Hemosuccus pancreaticus

— Report of a case associated with cyst forming undifferentiated carcinoma
of the pancreas and review of the literature —

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A rare case of gastrointestinal hemorrhage due to rupture of a pseudoaneurysm into the pancreatic duct in a patient with undifferentiated carcinoma of the pancreas is described. The diagnosis was made by recognizing bleeding from the papilla of Vater at duodenoscopy and was corroborated by the findings of selective angiography and CT scanning.

Review of the previously reported cases was also performed, and the pathogenesis, diagnosis, and treatment of hemosuccus pancreaticus are discussed in relation to the present case.

This unusual cause of gastrointestinal bleeding should be considered when the common diseases have been excluded.

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Hemorrhage through the pancreatic duct, a rare cause of gastrointestinal bleeding, usually results from rupture of an aneurysm or a pseudoaneurysm of the splenic / hepatic artery or their branches into the pancreatic duct. This condition was first reported in 1931 by Lower & Farrell¹⁾. In 1970, Sandblom²⁾ termed this condition "hemosuccus pancreaticus", while Longmire & Rose³⁾ suggested the name "hemoductal pancreatitis" in 1973 and Favriel⁴⁾ named it "Wirsungorrhagia" in 1979.

Inflammation of the pancreas is usually described as the underlying cause, but others have also been reported including ruptured pseudoaneurysm⁵⁾ and ulceration of the duplicated stomach and duodenum with bleeding through the papilla of Vater³⁾.

The following case report describes a patient with a cyst forming undifferentiated carcinoma of the pancreas who developed major upper gastrointestinal bleeding when a pseudoaneurysm in the cyst wall developed a communication with the pancreatic duct.

Case report

A 70-year-old man was admitted to Aichi Prefectural Hospital in May 1993 with melena and dyspnea on exertion. He had undergone right pneumonectomy 4 years previously

Fig. 1 Duodenoscopy detected the continuous oozing of blood from the papilla of Vater

for stage I large cell lung cancer. Physical examination on admission revealed marked conjunctival pallor. No abdominal pain or tenderness was noticed. Laboratory studies revealed that his hemoglobin value was 4.6 g/dL on admission. Evaluation at that time included endoscopic examination of the upper and lower gastrointestinal tract, barium meal follow through, and intestinal scintigraphy, the results of which were all negative. Because of his continuing melena and severe anemia, he received a transfusion of 12 units (1,560 ml) of packed red blood cells.

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Fig. 2 Ultrasonography revealed a 3.5 cm hypoechoic mass in the body of the pancreas

Fig. 3 Axial computerized tomography showed a pancreatic mass with a relatively high density area inside

Fig. 4 Selective celiac artery angiography showed a pseudoaneurysm of the dorsal pancreatic artery. Neither collateral vessels nor a tumor stain was noticed around the mass in the body of the pancreas

After transfusion, the source of the bleeding was again investigated by duodenoscopy and bright red blood was noticed oozing from the papilla of Vater (Fig. 1). Endoscopic retrograde cholangiopancreatography (ERCP) demonstrated obstruction of the main pancreatic duct at the body of the pancreas. Cytologic examination of the ERCP specimen was negative for cancer. Ultrasonography (US) revealed a hypoechoic, well-demarcated mass about 3.5 cm in diameter in the body of the pancreas and also showed moderate dilatation of the pancreatic duct distal to the mass (Fig. 2).

Simultaneous axial computed tomography (CT) also demonstrated a round mass in the body of the pancreas without contrast enhancement (Fig. 3). Selective celiac angiography showed tortuous vessels surrounding the mass and a large pseudoaneurysm arising from the dorsal pancreatic artery (Fig. 4).

A preoperative diagnosis of pancreatic pseudocyst or cyst-carcinoma as the cause of hemosuccus pancreaticus was made, and surgery was performed on the 49th hospital day. Upon exploration, a cystic mass in the body of the pancreas

Fig. 5 The resected specimen with the spleen at the lower right
The arrow indicates the cystic tumor in the body of the pancreas.

was confirmed, and the body and tail of the pancreas were removed together with the spleen. The postoperative course was uneventful and the patient was discharged after 4 weeks. Seven months later, he is progressing well and has healthy CT scans.

Pathologic findings

The resected specimen consisted of a portion of the pancreas measuring $9.0 \times 4.5 \times 3.0$ cm (Fig. 5). The cut surface showed black thrombus and clots in the cyst and in the pancreatic duct. In the tail of the pancreas distal to the cystic mass, the parenchymal tissue revealed fibrosis and inflammatory changes and the main pancreatic duct was mildly dilated. The cyst itself was a round mass measuring 3.5 cm in diameter and its wall was irregular with multiple small nodules of various sizes. Microscopic examination revealed cells with severe atypia in and around the cyst wall as well as organized thrombus and necrotic tissue. The cyst wall consisted of large nucleated atypical cells without gland formation and the pathological diagnosis was undifferentiated carcinoma of unknown origin (Fig. 6).

Discussion

Although hemosuccus pancreaticus has been considered a rare cause of bleeding from the duct of Wirsung, the improving sophistication and accuracy of the diagnostic modalities available may well increase the chance of identifying this condition.

A total of 33 cases were found by our review of the literature⁶⁻²⁰ (Table 1). The age varied between 15 and 86 years, and the male to female ratio was 2 : 1 (22 males and 11 females). There was a previous history of gastrointestinal bleeding before the diagnosis was confirmed in 79% of the patients. Signs of pancreatitis were also noted in 17 cases¹⁻²⁰. In some patients, pancreatitis was considered to have caused the aneurysm or pseudoaneurysm, and in other cases it may

Fig. 6 Histologic section of the cyst wall
Thrombus was recognized in the cyst and in the main pancreatic duct. The wall of the cyst consisted of undifferentiated carcinoma cells of unknown origin.

have been a secondary reaction to bleeding into the pancreatic duct. The source of bleeding was either an aneurysm or a pseudoaneurysm (with or without a pancreatic pseudocyst) that formed a fistula communication with the pancreatic duct.

Etiology and pathogenesis

In considering the pathogenesis of this condition, differentiation between aneurysm and pseudoaneurysm as the origin of the bleeding is important. In only two cases has the cause of hemosuccus pancreaticus been documented as a true aneurysm of the splenic artery, which was atherosclerotic in nature^{2,13}. In most of the other cases, the bleeding was from a pseudoaneurysm associated with pancreatitis or some other cause. These pseudoaneurysms probably arise as a result of enzymatic destruction of the arterial wall by a mechanism similar to the pancreatic necrosis that leads to hemorrhage and pseudocyst formation^{14,15}. In our present case, unlike the previous ones, the pseudoaneurysm was closely related to a cystic carcinoma. Although the thrombus in the cyst strongly suggested the rupture of a vessel at that site, there is also a possibility that necrosis of a tumor involving a branch of the splenic artery was the direct cause of ductal bleeding.

Concerning the origin of the carcinoma, it could be a primary tumor or a metastasis from the previous lung cancer. A relatively high incidence of pancreatic metastasis of small cell carcinoma of the lung has been reported²¹ and this taken together with the unusual pattern of growth of the cystic tumor suggests the possibility of metastasis. On the other hand, immunostaining with anti-sialyl-Lewis^x antibody (CA 19.9) demonstrated this antigen in the lung cancer but not in

Table 1 Cases of hemosuccus pancreaticus reported in the English literature

Case No.	Age (Yrs)	Sex	History #	Diagnostic Procedure	Definitive Operation*	Involved Artery+	Histological Pancreatitis	Prognosis	Author (Year)
1.	16	M	Abdominal pain & melena	Laparotomy	DP+ligation of artery	SpA	+	Survived	Lower (1931)
2.	80	M	Hematemesis	Autopsy	none	SpA	+	Died	Hentel (1966)
3.	59	M	GI bleeding (subtotal GTX)	Laparotomy	DP+SPX	SpA	+	Survived	Jones (1968)
4.	51	M	GI bleeding (GTX)	Arteriogram	DP+SPX	SpA	+	?	Boijesen (1969)
5.	51	M	Abdominal pain GI bleeding (GTX)	Arteriogram	DP+SPX	SpA	+	Survived	Sandblom (1970)
6.	69	M	Abdominal pain & hematemesis	Arteriogram	DP+SPX	SpA	-		
7.	15	F	Abdominal pain & hematemesis	Arteriogram	PD	PDA	-	Survived	Longmire (1973)
8.	68	M	Abdominal pain & melena	Arteriogram	DP+SPX+ GJstomy	SpA	-	Survived	Soeno (1974)
9.	54	F	GI bleeding	Arteriogram	PD?	IPDA	?	Died	Brintnall (1974)
10.	46	M	Abdominal pain & hematemesis	Arteriogram	DP+SPX	SpA	?	Survived	Lam (1975)
11.	50	F	Melena (colectomy)	Arteriogram	DP+SPX	SpA	+	?	Koehler (1976)
12.	62	F	Melena (colectomy)	Arteriogram	DP+SPX	SpA	+	?	Bowers (1976)
13.	57	M	Recurrent pancreatitis	Arteriogram	Not Stated	SpA	+	?	Walter (1977)
14.	62	F	GI bleeding	Arteriogram	DP+SPX ligation	SpA	+	Survived	Bivins (1978)
15.	59	M	Abdominal pain & hematemesis	Arteriogram	Ligation of artery	SpA	+	Survived	Starling (1979)
16.	73	M	GI bleeding	Arteriogram	DP+SPX	SpA	-	?	Favriel (1979)
17.	54	F	Abdominal pain	Arteriogram	DP+SPX	SpA	+	?	Lung (1980)
18.	57	M	Melena (colectomy)	Arteriogram	DP+SPX	SpA	+	Survived	Newstead (1980)
19.	65	M	Abdominal pain & hematemesis	Arteriogram	SPX+ ligation	SpA	?	Survived	Borjesson (1981)
20.	42	F	Hematemesis & melena	Arteriogram	SPX+ ligation	SpA	?	Survived	
21.	50	M	Abdominal pain & hematemesis	Laparotomy	DP+SPX	SpA	+	Survived	Hall (1982)
22.	29	M	Abdominal pain	Laparotomy+ Arteriogram	Subtotal PX +SPX	CHA	+	Survived	Cahow (1983)
23.	35	M	Abdominal pain	Arteriogram	DP+SPX	SpA	+	Died (liver failure)	
24.	47	M	Abdominal pain & melena	Arteriogram	Ligation of artery	CHA	?	Survived	
25.	38	M	Hematemesis & melena	Endoscopy & Arteriogram	DP+SPX	SpA	+	Survived	Yokoyama (1984)
26.	62	F	GI bleeding	Arteriogram	DP+SPX	PMA	+	?	Clay (1985)
27.	64	M	Abdominal pain	Arteriogram	DP+SPX	SpA	?	?	
28.	50	M	Abdominal pain	Arteriogram	DP+SPX	DPA	?	?	Yoshikai (1986)
29.	51	M	Melena	Laparotomy	DP+SPX+ ligation	SpA	-	Survived	Gutman (1991)
30.	59	F	Abdominal pain	Arteriogram	resection of aneurysm	PHA	?	Survived	Cruz (1992)
31.	86	F	Melena (colectomy)	Arteriogram	resection of aneurysm	SpA	?	Survived	Wagner (1994)
32.	54	F	Anemia	Arteriogram	PD	IPDA	?	Survived	Amaud (1994)
33.	70	M	Melena & dyspnea	Arteriogram	DP+SPX	DPA	- (carcinoma)	Survived	present case

Abbreviations :

#: GI=gastrointestinal, GTX=gastrectomy.

*: DP=distal pancreatectomy, SPX=splenectomy, PD = pancreaticoduodenectomy, GJstomy=gastrojejunostomy, PX=pancreatectomy

+: SpA=splenic artery, PDA=pancreaticoduodenal artery, CHA=common hepatic artery,

PMA=pancreatic magna artery, PHA=proper hepatic artery,

IPDA=inferior pancreaticoduodenal artery

the pancreatic tumors, suggesting that the undifferentiated cystic cancer was a primary tumor of the pancreas.

Diagnosis

The diagnosis of this condition on initial referral to hospital is very difficult to establish, mainly because the bleeding is intermittent in most cases and a definite angiographic or endoscopic diagnosis is possible only during active bleeding. Recently, duodenoscopy and ERCP have come to play an important role in marking the diagnosis. Active bleeding through the papilla of Vater may be visualized by duodenoscopy¹⁷⁾, and indirect evidence of bleeding may also be detected by ERCP demonstrating filling defects in the pancreatic duct after the injection of contrast medium. In our case, duodenoscopy revealed bleeding from the papilla of Vater and ERCP showed obstruction of the duct of Wirsung, suggesting the presence of blood clots. CT was also useful in defining the size and location of the mass and in displaying the dilated distal main pancreatic duct. All these modalities, however, were unable to determine whether the cystic mass was benign or malignant.

Treatment

Pancreatic resection including the pseudoaneurysm and pseudocyst is recommended by most authors as the treatment of choice^{4,8)}. Simple bipolar ligation with excision of the aneurysm¹³⁾, percutaneous transcatheter embolization²²⁾, or balloon occlusion¹²⁾ have also been reported as useful temporary measures. However, with recent progress in operative techniques and supportive therapy, the definitive procedure is presently considered to be distal pancreatectomy combined with splenectomy. Our patient underwent distal pancreatectomy and neither recurrence of the tumor nor recurrent bleeding has been observed to date.

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