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Aneurysm of Pancreatic Artery in Association with Celiac Axis Stenosis: Report of a Case and Review of the Literatures

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Abstract

A 63-year-old Japanese woman with a history of pemphigus was referred to us for abnormal findings of dynamic abdominal CT where three aneurysms of splenic artery and pancreaticoduodenal artery, celiac axis compression, and gall stone. Superior mesenteric artery supplied hepatic arterial flow via pancreaticoduodenal artery. Avoiding transarterial embolization to prompt arterial ischemia of liver/pancreas head/duodenum, she laparotomically underwent cholecystectomy, splenectomy, transection of median arcurate ligament, and ligation of splenic and inferior pancreaticoduodenal artery all at once. Postoperative course was uneventful except drainage of abdominal abscess, and she remained well without aneurysm recurring 40 months post. Important point of treatment for pancreaticoduodenal artery aneurysm associated with celiac artery occlusion/stenosis is both preventive solutions for rupture of aneurysm and hepatic/duodenal/pancreatic arterial ischemia. Remaining main arterial supply for the liver via pancreaticoduodenal artery from superior mesenteric artery would prompt recurrent aneurysm of pancreaticoduodenal artery.

When a clinician encounters a case of pancreatic aneurysm associated with celiac axis occlusion, the case should be treated using with multimodality such as interventional radiology, and vascular surgery.

Key Words: Pancreatic artery aneurysm; Celiac axis stenosis; Hepatic and duodenal ischemia

Introduction

Aneurysm of pancreatic artery in association with celiac axis compression is rare and little discussion has been made in clarifying the treatment, and prognosis. We recently encountered a case of pancreaticoduodenal artery aneurysm associated with splenic artery aneurysms, gallstone, and celiac axis stenosis. We report this case herein and review cases of pancreatic artery aneurysm documented in the English and Japanese literature, discussing the recent treatment, and prognosis.

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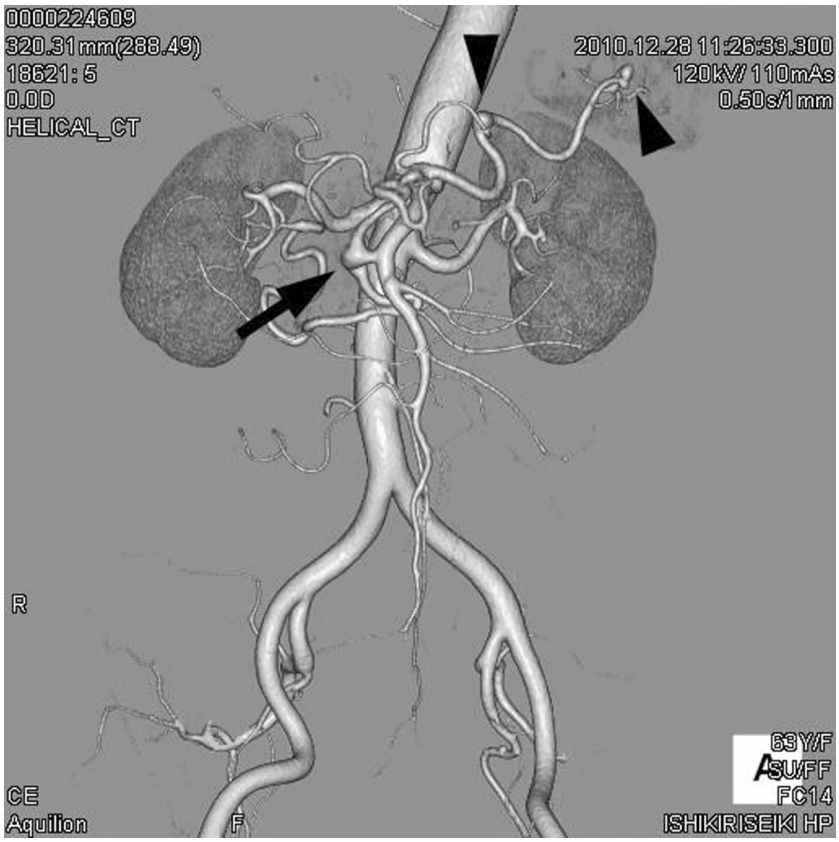

Figure 1. Dynamic computed tomography in early phase showed pancreaticoduodenal artery aneurysm (arrow), and splenic artery aneurysms (arrow heads). Proper and common hepatic artery was demonstrated as a thin line.

Case report

A 63-year-old Japanese woman, who had medical history of duodenal ulcer at 20-year-old, suffered from pemphigus at 59-year-old, and had been given predonisolone (5-50 mg/day). When she was 63-year-old, serum examination revealed liver dysfunction. Physical examination of abdomen demonstrated no abnormality. Although additional some serum examinations suggested that the steroid caused liver dysfunction, Doppler ultrasonography for a further examination, incidentally, demonstrated gall bladder stones, and 16 mm in diameter of aneurysm in pancreas head. Further, dynamic computed tomography demonstrated stenosis of celiac axis, two aneurysms of splenic artery, and aneurysm of pancreaticoduodenal artery. On the Doppler examination, the pancreatic aneurysm demonstrated peak systolic velocity 101 cm/second, flow volume 810 mL/minute, superior mesenteric artery (SMA); peak systolic velocity 92.6 cm/second, flow volume 730 mL/minute, celiac axis; 2.4 cm/second.

Dynamic computed tomography demonstrated aneurysms of pancreaticoduodenal and splenic arterys, and thin common and proper hepatic arteries (Fig. 1). Arterial angiography revealed hepatic arterial flow supplied by SMA via pancreaticoduodenal and dorsal pancreatic artery. On arterial angiography, the aneurysm 16 mm in diameter presented between inferior pancreaticoduodenal and dorsal pancreatic artery, and the aneurysms 6 and 8 mm in diameter presented in middle and hilum splenic artery (Fig. 2). Cardiovascular surgeon consulted our department and radiology. A radiologist suggested deviation of small coils from pancreatic artery prompting ischemic duodenal and, hepatic ischemia, and splenic infarction prompting splenic abscess when interventional radiologic treatment

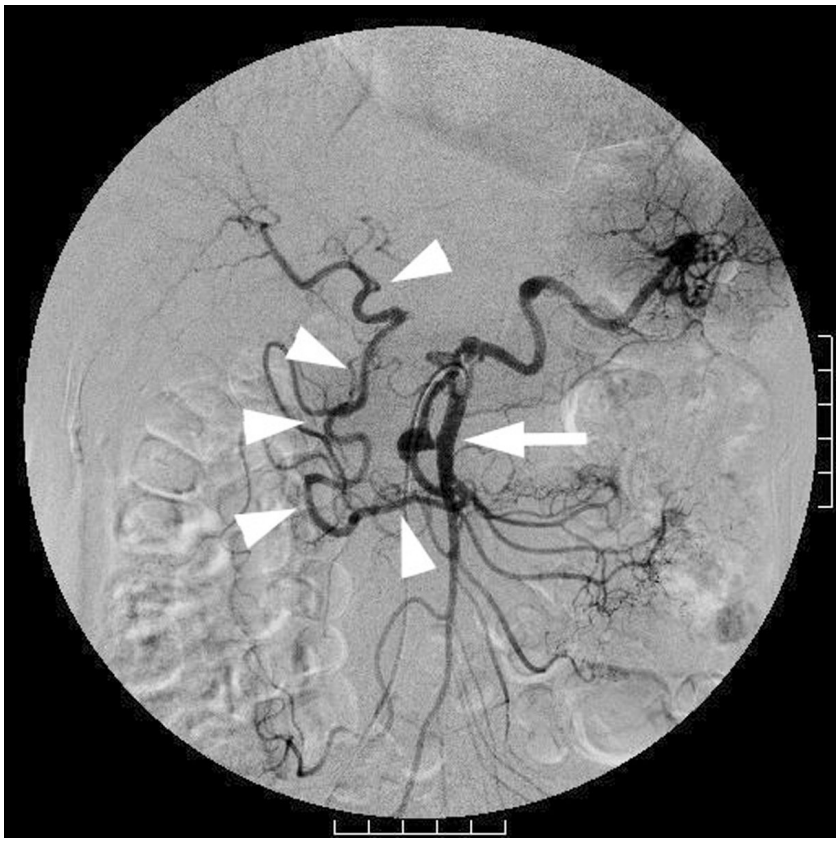

Figure 2. Celiac and superior mesenteric artery angiography demonstrated pancreaticoduodenal artery aneurysm (arrow), and splenic artery aneurysms. Common hepatic artery was demonstrated as thin arterial flow, and meanwhile, proper hepatic artery was normally demonstrated via superior mesenteric artery and pancreaticoduodenal artery (arrow heads).

would be perform.

After informed consent, the present patient, therefore, chose cholecystectomy, transection of median arcurate ligament, ligation of pancreatic aneurysm, and splenectomy with splenic artery under laparotomy. The operation performed by upper median incision. First of all, we performed the transection of the ligament, and confirmed arterial supply for the liver via common hepatic artery (Fig. 3). Second, we set taping the pancreas aneurysm of in/outlet from/to inferior pancreaticoduodenal artery and inlet from dorsal pancreatic artery, and then ligated the in/outlets of the aneurysm after we confirmed the orthodromic arterial flow of pancreas via gastroduodenal artery (Fig. 4). Then, we performed cholecystectomy, and splenectomy with splenic artery. On the Doppler examination, the common hepatic arterial flow demonstrated peak systolic velocity 110 cm/second. Although postoperative abscess formation of pancreatic tail developed, treatments of drainage and antibiotics resolved it.

Postoperative dynamic computed tomography demonstrated normal celiac arterial flow, and fine flow of pancreatic artery arcade. A 40-month follow-up did not reveal any splanchnic aneurysm.

Discussion

Pancreatic artery aneurysm (PAA) is a rare aneurysm, and accounts for 2% of all splanchnic artery aneurysms[1,2]. It is suggested that PAAs were induced by some factors, i.e., atherosclerosis,

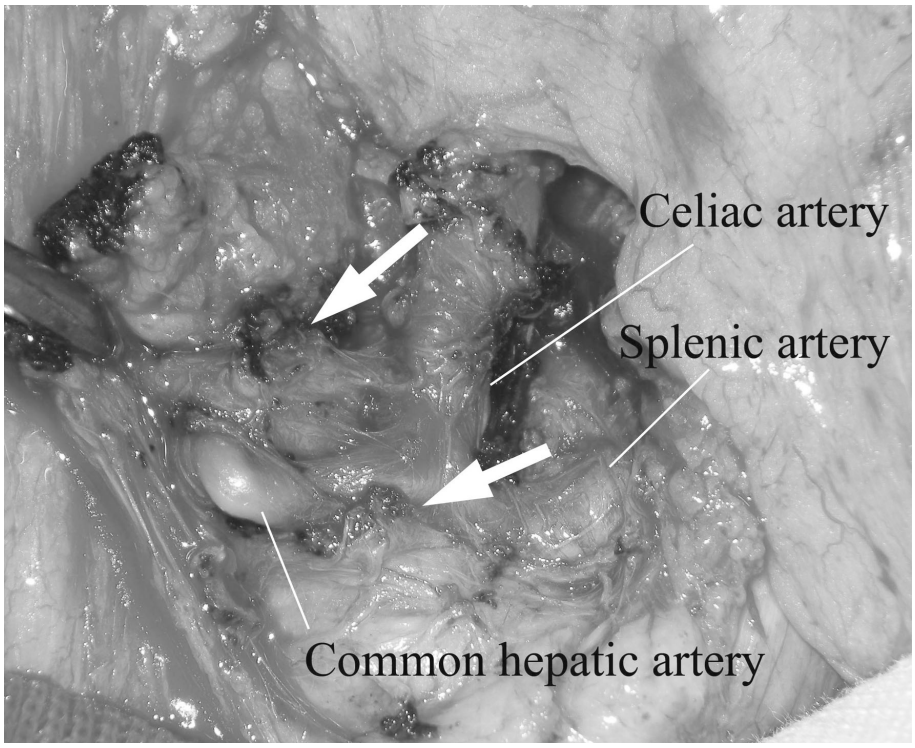

Figure 3. Operative macroscopic finding of celiac artery. Hard fibrous bundle compressed common hepatic artery. Hard fibrous string was cut (arrows), and common hepatic arterial flow increased.

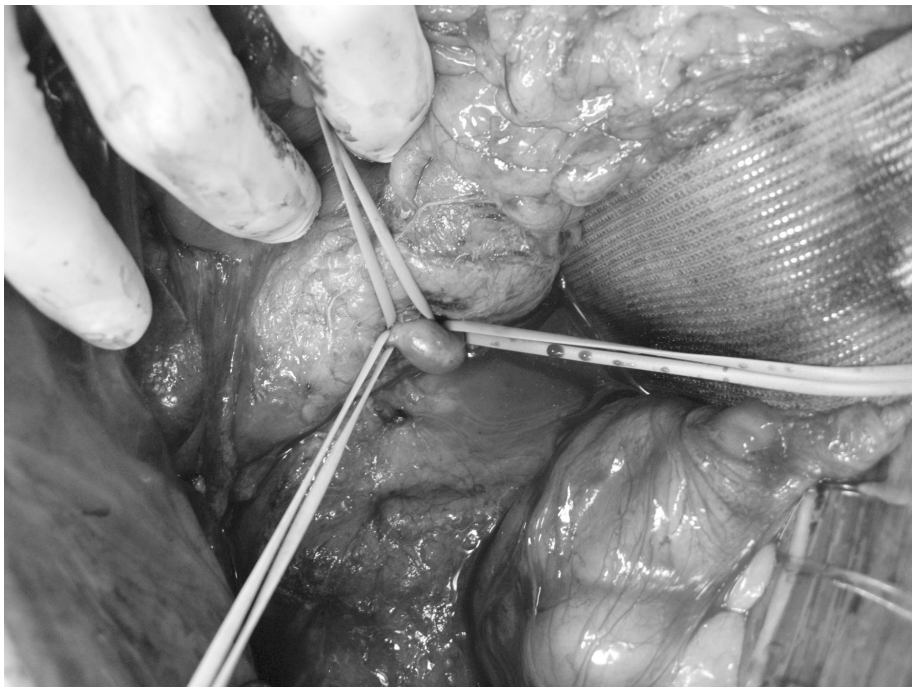

Figure 4. Operative macroscopic finding of pancreaticoduodenal artery aneurysm. After transection of median arcurate ligament, outlet/inlet artery was ligated.

pancreatitis, fibrodysplasia, trauma, congenital anomalies, and high arterial flow[3,4]. Approximately 10% cases of the aneurysm are however associated with celiac artery stenosis/obstruction (CAS) at

high frequency as far as we can review case reports of pancreatic aneurysm including pseudoaneurysm. Some investigators[4-6] suggest that disturbance of hepatic/duodenal/splenic arterial flow by CAS would induce detour of hepatic/splenic arterial flow via superior mesenteric artery and pancreatic artery, and that the increase of pancreatic arterial flow would deform construction of arterial wall and secondarily develop PAA. This mechanism and etiology also resembles intracranial aneurysm associated with blockage of the circle of Willis (moyamoya disease)[7]. In the English and Japanese literatures, 117 cases of PAA with CAS including the present case are introduced by radiologists, surgeons, gastroenterologists, and cardiovascular surgeons[1-6,8-85]. We reviewed and compiled the 117 cases. As some of the paper did not document the cases in detail sufficiently, we did not analyze the data of cases statically. The 117 cases consisted of range 21 to 83-year-old (average 58.6-year old), 58 male and 51 females (sex of 8 cases was not documented in the literatures). Eighty percent of all cases had solitary aneurysm, but 20% had double and more PAA. Five cases were solitary dorsal pancreatic artery aneurysm (DPAA), 106 were pancreaticoduodenal artery aneurysm (PDAA), 3 were unknown pancreatic artery, and 3 were both DPAA and PDAA. Nine cases of PAA (7.6%) had other aneurysm of the other organs. Chief complaint are various, abdominal pain/discomfort, back pain, epigastralgia, icterus, hematemesis, hematuria, gastrointestinal bleeding, intermittent hypertension, lumbago, syncope. Fifty-three cases (45%) are ruptured, 58 (49.6%) were not ruptured, and 6 were not documented at diagnosis. Aneurysms are 4 mm to 75 mm in diameter (mean, 21 mm). Mean diameter of ruptured aneurysm was 17.5 mm, and that of non-ruptured was 23.4 mm. The difference between the mean diameters suggests that PAA should be treated as soon as the aneurysm is found regardless of the tumor diameter.

Fifty-eight cases (49%) were treated interventional radiology (IVR) for PAA (transarterial embolization (TAE), 57 cases; endoaneurysmorrhaphy, one case), and 9 of the 58 were failed IVR. Forty-eight cases (41%) including the 6 cases failed IVR were treated surgical treatment (extirpation, 26 cases; ligation, 12 cases; pancreaticoduodenectomy, 6 cases; hematostasis, 3 cases; clipping, one case), and 18 cases (15%) were not treated for PAA. Thirty-one cases (26%) were treated for treatment for CAS (anastomosis, 16 cases; transaction, 13 cases; stenting, 2 cases; ballooning (failed), 2 cases). Twenty-five cases (21%) were treated for both PAA and CAS.

Twenty of 117 cases had postoperative complication (duodenal stenosis, 6 cases[8-13]; duodenal ischemia, one case[14]; intestinal ischemia, one case[2]; shock, 2 cases; ileus of small intestine, one case; abscess, one case; anastomotic leakage of intestine, hematoma, one case; one case; aortic dissection, one case; cholecystitis, one case; diarrhea, one case; pancreatic fistula, one case; wound infection, one case; and renal failure, one case). All 6 cases with duodenal stenosis were treated for only PAA. Eight cases were dead[2,3,6,15-19], 5 of the 8 dead cases were ruptured cases[2,3,15-17,19], 4 of the 8 cases were not performed any treatment[3,6,16,18]. PAA of 3 cases were persistently present[18,20,21], and PAA treated by only TAE recurred in one case[22].

Revealing prognosis of PAA with CAS, and correlation between treatment, complication, and mortality, the 117 cases were divided into several factors, and statically evaluated. Table 1 is summarized on the viewpoint of the correlation between treatment, postoperative complication, and prognosis (Table 1). There is a tendency to high rate of complication in cases of ruptured PAA (30%).

To reveal therapeutic trend of the contemporary times, dates of reports are classified into 1973-2000 and 2001-2013. The early group (28 years) consisted 51 cases, and the late group (14 years) did 66 cases. This suggests cases of PAA with CAS should tend to increase recently, or development

Table 1. Correlations between prognosis, complications, aneurysm status, and treatment

	Status of aneurysm		Treatment for aneurysm	
	Ruptured	Non-ruptured	Pancreatectomy	Ligation/embolization
Dead	5	3	2	6
Alive	43	30	2	74

	Status of aneurysm		Treatment for aneurysm	
	Ruptured	Non-ruptured	Pancreatectomy	Ligation/embolization
Complicated	14	6	2	18
No complication	33	41	4	84

CAS, celiac artery stenosis/obstruction; IVR, interventional radiology; and TAE, transarterial embolization.

Table 2. Summary of cases with pancreatic artery aneurysm and celiac axis stenosis

	Years of the reports (n)	
	1973-2000 (51)	2001-2013 (66)
Sex (n) (Male : Female)	28 : 18	30 : 33
Mean age±SD (y)	56.9±1.9	59.9±1.5
Mean diameter of aneurysms (mm±SD)	23.5±2.1	18.8±1.7
Mean number of pancreatic aneurysms (mean number±SD)	1.3±0.1	1.2±0.1
Timing of diagnosis (n) (not ruptured : ruptured)	28 : 23	29 : 30
Lesion of pancreatic aneurysm (n) (PDA : DPA : both)	48 : 1 : 2	58 : 4 : 1
Other aneurysm (n) (present : none)	4 : 47	6 : 60
IVR for aneurysm (n) (treated : non-treated)	32 : 16	42 : 23
IVR for CAS (n) (treated : non-treated)	1 : 46	3 : 62
Surgery for aneurysm (n) (treated : non-treated)	26 : 22	21 : 44
Pancreatomy for aneurysm (n) (performed : non-performed)	5 : 43	1 : 64
Surgery for CAS (n) (treated : non-treated)	10 : 37	20 : 45
Any treatment for aneurysm (n) (treated : non-treated)	37 : 11	58 : 7
Any treatment for CAS (n) (treated : non-treated)	10 : 38	22 : 43
Any treatment for both aneurysm and CAS (n) (both : aneurysm only : CAS only)	7 : 30 : 3	18 : 40 : 4
Outcome (n) (alive : dead)	27 : 8	49 : 0
Complications after treatment (n) (present : none)	11 : 31	9 : 50
Months of follow-up (mean months±SD)	13±3.6	17.2±3.3
Recurrent aneurysm (n)	1	0

n, number of cases; SD, standard deviation; PDA, pancreaticoduodenal artery; DPA, dorsal pancreatic artery; IVR, interventional radiology; and CAS, celiac artery stenosis/obstruction.

of recent imaging might find PAA easily. Table 2 is summarized on the viewpoint of some clinical factors. Number of treatment for CAS in 2001-2013 is more than 1973-2000 (Table 2), and a group of the early date is high mortality; in early group, numbers of cases performed IVR for PAA is fewer than those performed surgical treatment. Additionally, many PAA patients with CAS were recently treated both PAA and CAS. We guess that those results reflect recent progress of radiological technology such as spread of dynamic computed tomography, developments of small coil for embolization, small stent, and flexible thin catheter.

Meanwhile, as data of late date in Table 2 demonstrating, recently surgical treatment for CAS is often performed to improve hepatic/splenic/pancreatic arterial flow as well as the aforementioned. Many investigators would suspect that high pancreatic arterial flow as a detour of splenic/hepatic arterial flow would make pancreatic aneurysm. They, therefore, would radically perform reconstruction of hepatic artery connecting to aorta or common hepatic artery to decrease pancreatic arterial flow avoiding recurrent PAA. In fact, PAA recurred in one of 74 live cases not treated for CAS (1.4%).

In the present case, however, it was difficult to perform IVR for both splenic aneurysms and PDAA simultaneously because migration of coils for embolization might develop splenic infarction, hepatic ischemia, duodenal ischemia, and superior mesenteric ischemia. Because we must additionally performed cholecystectomy, splenectomy, and transection of median arcurate ligament, we selected simultaneous laparotomic multiple treatments and ultrasonographic examination, i.e., laparotomic ligations of splenic aneurysms and PDAA, splenectomy, cholecystectomy, transection of median arcurate ligament, and intraoperative Dopller method. If the present case had not cholelithiasis or splenic aneurysms, we would choose to perform IVR for PDAA, and surgical transection of CAS, following the recent trend.

As additional new knowledge obtained by the present analysis, postoperative complication of duodenal stenosis developed in seven PDAA cases not treated for CAS. Except one case caused by hematoma secondary to rupture of PAA[23], we guess that duodenal ischemia would cause duodenal stenosis of six cases[8-13]. This suggests that TAE would block arterial flow both aneurysm, pancreas head, and duodenum up when PDAA is treated TAE.

In summary, we encountered a rare case of PDAA with CAS, splenic artery aneurysms, and cholelithiasis, and performed unique laparotomic treatments simultaneously. Additionally, we revealed that recent trend in treatment for PAA with CAS should be simultaneous treatment for both PAA and CAS. It is the reason that only IVR for PAA should induce other complications, such as recurrent PAA, and hepatic/splenic/duodenal ischemia.

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