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Treatment Strategies for a Pancreaticoduodenal Artery Aneurysm with or without a Celiac Trunk Occlusive Lesion

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Objectives: A true pancreaticoduodenal artery aneurysm (PDAA) is a rare disease, and has some unique characteristics: a high rupture risk and a strong correlation with celiac trunk stenotic lesions (CTSL). We showed here that our treatment strategy for PDAA.

Materials and Methods: Seven consecutive patients with PDAA at our institution from 1998 to 2011 were retrospectively reviewed. Of the 7 patients, five were male and two were female, with a mean age of 55 ± 9.7 years. Three aneurysms were diagnosed incidentally, and the remaining four ruptured. The locations of the aneurysm were the anterior superior pancreaticoduodenal artery (ASPDA) in 3 patients and the inferior pancreaticoduodenal artery (IPDA) in four. CTSL found 3 patients in the IPDA.

Results: Of four ruptured patients, emergency catheter coil embolization was performed in three, and a simple ligation was performed in one. Three patients with non-ruptured aneurysms in the IPDA with a CTSL underwent direct aneurysm resection with arterial reconstruction. Six patients were successfully treated without complications or the appearance of new aneurysms during the follow-up period.

Conclusion: The treatment strategy for PDAA should be selected by the site of the aneurysm, the patients' condition, and the anatomical situation. A hybrid treatment could be considered a beneficial option for a CTSL.

Keywords: pancreaticoduodenal artery aneurysm, celiac lesion, coil embolization, reconstruction

INTRODUCTION

Although the prevalence of pancreaticoduodenal artery aneurysm (PDAA) is reported to be less than 2% of all splanchnic artery aneurysms (SAAs),[1] the number of incidental findings of the aneurysm is increasing as a result of the recent widespread use of diagnostic imaging modalities. One of the problems associated with PDAA is that this aneurysm, unlike other SAAs, possesses a

high risk of rupture irrespective of its diameter.[2] Many cases of ruptured PDAAs of <10 mm in diameter have been reported; therefore, the size of an aneurysm cannot be the predictor of its rupture.[2–6]

Another problem is that PDAA is occasionally found with a celiac trunk stenosis or occlusion, which complicates the treatment strategy.[2,3,7,9–11] Catheter embolization of both the inflow and the outflow artery of the aneurysm is currently most likely to be adopted for the treatment of SAAs because of its effectiveness and low degree of invasiveness. However, for PDAAs complicated with celiac trunk occlusive lesions, the approach to the aneurysm is difficult, and the risk of ischemic damage to the liver increases as a result of interruption of the collateral flow to the liver.[6,12] In such cases, open surgery remains an essential treatment option for the aneurysm. We have retrospectively reviewed the cases of 7 consecutive PDAA patients, who underwent treatment at our

Table 1   Characteristics of 7 patients with true PDAA

Case No.	Age/Sex	Location	Size	Status	Celiac axis	Treatment	Outcome	Follow-up (months)	CD
1	70/M	IPDA	NA	R	Normal	OS (ligation)	Death*	1	IHD
2	51/M	ASPDA	NA	R	Normal	Embolization	Alive	138	No
3	61/M	ASPDA	12	R	Normal	Embolization	Alive	96	HL
4	52/M	IPDA	35	NR	Stenosis	OS (Aneurysmectomy)	Alive	49	HT
5	40/M	IPDA	20	NR	Occlusion	OS (Aneurysmectomy)	Alive	33	No
6	51/F	IPDA	30	NR	Stenosis	OS (Aneurysmectomy)	Alive	30	No
7	61/F	ASPDA	10	R	Normal	Embolization	Alive	5	SAM

*Death of duodenal perforation. PDAA: pancreaticoduodenal artery aneurysm; CD: concomitant disease; IPDA: inferior pancreaticoduodenal artery; ASPDA: anterior superior pancreaticoduodenal artery; R: ruptured; NR: non-ruptured; NA: not available; OS: open surgery; IHD: ischemic heart disease; HL: hyperlipidemia; HT: hypertension; SAM: segmental arterial mediolysis

institution, to determine the treatment strategy for this unique aneurysm.

MATERIALS AND METHODS

Seven consecutive patients with PDAA who had been treated at our institution from 1998 to 2011 were retrospectively reviewed. Of the 7 patients, five were male and two were female, with a mean age of 55 ± 9.7 years, ranging from 40 to 70 years (**Table 1**). In 3 patients, the aneurysms were diagnosed incidentally without any symptoms, but in the remaining four, the aneurysms were ruptured at the time of diagnosis with the initial symptom of sudden abdominal pain. The locations of the aneurysm were the anterior superior PDA (ASPDA) in 3 patients and the inferior PDA (IPDA) in 4 patients. The diameter of the aneurysm ranged from 10 mm to 35 mm (median, 20 mm), excluding 2 aneurysms that could not be measured because of obscure demarcation between the aneurysm and hematoma, with extravasation of contrast agents. All of the celiac trunk occlusive lesions (found in 3 patients) were found in patients with aneurysms located in the IPDA. In 1 patient, multiple aneurysms were observed in the gastroduodenal artery, splenic artery, and ASPDA. None of the patients had a systemic background of arteritis, collagen disease, or hereditary disease. Comorbid risks in our series included ischemic heart disease, hypertension, and hyperlipidemia in 1 patient, respectively.

RESULTS

For ruptured aneurysms in the ASPDA in 3 patients and in the IPDA in 1 patient, emergency catheter coil embolization of the aneurysms and simple ligation with open surgery was performed, respectively. In all cases, hemostasis was successfully achieved. The 3 patients with non-ruptured aneurysms in the IPDA with celiac trunk occlusive lesions underwent direct aneurysm resection in combination with arterial reconstruction. Methods of arterial reconstruction consisted of bypass from the iliac to common hepatic artery in 2 patients, and direct anastomosis after aneurysm resection in 1 patient.

A patient who underwent aneurysm ligature in a shock state of rupture was the only mortality in our series. Although he recovered and was doing well immediately after the operation, sudden duodenal perforation caused by ischemia of the intestine occurred 1 week after the operation and led to death. Six patients were alive and followed until March 2012 with a mean follow-up period of 58 ± 49.3 months (range, 5–138 months). During the period, contrast enhanced CT was performed yearly to evaluate the post-operative morphological vascular changes. All of the arterial reconstructions have been patent to date. There were no procedure-related complications or recurrence of the aneurysms.

DISCUSSION

The association between PDAAs and the celiac occlusive lesion was first reported by Sutton in 1973,[13] and most of the subsequent reports of PDAAs strongly confirmed this relationship, with an incidence of up to 68%.[2,3,7–11] Given that PDAA is sometimes accompanied by celiac artery stenosis or occlusion, it is believed that the high-flow condition from the superior mesenteric artery (SMA), in compensation for the insufficient flow from the celiac artery, might be the cause of aneurysm formation.[2,3,6–16] A report of inferior mesenteric artery (IMA) aneurysm with occlusion of the celiac artery and severe atherosclerotic stenosis of the SMA may suggest a hemodynamic mechanism of aneurysm formation.[17] We suggest that such remodeling affects PDAA formation
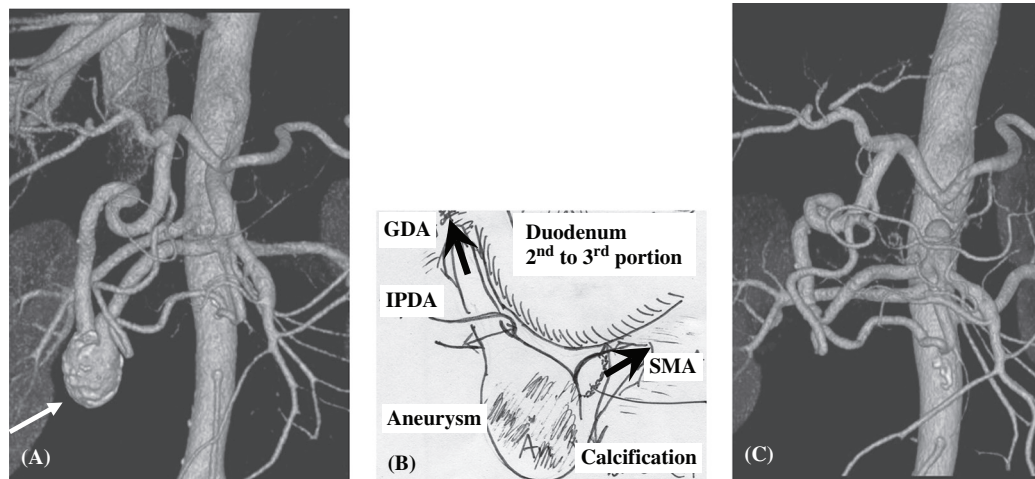
Fig. 1 Three-dimensional computed tomography (CT) shows the inferior pancreaticoduodenal artery aneurysm (IPDAA) with calcification (**A**; arrow). Direct anastomosis after aneurysmectomy was performed (**B**). Follow-up CT shows no recurrence of aneurysm 3 years after the surgery (**C**) (Case 4).
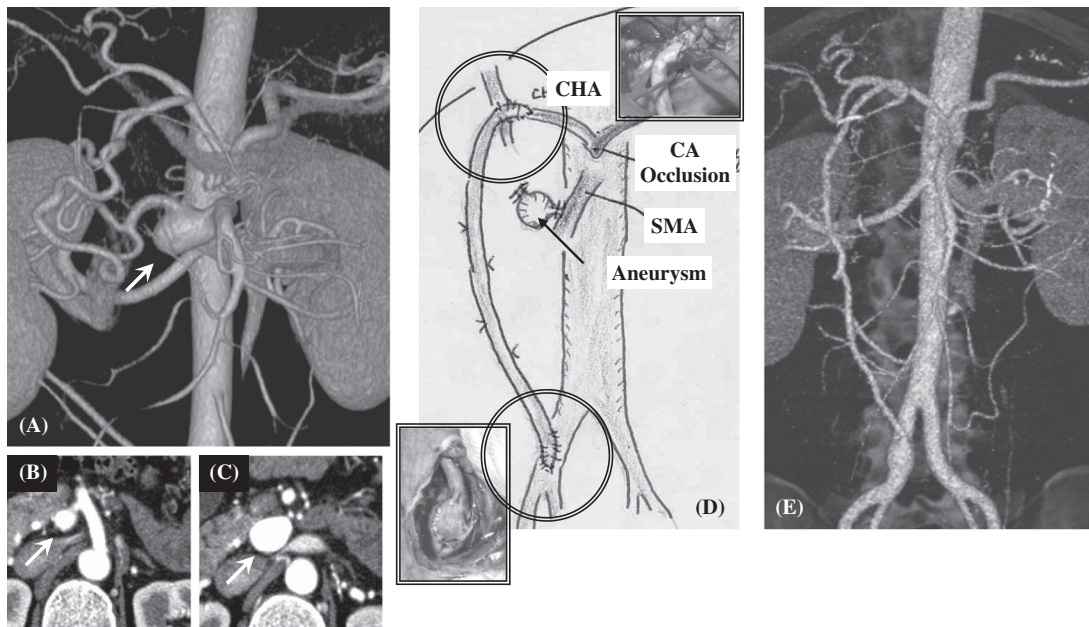

Fig. 2 Computed tomography (CT) scan shows the inferior pancreaticoduodenal artery aneurysm (IPDAA) near the trunk of the superior mesenteric artery (**A–C**; arrow). The right common iliac artery to the common hepatic artery bypass was performed after aneurysmectomy (**D**). Three-dimensional CT performed 15 months after surgery reveals the patency of the graft and disappearance of the pancreaticoduodenal arcade (**E**) (Case 5).

and that the pancreaticoduodenal arcade plays a significant role in this unique aneurysm. The absence of communication between the superior and inferior arteries (ASPDA–IPDA) is sometimes observed;[18] if this communication is absent, IPDA would theoretically be the main artery of aneurysm formation in the high-flow state via the SMA with its poor run-off. This hypothesis is supported by the fact that there are very few reports of

ASPDA aneurysm accompanied by celiac trunk lesion, and all 3 patients in our series also did not have celiac lesions. The pathogenesis of the IPDA aneurysm may be different from that of the ASPDA aneurysm.

Our treatment strategy for PDAA was based on the technical and pathogenetic perspective. First, we considered the less invasive transcatheter arterial embolization technique as the first line of treatment for ruptured PDAA,

if the patient was hemodynamically stable. For the patient in Case 1, we had no choice but to perform laparotomy for emergency bleeding control. The other 3 cases of rupture were fortunately not accompanied by celiac trunk lesion, and we successfully embolized the PDAAs without endovascular access problems.

Second, we selected open surgery for patients with the celiac trunk stenosis or occlusive lesion that may require an advanced endovascular technique. The successful embolization via SMA approach for such PDAAs with celiac lesion was reported to be only 10%–40%.[1] The outcomes of endovascular therapy alone were not satisfactory.[19] The technical success rate for ruptured PDAA with stable hemodynamics was reported to be 57%–79%, and there is still a risk of arterial occlusion.[12,20] Bageacu reported an excellent initial success rate of embolization with microcoils, accompanied by aorto-hepatic bypass for patients with celiac lesions.[6] Considering the blood supply for adjacent organs, such a hybrid therapy would be a beneficial option.

Third, the site of the aneurysm is crucial for the surgical strategy. Direct anastomosis after aneurysmectomy, as in our Case 4 would be ideal (**Fig. 1**); however, the PDA sometimes runs intrapancreatically,[18] which makes anastomosis difficult. Simple aneurysmectomy and extra-anatomical bypass reconstruction should be selected if the aneurysm is located near the pancreatic parenchyma or the feeding arterial branches of the SMA arise near the aneurysm, as in Cases 5 and 6 in our series (**Fig. 2**).

Fourth, we can choose both of the endovascular procedure and the open surgery for the non-ruptured PDAA without celiac lesion. The selection should be dependent on the patient's background, aneurysm site and morphology. In open surgery, we prefer the iliac artery as the inflow of the bypass to the celiac lesion. Although this technique requires a long bypass route, we can avoid touching the SMA or the splenic artery, which may be affected by undetected underlying disease such as segmental arterial mediolysis (SAM), or clamping the aorta and risking shower embolization and ischemic damage to important organs; easy access to the potential anastomotic aneurysm is another merit of this technique.

There are some reported cases of aneurysm regression or stability after simple reconstruction of the celiac lesion with stenting, division of the arcuate ligament, and revascularization.[14–16] However, there is no evidence that small aneurysms are safe and that the rupture of the PDA aneurysm is associated with high mortality.[2,3,4,6] Therefore, we do not think it appropriate to reconstruct only the celiac trunk lesion without management of the aneurysm.

Conclusion

In conclusion, we presented 7 cases of true PDA aneurysms treated by open surgery or endovascular repair at our department. These treatments should be selected on the basis of the site of the PDAA, patient condition, and the anatomical situation. A hybrid treatment can also be considered a beneficial option.

Disclosure Statement

None.

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