Spontaneous Resolution of Nontraumatic Bilateral Intracranial Vertebral Artery Dissections

—Case Report—

Atsushi FUKUNAGA, Masanao TABUSE, Heiji NARITAKA,
Tsuneo NAKAMURA, and Takenori AKIYAMA*

Department of Neurosurgery, Saiseikai Kanagawaken Hospital, Yokohama;

**Department of Neurosurgery, Keio University School of Medicine, Tokyo*

Abstract

A 49-year-old man presented with nontraumatic bilateral intracranial vertebral artery dissections without subarachnoid hemorrhage manifesting as Wallenberg's syndrome on the right. Magnetic resonance imaging revealed an infarct in the right dorsolateral aspect of the medulla oblongata. Antiplatelet therapy was administered. Vertebral angiography performed on the 9th hospital day (Day 9) revealed pearl and string sign in the right vertebral artery and narrowing of the left vertebral artery. Second angiography performed on Day 25 showed no change, but third angiography performed on Day 74 revealed spontaneous resolution of the bilateral vertebral artery dissections. Magnetic resonance angiography performed on Day 250 showed no evidence of dissection. However, magnetic resonance imaging revealed a small infarct in the splenium of the corpus callosum. Spontaneous resolution of stenotic dissections of the bilateral vertebral arteries is extremely unusual. Serial cerebral angiography and magnetic resonance angiography are very important for monitoring the time course of changes in patients with vertebral artery dissections.

Key words: resolution, bilateral vertebral arteries, dissection

Introduction

The most common angiographic findings in cases of dissecting aneurysms are fusiform dilatation, pearl and string sign, and narrowing. Serial angiography also reveals the time course of changes in cases of dissecting aneurysms; as gradual disappearance of the narrowing and irregularity, or progressive enlargement of the dilatation.¹⁴⁾ The mechanism and natural history of arterial dissection are still unclear, and the treatment also remains controversial. Use of antiplatelet or anticoagulant therapy as a nonsurgical method of treatment is one of the issues currently under debate. Here, we report a rare case of nontraumatic bilateral intracranial vertebral artery (VA) dissections, in which spontaneous resolution was observed following antiplatelet therapy.

Case Report

A 49-year-old man presented with a history of intermittent mild headache around the right orbital region since March 18, 2001. He suddenly developed numbness over the right side of the face and severe headache on March 26, 2001. He visited the Department of Internal Medicine at our hospital. Computed tomography of the head showed no abnormal findings, but the patient was immediately admitted to our hospital for further examination. Neither the family history nor his past history was remarkable. The patient was a smoker (smoking index = 400) and a habitual drinker (alcohol 100 g/day).

On admission, he was fully conscious and alert. His blood pressure was 154/110 mmHg. Neurological examination revealed diplopia, nystagmus, Horner's sign on the right, right facial numbness, poor corneal reflex on the right, hoarseness of voice,

Received January 8, 2002; Accepted July 26, 2002

Author's present address: A. Fukunaga, M.D., Department of Neurosurgery, Keio University School of Medicine, Tokyo, Japan.

Fig. 1 Axial T₂-weighted magnetic resonance image performed on March 29 (Day 3) demonstrating a high intensity area at the right dorsolateral part of the medulla oblongata (arrowhead).

Fig. 2 Right vertebral angiograms performed on April 4 (Day 9) showing pearl and string sign in the intracranial vertebral artery (arrowheads). A: anteroposterior view, B: lateral view.

Fig. 3 Left vertebral angiograms performed on April 4 (Day 9) showing stenotic change of the intracranial vertebral artery (arrowhead). A: anteroposterior view, B: lateral view.

Fig. 4 Third vertebral angiograms performed on June 8 (Day 74) demonstrating the diameter of the bilateral vertebral arteries had normalized (arrowhead). A: right anteroposterior view, B: left anteroposterior view.

swallowing disturbance, right ataxia, and disturbance of temperature and pain sensation on the left side of the body. Based on these findings, the diagnosis was Wallenberg's syndrome on the right. Blood examination revealed no abnormal findings. Magnetic resonance (MR) imaging performed on March 29 revealed an infarct in the right dorsolateral aspect of the medulla oblongata (Fig. 1), and MR angiography revealed poor visualization of the right VA and narrowing of the left VA.

On March 30, the patient was referred to our department. Antihypertensive treatment, bed rest, and antiplatelet drug (ozagrel sodium 160 mg/day) therapy were administered for 2 weeks. Cerebral angiography performed on April 4 (Day 9) showed

pearl and string sign in the right VA (Fig. 2) and narrowing of the left VA (Fig. 3). The patient's symptoms gradually improved and aspirin (81 mg/day) administration was started. Second cerebral angiography performed on April 20 (Day 25) showed no change, but third angiography on June 8 (Day 74) revealed dilation of the bilateral VAs to almost the original diameters (Fig. 4). MR angiography performed on December 1 (Day 250) at our outpatient clinic revealed no sign of dissection (Fig. 5), but MR imaging revealed a small infarct in the splenium of the corpus callosum (Fig. 6) that had not been detected on the MR images obtained earlier (Day 3). As of May 2002, the patient was back at work and doing well, except for slight dizziness when driving and mild disturbance of pain and temperature sensation

Fig. 5 Magnetic resonance angiogram performed on December 1 (Day 250) showing no signs of dissection of the bilateral vertebral arteries.

Fig. 6 Axial T₂-weighted magnetic resonance image performed on December 1 (Day 250) demonstrating a small high intensity area at the splenium of the corpus callosum (arrowhead).

in the left lower extremity.

Discussion

Intracranial dissection of the VA presenting without subarachnoid hemorrhage (SAH) is considered to be a frequent cause of Wallenberg's syndrome.^{7,14} Seventy-three (78%) of 93 patients with Wallenberg's syndrome had VA dissection.⁷ Therefore, a patient with Wallenberg's syndrome must undergo cerebral angiography or MR angiography for the exclusion of VA dissection.

The natural history of VA dissection remains unclear. Basilar impression, afibrinogenemia, a rare coagulation disorder, or cystic medial necrosis might be related to the development of bilateral VA dissections.^{3,4,6} Only eight cases of spontaneous bilateral intracranial VA dissections without SAH have been described.^{5,6,9,11-13,16} According to a nationwide survey in Japan, 14 (9.3%) of 151 patients with nontraumatic intracranial artery dissections presenting without hemorrhage had bilateral VA dissections.¹⁸ However, this may reflect the finding that VA dissection accounts for about 80% of all such cases in Japan, whereas internal carotid artery dissection accounts for 41% of cases in other countries.¹⁸

Intracranial arterial dissections of the posterior cerebral circulation are considered to be dangerous and life-threatening because of the risk of SAH. However, some case reports have revealed that conservative treatment of these dissections may result

in successful resolution.^{6,8,12,14} Thus, the question of whether intracranial dissections of the posterior circulation should be treated surgically or nonsurgically remains unresolved. Extracranial saphenous vein bypass was recommended in patients with bilateral carotid artery or VA disease.⁹ On the other hand, six consecutive patients with VA dissection presenting without SAH were treated only with antihypertensive drugs (to keep the blood pressure at a normotensive level) and bed rest for several weeks, indicating that such patients may be treated nonsurgically with careful angiographic follow-up or monitoring.⁸ If nonsurgical treatment is chosen for VA dissection without SAH, the use of antiplatelet or anticoagulant drugs still remains controversial. They consider that dissection is essentially a hemorrhagic disease, so intracranial dissection should be treated without antiplatelet or anticoagulant drugs.⁸ Mortality in the group treated with antiplatelet or anticoagulant therapy was 7.0% (3/43), whereas mortality in the group not receiving these treatments was only 3.3% (2/60), although the difference was not statistically significant ($p = 0.35$, Fisher's exact probability method).¹⁹ However, some authors contend that anticoagulants or antiplatelet agents may be beneficial in patients with VA dissections presenting without SAH.^{6,12,14}

In our case, spontaneous resolution of the bilateral VA dissections occurred during antiplatelet therapy. A previous case of bilateral VA dissections associated with Wallenberg's syndrome showed

spontaneous resolution of the aneurysmal dilation of the left VA and the pearl and string sign of the right VA during antiplatelet therapy.¹⁴⁾ However, the exact contribution of the antiplatelet therapy to the spontaneous resolution of these dissecting aneurysms remains unclear, and there have been no large controlled trials of such agents in the treatment of VA dissections.²⁾ The risks and benefits of these therapies are still unknown and require further investigation. A review of 24 (51%) of 47 cases of dissection without SAH showed resolution during follow-up angiography,¹⁴⁾ and five (29%) of 17 cases with unruptured dissecting aneurysms showed spontaneous resolution by serial angiography between Days 114 to 340.¹⁰⁾ Therefore, spontaneous resolution of unilateral dissecting aneurysm is not so rare. However, this case of spontaneous resolution of bilateral stenotic dissecting lesions is extremely unusual. The mechanism of the spontaneous resolution has not yet been clarified. Possibly, the hematoma between the internal elastic lamina and the media may be absorbed, followed by fibrous change of the vascular wall during the healing process,¹⁾ or the hematoma is washed out through the torn intima. In the present case, MR imaging (Day 250) revealed a small infarct in the splenium of the corpus callosum, fortunately without associated clinical symptoms, which was probably caused by a microthrombus from the pseudolumen of the VA dissections during the healing process, because MR imaging on Day 3 did not reveal any infarct in the corpus callosum.

Recurrent arterial dissection was found in five (4.2%) of 118 patients without SAH in a nationwide Japanese survey.¹⁷⁾ Recurrent arterial dissection developed in 16 (8.0%) of 200 patients with spontaneous cervical artery dissections, but the same vessel was not involved in any of the reported cases of recurrence, possibly because of the healing process of the connective tissue.¹⁵⁾ Younger patients were found to be at a greater risk of recurrent dissection. Therefore, we considered that long-term use of antiplatelet agents would be useful for protection against potential ischemic brain damage due to possible recurrent dissection during the rest of our patient's life. We also consider that long-term follow up by MR angiography in the outpatient clinic is very important.

References

- 1) Caplan LR, Baquis GD, Pessin MS, D'Alton J, Adelman LS, DeWitt LD, Ho K, Izukawa D, Kwan ES: Dissection of the intracranial vertebral artery. *Neurology* 38: 868-877, 1988
- 2) Chang AJ, Mylonakis E, Karanasias P, De Orchis DF, Gold R: Spontaneous bilateral vertebral artery dissections: case report and literature review. *Mayo Clin Proc* 74: 893-896, 1999
- 3) Chang CM, Ng HK, Leung SY, Fong KY, Yu YL: Fatal bilateral vertebral artery dissection in a patient with cystic medial necrosis. *Clin Neurol Neurosurg* 93: 309-311, 1991
- 4) Dickinson LD, Tuite GF, Colon GP, Papadopoulos SM: Vertebral artery dissection related to basilar impression: case report. *Neurosurgery* 36: 835-838, 1995
- 5) Felber S, Auer A, Schmidauer C, Waldenberger P, Aichner F: [Magnetic resonance angiography and magnetic resonance tomography in dissection of the vertebral artery]. *Radiologe* 36: 872-883, 1996 (Ger, with Eng abstract)
- 6) Garcia-Monco JC, Fernandez Canton G, Gomez Beldarrain M: Bilateral vertebral artery dissection in a patient with afibrinogenemia. *Stroke* 27: 2325-2327, 1996
- 7) Hosoya T, Nagahata M, Yamaguchi K: Prevalence of vertebral artery dissection in Wallenberg syndrome: neuroradiological analysis of 93 patients in the Tohoku District, Japan. *Radiat Med* 14: 241-246, 1996
- 8) Kitanaka C, Tanaki J, Kuwahara M, Teraoka A, Sasaki T, Takakura K: Nonsurgical treatment of unruptured intracranial vertebral artery dissection with serial follow-up angiography. *J Neurosurg* 80: 667-674, 1994
- 9) Morgan MK, Sekhon LH: Extracranial-intracranial saphenous vein bypass for carotid or vertebral artery dissections: a report of six cases. *J Neurosurg* 80: 237-246, 1994
- 10) Nakagawa K, Touho H, Morisako T, Osaka Y, Tatsuzawa K, Nakae H, Owada K, Matsuda K, Karasawa J: Long-term follow-up study of unruptured vertebral artery dissection: clinical outcomes and serial angiographic findings. *J Neurosurg* 93: 19-25, 2000
- 11) Plaza I, Diez-Tejedor E, Lara M, Barreiro P: [Spontaneous dissection of the vertebral artery]. *Rev Neurol* 24: 163-171, 1996 (Spa, with Eng abstract)
- 12) Pozzati E, Andreoli A, Limoni P, Casmiro M: Dissecting aneurysms of the vertebrobasilar system: study of 16 cases. *Surg Neurol* 41: 119-124, 1994
- 13) Rees JH, Valentine AR, Llewelyn JG: Spontaneous bilateral carotid and vertebral artery dissection presenting as a Collet-Sicard syndrome. *Br J Radiol* 70: 856-858, 1997
- 14) Sasayama T, Korosue K: [Spontaneous resolution of the unruptured dissecting aneurysms of bilateral vertebral arteries with conservative treatment: analysis of the transient enlargement of aneurysmal dilatation]. *No Shinkei Geka* 28: 47-51, 2000 (Jpn, with Eng abstract)
- 15) Schievink WI, Mokri B, O'Fallon WM: Recurrent spontaneous cervical-artery dissection. *N Engl J Med* 330: 393-397, 1994

- 16) Soper JR, Parker GD, Hallinan JM: Vertebral artery dissection diagnosed with CT. *AJNR Am J Neuroradiol* 16: 952-954, 1995
 - 17) Yamaura A, Ono J, Hirai S: Clinical picture of intracranial non-traumatic dissecting aneurysm. *Neuropathology* 20: 85-90, 2000
 - 18) Yamaura A, Yoshimoto T, Hashimoto N, Ono J: [Nationwide study of nontraumatic intracranial arterial dissection: Clinical features and outcome]. *Surgery for Cerebral Stroke* 26: 79-86, 1998 (Jpn, with Eng abstract)
 - 19) Yamaura A, Yoshimoto T, Hashimoto N, Ono J: [Nationwide study of nontraumatic intracranial arterial dissection: Treatment and its results]. *Surgery for Cerebral Stroke* 26: 87-95, 1998 (Jpn, with Eng abstract)
-
- Address reprint requests to: A. Fukunaga, M.D., Department of Neurosurgery, Keio University School of Medicine, 35 Shinano-machi, Shinjuku-ku, Tokyo 160-8582, Japan.
e-mail: fukunaga@sc.itc.keio.ac.jp.