Isolated Celiac Artery Dissection Caused by Segmental Arterial Mediolysis: A Case Report

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We encountered a case of intraabdominal hemorrhage caused by celiac artery dissection in a 74-year-old man. Angiography revealed the characteristic string-of-beads sign, irregular alternating arterial dilatations and stenoses giving a rosary-like appearance, which we suspected was caused by segmental arterial mediolysis（SAM）in the branches of the superior mesenteric artery（SMA）; these findings were observed in two locations along the pancreaticoduodenal artery and at one point along the small gastric branch. The patient was managed conservatively and the splanchnic arterial aneurysm believed to be the origin of the hemorrhage, as well as the aneurysm along the small gastric branch, resolved. There are no established criteria for invasive treatment of splanchnic artery aneurysms but this case suggests that thorough follow-up is necessary in cases of SAM.

Key words: celiac artery dissection, segmental arterial mediolysis, interventional radiology

INTRODUCTION

Known causes of celiac artery dissection include aortic dissection, trauma, and abdominal angiography. This report describes a case of isolated celiac artery dissection that we encountered and believe was caused by segmental arterial mediolysis（SAM）.

CASE

Patient: 74-year-old male; height: 162 cm; body weight: 41.8 kg.

Chief complaint: Right lower abdominal pain

Medical history: Hypertension, cerebral infarction

Current medication: Methyldigoxin 0.1 mg

Lifestyle and social history: Alcohol consumption, 1 serving/day for 50 years; tobacco smoking, previously smoked 20 cigarettes/day for 20 years（quit smoking 30 years earlier）.

History of present illness: Four days before being transported to our hospital, the patient was examined by his local physician for a chief complaint of epigastric pain. There were no other objective findings and he was observed and followed up. On the morning of the day that he was transported to our hospital, he visited his local physician again, and abdominal examination revealed epigastric tenderness and rebound tenderness. Blood pressure was 90/60 mmHg and heart rate（HR）was 110 beats/min; loading with 1000 mL of physiological saline raised his blood pressure to 115/86 mmHg and HR to 90 beats/min. Serum creatinine（Cr）and estimated glomerular filtration rate（eGFR）were 3 mg/dL and 15 mL/min/BSA, respectively, so contrast-enhanced computed tomography（CT）was contraindicated due to poor renal function. He was transferred to our hospital via emergency medical transport and we suspected celiac artery dissection based on findings

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of non-contrast-enhanced CT scan.

Condition on admission: Blood pressure was 124/57 mmHg, pulse was 86 beats/min, and body temperature was 36.5°C. On abdominal examination, there was marked tenderness extending from the epigastrium to the right hypogastric region, with muscle guarding and rebound tenderness.

Blood biochemistry: Anemia (Hb 8.5 g/dL, Ht 25.1%), excessive fibrinolysis (D-dimer 6.8 μg/mL), hypoproteinemia (TP 5.0 g/dL, Alb 2.8 g/dL), renal insufficiency (BUN 53.1 mg/dL, creatinine 2.56 mg/dL, eGFR 20.2 mL/min/BSA) (Table 1).

Electrocardiogram: HR 85 beats/min, negative T waves in leads II, III, aVF, and V1-V6.

Plain abdominal X-ray: Cardiothoracic ratio (CTR) was 43.2% and there was no pulmonary congestion.

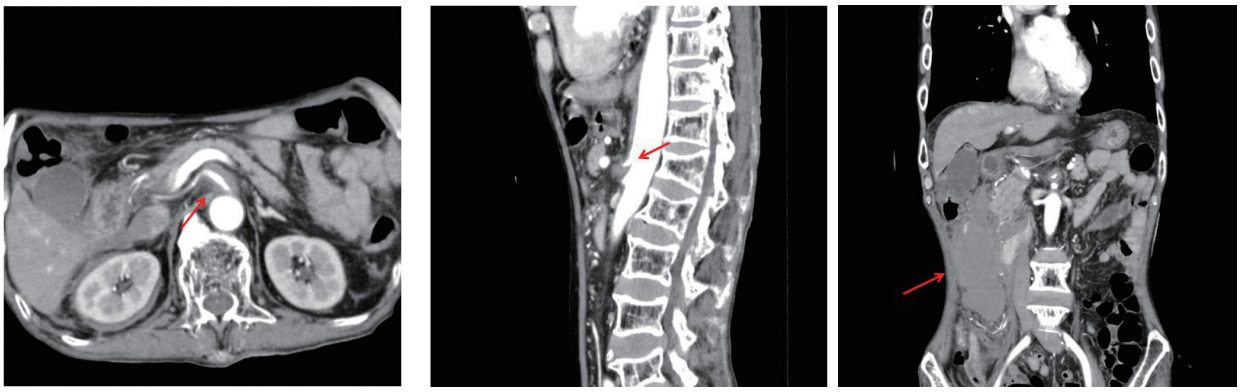
Abdominal contrast-enhanced CT: Signs of dissection were observed from the origin of the celiac artery to the common and left hepatic arteries (Fig. 1A, 1B). There were also mild signs of dissection at the origin of the splenic artery. The false lumina were all embolized by thrombus. There was no impairment of blood flow, and no organ ischemia was observed. A hematoma was observed from the area surrounding the descending limb of the duodenum to the center of the right retroperitoneum (Fig. 1C). There were no findings suggestive of a lesion indicative of a false aneurysm.

Clinical course after admission: Conservative treatment involved pain relief by administering fentanyl citrate and hypertension was treated with nicardipine hydrochloride. The patient developed aspiration pneumonia, although his general condition improved. On day 10 of admission, we performed angiography, which revealed the string-of-beads sign, a rosary-like appearance formed by irregular alternating dilatations and stenoses (Fig. 2).

On day 37 of admission, after the second episode of aspiration pneumonia had subsided, we performed a repeat angiography for the purpose of arterial embolization. Visualization of the region from the celiac artery to the gastroduodenal artery was much improved; the splanchnic aneurysm that was believed to the source of hemorrhage, and the aneurysm along the small gastric branch had resolved (Fig. 3), and embolization was not performed. The patient was discharged on day 50 of admission.

DISCUSSION

Celiac artery dissection most commonly occurs secondary to an aortic dissection, and isolated celiac artery dissection is rare. We performed a search of *Igaku Chuo Zasshi* (ICHUSHI) with the keyword "isolated celiac artery dissection" between 2010 and 2016 and found 276 cases, and only one case was due to SAM [1]; our case is the second. The most common chief complaint in celiac artery dissection is abdominal pain; epigastric pain was noted in our patient [2-8]. From previous reports, the mean age at the time of onset is 53.8 years (38-89 years) and the male-to-female ratio shows an overwhelmingly male preponderance, with 84.5% of patients being male [9]. Risk factors are the same



A B C

Fig. 1. Abdominal contrast-enhanced CT. (A) Axial view and (B) sagittal view showing celiac artery dissection (arrows), and (C) coronal view showing hematoma near the descending limb of the duodenum (arrow).

Table 1. Blood biochemistry

WBC	4690/μL	Alp	228 U/L
RBC	2.84×10^{6}/μL	γ-GTP	32 U/L
Hb	8.5 g/dL	CK	394 U/L
Ht	25.1%	Amylase	22 U/L
Plt	123×10^{3}/μL	BUN	53.1 mg/dL
INR	1.26	Crea	2.56 mg/dL
APTT	27.1 s	eGFR	20.2 mL/min/BSA
Fib	294 mg/dL	Na	136 mmol/L
D-dimer	6.8 μg/mL	K	4.7 mmol/L
AT	68%	Cl	107 mmol/L
TP	5.0 g/dL	Ca	8.1 mg/dL
Alb	2.8 g/dL	CRP	3.05 mg/dL
T-Bil	0.7 mg/dL	Glu	109 mg/dL
AST	54 U/L	HbA1c	5.6%
ALT	45 U/L	BNP	45.0 pg/mL
LDH	199 U/L		

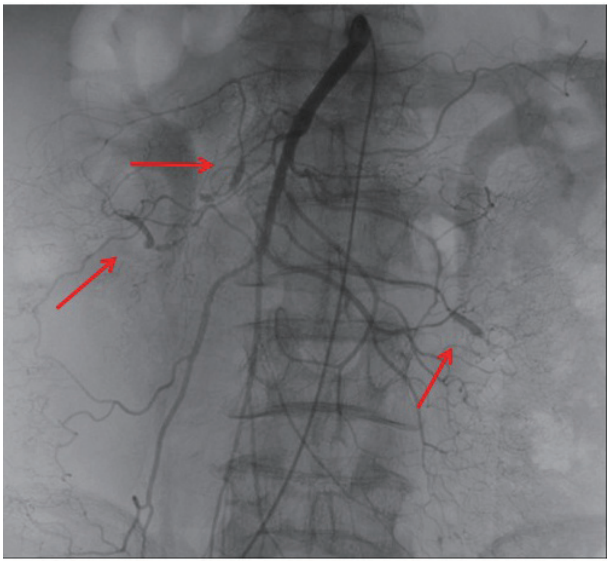


Fig. 2. First angiograph showing aneurysms in three different branches of the superior mesenteric artery (red arrows).

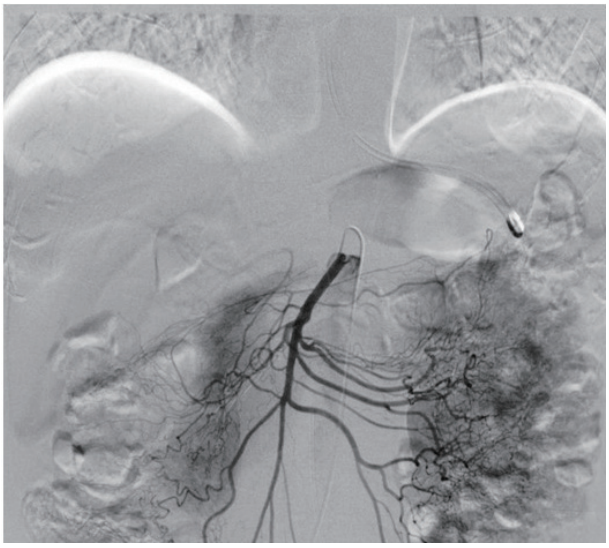


Fig. 3. Second angiograph showing the aneurysms had disappeared.

as coronary risk factors, and the relevant factors in the present case were a history of smoking and hypertension. Indications for invasive treatment include (1) arterial aneurysm and enlargement of the aneurysm diameter to 15-20 mm, (2) occlusion of the true lumen at the dissection, (3) progressive organ ischemia, (4) persistent pain, and (5) rupture/imminent rupture/intraabdominal hemorrhage [9]. In the present case, CT scan did not show signs of arterial aneurysm, and although we observed an intraperitoneal hematoma, there were no signs of active hemorrhage, so we decided that there were no urgent indications for invasive treatment. We therefore opted to first administer conservative treatment, focusing on controlling the blood pressure.

Vascular angiography performed on day 10 of admission showed findings suggestive of an SAM of the pancreaticoduodenal artery, one of the branches of the SMA. The concept of SAM as a definitive disease was first proposed by Slavin *et al.* in 1976; this disease frequently occurs in the splanchnic arteries, which are branches of the abdominal aorta,

causing segmental lysis of the arterial media, which in turn causes arterial aneurysm and dissection [10]. A report by Stanley *et al.* states that the site of onset is the splenic artery in 60% of cases, the hepatic artery in 20%, the SMA in 5.5%, the celiac artery in 4%, and the gastric or gastroepiploic artery in 3% [11]. In Japan, Matsumoto *et al.* reported that this condition occurs in the splenic artery in 31.6%, the hepatic artery in 24.8%, the submucosal arteries of the small bowel in 12.8%, the pancreaticoduodenal artery in 12.8%, the SMA in 6.8%, the gastric artery in 6.0%, the colic arteries in 6.0%, the celiac artery in 1.7%, and the inferior mesenteric artery (IMA) in 1.7% of cases [12]. A definitive diagnosis of SAM is made by means of histopathology, although in recent years, interventional radiology (IVR) has become a commonly treatment option for arterial aneurysms, and a histopathological diagnosis is obtained in very few cases. The diagnosis is based on four clinical criteria described by Uchiyama *et al*., namely (1) a middle-aged to elderly patient, (2) exclusion of underlying disease, such as inflammatory or arteriosclerotic changes, (3) sudden-onset intraabdominal hemorrhage, and (4) the string-of-beads sign, or a rosary-like arrangement of irregular alternating dilatations and stenoses [13]. According to Inada *et al*., the majority of splanchnic artery aneurysms are caused by SAM, with multiple onset occurring in 34.6% of cases [14]. The celiac artery dissection in our case occurred at multiple ectopic sites in addition to the pancreaticoduodenal artery, onset was sudden, there was no genetic background, and although there was history of hypertension and cerebral infarction, there were no other atherosclerotic changes observed. Thus, we believe that the cause was probably SAM.

Aggressive treatment is opted for in cases of SAM that have ruptured and present in a state of shock, and this treatment focuses on life-saving surgery and IVR. However, to date, there are no established criteria to determine the indications for elective invasive treatment, and the clinical course of cases of SAM that remain hemodynamically stable after rupture, or have not yet ruptured, is unclear. Takahashi *et al*. state that invasive treatment is indicated if the aneurysm diameter is ≥ 2 cm, there is a false aneurysm, a tendency towards aneurysm enlargement is observed, aneurysms arise in relatively small-caliber arteries, such as the pancreaticoduodenal artery, gastroduodenal artery, or branches of the SMA, or there are subjective symptoms [15]. The first choice of treatment is endovascular intervention, which is minimally invasive; laparotomy is performed in cases that are unable to undergo endovascular intervention or cases that require revascularization [15]. In our case, there were aneurysms of the branches of the SMA, and revascularization was not required. Consequently, we did not opt for surgical treatment but decided that arterial embolization was indicated. However, when we performed angiography on day 37 of admission, we found that the vascular aneurysms had all resolved, so we did not perform embolization. In this case, both angiographies were performed after aspiration pneumonia had improved. Thus, if the general condition of patients allows, it is desirable to perform angiography as early as possible.

Inada *et al*. reported that among the cases that were managed by treating only the ruptured aneurysm caused by SAM, in which other arteries remained untreated, the clinical course could only be established in five cases. However, it is possible that the size and number of the aneurysms were unknown during angiographic follow-up of these cases, and thus may have resolved spontaneously [14]. However, there are cases of secondary aneurysm rupture after SAM that presented with heterochronic onset [16], and thus it would be desirable to accumulate further cases in the future.

CONCLUSION

We encountered a case of isolated celiac artery dissection heralded by the onset of epigastric pain. SAM should be considered as the cause of rupture or dissection of splanchnic artery aneurysms occurring in middle-aged to elderly patients who do not have conspicuous atherosclerosis. There are no established standards for deciding on invasive treatment of splanchnic artery aneurysms, and this is left at the discretion of the managing institution. However, this case suggests that careful follow-up is necessary in cases of SAM to prevent recurrence or confirm spontaneous cure.

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