

# One-stage Stent-assisted Coil Embolization for Rupture-side-unknown Bilateral Vertebral Artery Dissecting Aneurysms in an Acute Stage: A Case Report

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Bilateral vertebral artery dissecting aneurysms (VADAs) with subarachnoid hemorrhage (SAH) are rare and their management is still challenging. In this report, we successfully performed one-stage stent-assisted coil embolization (SAC) for bilateral VADAs with SAH in an acute stage, because the ruptured side could not be diagnosed. A 47-year-old woman presented with a sudden onset of headache without laterality, and left-side dominant SAH with bilateral VADAs was noted on computed tomography (CT) scans. The size of aneurysmal dome and neck was similar between the two VADAs, and a bleb was observed only on the right VADA. In computational fluid dynamics (CFD) simulations, findings of wall shear stress (WSS), normalized WSS, and WSS gradient suggested that the left VADA was ruptured, while the oscillatory shear index and aneurysm formation indicator suggested the opposite-side one to be ruptured. Thus, we could not determine which VADA was ruptured by clinical data and CFD analyses. Therefore, we performed simultaneous treatment for the bilateral VADAs by using SAC technique 8 h after the onset under dual antiplatelet and anticoagulation therapies. There was no evidence of rebleeding and stent thrombosis. Stent thrombosis was monitored by duplex color-coded ultrasonography after the intervention. She was discharged without neurological deficits, and 6-month follow-up cerebral angiography demonstrated no recanalization of VADAs. This is the first report showing bilateral VADAs with SAH treated by one-stage SAC within 24 h of SAH, and the potential risks are discussed.

**Keywords:** bilateral vertebral artery dissecting aneurysms, coil embolization, computational fluid dynamics, stent, subarachnoid hemorrhage

## Introduction

Bilateral vertebral artery dissecting aneurysms (VADAs) with subarachnoid hemorrhage (SAH) are rare and their

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Received: May 9, 2017; Accepted: July 31, 2017  
Online March 9, 2018

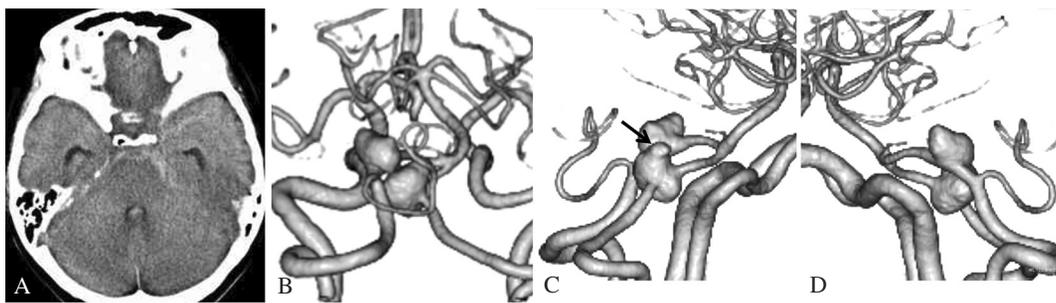
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treatment remains challenging. Redekop et al.<sup>1)</sup> reported successful staged bilateral vertebral artery (VA) occlusion for such lesions. For cases with bilateral VADAs with SAH that VA obliteration is impossible, Kono et al.<sup>2)</sup> and Zhao et al.<sup>3)</sup> reported successful treatment with stent-assisted coil embolization (SAC) from 2 days to 15 days after the onset. However, Mizutani et al.<sup>4)</sup> reported that about 40% of ruptured VADAs re-ruptured within 24 h after the onset if untreated. Thus, surgical or endovascular interventions should be promptly performed for ruptured VADAs, while there is no report describing one-stage SAC for bilateral VADAs with SAH within 24 h after the onset. Here, we present a case of bilateral VADAs with SAH, whose rupture side could not be determined but was successfully treated by one-stage SAC 8 h after the onset.

## Case Report

Appropriate informed consent was obtained from the patient and her relatives. A 47-year-old woman presented with severe headache without laterality. Computed tomography (CT) scans showed SAH dominant on the left side (Fig. 1A). The World Federation of Neurosurgical Societies grade was 2. CT angiography showed bilateral VADAs, whose size was similar (Figs. 1B–1D). A bleb was observed only on the right-side VADA. The right posterior inferior cerebellar artery (PICA) was not visible, and the left VADA was located distal to the PICA origin. The size of bilateral VAs was similar. There were no posterior communicating arteries visible on both sides, and the carotids and anterior circulation showed no abnormality. Since the ruptured side was suggested to be the left VADA by the distribution of SAH while the right VADA by the existence of a bleb, we performed computational fluid dynamics (CFD) simulations.

CFD analysis was performed as previously reported.<sup>5)</sup> From the simulated flow fields, we calculated wall shear stress (WSS) and the following WSS-related hemodynamic parameters as previously reported:<sup>6–11)</sup> normalized WSS (NWSS), oscillatory shear index (OSI), aneurysm formation indicator (AFI) and WSS gradient (WSSG). The morphological and hemodynamic parameters were summarized in Table 1. According to our previous study,<sup>6)</sup> lower WSS, NWSS and WSSG suggested that the left VADA was ruptured, but higher OSI and lower AFI suggested the right VADA to be ruptured.



**Fig. 1** Computed tomography scans (A) and 3D CT angiography (B, C, D) at admission. (A) CT scan shows left-side dominant subarachnoid hemorrhage. (B) CT angiography postero-anterior view; (C) right lateral view; (D) left lateral view shows bilateral vertebral artery dissecting aneurysms (VADAs) in the V4 segment. A bleb is observed only on the right-side VADA (arrow).

**Table 1** Summary of morphological and hemodynamic parameters of the bilateral vertebral artery dissecting aneurysms

Parameter	Right	Left
Dome size (mm)	14.0	14.0
WSS (Pa)	1.51	1.22
NWSS	0.445	0.366
OSI	0.0284	0.0267
AFI	0.977	0.988
WSSG (Pa/mm)	1.54	0.998

AFI: aneurysm formation index, NWSS: normalized WSS, OSI: oscillatory shear index, WSS: wall shear stress, WSSG: WSS gradient.

Since the ruptured side could not be determined even using CFD analyses, the bilateral VADAs were simultaneously treated using SAC technique 8 h after the onset on the admission day. The approval from the institutional ethical committee and written informed consent for off-label use of intracranial stents were obtained prior to the procedure.

Stent-assisted coil embolization was performed via a femoral artery approach under general anesthesia. Immediately before SAC, 300 mg clopidogrel and 100 mg aspirin were orally administered. A bolus of 3,000-unit heparin was intravenously injected, followed by continuous injection of 1,000-unit/h heparin during the procedure, under control of activated clotting time (approximately 250 s). First, SAC was performed for the right VADA with a bleb. A self-expanding stent (Enterprise 4.5 mm × 28 mm; Cordis Neurovascular, Inc, Miami, Florida, USA) was placed across the VADA, and soft coils were deployed into the sac. Then, the left VADA was obliterated in the same fashion (Fig. 2).

Argatroban was intravenously administered for 1 week. Oral intake of 75 mg/day clopidogrel and 100 mg/day aspirin were continued. No findings suspected of rebleeding from VADAs or other hemorrhagic complications were observed during the course of hospitalization, and follow-up MR images at 3 and 7 days post-SAC and digital subtraction angiograms at 15 days post-SAC (Fig. 3) showed no recanalization of the VADAs. Stent thrombosis was monitored by duplex color-coded ultrasonography from the day to 13 days post-SAC: peak systolic velocity, end-diastolic velocity, and mean velocity were stable between 35.5 and 84.0 cm/s, 12.2

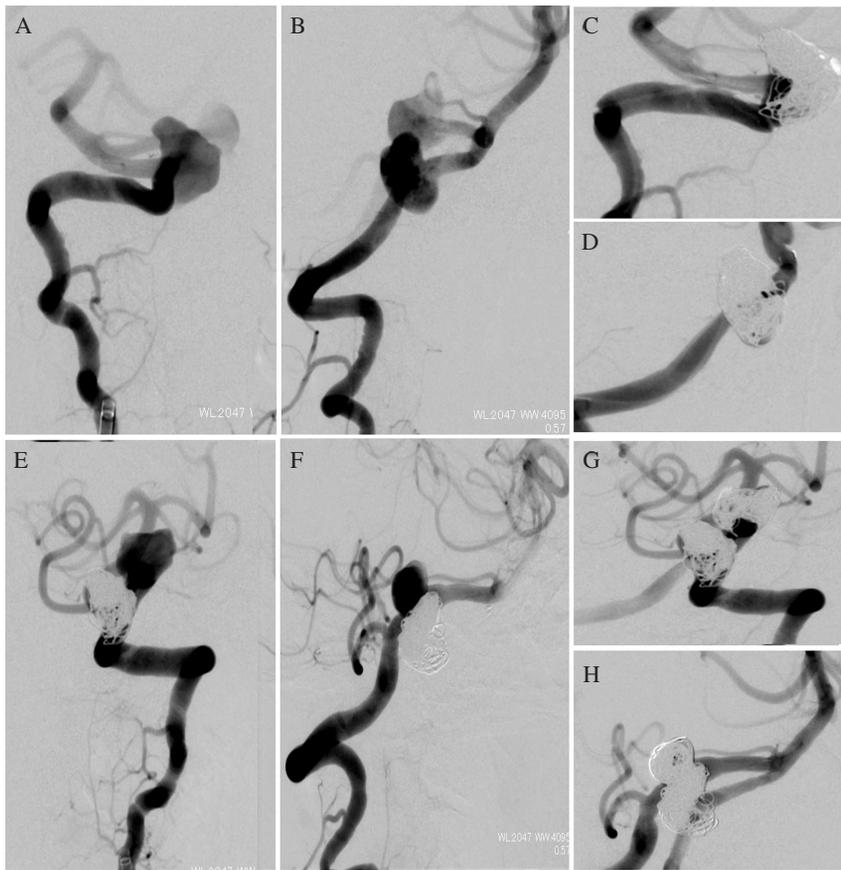
and 28.0 cm/s, and 21.9 and 49.4 cm/s, respectively, indicating no stent occlusion. Neither cerebral vasospasm nor hydrocephalus was developed, but a transient left facial palsy occurred associated with MR abnormalities possibly by an inflammatory reaction at 3 days post-SAC (Fig. 4). Her symptom and MR findings improved at 17 days post-SAC. She was discharged with modified Rankin Scale 1. Aspirin was stopped 4 months post-SAC and clopidogrel was continued. Six-month follow-up angiogram demonstrated no recanalization of the VADAs.

## Discussion

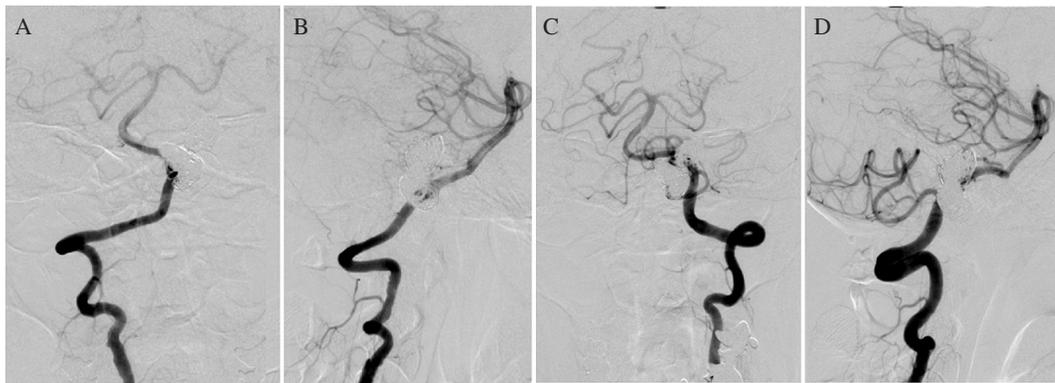
The natural history of ruptured VADAs is devastating. In a 42-patient series, Mizutani et al.<sup>4)</sup> found a 69% incidence of re-rupture before VADA obliteration, of which 56.7% occurred within 24 h, and 80% within 7 days. Thus, surgical or endovascular interventions should be promptly performed for ruptured VADAs. However, there is no report describing one-stage SAC for bilateral VADAs with SAH within 24 h after the onset. Here, we presented a case of bilateral VADAs with SAH, of which the ruptured side could not be determined, and were successfully treated by one-stage SAC 8 h after the onset.

Bilateral VADAs presenting with SAH are rare, and the treatment remains more challenging. A variety of therapeutic approaches have been reported including endovascular or surgical trapping of a ruptured VADA with the parent VA.<sup>12–14)</sup> However, parent artery obliteration of the ruptured side increases hemodynamic stress, causing a rupture of the contralateral unruptured VADA at 2 days to 5 months after the treatment.<sup>12–14)</sup> Therefore, early therapeutic intervention for the contralateral unruptured VADA should be considered after VA obliteration of the ruptured side.

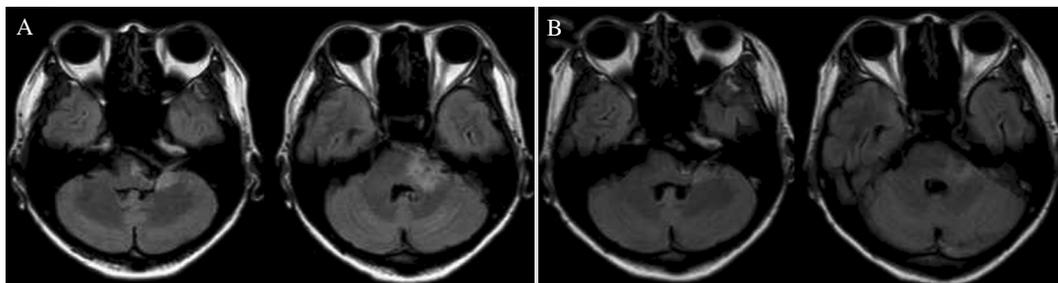
In bilateral VADAs, one of which is ruptured but unidentified, one possible treatment option is VA to VA bypass with interposed radial artery.<sup>15)</sup> However, the procedure is not easy and associated with a high risk. Another option is SAC that preserves VA blood flow. Wilkinson et al.<sup>16)</sup> reported a case in which bilateral VADAs with SAH were treated successfully by staged SAC: first, a ruptured VADA was treated and later the contralateral unruptured VADA was treated. Zhao et al.<sup>3)</sup> reported two cases of bilateral VADAs that were successfully treated by single-stage SAC from 2 to 15 days



**Fig. 2** Vertebral artery angiogram (VAG) before and just after stent-assisted coil embolization (SAC) for the bilateral vertebral artery dissecting aneurysms (VADAs). SAC was first performed for the right VADA (A–D), and then for the left VADA (E–H). Frontal view (A) and lateral view (B) of right VAG before SAC; frontal view (C) and lateral view (D) of right VAG after SAC of the right VADA; frontal view (E) and lateral view (F) of left VAG after SAC of the right VADA but before SAC of the left VADA; frontal view (G) and lateral view (H) of left VAG after SAC of the left VADA showing coil occlusion of the VADAs and patency of the parent arteries.



**Fig. 3** Follow-up vertebral artery angiogram (VAG) 15 days after stent-assisted coil embolization for the bilateral vertebral artery dissecting aneurysms (VADAs). Recanalization of the bilateral VADAs is not shown. Frontal view (A) and lateral view (B) of right VAG; frontal view (C) and lateral view (D) of left VAG.



**Fig. 4** Fluid attenuation inversion recovery MR images after stent-assisted coil embolization (SAC) for the bilateral vertebral artery dissecting aneurysms (VADAs). A high intense area is shown in the brain-stem and cerebellum in contact with the left VADA at 3 days (A) and is diminished at 17 days post-SAC (B).

after SAH because the site of rupture could not be identified. When a rupture-side VADA cannot be identified like our case, one-stage SAC for both VADAs may be a reasonable strategy.

However, it is important to remind that SAC for ruptured VADAs has some risks. First, since it requires anticoagulation or antiplatelet therapy to prevent stent-related thromboembolisms, hemorrhagic complications may occur. The incidence of post-SAC intracranial hemorrhagic complications was reportedly 6.9–8% for ruptured saccular aneurysms,<sup>17–19</sup> but standardized anticoagulation or antiplatelet therapies have never been established. In this case, we administered antiplatelets like for unruptured cases, and tried gentler catheterization and coiling into the aneurysms under strict control of blood pressure, resulting in no aneurysmal re-rupture. Second, during the postoperative period, patients with ruptured aneurysms are in a hypercoagulate state with hyperactive platelets due to not only surgical procedure, but also SAH itself. Reportedly, significant post-SAC thromboembolic events occurred in 5.3–6.9% of patients with ruptured saccular aneurysms.<sup>17–19</sup> In this case, stent thrombosis was daily evaluated by the algorithm using duplex color-coded ultrasonography.<sup>20</sup> As a result, we experienced no thromboembolic complications during the post-intervention period. However, a rigorous monitoring of platelet function is needed to prevent thromboembolic events and therefore platelet aggregation tests should be performed prior to the procedure of SAC if possible, although it was impossible for this case in our hospital at that time.

To determine the ruptured side of bilateral VADAs with SAH, the following determinants were reported: the side of headache,<sup>21</sup> hemiplegia,<sup>13</sup> thicker hematoma on CT,<sup>12,14</sup> larger and more irregular aneurysm,<sup>12,13</sup> pearl and string sign,<sup>13</sup> pooling of contrast media in a pseudoaneurysm on angiography,<sup>12,14</sup> and intramural hematoma in MR images.<sup>1</sup> More recently, CFD analysis is reported to be potentially useful to identify a ruptured saccular aneurysm, but the findings are conflicting.<sup>22–26</sup> Our previous study demonstrated that low WSS, low NWSS, high OSI, low AFI and low WSSG were associated with a ruptured state of middle cerebral artery aneurysms.<sup>6</sup> In this case, however, we failed to determine the ruptured side of VADAs even using clinical, morphological and CFD findings, although future CFD studies may reveal characteristic hemodynamic features of a rupture status of VADAs.

## Conclusion

This is the first to report that one-stage SAC is feasible for bilateral VADAs with SAH within 24 h after the onset. One-stage SAC may be reasonable for bilateral VADAs, in which the ruptured side cannot be determined.

## Conflicts of Interest Disclosure

All authors have no conflicts of interest.

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