

Case report

Pancreaticoduodenal artery aneurysms associated with celiac axis stenosis due to compression by median arcuate ligament and celiac plexus

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Abstract: Celiac axis stenosis is frequently associated with pancreaticoduodenal artery aneurysms. Although the cause of stenosis was not clear in most of the reported cases, compression of the median arcuate ligament of the diaphragm was found to be responsible for the stenosis in 7 of 42 reported cases of this type of aneurysm. We report a case of aneurysm caused by compression of the median arcuate ligament of the diaphragm and celiac plexus. An asymptomatic 43-year-old Japanese man was admitted with a low echoic lesion in the uncus of pancreas. Computed tomographic scan and angiogram revealed stenosis of the celiac axis and two aneurysms in the inferior posterior pancreaticoduodenal artery. The celiac plexus and median arcuate ligament were divided surgically and normal flow was reestablished in the celiac axis. One of the aneurysms was resected and the afferent artery of the other aneurysm was ligated. In the setting of pancreaticoduodenal artery aneurysm associated with celiac axis stenosis, management of stenosis should be considered in addition to local treatment of the aneurysm. In this context, division of median arcuate ligament and celiac plexus or aorto-celiac bypass may normalize the flows in the pancreaticoduodenal arcade and could be effective in preventing aneurysm reformation.

Key words: pancreaticoduodenal artery aneurysm, celiac axis stenosis, median arcuate ligament of diaphragm, celiac plexus

Introduction

Aneurysms of the pancreaticoduodenal arteries (PDA) are rare, accounting for only 2% of all splanchnic artery

aneurysms.¹ Atherosclerosis, infection, congenital defects, pancreatitis, fibromuscular dysplasia, connective tissue disorders, and trauma were found to be related to PDA aneurysms in many cases.^{1,2} Celiac axis stenosis-associated PDA aneurysms have also been reported. In 1973, Sutton and Lawton³ first described this association and there are 43 reported cases to date including the case we report here. The cause of celiac axis stenosis was not clear in most cases. But compression of the median arcuate ligament of the diaphragm was found to be responsible for 8 of the 43 reported cases. Therefore, PDA aneurysm with celiac axis stenosis should be considered as a separate entity in which the stenosis could be the key factor in aneurysm formation. We present a case of asymptomatic PDA aneurysms associated with celiac axis stenosis, and a review of the literature.

Case report

A 43-year-old Japanese man who had been in good health had a medical checkup and was found to have a low echoic lesion in the uncus of the pancreas. He was hospitalized for further investigations. There was no history of pancreatitis, atherosclerotic disease, abdominal trauma, or alcohol abuse. On physical examination no abnormality was found, and results of routine laboratory tests and radiological examinations of the chest and abdomen were normal.

Ultrasound examination revealed a 29 × 22-mm diameter clearly bordered sonolucent mass inside the uncus of the pancreas (Fig. 1). Color Doppler sonography revealed pulsatile arterial flow from the superior mesenteric artery (SMA) toward the sonolucent mass and celiac artery. A dynamic computed tomographic (CT) scan of the abdomen revealed two contrast-enhanced masses, a 30-mm mass inside the uncus of the pancreas and a 15-mm mass located to the right side of the first one (Fig. 2). A selective superior mesenteric angiogram

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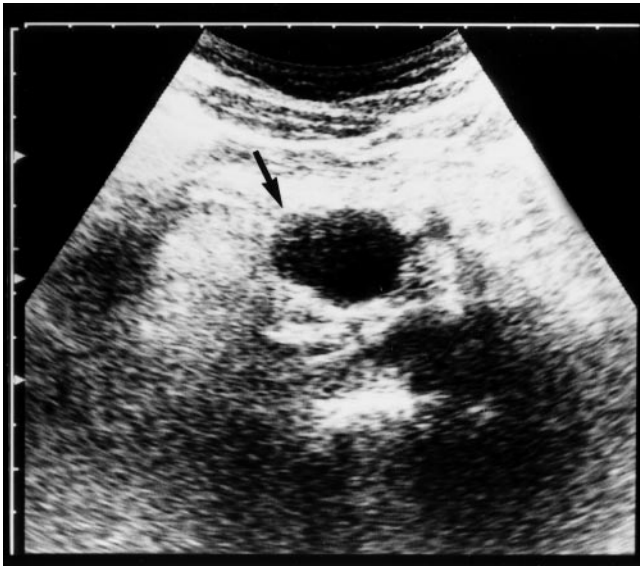


Fig. 1. Epigastric transverse ultrasound scan shows a clearly bordered sonolucent mass in the uncus of the pancreas (arrow)

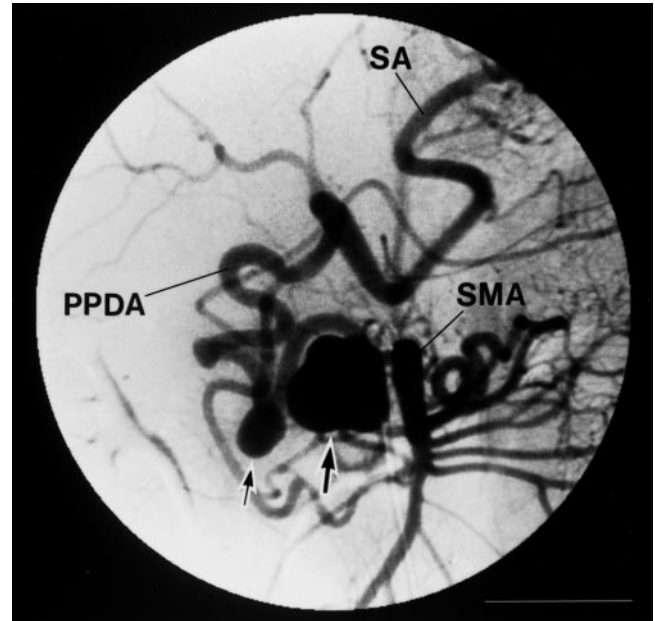


Fig. 3. Selective superior mesenteric angiogram shows a 30-mm aneurysm arising just distal to the origin of the inferior posterior pancreaticoduodenal artery (large arrow) and a 15-mm aneurysm at the distal side (small arrow). The posterior pancreaticoduodenal artery shows dilatation and tortuosity with retrograde filling of the celiac axis area as a collateral pathway. PPDA, Posterior pancreaticoduodenal artery; SA, splenic artery; SMA, superior mesenteric artery



Fig. 2. Dynamic computed tomographic scan of the abdomen shows two enhanced masses (large and small arrows) in the uncus of the pancreas. Ao, Abdominal aorta

revealed one aneurysm just distal to the origin of the inferior posterior pancreaticoduodenal artery and another at the distal side of the first one. The inferior posterior pancreaticoduodenal artery showed dilatation and tortuosity with retrograde filling of the celiac axis (Fig. 3) and well marked stenosis of celiac axis was detected at its origin (Fig. 4). Percutaneous transluminal angioplasty (PTA) was tried for dilatation of the stenosis, but was not effective. At laparotomy, the



Fig. 4. Percutaneous transluminal angioplasty, showing stenosis of the celiac axis. The constricted site in the balloon catheter (arrow) represents the site of stenosis

celiac axis was found to be compressed by the median arcuate ligament of the diaphragm and by the celiac plexus. Once these compression fibers were divided, normal flow at the celiac axis was observed with a flow meter. Since the first aneurysm was embedded in the pancreatic tissue, ligation only of the afferent artery was performed and the other aneurysm was resected. The postoperative course was uneventful and the patient was discharged after normal flow at the celiac axis was confirmed by color Doppler sonography. A 32-month follow-up did not reveal any reformation of the aneurysms.

Discussion

Recently, with advances in investigatory techniques, the association of celiac axis stenosis with PDA aneurysms has been increasingly diagnosed. We presented a case of asymptomatic pancreaticoduodenal artery aneurysms with celiac axis stenosis caused by compression from the median arcuate ligament and celiac plexus. The patient had no history of chronic pancreatitis, infection, or trauma. Histologically there was no evidence of atherosclerosis or abnormality of media and connective tissue. The aneurysms may have developed as a result of increased blood flow in the PDA caused by celiac axis stenosis.

PDA aneurysms associated with celiac axis stenosis have been reported in 43 cases, including ours (Table 1). With the stenosis of the celiac axis, the blood flow to the liver, spleen, and stomach is supplied by the SMA via the pancreaticoduodenal arcade as a collateral pathway. It is presumed that chronic increased blood flow and turbulence through the pancreaticoduodenal arcade weakens the arterial wall, causing dilatation and tortuosity that ultimately lead to aneurysm formation.²⁻⁴

The cause of celiac axis stenosis was not clear in most of the reported cases. A postmortem study revealed that the origin of the celiac axis in relation to the median arcuate ligament was extremely variable; therefore, a lower position of the median arcuate ligament or a higher position of the celiac axis could result in compression to the celiac axis.⁵ The same study demonstrated that the celiac plexus and its supplemental fibers, either alone or together with the median arcuate ligament, may also be a factor in the compression. Median arcuate ligament compression was responsible for celiac axis stenosis in 8 of the 43 reported cases.⁶⁻¹⁰ Atherosclerosis,⁴ thrombosis¹¹ and agenesis of the celiac axis¹² were also reported; in the remainder (32 cases), the causes remained unclear. In the absence of rupture most of the patients had vague symptoms such as abdominal or lumbar pain. In 18 patients, rupture

was the initial clinical manifestation. When rupture occurred, severe intraperitoneal or gastrointestinal bleeding, abdominal pain, shock, obstructive jaundice, or duodenal obstruction caused by expanding hematoma in the region of pancreatic head were reported.^{4,7,9,10,13-24}

In the 43 reported patients with PDA aneurysms associated with celiac axis stenosis, the age at the time of diagnosis ranged from 21 to 78 years (mean, 55 years) and the male/female ratio was 22:21. In the 40 aneurysms in which diameter was reported, it ranged from 4 to 70 mm (mean, 21.7 mm). In patients with rupture, the diameter ranged from 4 to 70 mm (mean, 22.2 mm), while in those without rupture it ranged from 5 to 42 mm (mean, 21.4 mm). Therefore, the size of an aneurysm did not seem to be a determining factor in the causation of rupture.

In 21 patients, only local treatment of the aneurysms was considered, and no attempt was made to resolve the celiac axis stenosis, which may have remained as a risk factor for aneurysm reformation. Although there was no reported case of aneurysm reformation, this may reflect lack of long-term follow-up. Management in these patients included local resection,^{2,8,12,21-30} exclusion,^{3,7} ligation,^{16,20} endoaneurysmorrhaphy,^{19,23} and embolization.²⁴

Revascularization of the celiac axis,³¹ and section of the median arcuate ligament⁶ without resection of the aneurysms led to a favorable outcome and postoperative angiography did not reveal any aneurysms, presumably because of thrombosis within the aneurysm. Recently, in atherosclerotic and nonsclerotic arterial stenosis, satisfactory results have been reported with PTA.^{32,33} In our patient, we tried PTA first to relieve the stenosis, but failure of PTA indicated that the stenosis was caused by severe external compression, thus, an operative approach was undertaken.

Treatment for both the celiac axis stenosis and the aneurysms was performed in only four patients, including ours. Aorto-hepatic¹¹ or aorto-celiac³⁴ bypass together with resection of the aneurysm were performed in two patients. Resection of the aneurysm and section of the median arcuate ligament were performed in one patient.¹⁰ In our patient, along with this procedure, we also sectioned the celiac plexus. In 8 patients with rupture, pancreaticoduodenectomy^{9,13} or segmental bowel resection was required.^{14,18} Another patient died from hematoma bleeding.¹⁵ Ruptured aneurysm was diagnosed only at autopsy in one patient,⁴ while spontaneous obliteration of the aneurysm after retroperitoneal hemorrhage was reported in another.¹⁷ In five patients, without rupture who were asymptomatic, no treatment was given, and none of these showed ruptures during the follow-up period,^{3,24,35} although two patients died from other causes.^{10,24} For three patients, no information

Table 1. Reported cases of pancreaticoduodenal artery aneurysms associated with celiac axis stenosis or occlusion

Author	Age years Sex	Cause of CA stenosis or occlusion	Rupture	Management	Outcome
1. Sutton, 1973 ³	21F	NS	NR	Follow-up	NS
2. Sutton, 1973 ³	69F	NS	NR	Exclusion	Favorable
3. Murase, 1973 ²⁵	48F	NS	NR	Resection	Favorable
4. Mora, 1976 ³¹	44F	NS	NR	Revascularization of CA	Favorable
5. Schefflan, 1977 ¹³	56M	NS	R	PD	Favorable
6. Kadir, 1978 ¹⁴	72M	NS	R	Colectomy	Died
7. Kadir, 1978 ¹⁴	36M	NS	NR	NS	NS
8. Kadir, 1978 ¹⁴	63F	NS	NR	NS	NS
9. Proud, 1978 ⁶	32F	MAL	NR	Section of MAL	Favorable
10. Roback, 1979 ¹⁵	56F	NS	R	Packing of the hematoma	Died
11. Ho, 1979 ⁴	64M	Atherosclerosis	R	Conservative	Died
12. Vermynck, 1979 ¹¹	44F	Thrombosis	NR	Resection, aorto-hepatic bypass	Favorable
13. Kaneko, 1980 ²¹	62M	NS	R	Resection	Favorable
14. Mariano, 1981 ¹⁶	67F	NS	R	Ligation of afferent artery	Favorable
15. Samson, 1981 ²⁶	56F	NS	NR	Resection	Favorable
16. Vernhet, 1982 ⁷	65M	MAL	R	Exclusion	Favorable
17. Vernhet, 1982 ⁷	53M	NS	R	Exclusion	Favorable
18. Thévenet, 1983 ⁸	37F	MAL	NR	Resection	Favorable
19. Matsumoto, 1983 ³⁵	75F	NS	NR	Follow-up	Favorable
20. Lois, 1983 ¹⁷	53M	NS	R	Conservative	Favorable
21. Gangahar, 1985 ²²	61M	NS	R	Resection	Favorable
22. Partensky, 1987 ¹²	38M	Agenesis	NR	Resection	Favorable
23. Ambrosetti, 1987 ⁹	58M	NS	R	PD	Favorable
24. Ambrosetti, 1987 ⁹	59F	MAL	R	PD, Section of MAL	Died of celio- mesenteric ischemia
25. Wrazidlo, 1987 ²⁷	67F	NS	NR	Resection	Favorable
26. Gaa, 1988 ³⁴	44F	NS	NR	Resection, aorto-celiac bypass	Favorable
27. Grech, 1989 ³⁶	50F	NS	NR	NS	NS
28. Grün, 1989 ²⁸	45F	NS	NR	Resection	Favorable
29. Quandalle, 1990 ¹⁰	42M	MAL	NR	Conservative	Died of hepatic failure
30. Quandalle, 1990 ¹⁰	62M	MAL	R	Exclusion, section of MAL	Favorable
31. Granke, 1990 ²³	62F	NS	R	Endoaneurysmorrhaphy	Favorable
32. Granke, 1990 ²³	44F	NS	NR	Resection	Favorable
33. Suto, 1991 ¹⁸	65M	NS	R	Ligation of afferent artery, duodenectomy	Died
34. Chiou, 1993 ²	59F	NS	NR	Resection	Favorable
35. Shibahara, 1993 ¹⁹	57M	NS	R	Endoaneurysmorrhaphy	Favorable
36. Chikamori, 1993 ³⁰	56F	NS	NR	Resection	Favorable
37. Taylor, 1993 ²⁰	67M	NS	R	Ligation of afferent artery	Died
38. Uher, 1995 ²⁴	52M	NS	NR	Resection	Favorable
39. Uher, 1995 ²⁴	69M	NS	R	Embolization	Favorable
40. Uher, 1995 ²⁴	50M	NS	NR	Follow-up	Favorable
41. Uher, 1995 ²⁴	48M	NS	NR	Follow-up	Died of sepsis
42. Iyomasa, 1995 ²⁹	78M	NS	NR	Resection	Favorable
43. This report, 1997	43M	MAL	NR	Resection, ligation of afferent artery, section of MAL and celiac plexus	Favorable

NR, Nonruptured; R, ruptured; CA, celiac axis; PD, pancreaticoduodenectomy; NS, not stated; MAL, median arcuate ligament; Favorable, No change in size observed during follow-up period in no. 19 and 40, disappearance of aneurysm by conservative management in no. 20, and no recurrence of aneurysm in rest of the cases

was given regarding the management and follow-up of the aneurysms.^{14,36}

In recent years, selective embolization of visceral artery aneurysms with microcoils, gelfoam, or other embolic materials has been performed.^{37,38} In the setting of PDA aneurysms associated with celiac stenosis, a favorable outcome was observed in only one patient²⁴ and in two others subsequent therapy was required.^{19,20}

The association of celiac axis stenosis with PDA aneurysm is not coincidental. In this setting the cause of stenosis should be routinely investigated. Together with local treatment of the aneurysm, management of celiac axis stenosis could be considered, to prevent aneurysm reformation.

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