · Sudden death · Pancreatitis · Great pancreatic artery · Rupture

Introduction

Sudden deaths due to factors of pancreatic origin are rare and are generally caused by acute haemorrhagic pancreatitis or diabetes mellitus [1]. Occasionally a massive haemorrhage into the peritoneal cavity, retroperitoneum or gastrointestinal tract can complicate the course of either acute or chronic pancreatitis [2, 3, 4]. Such bleeding is generally caused by erosion of a peri- or intrapancreatic vessel by a pseudocyst, severe inflammation, regional necrosis, or infection. Splenic, gastroduodenal, and superior pancreatoduodenal arteries are the most commonly involved vessels [2, 3, 5].

We report a sudden death due to massive intraperitoneal bleeding caused by rupture of the great pancreatic artery, a complication of a small immature pseudocyst, in a 49-year-old alcoholic male with chronic pancreatitis.

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Case report

Circumstances

On 1st July 2000 at 6:00 a.m. a 59-year-old alcoholic male woke up from his bed and his wife found him a few minutes later trembling and unable to stand up from the toilet bowl. At 8 : 15 a.m. she alerted an ambulance and soon after the man lost consciousness. The emergency doctor arrived at 8:28 a.m. and death was pronounced on site at 8 : 32 a.m. No resuscitation attempts were performed. No history of recent trauma was reported to the investigators and forensic pathologist.

Autopsy findings

On internal examination, there was a massive haemorrhage (3.5–4 l) in the peritoneal cavity with adipose necrosis of the omental fat. Grossly, the pancreas was diffusely fibrotic without significant foci of calcification or calculi. At the middle third of the pancreas body, a 1.5 × 2 cm dark-brown area opened to the inner sac (Fig. 1). A 2-mm probe demonstrated the communication of this small cavity with the great pancreatic artery (arteria pancreatica magna) at about 15–18 mm from its origin from the splenic artery. At the site of artery laceration there was a dark-red blood clot (Fig. 1). The cavity was not delimited by fibrotic tissue and no communication with major pancreatic ducts was found. Additional findings were mild hepatic fibrosis, moderate cardiac hypertrophy, mild coronary atherosclerosis, and mild nodular hyperplasia of the thyroid. No signs of mechanical trauma were found. Toxicological analysis was negative for alcohol and ethylene glycol (GC) in blood and urine. The drug screening (GC) and the search for analgesics, diabetic drugs, phenobarbital, phenytoin, and warfarin (HPLC) in blood and urine were negative.

Pancreas histology

The pancreas showed diffuse, moderate to severe perilobular fibrosis and mild diffuse intralobular fibrosis. The residual acinar tissue was atrophic, the ducts were distorted and contained variable amounts of inspissated proteinaceous material. No intraductal calculi or periductal inflammation were found. Chronic pancreatitis was consistent with stage 9–10 of the 12-stage histological scoring system proposed for pancreatic fibrosis by Klöppel and Mail-

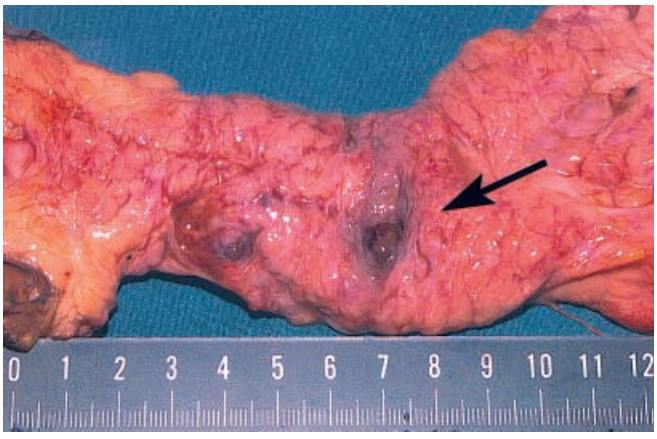

Fig. 1 Small necrotic area on the anterior side of the body of pancreas opened to the inner sac

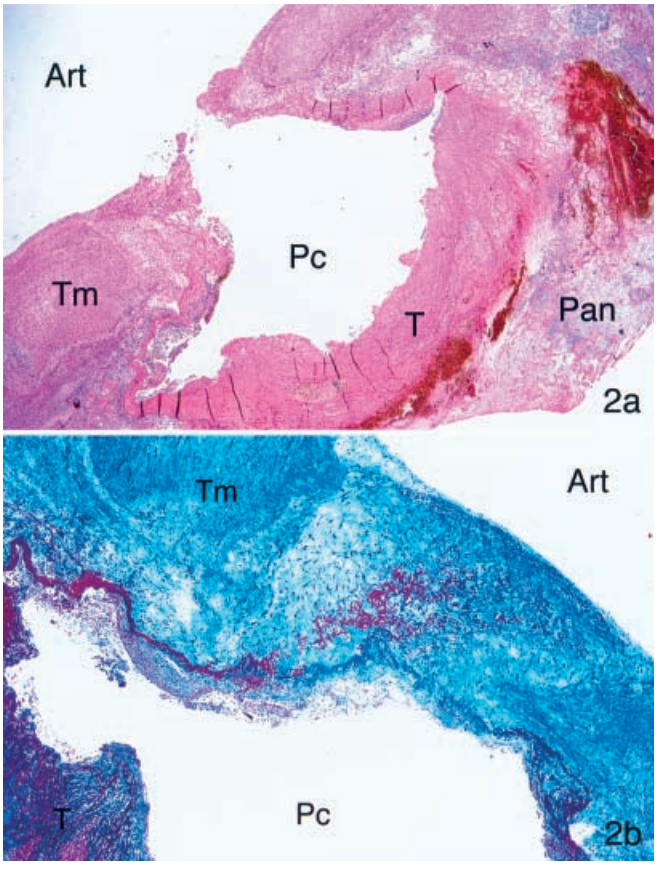

Fig. 2 **a** Wall of the artery at the site of rupture and the pseudocyst cavity with thrombus formation (Herovic, original magnification 10 ×). **b** Detail of the arterial wall at the site of rupture in a successive section showing the disarray of the structure and a recent infiltration of erythrocytes (Masson's trichrome staining; original magnification 40 ×). *Art* arteria pancreatica magna, *TM* tunica muscolaris, *Pc* pseudocyst cavity, *T* thrombus, *Pan* pancreatic tissue

let [6]. Scattered foci of fatty necrosis with resolution by foamy macrophages were observed in the tissue. The samples obtained from the site of vessel rupture were studied through multiple step sections (n = 30, thickness 5 μm, interval between sections 5–50 μm) and analysed with Herovic, PAS, Weigert, and Masson's trichrome staining methods. The laceration of the pancreatic artery was clearly visible (Fig. 2 a) together with recent haemorrhagic infiltration in adjacent sections of disarrayed wall structures (Fig. 2 b). No signs of arteritis were found. The ruptured vessel communicated with a cavity partially filled with a thrombus showing blood cells of varying degrees of freshness, fibrin strands, platelets, neutrophils, and lymphocytes and initial fibroblastic reaction. The thrombus merged with the adjacent fibrotic pancreatic tissue, where residual acinar tissue was still visible. No ductal epithelium, mature granulation tissue, or thick fibrotic capsule lined the wall of the cavity between the thrombus and the pancreatic tissue. On the basis of these histological features the cavity was defined as a post-necrotic immature pseudocyst [7, 8].

Discussion

Acute haemorrhagic pancreatitis is the most frequent cause of sudden death of pancreatic origin. Of 26,428 natural deaths autopsied between 1976–1998 at the Department of Forensic Medicine, University of Helsinki, 0.48% (n = 126) were caused by acute haemorrhagic pancreatitis. In a consecutive series of 1,000 natural deaths autopsied in a US Department of Forensic Medicine, the frequency was lower (0.2%) [1].

Sudden deaths caused by massive haemorrhages from rupture of peripancreatic or pancreatic vessels complicating acute or chronic pancreatitis have been sporadically reported [5, 6]. No data on the frequency of such massive fatal haemorrhages are currently available. A recent review covered 121 cases found in the literature [9]. The above mentioned material of the Department of Forensic Medicine, University of Helsinki, included only three massive bleeding pseudocysts, all in the setting of chronic pancreatitis. In two of these cases, massive bleeding occurred in the gastroenteric tract and death resulted from post-surgical complications, while the third case was a sudden death due to the rupture of the splenic artery caused by a mature pseudocyst.

The incidence of massive bleeding due to local vascular sequelae of acute or chronic pancreatitis ranges from 1.7 to 2.5% of cases [3, 9, 10]. Such complications are more frequently associated with chronic pancreatitis and pseudocysts [5]. A pancreatic pseudocyst is defined as an acute or chronic collection of pancreatic contents enclosed by a non-epithelialised wall, which arise as a consequence of acute pancreatitis, pancreatic trauma, or chronic pancreatitis [5]. Histologically, during the first 2–4 weeks the pseudocyst is not marginated, later the wall becomes lined by granulation tissue and then, in more advanced stages, by a thick dense fibrotic capsule [7, 8]. The pathogenesis, natural history, and relationship between acute and chronic pancreatic pseudocysts are under debate [3, 6, 7, 8, 11] but it is generally accepted that pseudocysts develop as a result of autodigestive tissue necrosis caused by activated pancreatic enzymes [5, 6, 8, 11]. In chronic pancreatitis, the enzyme leakage can be related to ductular

obstruction by protein plugs, calculi, or stenosis, followed by increased intraductal pressure and ductal rupture [7, 8, 11]. However, the view that a pseudocyst in chronic inflammation is a retention cyst secondary to ductal hypertension, has been questioned because of the recurrent lack of ductal epithelium in the pseudocyst cavity and the microscopic similarity between pseudocysts in chronic and acute pancreatitis [11].

The erosion of the arterial wall results from the proteolytic action of the enzymes present in the pseudocyst or, in the case of chronic pancreatitis, also as a consequence of the mechanical or ischaemic action of the pseudocyst [9, 12, 13]. Bleeding occurs either into the peritoneal cavity, retroperitoneum, and gastroenteric tube or into the pseudocyst with formation of a pseudoaneurysm, that can later rupture into the body cavities.

The gastroduodenal artery and its branches, the gastroepiploic and pancreatoduodenal arteries, as well as the splenic artery are vulnerable to erosion because of their superficial and close contact with the head and the upper margin of the tail of the pancreas, respectively. The distal branches of the splenic artery, the arteria pancreatica dorsalis, arteria pancreatica magna and arteria caudae pancreatis, are also critically embedded in the body and tail of the pancreas. The splenic artery is the most commonly damaged vessel followed by the gastroduodenal artery and then the pancreatoduodenal artery [2, 3, 5, 13] but cases involving the gastric and hepatic arteries have also been reported [3, 5, 14, 15]. Occasionally no rupture of major vessels is found and in some of these cases, the cause of haemorrhage is multiple bleeding from the small vessels of the pseudocyst wall [5, 16].

The involvement of the great pancreatic artery has not so far been reported in more than 300 cases of non-fatal and fatal massive haemorrhages recently reviewed [3, 9]. In this case the pseudocyst, in the early stage of its development, had eroded the pancreatic artery. It was about 2 × 1.5 cm in size, while most of pseudocysts producing vascular damage are reported to be 4–8 cm in diameter [8], with a few cases up to 25–30 cm [7, 8]. Moreover, the wall of the pseudocyst was not well marginated, whereas generally, complicating pseudocysts have a mature wall with clearly recognisable granulation tissue and a dense collagen layer or fibrous capsule [8, 11]. The development of such an inflammatory capsule takes nearly 4–6 weeks [7, 8]. In the case described there was only an initial fibroblastic reaction in the thrombus, which usually appears a few days after the onset of haemorrhage.

At autopsy, the demonstration of the source of massive bleeding from pancreatic vessels is easy when a large pseudocyst is involved or when a selective ante-mortem visceral angiography is available to the pathologist. When no clinical data are available, pseudocysts are small, or bleeding occurs from minor vessels, the source of bleeding may be difficult or impossible to determine. In our case, the adiponecrosis foci oriented the investigation to the inner sac, where the necrotic area on the anterior wall of the pancreas was easily detected. In all cases with suspicion of pancreatic vessel anomaly or rupture, the peri- and intrapancreatic arteries should be opened from the celiac trunk and, whenever possible, the usefulness of a post-mortem (PM) celiac angiography [17] carefully evaluated. In some cases this technique is indispensable, for instance, after surgery and with peritoneal and omental adhesions [17]. The introduction of the contrast medium into the vessels can however, remove the content of the pseudocyst thus impeding histological study and age-estimation of thrombi present at the site of haemorrhage. This reason, together with successful macroscopic identification of the source of bleeding, led to the decision not to perform a PM angiography.

In conclusion, this report demonstrates that small immature pseudocysts of the pancreas in the setting of chronic inactive pancreatitis can also serve as complicating factors in fatal haemorrhages involving minor pancreatic vessels. The differential diagnosis between fatal haemorrhages arising from pseudocysts complicating pancreatitis and those arising from post-traumatic pseudocysts or, more in general, pancreatic trauma [18, 19] is crucial from a medico-legal point of view.

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