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HOW I DO IT

Treatment strategies for unruptured pancreaticoduodenal artery aneurysms associated with celiac artery occlusion

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Abstract

Celiac artery (CA) occlusion or stenosis is identified in up to almost half of all patients undergoing abdominal angiography, and the resulting increased collateral blood flow from the superior mesenteric artery to the pancreaticoduodenal artery (PDA) may cause PDA aneurysms (PDAAs). PDAAs are rare but could be fatal if they rupture. However, treatment of the PDAA could block this important collateral blood flow pathway, leading to ischemic organ damage. Treatment of such aneurysms is therefore difficult, especially in patients with multiple PDAAs. Successful treatment of PDAAs requires establishing blood flow in the CA region and selecting which aneurysm(s) to treat. We present four patients who underwent surgery for unruptured PDAAs caused by CA obstruction. Blood flow in the CA region was established by bypassing the splenic artery and by anastomosing it either directly to the left renal artery (n = 1) or to the abdominal aorta using a graft (saphenous vein: n = 1; artificial vessel: n = 2). Three patients had multiple PDAAs: all PDAAs were treated in one patient with PDAAs of similar size and shape, but only the largest PDAA with the highest risk of rupture was treated in the other two patients to simplify the procedure. The median observation period was 19.5 months (range: 11-28 months), and all patients were alive without recurrence at the time of writing. Surgical treatment including splenic artery bypass may thus be a viable option for treating patients with unruptured PDAAs.

KEYWORDS
celiac artery obstruction, pancreaticoduodenal arcade, pancreaticoduodenal artery aneurysm, reconstruction, splenic artery bypass

1 | INTRODUCTION

Pancreaticoduodenal artery aneurysms (PDAA) are a subtype of visceral artery aneurysm (VAA).[1,2] The incidence of VAAs is extremely low, ranging from 0.01% to 0.2%,[3,4] and PDAAs comprise only 2% of all VAAs.[4–7]

PDAAs are associated with stenosis or occlusion of the celiac artery (CA) or superior mesenteric artery (SMA) in 50%-80% of patients.[2] CA occlusion results in increased blood flow from the SMA to the pancreaticoduodenal artery (PDA), which not only compresses the PDA and leads to aneurysm formation, but also makes the PDA itself an important collateral blood vessel.[8] Compensatory hyperdynamic flow through the SMA-PDA-gastroduodenal artery (GDA)-hepatic artery vasculature is thought to be the result of CA stenosis or occlusion.[7,9]

Because of the high risk of rupture, the 2020 Society for Vascular Surgery clinical practice guidelines recommend treatment for

PDAAs regardless of their size.[10] This is in contrast to the previous recommendation for VAA treatment of a measurement of >2 cm.[1,4] Additionally, for unruptured PDAAs, the guidelines recommend open surgery for revascularization and CA reconstruction in cases of CA stenosis or occlusion.[9]

However, because unruptured PDAAs are even rarer, there is no consensus on how to establish blood flow to the CA region. Furthermore, it is difficult to determine from cases of ruptured aneurysms measuring ≤2 cm whether treatment of all PDAAs is mandatory. To address this issue, in this study, we treated four surgical cases of unruptured PDAA with CA occlusion using splenic artery (SA) bypass to establish blood flow to the CA region. The key treatment principles that we followed in these cases were (1) maintaining or normalizing blood flow to organs originally supplied by the CA, and (2) selecting the aneurysm to be treated, in cases of multiple aneurysms; thus, simplifying the procedure. In this report, we discuss our experience with these four surgical cases of unruptured PDAA with CA occlusion or stenosis, using SA bypass.

2 | PATIENTS AND SURGICAL TECHNIQUE

2.1 | Patients

Four consecutive patients with PDAA treated at our institution between January 2020 and July 2021 were enrolled in this study. The patients' clinicopathological variables, preoperative imaging findings, and surgical procedures are summarized in Table 1.

2.2 | Surgical technique

There are two procedures for SA bypass to establish blood flow in the CA region: (1) Left renal artery (LRA)-SA bypass, which involves direct anastomosis of the SA to the LRA. First, the LRA is secured to evaluate calcification and atherosclerosis to confirm that the LRA is suitable for bypass. Next, the SA is identified at the cephalic side of the pancreas. The SA is sufficiently separated from the pancreas then ligated and resected. The SA is then passed through the dorsal side of the pancreas, and the LRA and SA are anastomosed side-to-end (Figure 1A, Video S1). (2) Abdominal aorta (AA)-SA bypass. This procedure is performed when LRA-SA bypass is impossible. AA-SA bypass requires a graft, and the great saphenous vein (GSV) (Figure 1B) or an artificial vessel (Figure 1C,D) are options.[11] In patients receiving artificial vessels, antiplatelet medications should be administered. The graft should pass dorsal to the mesentery. There are ventral and dorsal pancreatic routes, and the route that facilitates anastomosis between the graft and the SA should be selected. If the dorsal pancreatic route is chosen, it is important to note that a GSV graft may be pushed and bent by the pancreas, which is what occurred in Case 2 (Figure 1B). However, this was not a concern in the cases in which we used an artificial blood vessel with a ring

TABLE 1 Patients' characteristics and the surgical outcomes

Parameter	Total (*n* = 4)
Background characteristic	
Age, years[a]	67 (57-75)
Sex	
Male	2 (50)
Female	2 (50)
BMI, kg/m^2[a]	22.2 (18-24.9)
Symptoms	
None	4 (100)
Preoperative comorbidity	
Hypertension	3 (75)
Diabetes mellitus	2 (50)
Respiratory disease	1 (25)
Cerebrovascular disease	1 (25)
Social history	
Smoking	1 (25)
Alcohol drinking	1 (25)
Number of aneurysms	
1	1 (25)
2	1 (25)
3	1 (25)
≥4	1 (25)
Number of aneurysms measuring >15 mm	
1	3 (75)
2	1 (25)
Aneurysm size, mm[a]	20.5 (17-36)
Celiac artery obstruction	4 (100)
Surgical outcomes	
Surgical procedure for vascular reconstruction	
LRA-SA bypass	1 (25)
AA-SA bypass	2 (25)
LCIA-SA bypass	1 (25)
With or without a graft	
Direct artery anastomosis without a graft	1 (25)
With a graft	3 (75)
Great saphenous vein	1 (25)
Artificial vessel	2 (50)
Aneurysm treatment	
Resection	3 (75)
IVR (Coiling)	1 (25)
Planning of the procedure	
One-stage surgery	3 (75)
Two-stage surgery	1 (25)

Note: Values in parentheses are percentages unless indicated otherwise.

Abbreviations: AA, abdominal aorta; BMI, body mass index; IVR, interventional radiology; LCIA, left common iliac artery; LRA, left renal artery; SA, splenic artery.

[a]Median (range).

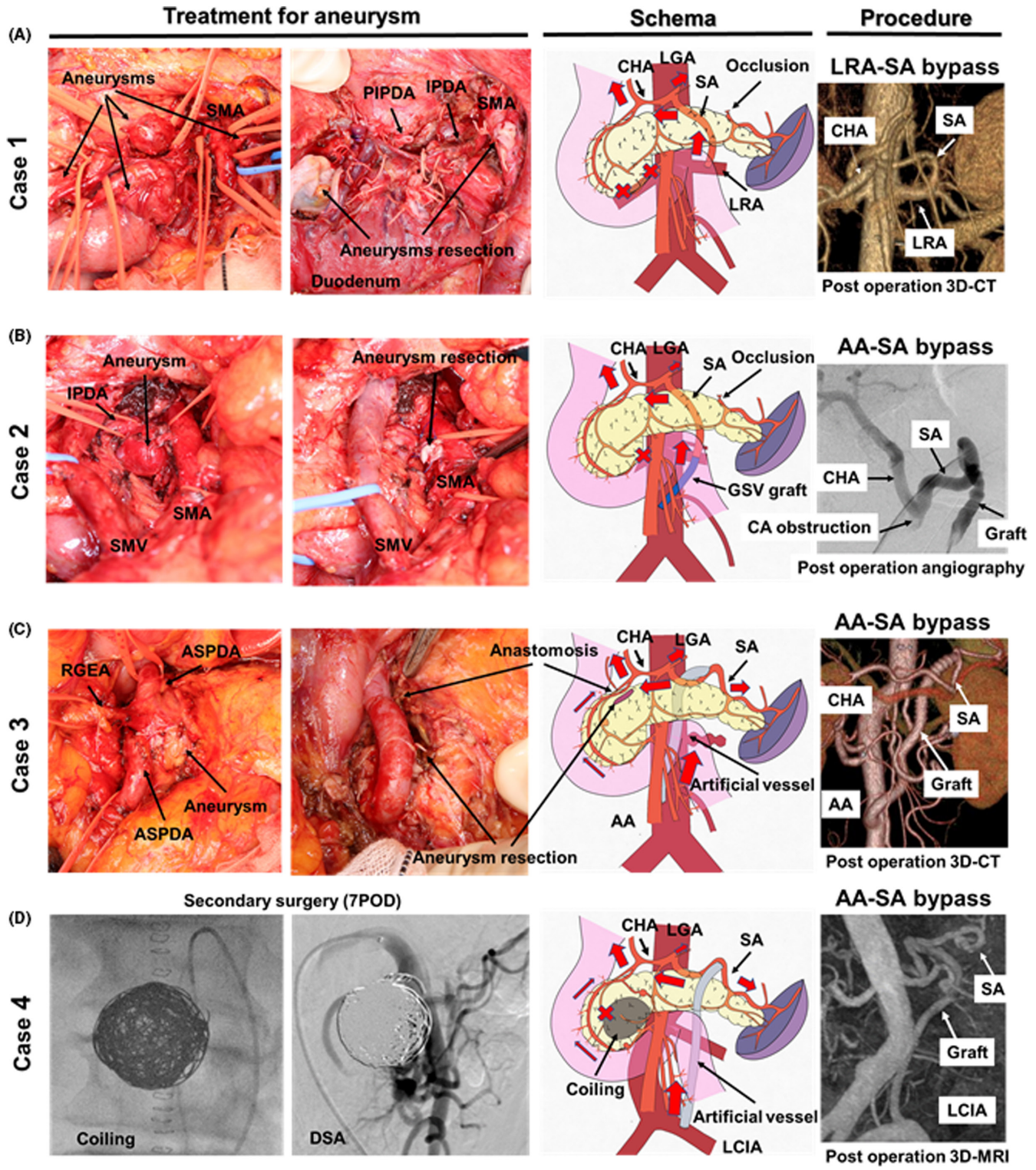

FIGURE 1 Surgical procedure for pancreaticoduodenal artery aneurysmal (PDAA) repair. Left panels: aneurysm treatment. Middle panels: schemas after surgery. Blood flow after the treatment is also illustrated. Right panels: postoperative imaging. A, Case 1. All PDAAs were ligated and dissected. Left renal artery (LRA) to splenic artery (SA) bypass procedure: The splenic artery was divided, and the SA and LRA were anastomosed end-to-side. B, Case 2. The PDAA was ligated and dissected, and the SA was divided. The SA and abdominal aorta (AA) were anastomosed end-to-side using a graft from the great saphenous vein (GSV). C, Case 3. The aneurysm was in the anterior superior pancreaticoduodenal artery (ASPDA). The inflow and outflow vessels were anastomosed after aneurysm dissection. An artificial blood vessel and the SA and AA were anastomosed end-to-side. D, Case 4. An artificial blood vessel and the SA and left common iliac artery were anastomosed end-to-side. Only the largest aneurysm was embolized by coiling 7 days after surgery. 3D, three-dimensional; CA, celiac artery; CHA, common hepatic artery; CT, computed tomography; DSA, digital subtraction angiography; IPDA, inferior pancreaticoduodenal artery; LCIA, left common iliac artery; LGA, left gastric artery; MRI, magnetic resonance imaging; PIPDA, posterior inferior pancreaticoduodenal artery; RGEA, right gastroepiploic artery; SMA, superior mesenteric artery; SMV, superior mesenteric vein

(Propaten®; W.L. Gore & Associates, Inc.) (Figure 1C). For anastomosis of the abdominal aorta, select a site that is without calcification and easy to anastomose. In Case 4, the aorta was tortuous, and the left common iliac artery was easy to anastomose because it was perpendicular to the graft. The next step is treating the PDAA. In this study, if interventional radiology (IVR) was possible, treatment of the PDAA with coiling was selected (Figure 1D). If this procedure was not possible, the PDAA was dissected directly, the inflow and outflow vessels were ligated, and the aneurysm wall was opened and then sutured to prevent bleeding (Figure 1A,B, Video S1). After the PDAA was treated, the inflow and outflow vessels were anastomosed, if possible (Figure 1C, Video S2). This procedure also ensured blood flow to the CA region. We believe that the risk of untreated aneurysmal rupture improves after bypass surgery because blood flow in the PDA is normalized. If there were multiple aneurysms and the aneurysm with the highest risk of rupture was clearly identified, we considered prioritizing the treatment of this aneurysm and not treating the other aneurysms (Figure 1C,D). This treatment strategy simplifies the surgery and preserves the collateral blood vessels. As a result, complications may be minimized. In such cases, it is always necessary to consider preoperatively how to treat the untreated aneurysm(s) if it worsens. Additionally, in this study, PDAA treatment was performed after confirming blood flow in the CA region. Resection of the PDAA(s) was performed after confirming that blood flow in the CA region was ensured by palpation, Doppler ultrasonography, and a flowmeter. Alternatively, angiography was performed from the artificial vascular graft to confirm blood flow in the CA region before coiling.

3 | RESULTS

The patients' and aneurysmal characteristics are shown in Table 1 and Figure 2. Imaging studies showed complete occlusion of the CA in all patients.

It is important to note that there are many collateral blood channels between the SMA and CA, which vary from patient to patient. We evaluated the preoperative status of the dorsal pancreatic artery (DPA), which connects the CA and SMA via vessels in the pancreas, and the left gastric artery (LGA), which supplies blood flow to the spleen and the tail of the pancreas after SA dissection. Digital subtraction angiography images of the SMA showed that both the LGA and DPA were thick and well-defined in Case 1. However, in Case 2, both the LGA and DPA were thin and fragile. In Case 3, the DPA could not be identified, and in Case 4, the LGA could not be identified. The vascular network of the LGA and DPA was different in each case (Figure 3A). Only one patient had a single aneurysm; the remaining three patients had multiple aneurysms (Figure 2). The bypass procedures to establish blood flow to the CA region were LRA-SA bypass (n = 1), AA-SA bypass with a GSV graft (n = 1), and AA (n = 1) or left common iliac artery (n = 1) to SA bypass with an artificial vessel graft. The PDAA treatment strategies were IVR coil embolization (n = 1) and direct aneurysm treatment (n = 3). Treatment of aneurysms in cases of multiple aneurysms consisted of treating all aneurysms (n = 1) and treating only the aneurysm with a high risk of rupture (n = 2) (Figures 1 and 2). Postoperative changes in the PDA dilation and PDAA were evaluated in the two cases (Case 3 and Case 4) in which we treated only the aneurysm with a high risk of rupture. The PDA dilation improved from 5.5 to 2.8 mm in Case 3 and from 7.1 to 3.2 mm in Case 4, pre- and postoperatively, respectively. In addition, the diameters of the aneurysms that were not directly treated shrank from 9.6 to 6.2 mm in Case 3, and in Case 4, one aneurysm could no longer be identified, and the diameter of the other aneurysm improved from 9.0 to 7.6 mm, pre- and postoperatively, respectively (Figure 3B,C). Postoperative complications varied from case to case and are detailed in Table S1. Case 2 developed graft occlusion, resulting in ischemic organ damage to the liver, spleen, and pancreas during reoperation 1 day after surgery, inducing a pancreatic fistula and postoperative bleeding. This complication was a stark reminder of how important it is to ensure reliable blood flow in the CA region when treating PDAA with CA occlusion. There were no complications resulting from the separation of the SA from the pancreatic parenchyma. All patients are currently under follow-up, with a median follow-up duration to date of 19.5 months (range: 11-28 months). Radical cure was achieved in all patients, and there has been no recurrence of an aneurysm.

4 | DISCUSSION

The most important treatment strategy concern for unruptured PDAAs with an occluded CA is how to treat aneurysms that may rupture while maintaining blood flow to the CA region.[4,6]

Although the causes of PDAA are varied, the majority of PDAAs are caused by stenosis or occlusion of the CA.[3,4,7,10] CA occlusion has been reported to increase blood flow from the SMA to the inferior PDA and pancreatic arcades, resulting in increased arterial pressure and aneurysm formation.[12] If the PDAA is treated first, blood flow from the SMA through the PDA becomes blocked, and organs in the CA region may suffer ischemic damage.[13] First, it is necessary to diagnose whether the CA obstruction is median arcuate ligament syndrome (MALS). In Case 3, the cause was diagnosed as fibromuscular dysplasia, and in Case 4, the cause was atherosclerosis. Cases 1 and 2 were diagnosed as having MALS, and the selection of treatment by MAL division or arterial reconstruction was in accordance with the report of Sugae et al.[14] Bypass was indicated when the stenosis rate was ≥80%, the length of the stenosis was ≥8 mm, and the distance between the stenosis and the aorta was <5 mm (Figure S1). At our institution, we perform LRA-SA bypass.[10] If anastomosis with the LRA is difficult, an application of this surgical technique is to bypass the AA and SA using a graft.

The indication for treatment of some VAAs is an aneurysmal diameter of ≥2 cm.[10] However, it has been suggested that all PDAAs should be treated regardless of size because there are reports of rupture of PDAAs smaller than 2 cm.[11,15] However, these reported

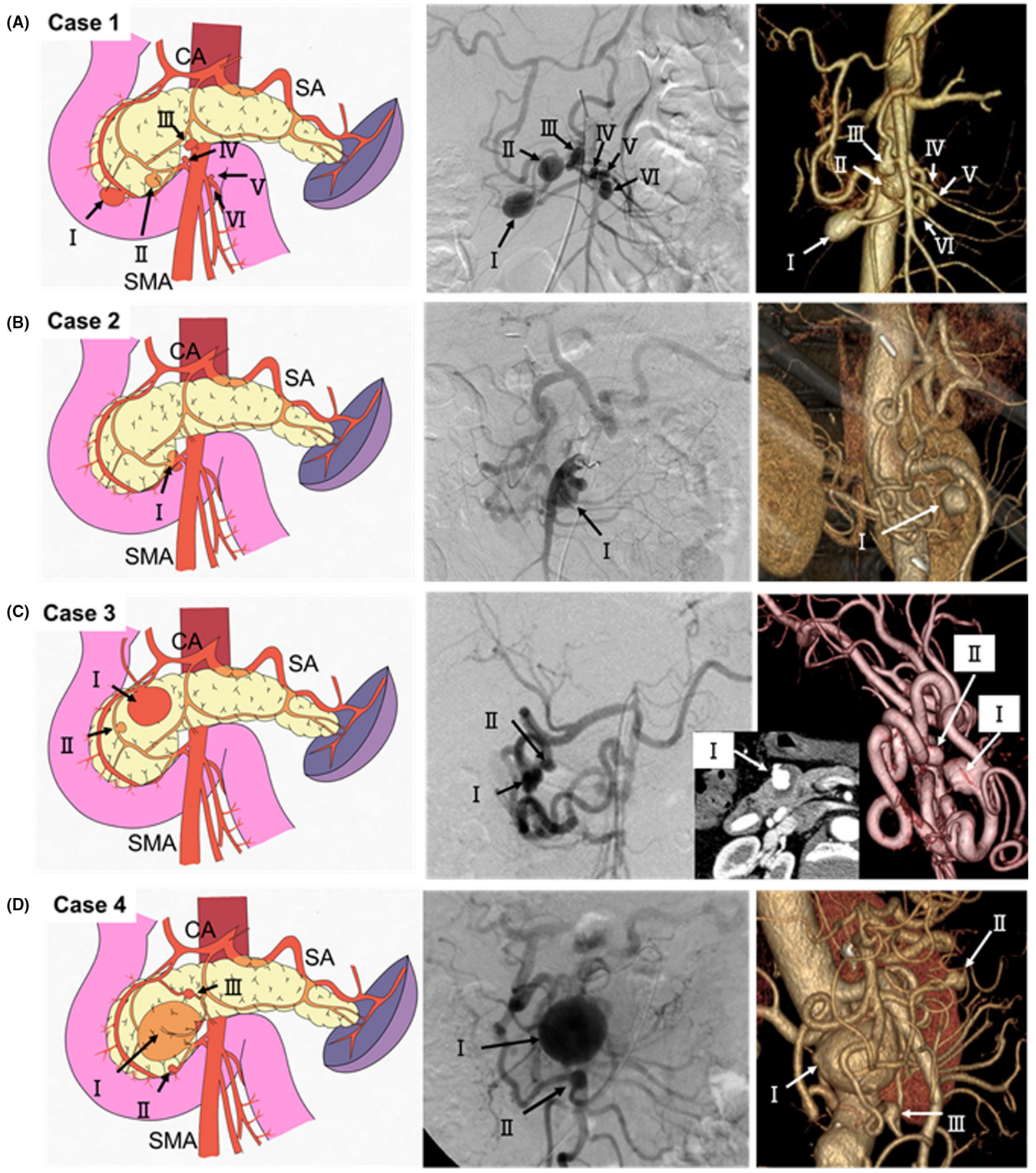

FIGURE 2 Upper panels: Characteristics of the pancreaticoduodenal artery aneurysms (PDAA). A, Case 1. I: AIPDA, 24.0 mm; II: PIPDA, 17.0 mm; III: DPA, 11.2 mm; IV: IPDA, 10.1 mm; V: IPDA, 7.5 mm; VI: J1A, 6.3 mm. B, Case 2. I: IPDA, 17.0 mm. C, Case 3. I: ASPDA, 30.3 mm; II: PSPDA, 9.6 mm. D, Case 4. I: PIPDA, 45.6 mm; II: AIPDA, 9.0 mm; III: TPA, 11.3 mm. Lower panels: Left panels: schemas of the PDAAs. Middle panels: digital subtraction angiography. Right panels: 3D-CT angiography. Case 3 underwent contrast-enhanced CT to identify the thrombus within the aneurysm. 3D, three-dimensional; AIPDA, anterior inferior pancreaticoduodenal artery; ASPDA, anterior superior pancreaticoduodenal artery; CT, computed tomography; DPA, dorsal pancreatic artery; IPDA, inferior pancreaticoduodenal artery; J1A, first jejunal artery; PIPDA, posterior inferior pancreaticoduodenal artery; PSPDA, posterior superior pancreaticoduodenal artery; TPA, transverse pancreatic artery

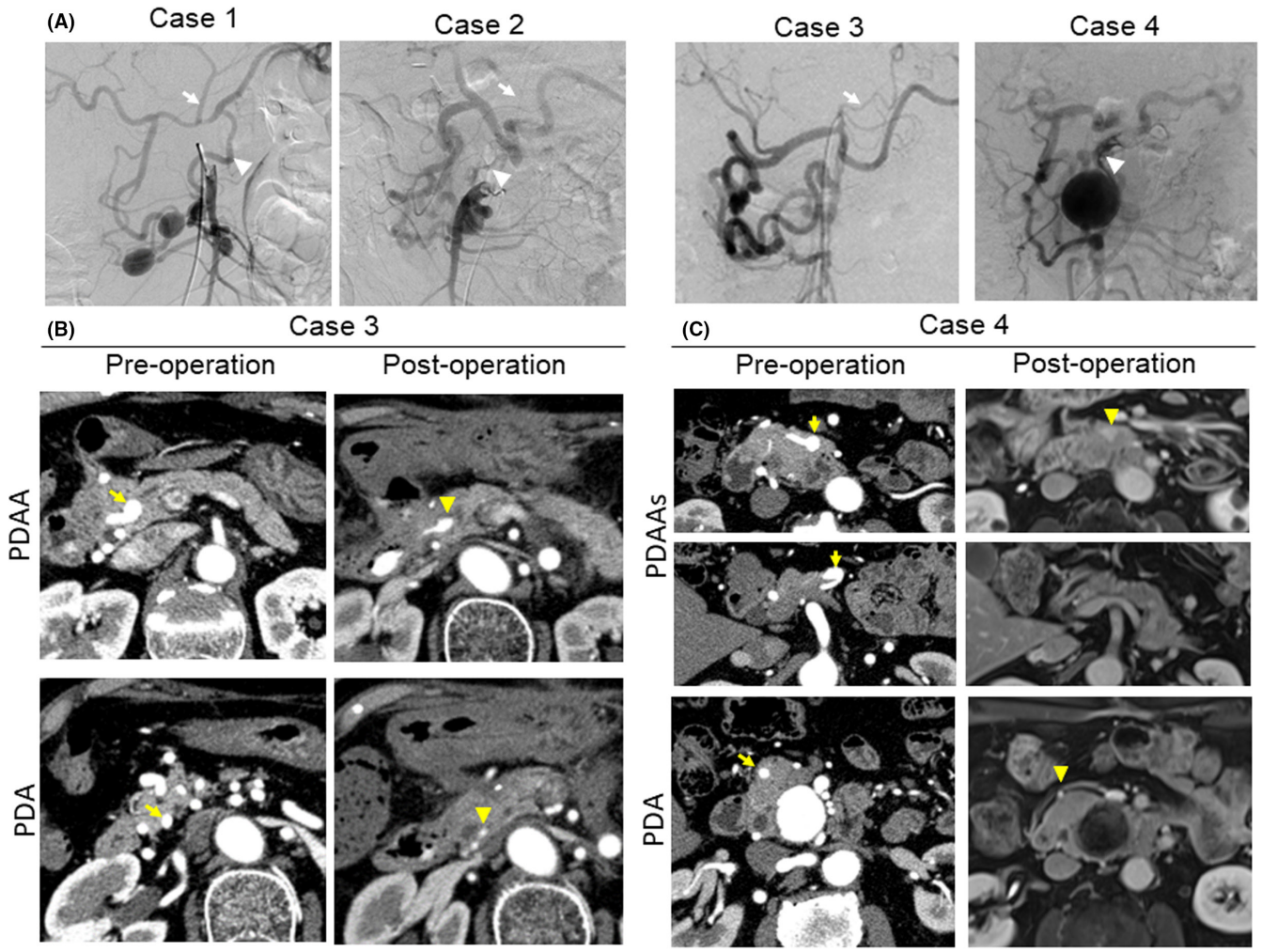

FIGURE 3 A, Comparison of the left gastric artery (LGA) and the dorsal pancreatic artery (DPA) in four cases by superior mesenteric artery digital subtraction angiography. Vessel diameter for each case: Case 1. LGA: 3.1 mm, DPA: 4.2 mm. Case 2. LGA: 1.2 mm, DPA: 1.1 mm. Case 3. LGA: 2.3 mm, DPA: not detected. Case 4. LGA: not detected, DPA: 2.3 mm. Arrowhead: DPA; arrow: LGA. B and C, Postoperative changes in the pancreaticoduodenal artery aneurysm (PDAA) and the pancreaticoduodenal artery (PDA). B, Case 3. The PDAA improved from a preoperative diameter of 9.6 mm (arrow) to a postoperative diameter of 6.2 mm (arrowhead). The PDA also improved from a preoperative diameter of 5.5 mm (arrow) to a postoperative diameter of 2.8 mm (arrowhead). C, Case 4. Upper panels: The PDAA improved from a preoperative diameter of 9.0 mm (arrow) to a postoperative diameter of 7.6 mm (arrowhead). Lower panels: The PDAA measured 11.3 mm preoperatively (arrow) and could not be detected postoperatively. The PDA improved from a preoperative diameter of 7.1 mm (arrow) to a postoperative diameter of 3.1 mm (arrowhead)

cases were diagnosed after rupture; therefore, blood flow in the PDA remained high, and intravascular pressure also remained high.

An SA or common hepatic artery bypass supplies blood flow in the CA region and returns PDA blood flow and intravascular pressure closer to normal.[4,7] In Cases 3 and 4, we treated only the largest aneurysm in each case, which was a giant aneurysm that was considered at high risk of rupture, and we left the other smaller aneurysms untreated, to simplify the procedure. Postoperative follow-up computed tomography (CT) and magnetic resonance imaging showed that the PDA dilation had disappeared, probably because the SA bypass reduced the blood flow and pressure in the PDA (Figure 3B,C). Notably, the remaining aneurysms also tended to shrink. Aneurysm rupture is the worst possible outcome and preventing rupture must be a top priority. To minimize both the risk of rupture and the risk of organ ischemia, treating only the largest aneurysm after SA bypass may simplify treatment and reduce complications.

It is likely that some cases are relatively resistant to ischemia of the CA region and others are susceptible, on the basis of our findings in this study. There was a clear difference in the degree of development of the LGA and the DPA in each case. Case 1 had a well-developed LGA and DPA, while Case 2 had a thin and weak LGA and DPA. The digital subtraction angiography findings did not identify a DPA in Case 3 or LGA in Case 4. In fact, Case 2 was complicated by ischemic organ damage in the CA region owing to postoperative graft occlusion. Vascular structures such as those in Case 2 are prone to hemodynamic disturbance, which may lead to significant ischemic organ damage due to perioperative complications or graft failure.[5] In similar cases, it is effective to perform a revascularization procedure

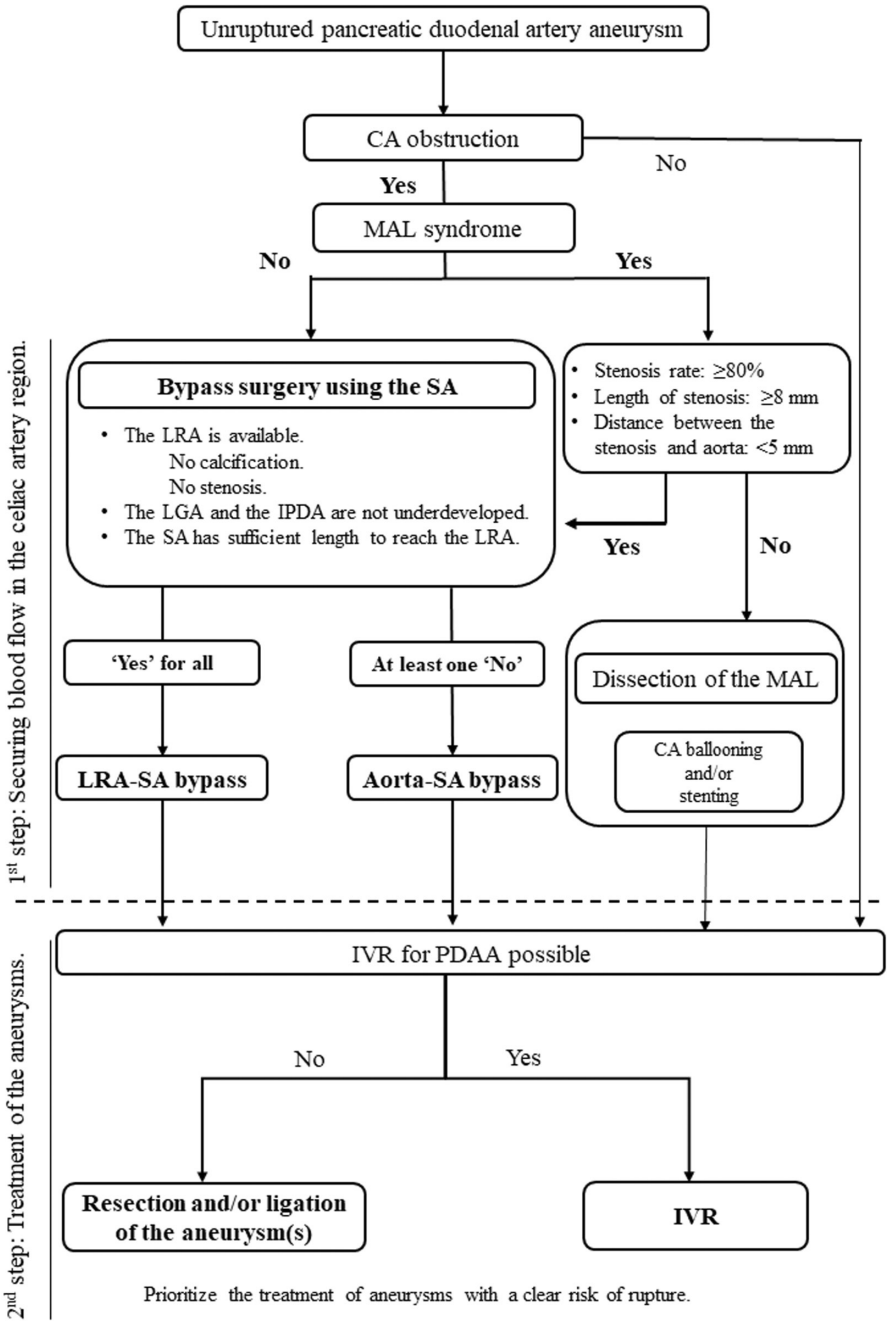

FIGURE 4 Study flowchart of the treatment of the pancreaticoduodenal artery aneurysm (PDAA). CA, celiac artery; IPDA, inferior pancreatoduodenal artery; IVR, interventional radiology; LGA, left gastric artery; LRA, left renal artery; MAL, median arcuate ligament; SA, splenic artery

to ensure blood flow in the CA region and treat the aneurysm on a different day.[5] In Case 4, as in Case 2, with a weak LGA (Figure 3B), the PDAA was treated on a different date. An acceptable outcome and no complications were observed after either procedure. Figure 4 presents a flowchart of our approach to the treatment of PDAA.

As a limitation in our study, we acknowledge that this was a retrospective observation with a limited number of cases from a single institution, which may have affected the efficacy of the treatment and the development of complications. However, because PDAAs are rare and unruptured cases are even rarer,[4] we believe that our case series clarifies the characteristics of unruptured PDAAs due to CA occlusion.

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Ethics Statement: The protocol for this research project has been approved by a suitably constituted Ethics Committee of the institution and it conforms to the provisions of the Declaration of Helsinki (Ethics Committee of Shinshu University, Approval Number: 5283).

Data Accessibility Statement: Data supporting the results of this study are available upon request to the corresponding author, AS.

Informed Consent: Informed consent was received from all participants.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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