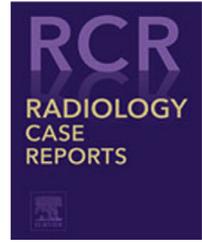


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Case Report

Successful coil embolization of a ruptured aneurysm of the arc of Riolan artery [☆]

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ABSTRACT

The arc of Riolan (AOR) is an anastomosis between the middle and left colic arteries. Aneurysms of the AOR are very rare visceral artery aneurysms. A 44-year-old man presented with abdominal pain and loss of consciousness. Computed tomography and angiography showed hemorrhagic ascites around the liver and spleen. An irregularly dilated artery was visible within a hematoma in the upper left region of the abdomen, consistent with a ruptured pseudoaneurysm of the AOR. Transcatheter arterial embolization was performed with microcoils. The patient's abdominal pain disappeared after embolization, and no symptoms of intestinal ischemia were observed. To our knowledge, this is the first case of an AOR aneurysm with AOR dilation due to dissection of the celiac artery that was successfully treated by coil embolization.

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Introduction

Visceral artery aneurysms (VAAs) are rare vascular pathologies with reported incidence rates of 0.01–0.2% [1]. Because rupture of a VAA is a potentially life-threatening event, immediate attention and treatment are necessary. They are more

frequent in the splenic (60%) and hepatic (20%) arteries, and are rare in the jejunal, ileal, and colic branches (about 3%) [2]. The arc of Riolan (AOR) is an anastomosis between the middle colic artery (MCA) and the left colic artery (LCA). There have been few published reports of AOR aneurysms [3–6], and even fewer of rare VAAs. To our knowledge, this is the first case of an AOR aneurysm with AOR dilation due to dissection of the

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celiac artery that was successfully treated by coil embolization.

Case report

Prior to preparing this report, informed consent was obtained from the patient. The 44-year-old man was transported to another hospital by ambulance due to abdominal pain and loss of consciousness. He had a history of untreated hypertension, but no significant family history. Upon arrival at the hospital, his systolic blood pressure was 60 mmHg and his pulse rate was 100 beats/min. His abdomen was rigid and distended with rebound tenderness.

Unenhanced and contrast-enhanced computed tomography (CT) revealed hemorrhagic ascites around the liver and spleen, and an irregularly dilated artery within the hematoma in the upper-left region of the abdomen, consistent with a ruptured pseudoaneurysm (Figs. 1A and B). In addition, localized dissection of the celiac artery was observed (Fig. 1C). He was admitted to the hospital and treated conservatively with fluid infusion and vasopressors. However, 3 days after admission, his hemoglobin level dropped to 5.9 g/dL from 14.7 g/dL on arrival.

He was transferred to our hospital and underwent emergency embolization using metallic coils. A 4 Fr sheath (Terumo, Tokyo, Japan) was inserted into the right femoral artery and a 4 Fr pigtail catheter (Medikit, Tokyo, Japan) was used to perform CT during aortography. Then, a 4 Fr 1CJ catheter (Medikit) was inserted into the inferior mesenteric artery (IMA). CT during aortography and IMA angiography revealed that the pseudoaneurysm was in the AOR (Figs. 2 and 3A). The dorsal pancreatic artery (DPA) branching from the celiac artery was dilated and connected to the AOR. The blood flow was directed from the LCA and the MCA to the celiac artery through the DPA.

A 1.9 Fr microcatheter (Carnelian Si, Tokai Medical Products, Aichi, Japan) was advanced from the IMA to the MCA beyond the pseudoaneurysm, which was embolized using an isolation technique with 10 microcoils (Target XL, Stryker Neurovascular, CA, USA; Fig. 3B). Postprocedural SMA and IMA angiographies revealed the disappearance of the pseudoaneurysm and the preservation of the intestinal blood flow through the marginal artery (MA). The patient's abdominal pain disappeared after embolization and no symptoms of intestinal ischemia were observed. Five days after admission to our hospital, the patient was transferred back to the original hospital.

Discussion

The definition of the AOR is ambiguous. According to Lange et al. [7], the incidence of AOR varies, from >50% of patients in some studies to <20% in other studies. The former studies simply referred to the MA, called the MA of Drummond, whereas the latter studies referred to rarer cases of anastomosis running more centrally than the MA. Although many

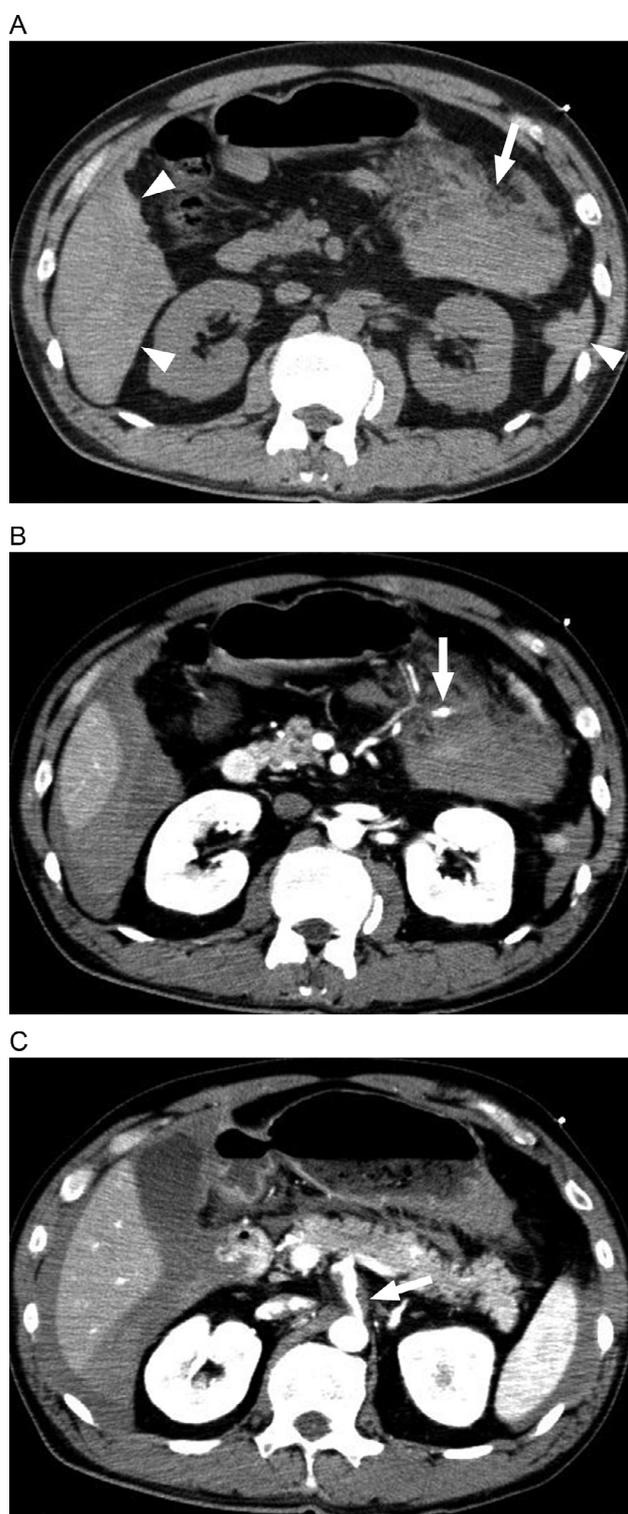


Fig. 1 – Computed tomography (CT) images obtained at the original hospital. (A) Unenhanced CT showing hemorrhagic ascites around the liver and spleen (arrowheads), and the hematoma in the upper left region of the abdomen (arrow). (B) Contrast-enhanced CT (arterial phase) showing a pseudoaneurysm (arrow) with a diameter of 5.4 mm within the hematoma. (C) Contrast-enhanced CT (arterial phase) showing the dissection (arrow) of the celiac artery.

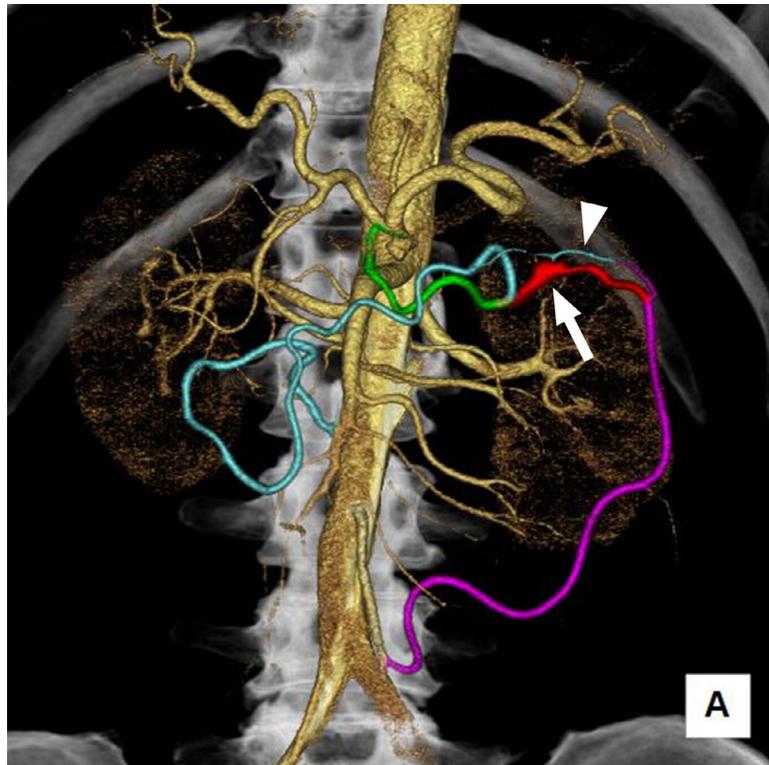


Fig. 2 – Volume rendered image created from computed tomography during aortography. A pseudoaneurysm (arrow) is seen in the arc of Rioloan artery (red) anastomosing the middle colic artery (light blue) and the left colic artery (purple). An anastomosis via the marginal artery is also seen on the intestinal side (arrowhead). The dorsal pancreatic artery (green) is dilated and connected with the arc of Rioloan artery.

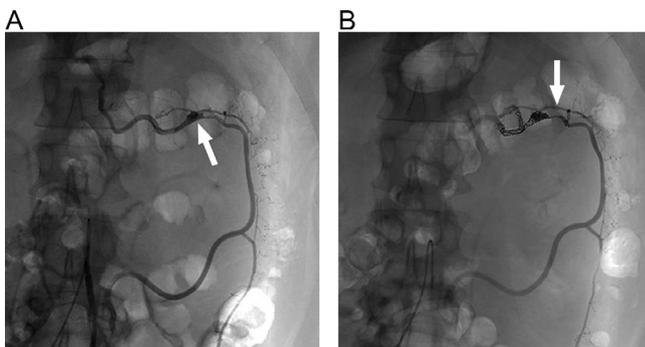


Fig. 3 – Embolization of the pseudoaneurysm. (A) Inferior mesenteric arteriography before embolization showing a pseudoaneurysm (arrow) of the arc of Rioloan. The blood flow is directed from the left colic artery to the celiac artery through the dorsal pancreatic artery. (B) Inferior mesenteric arteriography after embolization using metallic coils showing the disappearance of the pseudoaneurysm. The intestinal blood flow is maintained through the marginal artery (arrow) of the colon.

reports have described the latter as the AOR [8], a dilated anastomosis between the MCA and LCA is currently recognized as the AOR. In our patient, a pseudoaneurysm was found in

an artery running slightly more centrally than the MA in the splenic flexure of the colon. Therefore, this artery was considered to be the AOR.

Causes of VAAs include atherosclerosis, inflammation, connective tissue diseases (Marfan syndrome and Ehlers-Danlos syndrome), fibromuscular dysplasia, and segmental arterial mediolysis (SAM) [1,9]. In this case, SAM and increased blood flow were the most likely causes.

SAM is a non-atherosclerotic, non-inflammatory arterial lesion that causes arterial dilation, single or multiple aneurysms, dissecting hematomas, arterial stenosis, and arterial occlusion, mainly in the visceral arteries, in middle-aged patients due to lysis of the outer media of the arterial wall [10]. Because of the high frequency of celiac artery lesions (60%) [9], it is possible that SAM led to the celiac artery dissection and the AOR aneurysm in this case. However, because surgery was not performed, a definitive pathological diagnosis could not be made.

Increased blood flow can also result in aneurysms. Pancreatoduodenal arcade aneurysms associated with celiac artery stenosis due to median arcuate ligament syndrome are well known [11]. Aneurysms may occur as a result of hemodynamic stress on these small vessels due to increased blood flow and this theory is supported by reports showing that aneurysms shrank after treating the celiac artery stenosis [11,12].

In this case, there was stenosis of the celiac artery due to dissection. Although there was mild dilation of the pancreati-

coduodenal arcade, the DPA branching from the celiac artery was markedly dilated. Dilation of the DPA was observed in 9.1% of patients with celiac artery stenosis [13]. The DPA connected with the AOR on its caudal side, and the AOR was dilated to a diameter of up to 5.4 mm. Causes of AOR dilation include SMA or IMA occlusion or stenosis, colorectal cancer, and ulcerative colitis [14]. In the present case, AOR dilation was due to stenosis of the celiac artery caused by dissection, an atypical cause.

Angiography showed that blood flow was directed from the LCA and the MCA toward the DPA. Dilation of the AOR involving 2 arteries, the MCA and LCA, is often seen after endovascular aneurysm repair for abdominal aortic aneurysms. However, in such cases, aneurysm formation is thought to be very rare [4]. Therefore, in the present case, it is possible that more complex hemodynamics involving 3 arteries (DPA, MCA, and LCA) probably contributed to the formation of the AOR aneurysm.

Considering the other causes of aneurysms, atherosclerosis was not strongly suspected because the patient was relatively young and the arteriosclerotic lesions of the aorta or its branches were not prominent. Inflammation and connective tissue diseases were also not suspected because there were no clinical or blood laboratory findings associated with these diseases. Fibromuscular dysplasia usually affects the renal and carotid arteries of young to middle-aged women, and rupture is rare [9], inconsistent with the findings of the present case.

There are 4 prior reports of AOR aneurysms [3–6]. Endovascular treatment using coils was performed in 3 reports [4–6], and conservative treatment was performed at the patient's request in the other report [3]. In the 3 reports describing coil embolization, all of the patients were female (aged 45–61 years old). Their main symptom was abdominal pain, and the diameters of the aneurysms ranged from 8.8 to 20 mm. The causes of the aneurysms included dilation of the AOR after endovascular aneurysm repair [4] and fibromuscular dysplasia [6]. There were no complications after coil embolization and the postoperative course was uneventful. The fourth report described conservative treatment in a female patient (aged 65 years old) whose main symptom was abdominal pain, and the diameter of the aneurysm was 30 mm. The cause of the aneurysm was thought to be medial degeneration due to increased blood flow. The patient died following rupture of the aneurysm. Despite the small number of reports, the results suggest that aggressive treatment should be considered in patients with AOR aneurysms.

In patients with VAAs, there is no significant difference in the mortality rate between surgery and endovascular treatment [1]. Although endovascular treatment may be the safer option, especially for older patients or those with severe comorbidities, the risk of intestinal ischemia after treatment must be carefully assessed by predicting the candidate artery for collateral blood flow and reviewing the patient's history of previous abdominal surgery, as mentioned in prior reports [3,6]. Because new aneurysms may occur, periodic follow-up visits are essential to detect them at an early stage.

In conclusion, endovascular treatment can be an effective option for VAAs, including AOR aneurysms.

Data availability

Data may be obtained from the corresponding author.

Past presentation

None.

Patient consent

The patient provided informed consent for publication of this case report (05/05/2022).

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