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Case Report

A Case of Brainstem Infarction That Was Found to Be Vertebral Artery Dissection in a Short Period after the Diagnosis of Atherothrombotic Infarction

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Objective: We report a case of vertebral artery dissecting aneurysm that caused right lateral medullary infarction, which was treated by endovascular therapy.

Case Presentations: A 57-year-old man developed right-side headache and dysarthria on the day before presentation, and exhibited mouth dropping and dysphagia the following day. Initial MRI demonstrated right lateral medullary infarction with atherothrombotic change with no vessel lesion, and we started infusion and medication administration. Later MRI revealed bilateral vertebral artery dissection, and we treated the growing right vertebral artery dissecting aneurysm by stenting and coils.

Conclusion: The possibility of dissecting lesions should be considered in cases of medullary infarction. Stenting and coil treatment is a useful option for bilateral dissecting vertebral aneurysms.

Keywords ▶ brain stem infarction, vertebral artery dissecting aneurysm, stent and coil

Introduction

The course of vertebral artery dissection varies: the exacerbation of ischemia and rupture of aneurysms; therefore, the treatment strategy is controversial. In this study, we report a patient in whom ischemic symptoms developed but no abnormality in the main cerebral artery was initially found; however, the gradual change in vascular morphology suggested bilateral vertebral artery dissection, and stent-assisted coil embolization of an enlarging right dissecting vertebral artery aneurysm was performed. We also present a literature review.

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Case Presentation

In a 57-year-old male, right temporal headache and dysarthria were noted the day before consultation. Mouth dropping and dysphagia developed the following day, and he consulted our hospital.

On consultation, consciousness was clear and there was no paralysis of the limbs. However, he complained of mild sensory disturbance. In addition, right mouth dropping, dysarthria, slight hoarseness, and thermal hypoalgesia of the left half body involving the cervical or lower regions were observed.

MRI-diffusion-weighted imaging (DWI) revealed a high-signal-intensity area on the lateral side of the right medulla oblongata, but MRA did not demonstrate stenosis or dissection of the main artery (**Fig. 1**). A diagnosis of right medullary infarction related to ischemia of the penetrating vessel area was made. The administration of ozagrel sodium and edaravone was started. As oral drug swallowing became possible, the oral administration of clopidogrel at 75 mg was started on Day 3.

On Day 12, no increase in the infarcted focus size was observed on MRI, but MRA and contrast-enhanced CT

Fig. 1 MRI on the initial consultation. (**A**) DWI revealed an infarcted focus on the lateral side of the right medulla oblongata (solid arrow). (**B**) MRA showed no wall irregularity in the left or right vertebral arteries. DWI: diffusion-weighted imaging

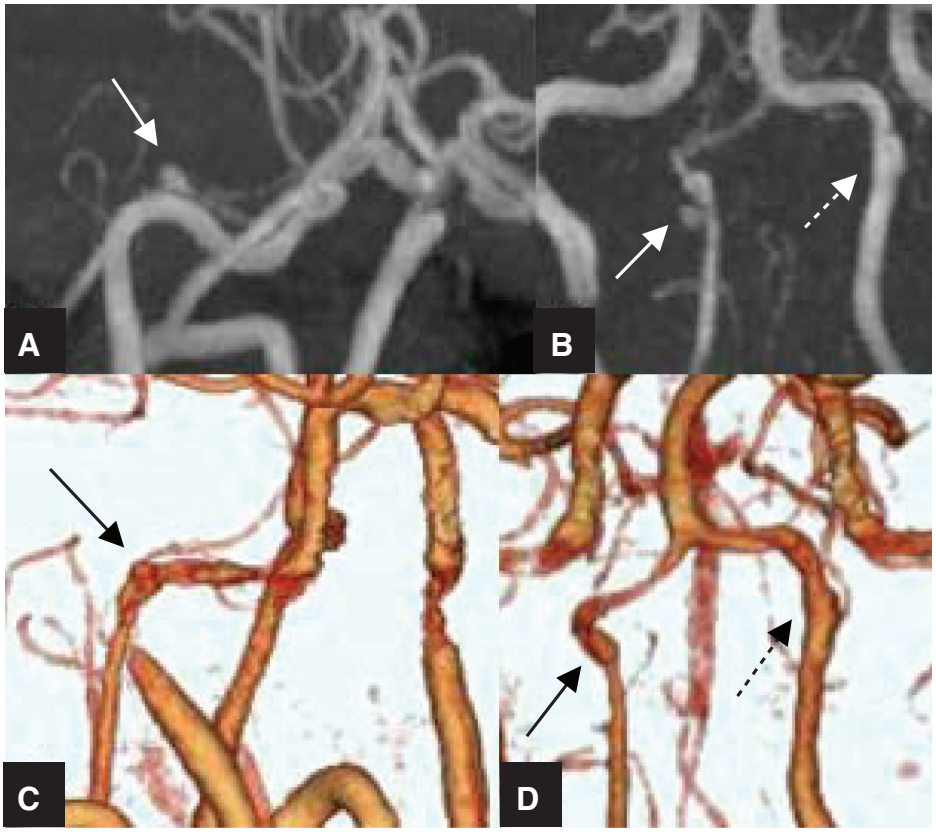

Fig. 2 MRI (**A** and **B**) and CT (**C** and **D**) on Day 12. (**A** and **C**) Lateral view showing a dissecting right vertebral artery aneurysm (3 × 4 mm) and narrowing (pearl and string sign) (solid arrows). (**B** and **D**) Frontal view showing a dissecting right vertebral artery aneurysm (solid arrows) and wall irregularity in the left vertebral artery (dotted arrows).

revealed an aneurysm-like deformity (approximately 3 × 4 mm) and narrowing of the right vertebral artery, in addition to wall irregularity in the left vertebral artery (**Fig. 2**). Based on these findings, the patient was diagnosed with bilateral vertebral artery dissection and dissecting vertebral artery aneurysms, but not with atherothrombotic change-related

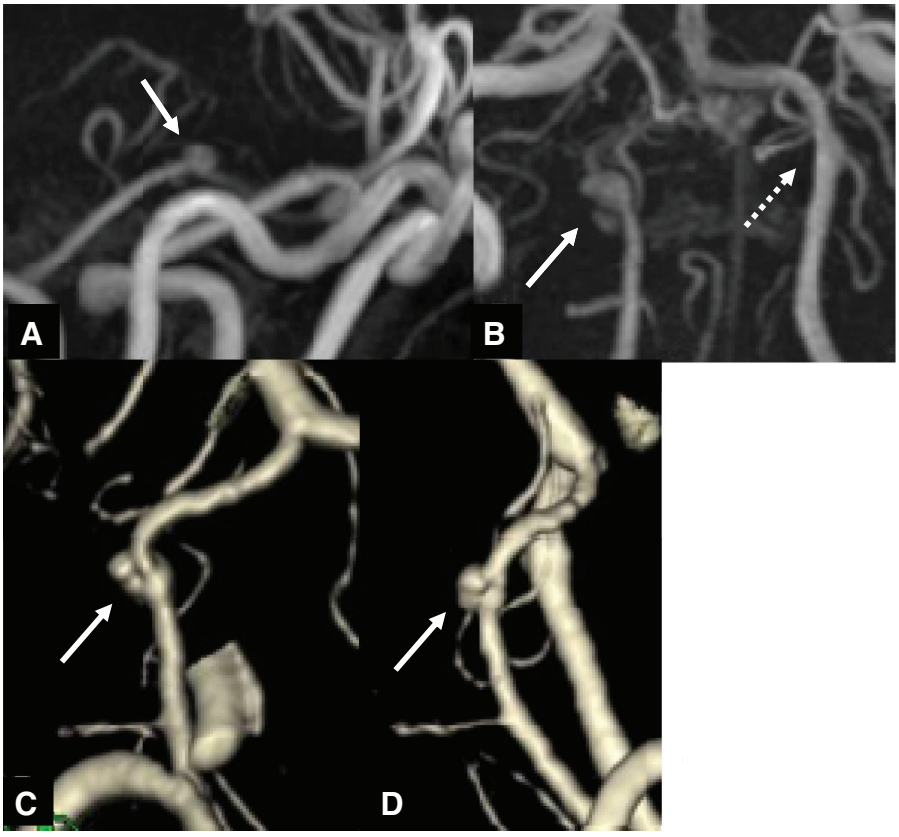

Fig. 3 MRI (**A** and **B**) and CT (**C** and **D**) on Day 19. (**A**) The size of the dissecting right vertebral artery aneurysm had increased to 3.5 × 4.5 mm (solid arrow). (**B**) Enlargement of the dissecting right vertebral artery aneurysm (solid arrow) was noted, whereas there was no change in wall irregularity in the left vertebral artery (dotted arrow). (**C** and **D**) Bleb formation was observed in the dissecting right vertebral artery aneurysm site (solid arrow).

ischemia. Right vertebral artery dissection was considered to be a preceding, responsible lesion for the following reasons: right temporal headache was noted the day before consultation and wall irregularity in the right vertebral artery was more marked. At the site of dissection, aneurysm formation was observed, and the oral administration of the antiplatelet drug was promptly discontinued considering the risks of deterioration of dissection/subarachnoid hemorrhage. A strategy to strictly examine the presence of morphological changes in the aneurysms under blood pressure control was implemented.

On Day 19, an increase in the right dissecting vertebral artery aneurysm size (approximately 3.5 × 4.5 mm) was observed on MRI, but there was no change in the left dissecting vertebral artery aneurysm. The right vertebral artery aneurysm had rapidly increased in size in the 3 weeks after onset. In addition, bleb formation was noted. Considering the risk of hemorrhage, surgical treatment was selected. Concerning the left vertebral artery aneurysm, there was no aneurysmal deformity during the course and follow-up was continued.

Contrast-enhanced CT and DSA revealed that the right posterior inferior cerebellar artery (PICA) had branched from an area proximal to the dissecting vertebral artery aneurysm, and that a sufficient distance was maintained (**Figs. 3 and 4**). Embolization of the aneurysm was considered to be safe, and endovascular treatment was selected. In addition, we adopted stent-assisted coil embolization, but not internal trapping, considering the possibility of the contralateral aneurysm needing to be blocked when contralateral vertebral artery dissection progressed or its area increased. On Day 25, this procedure was performed. To prevent stenting-related thrombosis, aspirin at 300 mg and clopidogrel at 300 mg were infused through a gastric tube after general anesthesia was induced. Right femoral artery puncture was conducted, and a 6 Fr Roadmaster (Goodman, Aichi, Japan) was guided into the right vertebral artery. An LVIS Jr 3.5 × 23 mm (Terumo, Tokyo, Japan) was deployed through a Headway 17 (MicroVention, Aliso Viejo, CA, USA) such that its distal and proximal ends reached an area before the union and beyond the PICA,

Fig. 4 Right vertebral arteriography on Day 22. (**A** and **B**) The PICA (dotted arrows) was observed at an area proximal to the aneurysm (solid arrows) with a sufficient distance. PICA: posterior inferior cerebellar artery

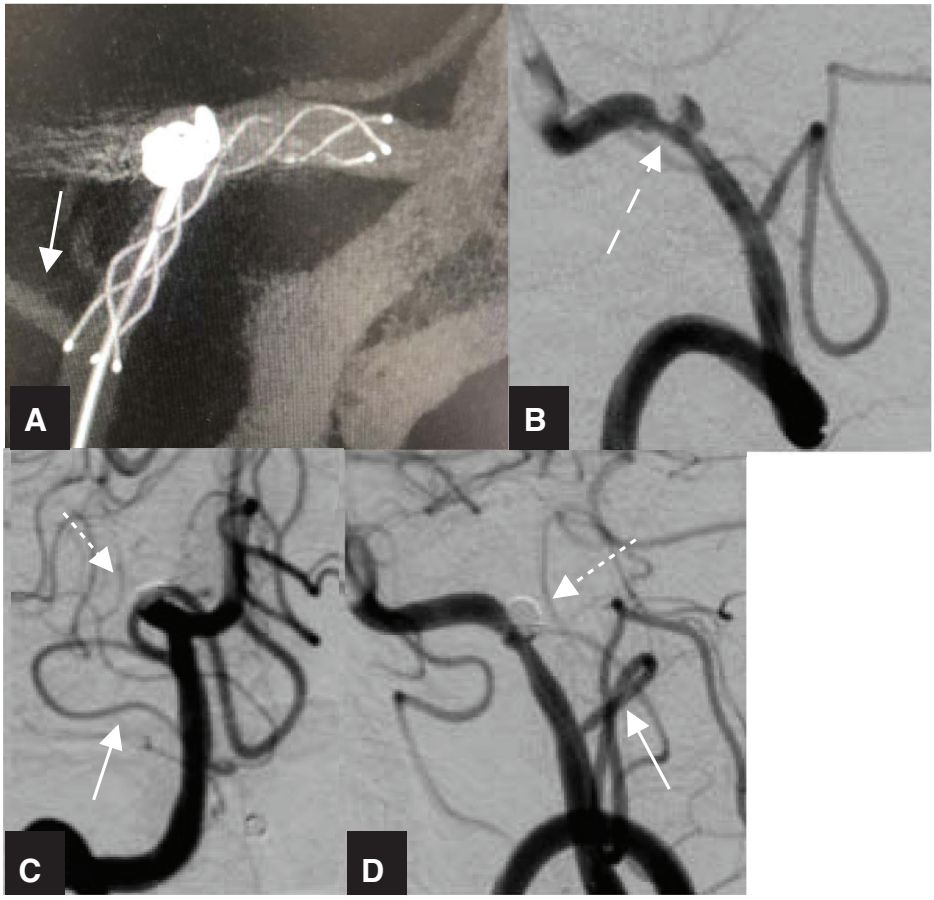

Fig. 5 DSA at the time of treatment on Day 25. (**A**) An LVIS Jr 3.5 × 23 mm was deployed such that its distal and proximal ends reached an area before the union and beyond the origin of the PICA (solid arrow), respectively. (**B**) Immediately after stenting, there was a delay in intra-aneurysmal blood flow (dashed arrow). (**C** and **D**) Immediately after coil embolization. There was no intra-aneurysmal blood flow (dotted arrows) and the PICA remained (solid arrows). PICA: posterior inferior cerebellar artery

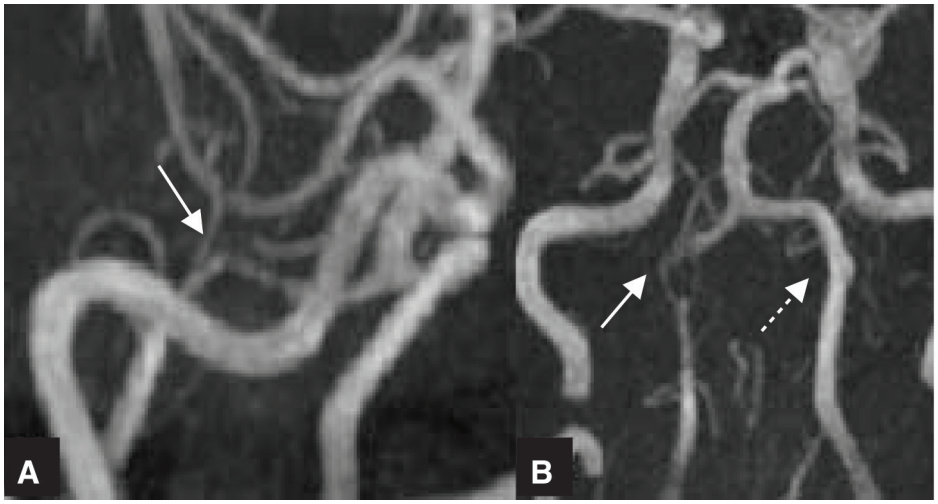

Fig. 6 MRI on Day 26. (**A**) The dissecting right vertebral artery aneurysm was not visualized (solid arrow). (**B**) The dissecting right vertebral artery aneurysm was not visualized (solid arrow). There was no progression of wall irregularity in the left vertebral artery (dotted arrow).

respectively. After stent deployment, angiography confirmed a reduction in intra-aneurysmal blood flow related to the flow diversion effects of the stent. Two Target 360 Ultra 3 × 6 (Stryker, Kalamazoo, MI, USA) were used as framing coils. As a filling coil, a HydroSoft 3D 2.5 × 6 (Terumo) was used. As finishing coils, Target 360 Nano 2.5 × 4, 2.5 ×4, and 2.5 × 3 (Stryker) were used. A total of six coils were inserted to completely occlude the aneurysm. After embolization, PICA blood flow was maintained well (**Fig. 5**). MRI on the following day demonstrated no fresh infarcted focus (**Fig. 6**). Although sensory disturbance remained, the patient was referred to a rehabilitation hospital on Day 43 with a modified Rankin Scale score of 2. After discharge, no recurrent aneurysm was observed on MRI at the outpatient clinic 3 months after the start of treatment. Furthermore, the shape of the left vertebral artery, in which wall irregularity was initially observed, was normalized.

Discussion

In Japan in the 1990s, Yamaura et al.[1] reported that intracranial artery dissection is frequent in the vertebral–basilar system and that patients with hemorrhage accounted for >50%. However, in 2011, Mizutani[2] investigated 93 patients with unruptured dissecting vertebral artery aneurysms, with a mean follow-up of 3.44 years, and reported that morphological changes were observed in 83.9% patients, whereas recovery to a normal morphology was achieved in 18.3% patients. They also noted that only one of these patients developed subarachnoid hemorrhage, emphasizing that the risk of hemorrhage is low in unruptured aneurysm patients. In addition, according to a retrospective study of a multicenter, nationwide survey involving 149 institutions (632 patients) regarding nontraumatic intracranial artery dissection in 2013,[3] the types of onset consisted of hemorrhage (subarachnoid hemorrhage) in 193 patients (30.5%), ischemia (cerebral infarction/transient ischemic attack) in 209 patients (33.1%), and headache (no hemorrhage/ischemia on the initial consultation) in 230 patients (36.4%). When focusing on the ischemia group, to which the present patient may be assigned, the median age was 50 years and there were 164 males (78.5%); the rate of males was significantly higher. Such epidemiological information may be related to the possibility that lesions are incidentally detected by recent diagnostic imaging techniques such as MRI. However, considering that a specific number of patients with arterial dissection are included among those initially diagnosed with atherothrombotic cerebral infarction, as demonstrated in the present case, the incidence of vertebral artery dissection in ischemia- or headache-onset patients may further increase.

On the other hand, to our knowledge, no case report in which abnormalities in the vascular wall were absent on MRI on the initial consultation and subsequent imaging suggested abnormality in the vascular wall, as demonstrated in the present case, has been published. Therefore, the specific rate of patients with brainstem infarction may include those with vertebral/basilar artery dissection. Follow-up using imaging procedures, such as 3D -CTA and MRI, in consideration of this possibility may be necessary.

Furthermore, bilateral vertebral artery dissection, as observed in the present case, is not rare. The mechanism of bilateral dissection is hypothesized as follows: 1) dissection simultaneously develops on the bilateral sides, 2) dissection of the unilateral vertebral artery retrogradely progresses to the contralateral side through the vertebrobasilar junction, and 3) after dissection of the unilateral vertebral artery causes stenosis/occlusion, hemodynamic stress on the contralateral side, which is essentially normal, induces dissection.[4] In the present case, there were no marked morphological changes in the left or right vertebral arteries when ischemia of the right vertebral artery region developed. However, MRI 12 days after admission revealed aneurysm-like changes in the bilateral vertebral arteries; based on the time course, a lesion in the right vertebral artery may have preceded these changes. Based on the above 3) hypothesis, right vertebral artery dissection-related change in hemodynamics may have caused dissection of the left vertebral artery. In such patients with bilateral vertebral artery dissection, it may be necessary to examine the condition while considering treatment options that are available for further lesion progression/enlargement.

Concerning treatment, conservative treatment involving blood pressure control and follow-up imaging is primarily performed for ischemia-onset-type patients, but a consensus regarding treatment using antithrombotic therapy has not been reached. However, antithrombotic therapy cannot be recommended for patients with aneurysm formation,[5] such as our patient; endovascular treatment should be considered.

When selecting therapeutic strategies, the positional relationship between the aneurysm and PICA is important. Regarding distal-PICA-type aneurysms, as demonstrated in the present case, proximal occlusion or trapping under craniotomy has been conducted, but internal trapping to occlude a parent blood vessel by endovascular treatment is currently routinely performed based on the results of a previous study,[6] the efficacy of this procedure.

However, these treatments are difficult in the absence of ischemic tolerance. Stenting was reported to be useful when it was necessary to preserve a parent blood vessel. Treatment using a stent alone utilizing its flow diversion effects and stent-assisted coil embolization, as conducted in the present case, have been reported. Park et al.[7] performed treatment using a stent alone on 29 patients with dissecting vertebrobasilar aneurysms, consisting of 11 patients with hemorrhage and 18 patients without hemorrhage, and reported that there were no treatment-associated complications, suggesting the safety of this treatment. In addition, results of treatment with several stents were more favorable than those of treatment with a single stent. Furthermore, Isaka et al.[8] reported favorable long-term results based on the results of the 10-year follow-up of two patients who had undergone treatment with stenting alone for nonhemorrhagic dissecting vertebral artery aneurysms. On the other hand, stent-related dilation of fragile stenotic lesions may induce subarachnoid hemorrhage in patients with arterial dissection, considering the mechanism of this disorder. However, Miyamoto et al.[9] suggested that stenting involving false-cavity entry and reentry can prevent the progression of dissection, leading to the disappearance of aneurysms through repair of the dissected wall, a change in intra-aneurysmal flow dynamics, and the promotion of neointima outgrowth. The merits of combining coil embolization with stenting have been reported; one study found that intra-aneurysmal coil promoted early thrombosis,[10] and another study found that the use of a coil prevented posttreatment aneurysmal rupture in flow diverter-adopted patients.[11] Thus, in our patient with a gradually enlarging aneurysm despite ischemia onset, stent-assisted coil embolization may have been appropriate to promote aneurysmal thrombosis earlier and prevent a fatal course. In Japan, stents for supporting coil embolization of unruptured cerebral aneurysms, such as the Neuroform Atlas (Stryker), Enterprise (Codman & Shurtleff, John & Johnson, Raynham, MA, USA), and LVIS/LVIS Jr. (Terumo), have been approved. The purpose of their development was to prevent coil deviation into a parent blood vessel on embolization of aneurysms, but their flow diversion effects have also been noted. Wang et al.[12] analyzed computational fluid dynamics in 3 patients and reported that the flow diversion effects of an LVIS stent were more marked than those of other stents. In the present case, an LVIS stent was selected to reduce vascular wall hemodynamic stress.

Furthermore, in the present case, MRI 4 months after right vertebral artery treatment confirmed normalization of the left vertebral artery. As the mechanism, early repair of right vertebral artery dissection through the flow diversion effects of the stent inserted into the right vertebral artery may have led to a reduction in contralateral hemodynamic stress.

In addition, recently, treatment using a flow diverter has been emphasized such that more potent flow diversion effects in comparison with those of stents and aneurysmal thrombosis may be achieved. Yeung et al.[13] performed treatment using a Pipeline alone on 4 patients with unruptured dissecting vertebral artery aneurysms and reported

favorable results in the absence of complications or recurrence. According to their report, examination 2 years after surgery confirmed repair of the vascular wall. Furthermore, Narata et al.[14] overlapped 3 Pipelines in 2 patients with hemorrhagic dissecting vertebral artery aneurysms, leading to radical cure. Treatment using a flow diverter for dissecting vertebral artery aneurysms may be a new treatment option, but the number of patients is small. In the future, the results of treatment and long-term outcomes should be examined.

Conclusion

Even in patients with brainstem infarction in whom imaging on the initial consultation reveals no abnormality in the vascular morphology, such as ours, several sessions of imaging must be performed considering the presence of a latent dissecting lesion. Furthermore, stent-assisted coil embolization may be a useful treatment option for bilateral dissecting vertebral artery aneurysms.

Disclosure Statement

We declare no conflicts of interest during the past 3 years.

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