—Case Reports—

Successful Coil Embolization of an Aneurysm in the Arc of Bühler

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In the present report, we describe a case of a patient with an asymptomatic aneurysm in the arc of Bühler (AOB), which was successfully treated by transcatheter arterial embolization. The patient presented with severe stenosis of the celiac trunk, which was suspected to be due to median arcuate ligament syndrome. Arteriography of the superior mesenteric artery indicated a rapid stream in an aneurysm in the AOB. Hence, embolization was carefully performed using detachable coils and microcoils. An arteriography performed after embolization did not show any aneurysm, and the hepatic artery and splenic artery could be detected via the pancreatic arcade, originating from the superior mesenteric artery. The AOB is a persistent embryonic ventral anastomosis present between the superior mesenteric artery and the celiac artery. This anastomotic artery is independent of the gastroduodenal artery and the dorsal pancreatic artery, and is extremely rare, with an incidence of <4%. Aneurysms of the AOB are even more uncommon, and such cases have been reported in association with stenosis or occlusion of the celiac trunk. Open surgical aneurysmectomy, with or without reconstruction, is the conventional treatment for such aneurysms. However, rapid advances in interventional radiology have enabled the safe and effective treatment of visceral aneurysms via transcatheter arterial embolization. Based on the current findings, we believe that transcatheter arterial embolization is a minimally invasive and valuable method that may serve as an initial treatment option for aneurysms of the AOB.
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Key words: arc of Bühler, aneurysm, transcatheter arterial embolization

Introduction

The arc of Bühler (AOB) is a persistent embryonic ventral anastomosis present between the superior mesenteric artery (SMA) and the celiac artery (CA)[1,2]. The AOB is independent of the pancreaticoduodenal and dorsal pancreatic arteries which are the most common anastomoses between the SMA and CA[3]. This condition is usually detected incidentally in asymptomatic patients undergoing imaging for other reasons as well as in symptomatic patients with celiac trunk stenosis related to arterial sclerosis or median arcuate ligament syndrome. This vascular anomaly is very rarely encountered, and aneurysms of the AOB are even more uncommon. The rupture of these AOB aneurysms may lead to a life-threatening condition. Although the management of this aneurysm remains unclear, a few reports[4–6] have described successful transcatheter arterial embolization of the aneurysm. In the present report, we describe a case of a patient with AOB that was successfully treated with coil embolization, and review the other reports.

Case Report

A 35-year-old man presented to our hospital with a feeling of abdominal enlargement. The patient had no history of abdominal trauma, surgery, or pancreatitis or any family history of aneurismal disease.

Enhanced abdominal computed tomography (CT) indicated the presence of a 2.7×2.0 cm aneurysm with clustered peripheral calcification located behind the pancreas (**Fig. 1a**), along with severe stenosis of the celiac trunk

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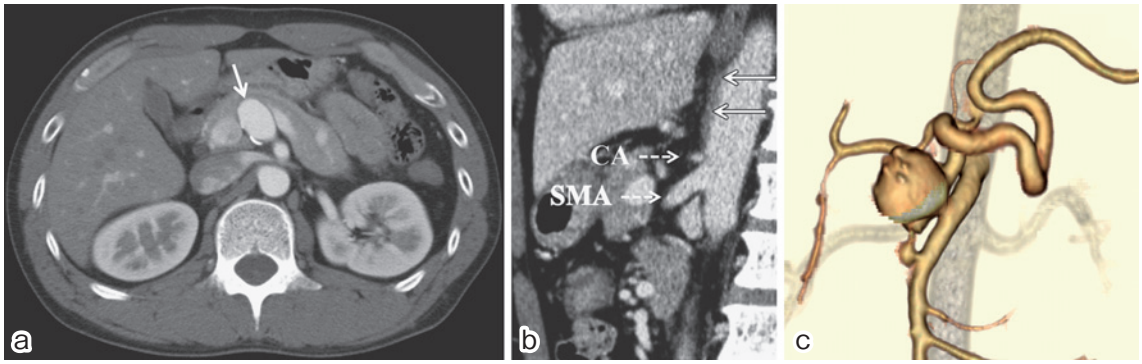

Fig. 1 Computed tomography (CT) findings.
a. Enhanced abdominal CT shows a 2.7×2.0 cm aneurysm with clustered peripheral calcification located behind the pancreas (**arrow**).
b. Reconstructed sagittal CT image shows severe stenosis of the celiac artery, which is suspected to be due to median arcuate ligament syndrome (**arrow**).
c. Three-dimensional CT angiography shows an aneurysm arising from an independent anastomotic artery located between the celiac artery and superior mesenteric artery.

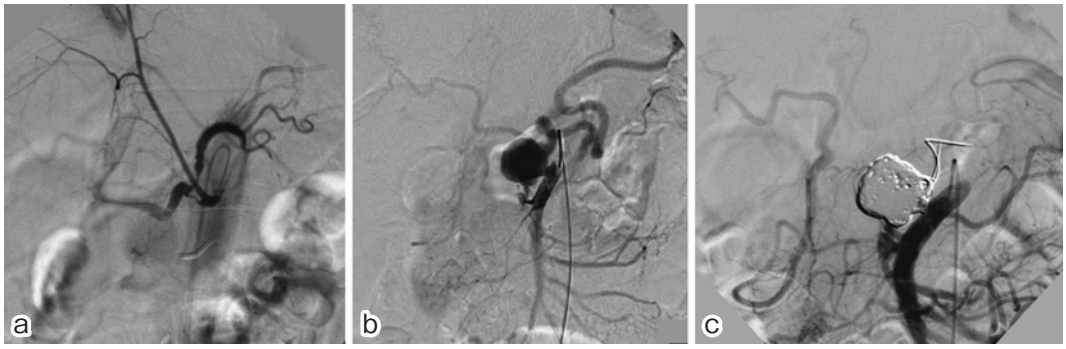

Fig. 2 Angiographic findings and treatment.
a. Celiac arteriography shows severe stenosis of the celiac artery.
b. Superior mesenteric arteriography shows a saccular aneurysm that is independent of the dorsal pancreatic artery and pancreatic arcade.
c. Post-embolization arteriography via the superior mesenteric artery shows complete occlusion of the aneurysm.

that may have been due to median arcuate ligament syndrome (**Fig. 1b**). Three-dimensional CT angiography showed that the aneurysm arose from an independent anastomotic artery between the CA and SMA (**Fig. 1c**). Moreover, celiac arteriography indicated severe stenosis of the CA (**Fig. 2a**). Superior mesenteric arteriography showed that the saccular aneurysm was independent of the dorsal pancreatic artery and pancreatic arcade, and a rapid stream was observed in the aneurysm. Based on these findings, we diagnosed it as an AOB aneurysm (**Fig. 2b**).

The rupture of AOB aneurysms can lead to life-threatening bleeding, and hence, it is preferable that such patients should receive early treatment. Moreover, as the patient in the present case desired to undergo treatment, we decided to isolate the aneurysm as a first-line therapy, and attempted to approach the AOB aneurysm through two routes, via the CA and SMA. However, a catheter could not be advanced due to the stenosis of the CA. Therefore, we decided to isolate the aneurysm via the SMA. Accordingly, a 2.3-Fr microcatheter (Prowler Plus, Codman & Shurtleff, Inc., MA, USA) was advanced into the efferent artery of the AOB aneurysm via the SMA. A 15 mm×30 cm detachable coil (DETACH™, Cook Medical, Bloomington, IN, USA) was then delivered into the efferent artery. However, this was not successful as the detachable coil had spread to the splenic artery due to the rapid stream. Therefore, we did not detach and pull back the coil, and instead changed the procedure for packing of the aneurysm. Framing was attempted with 13 detachable coils (five 15 mm×30 cm coils, two 12 mm×30 cm coils, five 10 mm×30 cm coils, and one 8 mm×20

cm coil, from DETACH™, Cook Medical, Bloomington, IN, USA), and packing of the aneurysm was attempted with 12 microcoils (10 MWCE-18S-10/5 mm coils, one MWCE-18S-8/4 mm coil, and one MWCE-18S-7/3 mm coil, from TORNADO™, Cook Medical, Bloomington, IN, USA).

After the embolization, an arteriography via the SMA showed complete occlusion of the aneurysm, as well as opacification of the hepatic artery and splenic artery via the pancreatic arcade (**Fig. 2c**).

On a follow-up CT performed after 2 months, the aneurysm was difficult to evaluate due to the metallic artifact from the embolization coils. However, no infarction of the liver and spleen, and no bowel ischemia were noted. Furthermore, no deterioration in laboratory data, including liver function results, was observed. One year after the embolization, superior mesenteric arteriography indicated the exclusion of the aneurysm along with a patent pancreatic arcade and no aneurysm recurrence. During the 5-year follow-up period, no aneurysm recurrence or other symptoms were noted.

Discussion

The AOB is formed by the failure of regression of the 10th and 13th ventral segmental arteries of the aorta in prenatal life[2]. The failure of such regression results in the presence of a persistent arterial connection, which forms the AOB and serves as a communication between the SMA and the CA artery or one of their branches[3]. However, this vascular anomaly is extremely rare, with an incidence of 1–4%[3,7]. Aneurysms of the AOB are even more uncommon, and to our knowledge, only five such cases have been reported in the literature[4–6,8,9]. The diameter of these reported aneurysms has ranged from approximately 2.0 to 6.0 cm. Surgical treatment was performed in 2 cases[8,9], including one case wherein transcatheter arterial embolization of the aneurysm was unsuccessful. The other three cases were successfully treated with transcatheter arterial embolization[4–6]. In most cases, aneurysms of the AOB have been reported in association with stenosis or occlusion of the celiac trunk. In the present case, the aneurysm of the AOB was thought to have developed as a collateral vessel due to the occlusion of the celiac trunk. Therefore, the artery of the AOB showed significantly large and rapid flow, and then showed complex dilatation as a collateral vessel. Among the cases reported thus far, the embolization procedure has involved packing of the aneurysm with detachable coils and micro coils. In cases of rapid flow, as in the present case, framing should be attempted first, with detachable coils, followed by packing of the aneurysm with micro coils. On the other hand, as embolization of this aneurysm may result in a significantly greater blood flow at other vessels such as a pancreaticoduodenal artery, it may lead to recurrence of the aneurysm. Hence, careful and long-term follow-up is necessary. Depending on the circumstances, resection of the median arcuate ligament or revascularization should be considered for severe stenosis or occlusion of the celiac trunk.

In conclusion, rapid advances in interventional radiology have enabled the safe and effective treatment of visceral aneurysms via transcatheter arterial embolization. We believe that transcatheter arterial embolization is a minimally invasive and valuable method that may serve as an initial treatment option for aneurysms of the AOB.

Conflict of Interest: The authors declare no conflict of interest.

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