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*Case Report*

# Endovascular treatment of a ruptured blister-like aneurysm at an azygos anterior cerebral artery: A case report and review of the literature

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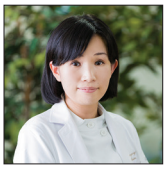

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## ABSTRACT

**Background:** Endovascular treatment for a ruptured blister-like aneurysm (BLA) has recently become a hopeful approach. BLAs are usually located on the dorsal wall of the internal carotid artery, whereas one located on the azygos anterior cerebral artery (ACA) is so rare, it has never been reported. We report a case of a ruptured BLA arising at the distal bifurcation of an azygos ACA treated by stent-assisted coil embolization.

**Case Description:** A 73-year-old woman presented with a disturbance of consciousness. Computed tomography showed diffuse subarachnoid hemorrhage, which was observed to be particularly dense in the interhemispheric fissure. Three-dimensional rotation angiography showed a tiny and conical bulge on the distal bifurcation of the azygos trunk. Follow-up digital subtraction angiography performed on day 4 showed enlargement of the aneurysm, and a BLA arising at the azygos bifurcation was diagnosed. Stent-assisted coiling (SAC) was performed using a low-profile visualized intraluminal support (LVIS) Jr. stent, which was implanted from the left pericallosal artery to the azygos trunk. Follow-up angiography showed that the aneurysm thrombosed gradually and reached complete occlusion 90 days after onset.

**Conclusion:** SAC for a BLA at the distal bifurcation of an azygos ACA might be an effective treatment option leading to early complete occlusion, but thrombus formation as an intraoperative complication should be noted in the BLA at the bifurcation or the peripheral artery, as in the present case.

**Keywords:** Blister-like aneurysm, Azygos anterior cerebral artery, Endovascular treatment

## INTRODUCTION

Blister-like aneurysms (BLAs) were described for the 1st time in 1986 as rare pathologic entities, accounting for 0.3–1% of all intracranial aneurysms.[7,12] They are small hemispherical bulges lacking a distinct aneurysmal neck with an extremely fragile wall.[18] The typical location of BLAs is the dorsal wall of the internal carotid artery (ICA) at non-branching sites. Several case reports and series have described aneurysms with similar characteristics to BLAs arising in atypical locations, such as the anterior cerebral artery (ACA), middle cerebral artery (MCA), posterior cerebral artery (PCA), and basilar artery (BA).[10,13,15-17] Most BLAs located at atypical sites were in the anterior communicating artery (AcomA). Only two cases of BLAs on the ACA

were reported,[15,17] with no reports of BLAs associated with an azygos ACA. In this report, a rare case of a ruptured BLA arising at the distal bifurcation of an azygos ACA treated by stent-assisted coil embolization is presented.

## CASE DESCRIPTION

A 73-year-old woman presented with subarachnoid hemorrhage (SAH). She was found unconscious at home and transferred to the hospital. She had a history of hypertension and hyperlipidemia. Her consciousness level was Glasgow Coma Scale E3V4M5. The SAH grade was Hunt and Hess Grade 3 and WFNS Grade 4. The thickest hematoma of the SAH was noted in the interhemispheric fissure on computed tomography (CT) [Figure 1a]. CT angiography and digital subtraction angiography (DSA) showed an azygos ACA with hypoplasia of the A1 segment of the right ACA [Figure 1b]. Three-dimensional rotational angiography indicated a small and conical aneurysm measuring 0.7 mm in height, with a 1.2 mm neck located on the bifurcation of the distal azygos trunk. This small aneurysm was arising from the distal end of the azygos trunk to the left pericallosal artery [Figure 1c]. This aneurysm was considered to be ruptured because of the absence of obvious aneurysms on other angiography and the distribution of the SAH in the CT. DSA after waiting for 4 days under sedation showed obvious enlargement of the aneurysm [Figure 1d]. This small aneurysm was diagnosed as a ruptured BLA located on the distal bifurcation of an azygos trunk based on its size, shape, and growth. Direct clipping and endovascular treatment using simple coiling and balloon-assisted coiling seemed to have a high risk for intraoperative rupture due to the BLA. Trapping with an anastomosis was also difficult because the aneurysm was located on the azygos trunk. Therefore, it was decided to perform stent-assisted coiling (SAC) on the same day as the follow-up DSA. Immediately after the follow-up DSA, dual antiplatelet agents were administered in loading doses (aspirin 200 mg and clopidogrel 300 mg) 4 h before the start of the endovascular treatment. First, a 6-Fr FUBUKI guiding sheath (Asahi Intec, Aichi, Japan) was inserted into the right femoral artery and advanced to the left cervical ICA. A Headway 17 microcatheter (Microvention, Aliso Viejo, CA, USA) was navigated over a CHIKAI 0.35-mm (0.014-inch) microwire (Asahi Intec, Aichi, Japan) into the left pericallosal artery with a 3.4-Fr Tactics (Technocrat, Aichi, Japan) placed on the A1 segment of the left ACA as an intermediate catheter [Figure 2a]. An Excelsior SL-10 (Stryker Neurovascular, Kalamazoo, MI, USA) shaped by steam on a 45-degree curve was placed in the proximal part of the aneurysm using a Traxcess 0.30-mm (0.012-inch) microwire (Microvention) because of the difficulty of placing the microcatheter inside the aneurysm. After a 2.5-mm * 17-mm low-profile visualized intraluminal

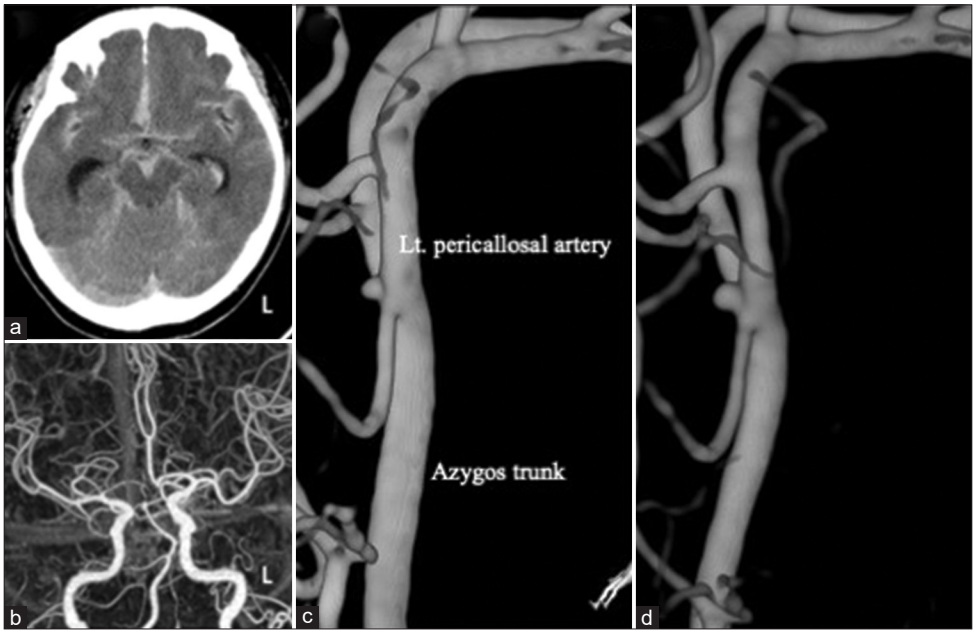


**Figure 1:** (a) Axial computed tomography (CT) on the initial visit shows diffuse subarachnoid hemorrhage distributed mainly at the interhemispheric fissure. (b) CT angiography shows an azygos anterior cerebral artery (ACA) with hypoplasia of the A1 segment of the right ACA. (c) Three-dimensional rotational angiography (3D-RA) with injection from the left internal carotid artery shows a tiny bulge on the distal bifurcation of an ACA on day 0. This small aneurysm rides from the distal end of the azygos trunk to the left pericallosal artery. (d) The follow-up 3D-RA on day 4 shows obvious enlargement of the aneurysm.

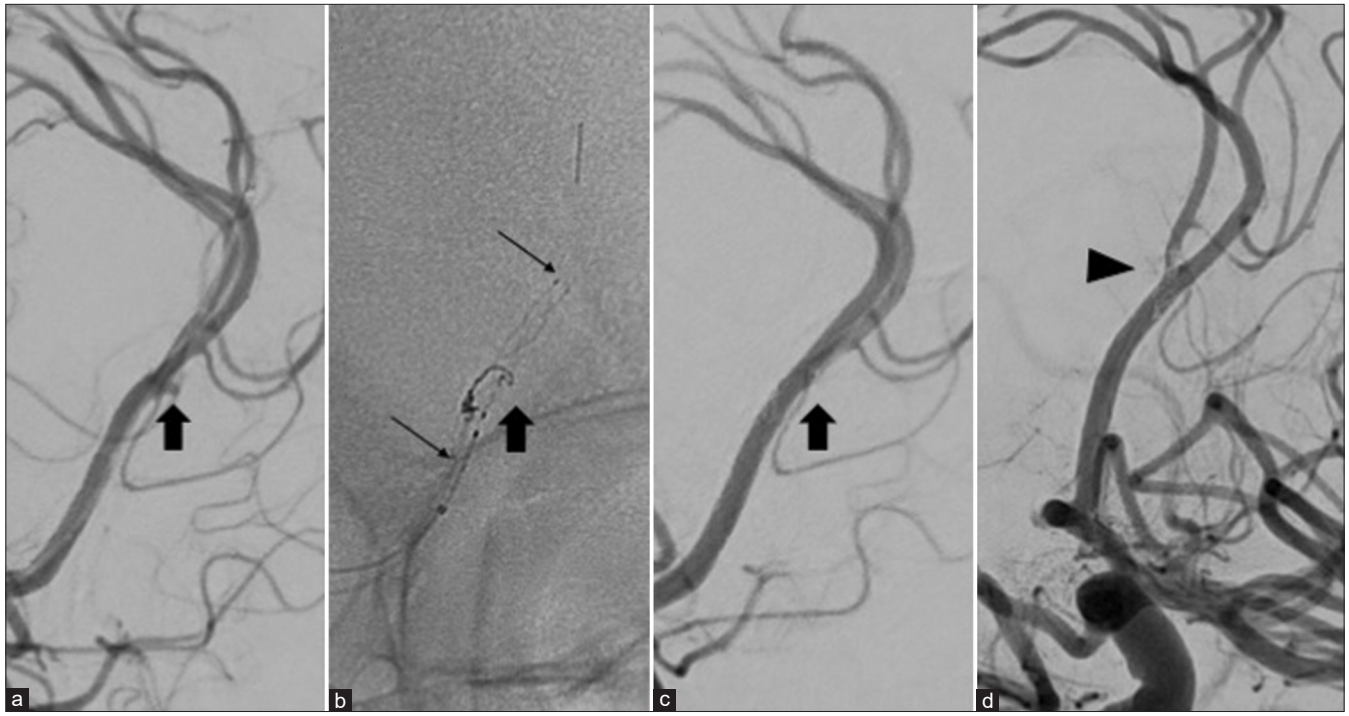


**Figure 2:** Stent-assisted coiling was subsequently performed on the same day as the follow-up digital subtraction angiography (DSA). (a) Left carotid angiography at a working angle shows a blister-like aneurysm (BLA) (black arrow) at the distal bifurcation of the azygos trunk and a Headway 17 microcatheter placed on the left pericallosal artery for stenting. (b) The fluoroscopic image demonstrates two loops of a single GalaxyG3 coil inside the BLA (black arrow) and the implanted LVIS junior stent from the left pericallosal artery to the azygos trunk (long thin black arrow). (c and d) The final DSA shows immediate complete occlusion of the BLA (black arrow) and thrombus at the origin of a right pericallosal artery (black arrowhead).

support (LVIS) Jr (Microvention) was deployed to just the distal neck of the aneurysm, a Galaxy G3 mini 1-mm * 20-mm (Cerenovus, Irvine CA, USA) was poured into the aneurysm. Two loops of the coil were placed inside the aneurysm by the microcatheter placed in the proximal of the aneurysm, which moved gradually with the coil protruding from the tip. The half-deployed stent was placed across the neck of the aneurysm to the end [Figure 2b]. Final postembolization DSA showed that the contrast effect of the aneurysm disappeared [Figure 2c]. Because a thrombus was found at the origin of the right pericallosal artery [Figure 2d], the patient was administered 80 mg of ozagrel sodium, and no blood vessels occluded. No new postoperative neurological deficits were found, and magnetic resonance imaging–diffusion-weighted imaging showed multiple spotty high intensities in the ACA region bilaterally [Figure 3], and magnetic resonance angiography showed no vessel occlusion. Follow-up DSA 14 days after onset showed residual aneurysm with contrast inside the dome. However, the findings showed stagnation of contrast in the venous phase and did not show contrast along the aneurysm wall, so follow-up was continued [Figure 4a]. Follow-up DSA 30 and 90 days after onset showed that the aneurysm thrombosed gradually and reached complete occlusion 90 days later [Figures 4b and c]. The patient had a good outcome, with a 90-day modified Rankin Scale (mRS) score of 2 and mild disturbance of memory as a sequela.

## DISCUSSION

A very rare case of a ruptured BLA at an azygos ACA that was successfully treated with stent-assisted coil embolization and achieved to complete occlusion was reported.

The most frequent typical site of origin of BLAs is the dorsal wall of the ICA at a non-branching site, which was first described as "chimame."[12] Recently, however, BLAs formed at atypical locations have been reported. BLAs often rebleed and recur even within a short period, and the rates of intraoperative and early postoperative reruptures are 33–80% in most surgical studies.[16] Because of the characteristics of BLAs, treatment of these lesions is challenging and risky. Andaluz *et al.* stated that two of five cases of clipping for BLAs located on AcomA developed intraoperative rupture.[1] Hellstern *et al.* reported that a case of BLAs at the MCA treated by clipping resulted in intraoperative rupture and dissection of M1.[6] The walls of the BLAs at atypical sites are also considered too fragile to clip preserving a parent artery, similar to ICA lesions.

Endovascular treatments for BLAs include parent artery occlusion (PAO), coil embolization, SAC, stenting, and flow

diverter (FD). A total of 23 cases of BLAs in atypical locations treated by intervention were found with the details reported; they included PAO in three cases, coiling in one case, SAC in five cases, stenting in seven cases, and FD in eight cases [Table 1].[6,10,13,15-17] Two of the three BLA cases treated with PAO resulted in severe infarction and poor outcomes.[13,15] PAO is the most reliable means to prevent rebleeding, but embolization of the parent artery with no collateral flows or perforators is impossible. A case of a BLA treated by coiling alone was reported to cause postoperative rebleeding,[16] and we thus considered it to be that, in small BLAs with a broad base, it is difficult to place enough coils to prevent blood flow in their dome, as in the present case. Moreover, we considered that the risk of intraoperative rupture is increased if the coils are embolized tightly for the BLA with or without balloon assistance. Since many coils cannot be inserted, we selected SAC in anticipation of the of the flow diversion effect and the effect of blocking the re-entry site.

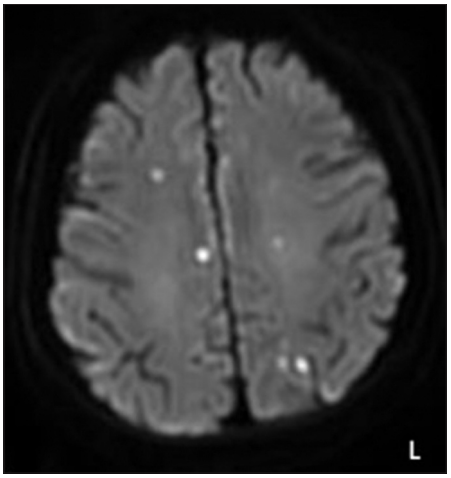


**Figure 3:** Magnetic resonance imaging-diffusion-weighted imaging shows multiple spotty high intensities in the bilateral anterior cerebral artery region.

Other endovascular treatment options, SAC, stenting, and FD, have good clinical and angiographical outcomes without serious perioperative complications related to the procedure, except in one case treated by FD for an MCA BLA that developed a major stroke.[15] Scerrati *et al*. reviewed 32 studies reporting 684 patients (707 BLAs) treated by endovascular approaches (stenting 282, SAC 256, and FD 155 patients).[20] The long-term complete occlusion rate was 76.9% (95% CI, 69.2–83.9%). The perioperative complication rate was 8.9%, and the clinical outcome at final follow-up was a mRS score <2 in 76.6% (95% CI, 68.2–84.2%) of patients. This study suggests that endovascular treatment of BLAs is associated with good long-term occlusion rates and reasonable complication and mortality rates. Moreover, the occlusion rate at the final follow-up was comparable among the different techniques, and there is no consensus on the best endovascular options for BLAs.

Some studies on the treatment of BLAs using a Pipeline Embolization Device (Medtronic, Minneapolis, MN, USA) have shown good clinical and angiographical outcomes.[9] Moreover, Mohlenbruch *et al*. reported the safety and effectiveness of treatment for BLAs with the Flow-Redirection Endoluminal Device (FRED; Microvention).[11] Of the 26 patients with 26 ruptured BLAs located on the ICA, BA, VA, AcomA, and PCA with follow-up, complete obliteration was achieved in 24 aneurysms (92%), and there was no case of rebleeding after FRED implantation. Major stroke occurred in two patients and asymptomatic FRED occlusion occurred in one case. Of the FD stents approved in Japan, FRED is suitable for the proximal MCA or proximal ACA, which can be the parent artery of BLAs in an atypical location. However, the diameter (from 1.8 to 2.2 mm) of the

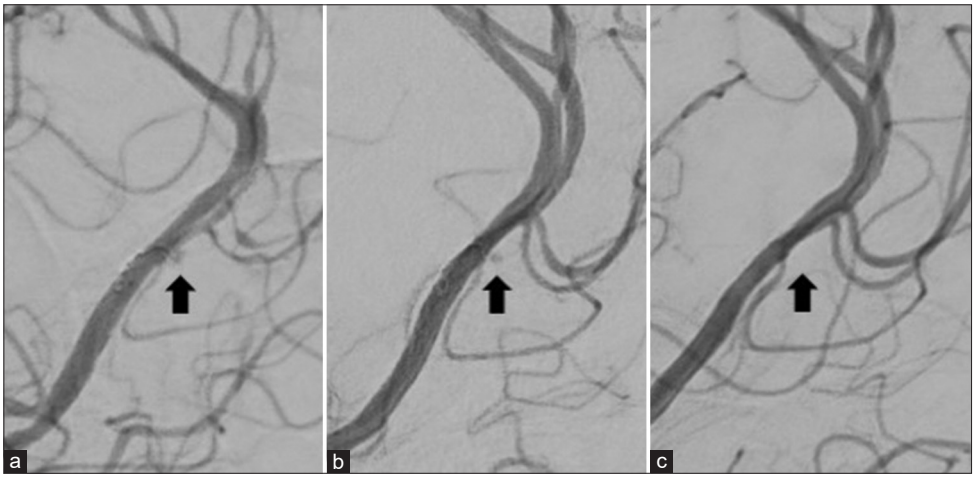


**Figure 4:** The follow-up digital subtraction angiography (DSA). (a and b) The DSA 14 (a) and 30 (b) days after onset shows the residual contrast effect and gradual thrombosis within the dome of a blister-like aneurysm (black arrow in (a) and (b)). (c) The DSA at 90 days shows complete obliteration (black arrow).

**Table 1:** Literature review of BLAs on atypical sites treated by an intervention.

| Author | Case | Location | SAH grade | Endovascular treatment | Device | Perioperative complication | Final follow-up mRS |
|---|---|---|---|---|---|---|---|
| Meckel *et al*., 2011 | 1 | BA | HH 4 | SAC | Unknown | None | 0 |
| | 2 | BA | HH 3 | SAC | (Neroform, Enterprise or Wingspan) | None | 1 |
| Pistocchi *et al*., 2012 | 3 | ACA (A2) | HH 2 | FD | Silk×2 | None | 0 |
| | 4 | MCA (M1) | HH 2 | FD | PED | None | 0 |
| Consoli *et al*., 2012 | 5 | PICA | HH 2 | FD | PED | None | 0 |
| Bulsara *et al*., 2013 | 6 | MCA | HH 1 | Stenting | Neuroform | None | 1 |
| | 7 | PCA | HH 2 | Stenting | Neuroform | None | 1 |
| | 8 | PCA | HH 1 | Stenting | Neuroform | None | 1 |
| Lim *et al*., 2013 | 9 | BA | HH 2 | SAC | Neuroform×3 | None | 0 |
| Rouchaud *et al*., 2013 | 10 | AcomA | HH 4 | FD | PED×2 | None | 2 |
| Peschillo *et al*., 2014 | 11 | MCA (bifurcation) | HH 3 | Coiling | | Rebleeding | 6 |
| Grant *et al*., 2014 | 12 | MCA | HH 1 | Stenting | Neuroform | None | 0 |
| | 13 | PCA | HH 1 | Stenting | Neuroform | None | 0 |
| | 14 | PCA | HH 1 | Stenting | Neuroform | None | 0 |
| Aydin *et al*., 2014 | 15 | BA | HH 2 | FD | Silk | None | 0 |
| Chaloui *et al*., 2014 | 16 | BA | HH 2 | FD | PED | None | 1 |
| Peschillo *et al*., 2015 | 17 | ACA (A1) | HH 2 | PAO (SAC) | p64 | Symptomatic infarction | 0 |
| | 18 | PICA | HH 4 | SAC | Enterprise | Pneumonia | 6 |
| | 19 | MCA (M2) | HH 4 | FD | Silk | Symptomatic infarction, increased ICP | 5 |
| Hellstern *et al*., 2018 | 20 | MCA (M3) | HH4 | PAO | | Symptomatic infarction | 4 |
| | 21 | AcomA | HH3 | FD | p64 | none | 1 |
| Park *et al*., 2020 | 22 | PCA (P1) | HH 4/WFNS 4 | PAO | | Symptomatic infarction related to clipping | 4 |
| Miyashita *et al*., 2021 | 23 | AcomA | WFNS 4 | Stenting | LVIS junior | none | 0 |
| Present Case | 24 | Azygos ACA (bifurcation) | HH 3/WFNS 4 | SAC | LVIS junior | Asymptomatic intraoperative thrombosis | 2 |

SAH: Subarachnoid hemorrhage, ICP: Intracranial pressure, BLAs: Blister-like aneurysms, BA: Basilar artery, ACA: Anterior cerebral artery, MCA: Middle cerebral artery, PCA: Posterior cerebral artery, PICA: Posterior inferior cerebellar artery, AcomA: Anterior communicating artery, HH: Hunt and Hess, WFNS: World Federation of Neurosurgical Society grade, SAC: Stent-assisted coiling, FD: Flow diverter, PAO: Parent artery occlusion, mRS: modified Rankin Scale, PED: Pipeline Embolization Device, LVIS: Low-prpfile visualized intraluminal support

azygos trunk, which was the parent artery in the present case, is too small and peripheral, because the recommended vessel diameters for FRED are from 3.0 to 5.5 mm. Therefore, treatment using the FD may not be encouraged in such cases.

Both stenting alone and SAC seemed to be good treatment options for the present case. The advantage of stenting over SAC is safety, since the risk of rupture is low during coil embolization. In retrospective multicenter study, Fang *et al*. sail that there were 12 (5.7%) perioperative hemorrhagic complications among 212 SAC procedures.[4] Scerrati *et al*. found that intraoperative hemorrhage occurred in 27/684 procedures (1.3%; 95% CU, 0.31–2.85%), and the hemorrhage rate was low in all groups; stenting (0%), SAC (0.61%), and FD (0.63%). The three cases of intraoperative bleeding occurred in only cases of flow-diverter positioning without associated coiling.[20] On the other hand, SAC was reported to have a higher rate of immediate complete occlusion (63.4%) compared with stand-alone stent placement (42%) and FD (53.7%). The same trend was confirmed at radiologic follow-up when SAC reached a rate of complete occlusion of 80.3%, with rates of 77.1% for stand-alone stent placement and 75.6% for FD. Moreover, the perioperative death rate was >2 times higher in the FD group (5.2%) compared with the stand-alone stent placement (2.1%) and SAC (0%) groups.

Since SAC achieved complete occlusion immediately at a higher rate than stenting and FD, it is considered to be beneficial for the treatment of BLAs. In SAC, the coil that fills in the aneurysm appears to rapidly promote thrombosis of the ruptured part of a BLA and facilitate healing of the vessel wall of the parent artery by stent placement. The opportunity to pack the coil into the aneurysm is only at the initial endovascular treatment using the stent, and it may be difficult to add coils in case of aneurysmal growth due to the size and shape of the BLA. The most important thing to avoid during SAC seems to be intraoperative rupture due to coil filling. In the present case, a microcatheter was placed in the proximal side of the aneurysm, and the coil was poured into the dome. Although only two loops of coil fit inside the aneurysm, immediate thrombosis of the dome was confirmed, and tighter coil insertion was avoided to prevent intraoperative rupture. However, a thrombus that did not cause a vessel occlusion and a symptomatic infarction was formed on the branch where the stent was not placed. It was considered to be an intraoperative complication that should be noted, in cases such as the present BLA located on the bifurcation of peripheral blood vessels, unlike a non-branching site of the ICA.

The pathophysiology of BLAs is not well understood. BLAs are said to consist of a fragile fibrin layer in a focal arterial segment, unlike true aneurysms.[18] Some reports indicated that arterial dissection is associated with the formation of BLAs.[3,6] Peschillo *et al*. reviewed BLAs at atypical sites and reported their features: Small size, <3 mm immediately after initial bleeding; located at a non-branching site, proximal to the circle of Willis; tendency to grow during a brief period; tendency to rebleed; and a conical and wide-necked shape.[14] Regarding the sites of BLAs arising, aneurysms of branching sites such as AcomA and MCA bifurcation, which usually cause saccular aneurysms, have been reported and were confirmed by the surgical findings at clipping.[6,16] Since almost all BLAs at atypical sites have been treated by the endovascular treatment in recent reports, they are diagnosed from the morphological finding of their small size, conical and wide-neck shape, and their rapid growth. The present aneurysm showed a maximum diameter of 1.4 mm at the time of initial diagnosis, a hemispherical bulge with a wide neck, and a rapid shape change in a few days. Therefore, the aneurysm was diagnosed as a BLA because of its characteristic features, excluding the branching site, and no cases of BLAs located on the azygos ACA have been reported.

An azygos ACA is a rare anatomical anomaly with an incidence of approximately 1.1%.[19] Baptista listed unpaired (Type 1), bihemispheric (Type 2), and triplicate (Type 3) types of distal ACA variations.[2] The azygos ACA is categorized as a Type 1 anomaly. The frequency of aneurysms associated with an azygos ACA is high, at 41–71%. Most aneurysms on the azygos ACA arise at the distal bifurcation of the parent artery, with a few exceptional cases located at the proximal part.[5] Kaspera *et al*. stated that hemodynamic stress related to the azygos bifurcation geometry with a bent course of the artery around the genu of the corpus callosum predisposes to aneurysm formation, as well as congenital factors such as vascular malformation.[8] Although the azygos ACA aneurysm in the present case arose from the distal end of the azygos trunk to the left pericallosal artery, its location is usual for an azygos ACA aneurysm, but not for a BLA. The hemodynamic vascular stress and congenital factors due to the structure of the azygos ACA may affect the occurrence of ruptured BLAs, but this report alone cannot lead to any conclusions regarding this.

Because BLAs are presumed to be a type of vascular dissection or pseudoaneurysm, their treatment should not focus solely on the aneurysm sac but should also address the reconstruction of the wall of the affected vessel. The present aneurysm was completely occluded by placing a LVIS junior stent and only two loops of a coil in the dome. If this aneurysm was a saccular aneurysm, tighter coil embolization may have been needed to achieve complete occlusion. It was highly likely that the coil outside the aneurysm was sandwiched between the vascular walls by the stent, blocking the re-entry site of the BLA and completely eradicating the BLA. In our view, because the present aneurysm was a BLA, it had been completely occluded by the repairment of the parent artery by the stenting, even though it was not tightly packed by the coil. Based on this finding, the present aneurysm was diagnosed as a BLA at the azygos ACA.

## CONCLUSION

This is the first report of a ruptured BLA arising on the bifurcation of the azygos trunk treated by SAC. The present case showed good clinical and radiological outcomes. SAC might be an effective treatment for BLAs leading to early complete occlusion, but thrombus formation as an intraoperative complication should be noted in BLAs at the bifurcation or a peripheral artery, such as the present case.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

### Conflicts of interest

There are no conflicts of interest.

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