Pancreaticoduodenal Artery Aneurysm Formation with Superior Mesenteric Artery Stenosis

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Celiac stenosis or occlusion is attributed partly to increased blood flow at pancreatic arcade from the superior mesenteric artery (SMA) system and may play a causal role in true aneurysm of pancreaticoduodenal artery (PDAA) formation. However, despite possible increased blood flow in the pancreatic arcades like celiac stenosis, PDAA with a stenotic SMA are extremely rare, with only three cases have been reported in the literature. We report a case of PDAA with SMA stenosis and review the literature.

Keywords: pancreaticoduodenal artery aneurysm, superior mesenteric artery, true aneurysm

Introduction
Pancreaticoduodenal artery aneurysm (PDAA) is a rare condition, that accounts for only 2% of splanchnic aneurysms.1) PDAA are classified as either true or false aneurysms. While false PDAA results from surrounding inflammation such as pancreatitis or trauma, true PDAA is degenerative or related to atherosclerosis and remains elusive in origin.1) Sutton et al. reported that true PDAA is associated with celiac stenosis or occlusion, and that 80% of patients with true PDAA had celiac stenosis.2) Celiac stenosis or occlusion, which is a result of atherosclerosis, fibromuscular hyperplasia or compression by the median arcuate ligament may induce hemodynamic changes in the pancreatic arcade. Increased blood flow in the pancreatic arcades is speculated to induce some of true PDAA.

However, despite possible increased blood flow in the pancreatic arcades, PDAA with a narrowed superior mesenteric artery (SMA) are rare, with only three cases have been reported in the literature, to the best of our knowledge.3–6)

The present report describes a case of PDAA with stenosis at the origin of the SMA.

Case Report
A 73-year-old woman was admitted to our hospital for detailed examination of a right abdominal mass detected during medical checkup. She had no symptoms such as pain, poor appetite and weight loss. The physical findings were not applicable except pulsatile abdominal mass at the right subcostal region. She suffered from chronic hepatitis C infection, which she contracted from a blood transfusion given during the delivery of one of her children. Abdominal computed tomography (CT) using contrast medium revealed an aneurysm 2-cm in diameter in the anterior superior pancreaticoduodenal artery (Fig. 1A). Blood testing showed the following, white blood cells 3500/µl, red blood cells 361×10⁴/µl, platelets 62000/µl, activated partial thromboplastin time 32.7 s, prothrombin time 13.2 s, AST 89 U/L, ALT 96 U/L, serum amylase 131 U/L.

Selective abdominal angiography from the celiac axis revealed excessive development of the pancreatic
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arcade and an aneurysm of 2 cm in diameter in the anterior superior pancreaticoduodenal artery without celiac stenosis. It also showed blood flow circulated smoothly from the celiac to the SMA. A selective angiogram from the SMA showed a narrowed orifice of the SMA and contrast medium did not perfuse into the distal SMA, which indicated that blood flow of the distal SMA comes from the celiac artery (Fig. 1B).

The patient underwent open surgery because aneurysm resection might decrease mesenteric circulation and need potential bypass to the SMA. An abdominal transverse incision was made under general anesthesia. She had liver cirrhosis and a hemorrhagic tendency on gross findings. After proximal and distal artery control was obtained, the aneurysm was completely isolated and resected under systemic heparinization. During clamping of the pancreaticoduodenal artery, pulsation of the SMA had ceased, but resumed after declamping. The pulsation of the aneurysm disappeared when clamping of the pancreaticoduodenal artery was made at the celiac axis side, which indicated that direction of blood flow was from celiac axis to the SMA. Histological specimens revealed a saccular aneurysm with layers of destroyed media and stretched adventitia.

Except formation of retroperitoneal hematoma around the ascending colon, which occurred due to traction and extravasation from small vessels during the operation, her postoperative course was uneventful. Postoperative CT displayed patent pancreaticoduodenal artery without aneurysm (Fig. 2). She
was discharged from the hospital on the 13th postoperative day.

**Discussion**

The hemodynamic stress of pancreaticoduodenal arcades induced by a narrowed or occlusive celiac trunk might lead to the development of true PDAAs. Mano, et al. reported that patients of true PDAAs with occlusive celiac trunk had increased blood flow in the pancreaticoduodenal arcades and that the direction of the flow was from the SMA to the hepatic artery. The present patient with no history of pancreatitis or trauma had a true PDAA, which was confirmed by histologic specimens. She did not have narrowed celiac trunk but had stenosis of the SMA, the main blood flow of which was supplied via the pancreaticoduodenal arcade from the celiac axis. The cause of SMA stenosis in the present patient was not identified; however, based on the findings from CT and angiography, it can be assumed that her pancreaticoduodenal arcades had increased blood flow. The SMA stenosis may also induce hemodynamic stress in the pancreaticoduodenal arcades and lead to develop a true PDAA. However, despite the fact that chronic mesenteric ischemia is not so rare, there were only three cases of patients with PADDs and SMA stenosis or occlusion have been published (Table 1). Moreover, Kimura, et al. reported a case of PDAA with both celiac and SMA stenosis. It remains elusive whether there is direct association between hemodynamics of pancreaticoduodenal arcades and development of a true PDAA.

Brocker et al. reviewed 93 cases of true PDAAs with celiac stenosis or occlusion and reported both that 52% were ruptured at the time of presentation and that aneurysm size did not correlate with rupture. These results suggest that PDAAs should be treated at the time of diagnosis. Treatment options include surgery (e.g., ligation, resection, or pancreatectoduodenectomy) and endovascular embolization. On the other hand, the optimal treatment for coexisting stenosis or occlusion of trunk arteries remains unclear. Brocker, et al. also did not observe PDAA recurrence secondary to residual celiac stenosis, on the other hand, 21% of the patients underwent treatment of their celiac stenosis with a corresponding mortality rate of 7.6%. We reviewed four cases of patients with PDAA and SMA stenosis, and only one of those patients underwent treatment of coexisting trunk stenosis.

The present patient had liver cirrhosis and underwent resection of an aneurysm without treatment of the narrowed SMA. She had good nutritional status and did not abdominal pain or diarrhea. We plan on following her and performing endovascular treatment for SMA stenosis if necessary in the future.

In conclusion, this report described a rare case of a patient with pancreaticoduodenal artery aneurysm and stenosis of the SMA. This disease entity remains poorly defined and requires additional investigation to determine the optimal treatment strategy.

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Table 1  A list showed reference related to pancreaticoduodenal artery aneurysm with narrowed superior mesenteric artery

<table>
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<th>Location</th>
<th>Size (mm)</th>
<th>Rupture</th>
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<th>Tx to aneurysm</th>
<th>Tx to SMA stenosis</th>
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CA: celiac axis; Tx: treatment; SMA: superior mesenteric artery
Disclosure Statement

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References