Blood flow modification might prevent secondary rupture of multiple pancreaticoduodenal artery arcade aneurysms associated with celiac axis stenosis

Fumio Yamana, MD,[a] Toshihiro Ohata, MD, PhD,[a] Mutsunori Kitahara, MD, PhD,[a] Masahisa Nakamura, MD, PhD,[b] Hideaki Yakushiji, MD, PhD,[b] and Shin Nakahira, MD, PhD,[c] *Sakai, Osaka, Japan*

ABSTRACT

A pancreaticoduodenal artery arcade aneurysm (PDAA) is rare and often associated with celiac axis stenosis by the median arcuate ligament. Although rupture risk of the PDAA is not related to its size, treatment guidelines are absent. Here we describe a 59-year-old woman with multiple ruptured PDAAs associated with celiac axis stenosis who was successfully treated with coil embolization. As follow-up computed tomography revealed rapid expansion of residual PDAAs and new gastric artery dissection, median arcuate ligament resection was followed by aorta-common hepatic artery bypass, which resulted in aneurysmal regression. Blood flow modification might prevent secondary rupture of PDAA associated with celiac axis stenosis. (J Vasc Surg Cases and Innovative Techniques 2020;6:41-5.)

Keywords: Pancreaticoduodenal artery aneurysms; Celiac axis stenosis; Median arcuate ligament syndrome

Pancreaticoduodenal artery arcade aneurysms (PDAAs) are rare and account for <2% of all aneurysms in visceral arteries.[1] PDAAs are reported to be associated with many causes, including trauma, pancreatitis, infection, autoimmune disease, and congenital anomalies.[2]

It is suggested that celiac artery stenosis due to compression by the median arcuate ligament (MAL) causes rupture of PDAAs,[3] which are thought to result from hemodynamic turbulence and an increase in peripancreatic collaterals.[4] Recently, the development of endovascular treatment has made it possible to perform transcatheter arterial embolization (TAE) of visceral aneurysms safely and effectively.[1,2] Moreover, TAE has been performed for first-line treatment of ruptured PDAAs.

Several choices for treatment of celiac artery stenosis in the presence of PDAAs have been reported,[1-4] but details of the therapeutic strategy, such as the follow-up term and treatment to prevent recurrence and secondary rupture of PDAAs, remain controversial. We describe a patient with multiple ruptured PDAAs associated with MAL compression. MAL resection followed by aorta-

https://doi.org/10.1016/j.jvscit.2019.10.005

common hepatic artery bypass resulted in aneurysmal regression after initial TAE for rupture. Informed consent for publication of this case report was obtained.

CASE REPORT

A 59-year-old nonsmoking woman was admitted to the emergency department of our hospital with continuous abdominal pain and vomiting. She reported no history of acute or remote abdominal trauma. She did not take any medications regularly, had never smoked or consumed alcohol, and did not have any family history of aneurysms. Enhanced computed tomography (CT) showed a large retroperitoneal hematoma (peripancreatic and periduodenal) induced by rupture of multiple visceral artery aneurysms (Fig 1, *A*). Significant stenosis of the celiac axis root origin and post-stenotic dilation, most likely because of MAL compression, were also found (Fig 1, *B*). Selective catheter angiography of the superior mesenteric artery (SMA) showed retrograde flow from the SMA to the celiac artery through the pancreaticoduodenal artery arcades and multiple inferior PDAAs.

Two 6-mm aneurysms in the posterior inferior pancreaticoduodenal artery arcade showed signs of rupture (Fig 1, *C*). Emergency endovascular treatment of both aneurysms was successfully performed with coil embolization. No additional treatment of celiac trunk stenosis was performed because of the emergency setting of the case.

Postoperative follow-up CT at 1 month revealed rapid expansion of the residual PDAAs and a new left gastric artery dissection (Fig 2). We considered that a change in hemodynamic turbulence and an increase in peripancreatic collateral flow between the celiac artery and SMA resulted in secondary arterial events. To prevent secondary PDAA rupture, normalization of antegrade blood flow was desirable in the celiac and SMA arcade.

There were two treatment options, including endovascular treatment and open surgery for celiac axis stenosis.
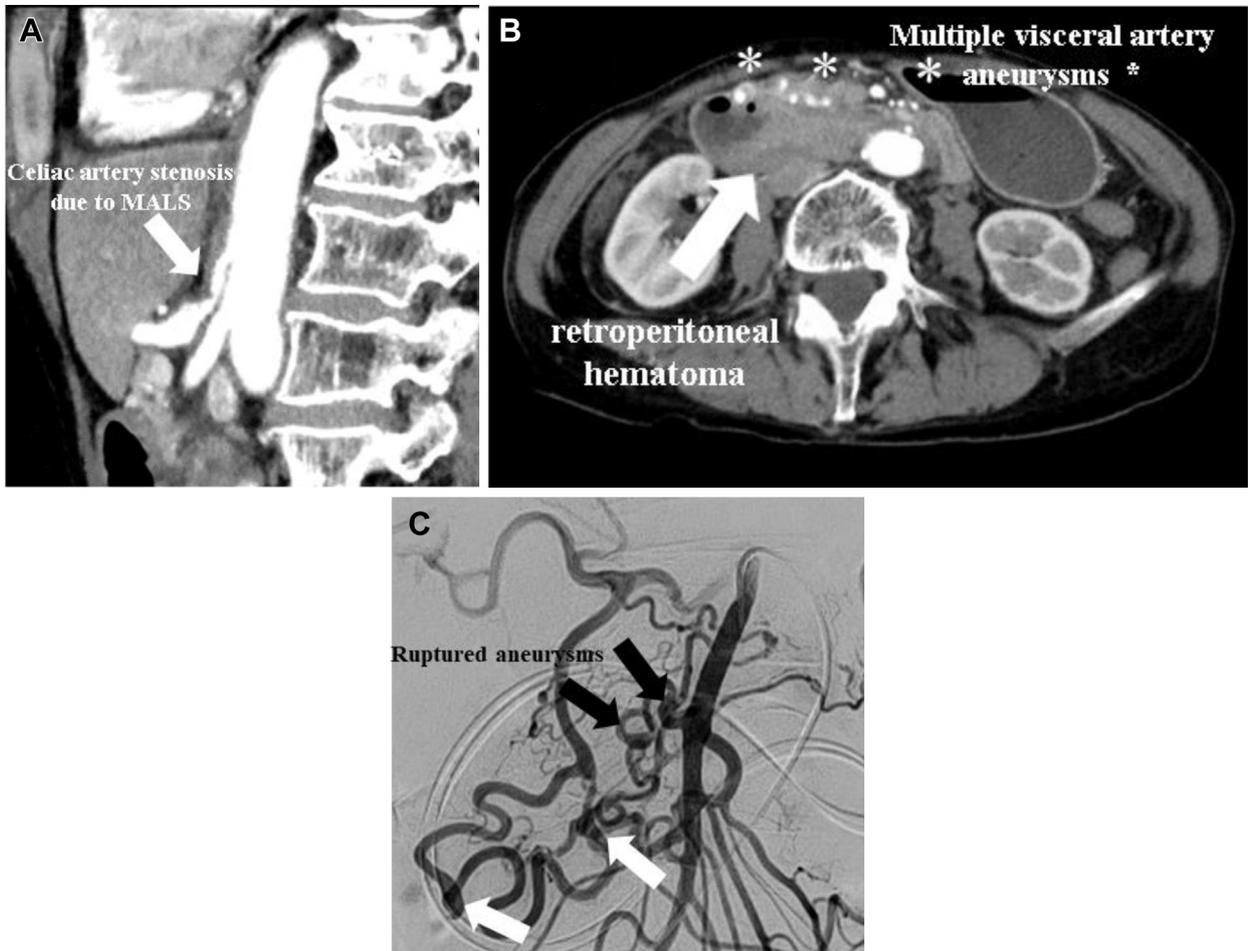
Fig 1. Preoperative computed tomography (CT) and intraoperative superior mesenteric artery (SMA) angiography. **A,** Enhanced CT showed significant stenosis of the celiac axis root origin and post-stenotic dilation, most likely because of median arcuate ligament syndrome (*MALS*) compression (*arrow*). **B,** CT showed a large retroperitoneal hematoma (peripancreatic and periduodenal; *arrow*) induced by rupture of multiple visceral artery aneurysms (*asterisks*). **C,** Angiography showed two 6-mm ruptured aneurysms (*black arrows*) and multiple pancreaticoduodenal artery arcade aneurysms (PDAAs; *white arrows*).

Endovascular treatment is less invasive, but there is a risk of distal occlusion and intraoperative rupture of the celiac artery, which may result in a fatal hemodynamic state. Open surgery is a safe and reliable method despite being more invasive compared with endovascular treatment.

We scheduled two-stage surgery after her complete recovery. Ligament release was tried first for the MAL, then aorta-common hepatic artery bypass was additionally performed if antegrade blood flow of the celiac artery was not improved angiographically. Intraoperative SMA angiography showed retrograde blood flow through the peripancreatic arcade from the SMA to the common hepatic and splenic arteries (Fig 3). Blood flow volume from the inferior pancreatic duodenal artery was thought to be significantly increased by severe celiac axis stenosis.

Ligament release for the MAL was performed through laparotomy, and selective SMA angiography revealed residual blood flow of the common hepatic artery from the SMA. Because the increase in antegrade blood flow from the celiac artery was insufficient, it was subsequently decided to perform aorta-common hepatic artery bypass. The infrarenal abdominal aorta and common hepatic artery were exposed, and a tunnel also provided retromesenteric access. We selected a 6-mm outer ring polytetrafluoroethylene graft (Propaten; W. L. Gore & Associates, Flagstaff, Ariz), not saphenous vein, for the graft conduit to obtain adequate blood flow in the celiac artery circulation. The bypass was performed in an end-to-side fashion through the retroperitoneal and mesentery space. Aortography depicted sufficient hepatic and splenic blood flow through the 6-mm graft, and retrograde blood flow of the common hepatic artery from the SMA disappeared (Fig 4). The operative time was 363 minutes, and total blood loss was 660 mL.

The patient was discharged 12 days postoperatively without complications. Follow-up CT confirmed no recurrence of PDAAs and good bypass flow 7 days after the operation (Fig 5) and 6 and 12 months postoperatively.

DISCUSSION

Multiple PDAAs have been rarely reported in the international literature.[5] PDAAs have been related to celiac
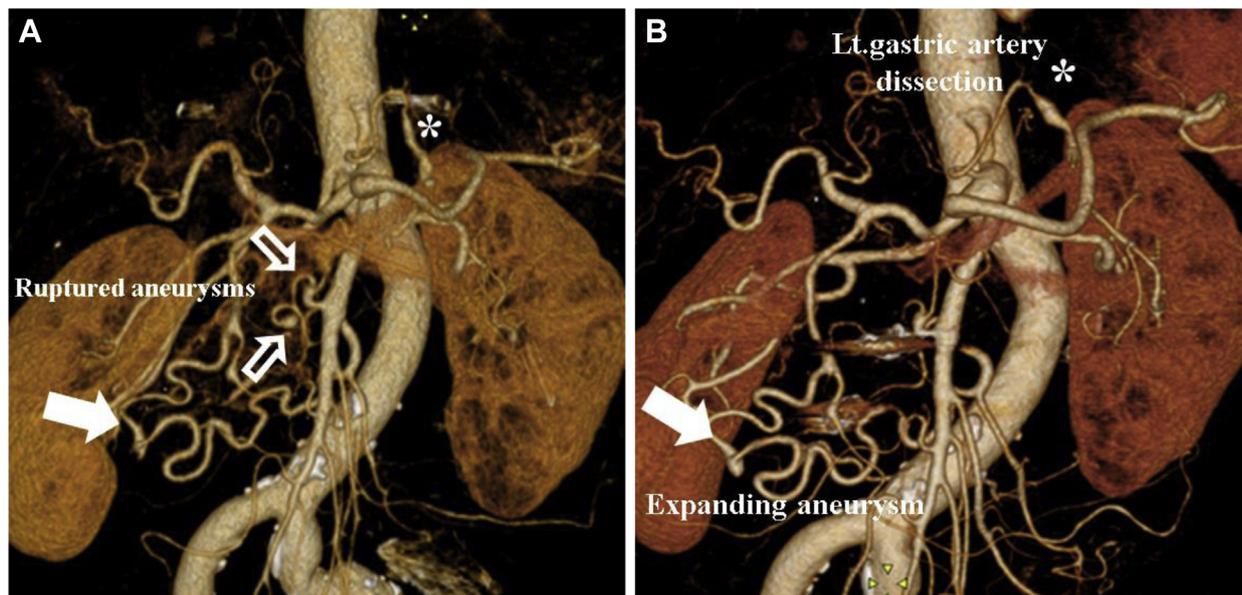
Fig 2. Comparison of the follow-up three-dimensional computed tomography (CT) image after the initial embolization. **A,** CT image immediately after the initial embolization showing two ruptured aneurysms (*open arrows*). **B,** Follow-up CT image at 1 month showing rapidly expanding aneurysm formation of the residual pancreaticoduodenal artery arcade aneurysms (PDAAs; *arrow*) and a new left gastric artery dissection (*asterisk*).
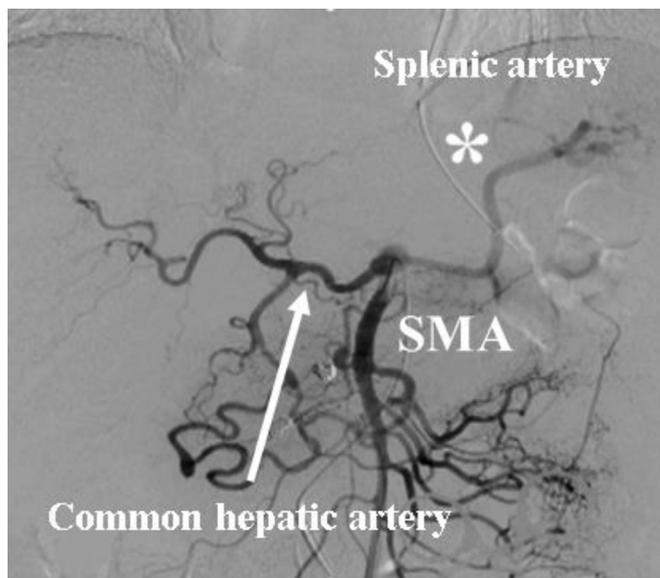

Fig 3. Selective superior mesenteric artery (*SMA*) angiogram shows retrograde blood flow through the peripancreatic arcade from the SMA to the common hepatic artery (*arrow*) and splenic artery (*asterisk*) before ligament resection.

axis stenosis or occlusion due to either atherosclerosis or MAL compression with a prevalence ranging from 63% to 74% of cases in different series.[2,6] The relationship between PDAAs and celiac axis lesions was first hypothesized by Sutton and Lawton[7] in 1973, and it has been reported recently by several other authors.[2,6,8] Compression of the celiac axis by the MAL causes decreased

blood flow in the celiac axis trunk. To compensate for this hypoperfusion, aneurysm formation of the pancreaticoduodenal artery arcade seems to be related to an increased retrograde collateral blood flow from the SMA to the celiac artery through the peripancreatic collaterals. Rupture of the PDAAs is the most worrisome because it can be life-threatening. In recent literature reviews, rupture has been reported in about 45% of all PDAAs due to celiac axis stenosis, and the estimated mortality of ruptured PDAAs ranges from 17.5% to 33.3%.[2,9] The size of PDAAs is not associated with the risk of rupture, and most ruptured aneurysms are <10 mm in diameter.[3]

TAE for ruptured PDAAs has almost been established for first-line treatment because of less invasiveness and the poor clinical hemodynamics.[1,9] On the other hand, second-line treatment of a residual PDAA and celiac axis stenosis after TAE remains controversial. In the case of PDAAs with celiac artery stenosis, TAE without revascularization of the celiac artery may lead to recurrence of PDAAs or ischemic dysfunction of the liver, spleen, or duodenum as a result of the absence of major collateral vessels.[1] Takeuchi et al[4] described four patients treated for celiac stenosis after TAE. Endovascular repair with stent was performed in 2 patients, open surgery for decompression in 1, and bypass grafting with saphenous vein in 1. They recommended surgical treatment of ligament release or arterial flow reconstruction because the endovascular approach might be difficult. In contrast, several authors have reported that revascularization for celiac stenosis after TAE may not be necessary
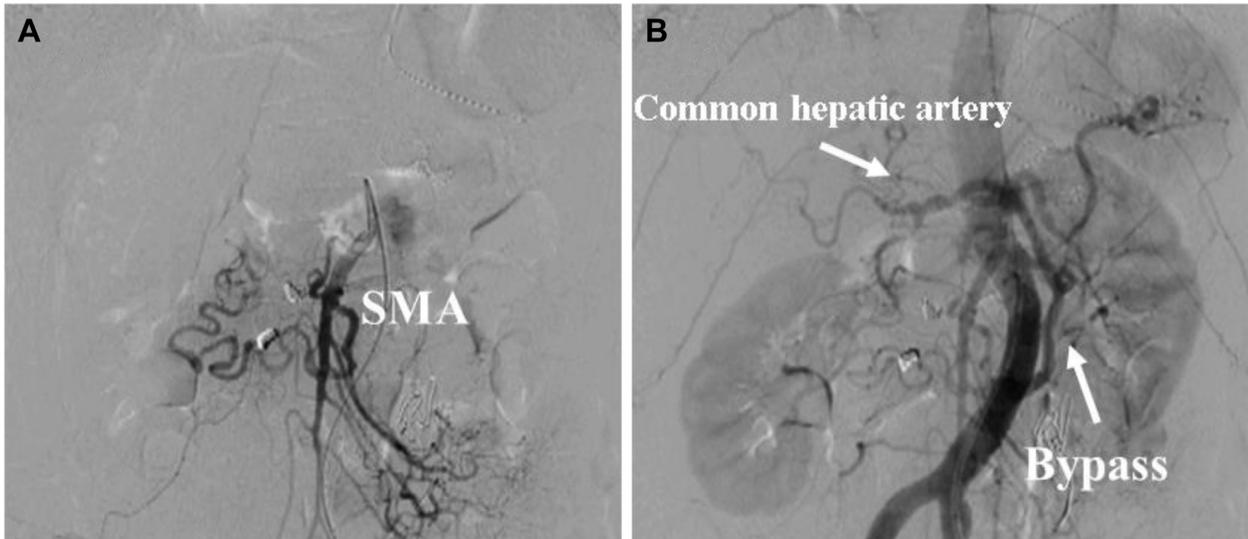
Fig 4. Superior mesenteric artery (*SMA*) and aorta angiography after the bypass. **A,** After aorta-common hepatic artery bypass, retrograde flow from the SMA is decreased. **B,** The aortogram shows antegrade blood flow from infrarenal aorta to the common hepatic artery by bypass.
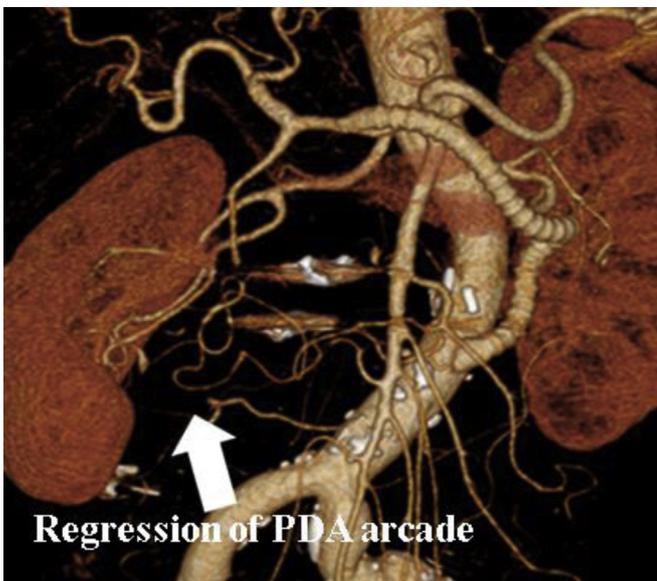

Fig 5. Postoperative follow-up three-dimensional computed tomography (CT) 7 days later shows regression of the pancreaticoduodenal artery (*PDA*) arcade (*arrow*).

because there was no recurrence of the aneurysm after TAE during the follow-up term.[10]

In our case, follow-up CT 1 month after initial TAE for ruptured PDAA showed rapid expanding residual aneurysm and new left gastric artery dissection. Additional therapy was necessary for the prevention of rerupture. We obtained regression of the residual PDAAs and normalization of the peripancreatic arcade artery by the aorta-common hepatic artery bypass after MAL release.

Insufficient bypass flow is caused by residual retrograde SMA flow and does not prevent residual PDAA rupture.

Liver and splenic circulation is reported to be about 359 mL/min[11] and 313 mL/min, respectively[12]; therefore, bypass conduit is the preferred artificial graft because saphenous vein is not enough to supply the liver and splenic circulation. In situ graft reconstruction could be a short bypass; however, an aortic maneuver, such as a partial clamp, is technically difficult around the celiac axis because of the deep and small working space. The infrarenal abdominal aorta is easy to manage and suitable for inflow of the graft. The bypass route is determined to be not retroperitoneal but transmesenteric because of the natural flow configuration through the graft.

Surgical treatment of celiac stenosis after TAE might be the best strategy if it is necessary and the patient's general condition is good. When subsequent follow-up CT reveals a new arterial event after blood flow modification, additional intervention for residual aneurysms might be necessary.

Intensive follow-up may also be recommended for a patient in poor condition or with no change in aneurysmal morphology although there is some possibility for sudden rupture.

Bypass surgery is a safe and reliable method for blood flow modification in the celiac artery arcade.

CONCLUSIONS

Close follow-up CT is essential to evaluate new aneurysm formation and arterial dissection after initial TAE of ruptured PDAA. Blood flow modification might prevent secondary rupture of PDAA associated with celiac axis stenosis.

REFERENCES
1. Murata S, Tajima H, Fukunaga T, Abe Y, Niggemann P, Onozawa S, et al. Management of pancreaticoduodenal

artery aneurysms: results of superselective transcatheter embolization. AJR Am J Roentgenol 2006;187:W290-8.

2. De Santis F, Bruni A, Da Ros V, Chaves Brait CM, Scevola G, Di Cintio V. Multiple pancreatoduodenal artery arcade aneurysms associated with celiac axis root segmental stenosis presenting as aneurysm rupture. Ann Vasc Surg 2015;29:1657.e1-7.

3. Miyayama S, Terada T, Tamaki M. Ruptured pancreaticoduodenal artery aneurysm associated with median arcuate ligament compression and aortic dissection successfully treated with embolotherapy. Ann Vasc Dis 2015;8:40-2.

4. Takeuchi Y, Morikage N, Samura M, Harada T, Yamashita O, Suehiro K, et al. Treatment options for celiac stenosis and pancreaticoduodenal artery aneurysms. Ann Vasc Surg 2017;41:281.e21-3.

5. Sakpal SV, Addis M, Chamberlain RS. Rapid progression of multiple splanchnic artery aneurysms. Surgery 2009;145:573-4.

6. Ducasse E, Roy F, Chevalier J, Massouille D, Smith M, Speziale F, et al. Aneurysm of the pancreaticoduodenal arteries with celiac trunk lesion: current management. J Vasc Surg 2005;39:906-11.

7. Sutton D, Lawton G. Coeliac stenosis or occlusion with aneurysm of the collateral supply. Clin Radiol 1973;24:49-53.

8. Flood K, Nicholson AA. Inferior pancreaticoduodenal artery aneurysms associated with occlusive lesions of the celiac axis: diagnosis, treatment options, outcomes, and review of the literature. Cardiovasc Intervent Radiol 2013;36:578-87.

9. Sugiyama K, Takehara Y. Analysis of five cases of splanchnic artery aneurysm associated with celiac artery stenosis due to compression by the median arcuate ligament. Clin Radiol 2007;62:688-93.

10. Sgroi MD, Kabutey NK, Krishnam M, Fujitani RM. Pancreaticoduodenal artery aneurysms secondary to median arcuate ligament syndrome may not need celiac artery revascularization or ligament release. Ann Vasc Surg 2015;29:122.e1-7.

11. Carlisle KM, Halliwell M, Read AE, Wells PN. Estimation of total hepatic blood flow by duplex ultrasound. Gut 1992;33:92-7.

12. Koyama K. Hemodynamics of the spleen in Banti's syndrome. Tohoku J Exp Med 1967;93:199-217.