








Embolization of Inferior Pancreaticoduodenal Artery Aneurysm with Celiac Stenosis or Occlusion: A Report of Three Cases and a Review of Literature

복강동맥 협착 또는 폐색을 동반한 하체십이지장동맥 동맥류의 색전술: 세 개의 증례 보고와 문헌 고찰

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True pancreaticoduodenal artery aneurysms are relatively rare, approximately 50% of which are associated with stenosis or occlusion of the celiac axis. It is imperative to treat the condition immediately after diagnosis, considering that its rupture has a mortality rate of approximately 50%. The current most commonly used method to treat pancreaticoduodenal artery aneurysms is transcatheter arterial embolization. Here, we report three cases of embolization of inferior pancreaticoduodenal artery aneurysm with celiac stenosis or occlusion along with a literature review.

Index terms Aneurysm; Celiac Artery; Embolization, Therapeutic; Median Arcuate Ligament Syndrome; Abdominal Pain

INTRODUCTION

Pancreaticoduodenal artery (PDA) aneurysms are rare, representing only 2% of all visceral artery aneurysms (1). In almost 50% of reported cases, formation of PDA aneurysm is associated with celiac axis stenosis or occlusion (2, 3). Similar to other aneurysms, rupture is the major complication in PDA aneurysms as well. The overall rup-

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Table 1. Patient Demographic, Disease, and Treatment Characteristics of the Three Cases

Case	Age (years)/ Sex	Symptoms	Findings of Celiac Axis	Findings of Aneurysm (number, size*)	Treatment	Follow Up
1	54/F	Epigastric pain	Stenosis d/t MAL compression	Multiple, ruptured AIPDAA, 11.7 mm	TAE with coils	No complication for 6 months
2	53/F	Abdominal pain	Stenosis d/t MAL compression	Multiple, ruptured AIPDAA, 3.1 mm	TAE with coils	No complication for 12 months
3	76/M	None	Occlusion d/t suspicious MAL compression	Single, unruptured AIPDAA, 22 mm	TAE with coils	No complication for 8 months

*If multiple aneurysms, size of the largest one.

AIPDAA = anterior inferior pancreaticoduodenal artery aneurysm, d/t = due to, MAL = median arcuate ligament, TAE = transarterial embolization

ture rate is approximately 40% and the mortality rate of rupture is up to 50% (1, 4, 5). Therefore, it is important to treat PDA aneurysm immediately on detection.

With advances in imaging and interventional techniques, transcatheter arterial embolization of the aneurysm has replaced surgery as the treatment of choice (2). In this article, we report three cases of embolization of inferior PDA (IPDA) aneurysm with celiac stenosis or occlusion. We also include a literature review of IPDA aneurysm associated with celiac artery stenosis or occlusion.

CASE REPORTS

CASE 1

Refer to Table 1 for patient's information and characteristics of disease and treatment. A 54-year-old female presented acute onset epigastric pain and had no significant underlying disease such as hypertension nor connective tissue disease. For evaluation, she underwent abdominopelvic CT (AP CT). On AP CT, there was retroperitoneal hematoma with a small aneurysm in the IPDA and celiac artery stenosis (Fig. 1A). The surgery department suggested endovascular treatment because of the high failure rate of surgery. So, she was scheduled to undergo endovascular treatment. The access was achieved via right femoral artery puncture with a 5-Fr vascular sheath (Terumo, Tokyo, Japan). Selective celiac and superior mesenteric artery (SMA) angiography was performed with a cobra catheter (Terumo), which revealed multiple aneurysms in the anterior IPDA, with the largest one approximately 11.7 mm (Fig. 1B). The celiac axis showed a hook-like appearance, suggesting median arcuate ligament (MAL) compression. The aneurysms, also with efferent and afferent arteries were embolized using fourteen metallic detachable coils (IDC; Boston Scientific, Marlborough, MA, USA) (Fig. 1C). The angiogram obtained after embolization showed no definite evidence of contrast filling in the aneurysm and persistent blood flow to the common hepatic artery (Fig. 1D). Further treatment was recommended, if needed. In the immediate follow-up, no hepatic ischemic symptoms or laboratory abnormalities were seen. On 6-month follow-up, CT showed no recurrent aneurysm nor infarction of visceral organ.

Fig. 1. (Case 1) 54-year-old female presenting with acute onset epigastric pain.

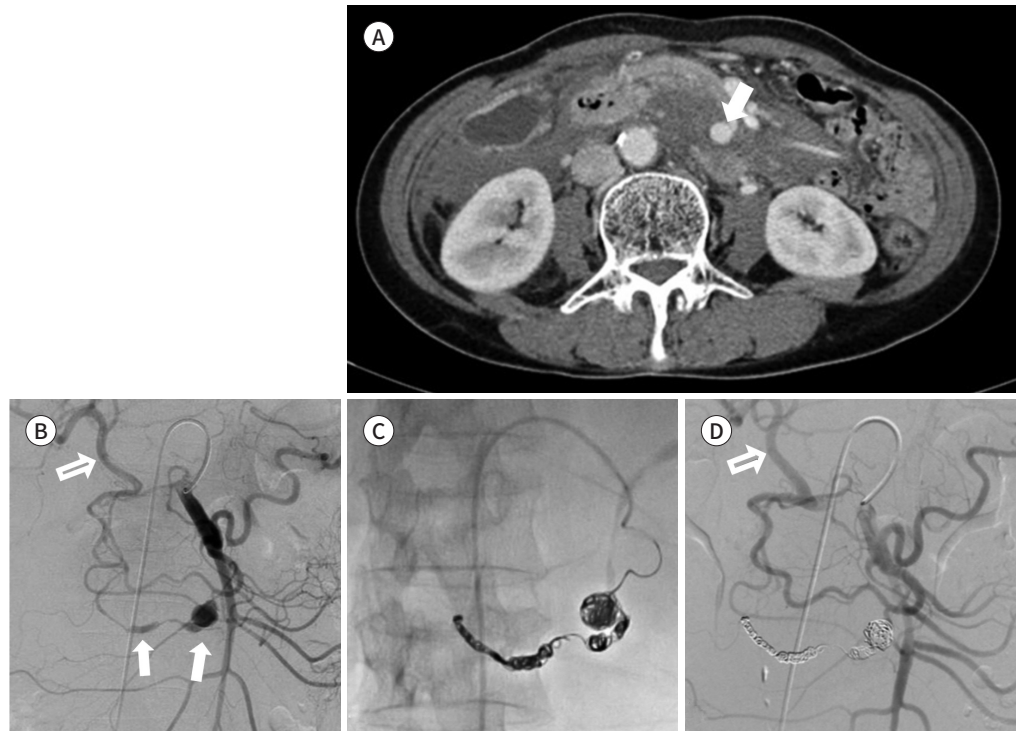
A. Pre-procedural CT shows aneurysm (arrow) on IPDA with retroperitoneal hematoma.

B. SMA arteriogram shows multiple small aneurysms in IPDA (arrows) with collateral blood flow to the common hepatic artery (open arrow) suggesting celiac trunk stenosis or occlusion.

C. IPDA aneurysm is embolized with detachable coil.

D. After embolization of the IPDA aneurysm, an SMA angiogram shows no contrast filling in the PDA aneurysm and persistent blood flow to the common hepatic artery (open arrow).

IPDA = inferior PDA, PDA = pancreaticoduodenal artery, SMA = superior mesenteric artery



CASE 2

Refer to Table 1 for patient's information and characteristics of disease and treatment. A 53-year-old female visited the emergency department with acute onset abdominal pain and had no significant underlying disease such as hypertension nor connective tissue disease. She underwent AP CT. On AP CT, there was retroperitoneal hematoma with a suspicious small aneurysm in the IPDA. The celiac axis showed a hook-like appearance, which indicated MAL compression (Fig. 2A, B). The surgery department suggested endovascular treatment because of the high failure rate of surgery. So, she was scheduled to undergo endovascular treatment. The access was achieved via right femoral artery puncture with a 5-Fr vascular sheath. Selective celiac and SMA angiography was performed with a cobra catheter. It showed a saccular aneurysm at the anterior IPDA with diffuse dilatation of PDA (Fig. 2C). Severe stenosis at the proximal celiac artery was also seen. Transarterial embolization for IPDA aneurysm was done with seven metallic detachable coils (IDC, Boston Scientific) (Fig. 2D). The angiogram obtained after embolization showed no definite evidence of contrast filling in the aneurysm (Fig. 2E). Further treatment was recommended, if needed. In the immediate follow-up, no hepatic ischemic symptoms or laboratory abnormalities were seen. But 5 days later, the patient complained of dizziness. Brain MRI showed acute infarction in the cerebel-

Fig. 2. (Case 2) 53-year-old female presenting with acute onset abdominal pain.

A. CT angiography shows a small aneurysm in the IPDA (arrow).

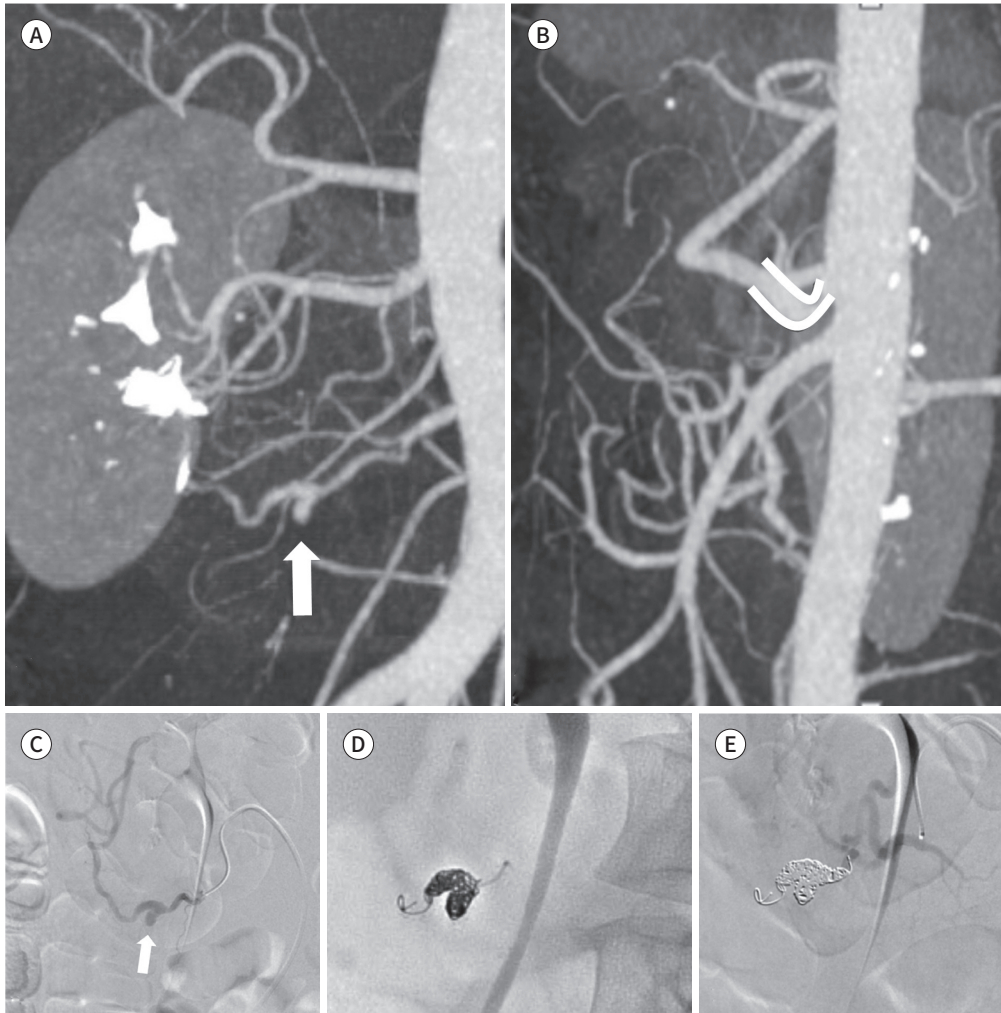
B. On CT angiography, the celiac axis shows a hook-like appearance (curved line), indicating MAL compression.

C. On SMA arteriography, a saccular aneurysm approximately 3.1 mm in size is seen (arrow). Diffuse dilatation of the IPDA is also noted.

D. The IPDA aneurysm is embolized with isolation.

E. Post-embolization angiography shows no definite blood flow in the aneurysm.

IPDA = inferior pancreaticoduodenal artery, MAL = median arcuate ligament, SMA = superior mesenteric artery



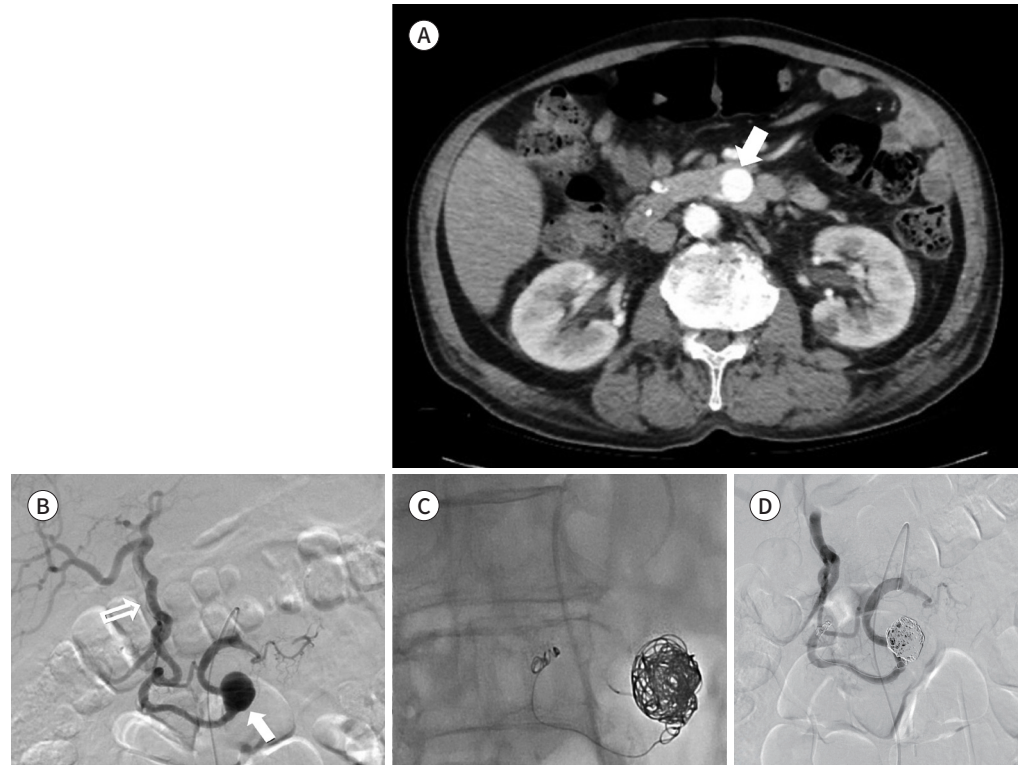
lum and the chest CT angiography revealed arteriovenous malformation (AVM) in left lower lobe of lung. So pulmonary angiography and endovascular coil embolization for pulmonary AVM were done with ten metallic detachable coils. On 12-month follow-up, CT showed no recurrent aneurysm nor infarction of visceral organ.

CASE 3

Refer to Table 1 for patient's information and characteristics of disease and treatment. A 76-year-old male came to our hospital due to uncontrolled fever and had no significant underlying disease such as hypertension nor connective tissue disease. For evaluation of fever focus, he underwent AP CT. AP CT revealed a suspicious large aneurysm in the IPDA without

Fig. 3. (Case 3) 76-year-old male presenting with uncontrolled fever.

- A.** Pre-procedural CT reveals an incidental aneurysm (arrow) in the IPDA without retroperitoneal hematoma.
B. SMA arteriogram shows a single aneurysm in the IPDA (arrow) with collateral blood flow to the proper hepatic artery (open arrow).
C. IPDA aneurysm is embolized with detachable coils.
D. SMA arteriogram after embolization of IPDA aneurysm shows that the aneurysm has disappeared.
 IPDA = inferior pancreaticoduodenal artery, SMA = superior mesenteric artery



retroperitoneal hematoma, indicating no definite rupture of aneurysm (Fig. 3A). The surgery department suggested endovascular treatment because of the high failure rate of surgery. So, he was planned to undergo endovascular treatment. The access was achieved via right femoral artery puncture with a 5-Fr vascular sheath. Selective celiac and SMA angiography was performed with a cobra catheter. A selective SMA angiography revealed a single aneurysm with a broad base at anterior IPDA and collateral blood flow to the proper hepatic artery (Fig. 3B). Transarterial embolization for IPDA aneurysm was done with seventeen detachable coils and five pushable metallic coils (Tornado; Cook, Bloomington, IN, USA) (Fig. 3C). The angiogram obtained after embolization showed no definite evidence of contrast filling in the aneurysm as well as intact hepatic flow through another PDA arcade (Fig. 3D). Further treatment was recommended, if needed. In the immediate follow-up, no hepatic ischemic symptoms or laboratory abnormalities were seen. On 8-month follow-up, CT showed no recurrent aneurysm nor infarction of visceral organ.

DISCUSSION

In 1973, Sutton and Lawton first suggested that celiac axis occlusion or stenosis was the

underlying cause of true PDA aneurysm. Stenosis or occlusion of the celiac axis increased blood flow in the peri-pancreatic arterial network and the chronic increase in blood flow through the PDA weakens and dilated the arterial wall, which, in turn leads to the formation of aneurysm (2, 3, 6).

The underlying cause of celiac axis stenosis or occlusion is frequently unknown, although atherosclerosis, fibromuscular dysplasia, aortic dissection, and MAL compression are possible etiologies (2). Among these etiologies, entrapment by the MAL (MAL compression) at the aortic hiatus has been cited as the cause more often than other etiologies (4). All three cases in this report were suspected to have the aneurysm due to celiac axis stenosis or occlusion. Furthermore, a hook-like appearance of the celiac axis, indicating MAL compression, was seen in two cases.

Clinical presentation of PDA aneurysm is variable and non-specific. The diagnosis of incidental PDA aneurysm is currently increasing as CT and ultrasonography are being increasingly used. Duplex ultrasound, CT angiography, and magnetic resonance angiography often yield this incidental diagnosis (2). However, conventional catheter angiography remains the gold standard (6).

Unlike other aneurysms of the visceral arteries, it seems that no correlation exists between the size of true PDA aneurysms and their propensity to rupture (3, 7). There are no predictive factors for rupture (2). Because it has unpredictability of rupture and has high mortality rate of rupture as up to 50%, it is important to treat this aneurysm when detected, regardless of its size (1).

In the past, surgery was the standard treatment, including ligation, resection, exclusion, and endoaneurysmorrhaphy (2). However, mortality rate for open repair has been reported to be as high as 50% (1, 3, 8). With the field of interventional radiology has continued to advance, endovascular treatment is now considered the first line therapy (2).

Until now, no definite guidelines have been established for treatment of PDA aneurysm. Some cases reported that patients only treated for celiac stenosis by stent ended up auto-occlusion of PDA aneurysm (2, 4, 7). However, when aneurysm ruptured like our two cases or when we considered unpredictability of aneurysm rupture, we therefore concluded that PDA aneurysm treatment is more reasonable with coil embolization.

Two major concerns of endovascular embolization for PDA aneurysm with celiac stenosis or occlusion are supposed. The first one is aneurysmal recurrence after aneurysm embolization; without celiac axis treatment, aneurysm recurrence is postulated to occur due to remaining blood flow via the PDA. However, in recent review, no cases of recurrence have been reported in patients who did not undergo treatment of celiac axis stenosis (1). The other concern is that interruptions in the arterial circulation to the liver may contribute to the development of hepatic failure. Generally, occlusion of the proximal hepatic artery from embolization is tolerated well in the presence of intact portal blood flow. In our cases, the hepatic arterial flow was observed to be normal after embolization and the portal venous flow was intact. Therefore, we presumed a low possibility of hepatic ischemia and infarction (9).

Treatment for concomitant celiac stenosis or occlusion is controversial (6, 8). Based on pathophysiology of PDA aneurysm, PDA aneurysm coil embolization without celiac axis treatment may remain increased flow via peri-pancreatic blood flow and leave possibility of

aneurysm recurrence (1, 3, 6). However, no cases of recurrences have been reported in the patient with untreated celiac axis (1, 2, 3). Also, endovascular treatment for celiac axis has many limitations. Percutaneous transluminal angioplasty and insertion of a stent for MAL compression do not solve the underlying problem of extrinsic compression of the celiac trunk and often require further open procedures due to stent occlusion by thrombus formation of neo-intimal hyperplasia, stent fracture, or hemorrhage induced by systemic antiplatelet therapy (7, 10). So, the treatment of celiac axis is considered when the patient's anatomy (on angiography) generates concern of potential hepatic or duodenal ischemia, the patient develops ischemic symptoms after initial definitive treatment, or the patient has continued symptoms similar to those on initial presentation (1).

Author Contributions

Writing—original draft, all authors; and writing—review & editing, all authors.

Conflicts of Interest

The authors have no potential conflicts of interest to disclose.

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복강동맥 협착 또는 폐색을 동반한 하체십이지장동맥 동맥류의 색전술: 세 개의 증례 보고와 문헌 고찰

곽민하¹ · 정현석^{2*} · 허영진¹ · 백진욱¹ · 신기원¹

하체십이지장동맥 동맥류는 드물다. 진성 하체십이지장동맥 동맥류의 약 50%가 복강동맥의 협착 또는 폐색과 관련이 있는 것으로 알려져 있다. 하체십이지장동맥 동맥류는 발견되는 즉시 치료해야 하는데, 파열 시 사망률이 50%에 이르기 때문이다. 최근에 가장 널리 쓰이는 치료법은 카테터 경유 동맥 색전술이다. 저자들은 복강동맥의 협착 또는 폐색을 동반한 하체십이지장동맥 동맥류를 색전술로 치료한 세 개의 증례를 소개하고, 이에 대한 문헌을 고찰하고자 한다.

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