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Embolic Cerebellar Infarction Caused by Spontaneous Dissection of the Extracranial Vertebral Artery

—Two Case Reports—

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Abstract

Spontaneous dissection of the extracranial vertebral artery (VA) may cause ischemic stroke in the posterior circulation. A 22-year-old female and a 38-year-old male presented with sudden onset of vertigo and nausea without trauma. Angiography was initially interpreted as normal, but retrospective examination disclosed extracranial VA dissection in the V_3 segment in both cases. Arterial dissection resulting in embolic stroke in the territory of the ipsilateral posterior inferior cerebellar artery was highly suspected. Both patients were treated conservatively without sequelae. Careful angiographic interpretation is important for the diagnosis of extracranial VA dissection. Spontaneous extracranial VA dissection should be suspected in young patients presenting with ischemic stroke but without predisposing risk factors or associated trauma.

Key words: angiography, cerebellar infarction, embolic stroke, vertebral artery dissection

Introduction

Vertebral artery (VA) dissection can occur in *extracranial* and *intracranial* locations. Intracranial VA dissection is increasingly recognized as a cause of subarachnoid hemorrhage and ischemic stroke in the posterior circulation. Extracranial VA dissection is not uncommon, and occurs predominantly in the young or middle-aged adults,[2-4,7-9,11,13] but is still poorly understood. We treated two patients with spontaneous extracranial VA dissection resulting in embolic cerebellar infarction in the territory of the ipsilateral posterior inferior cerebellar artery (PICA), and discuss the importance of careful interpretation of the angiograms for the evaluation of extracranial VA dissection.

Case Presentation

Case 1: A 22-year-old female suddenly developed vertigo, nausea, and vomiting. These symptoms subsided gradually over 2 days, but 1 week later she developed bilateral otalgia and occipitalgia followed by vertigo and nausea. The patient presented about 2 weeks after the initial symptoms. Neurological examination found no abnormalities. Her past and family history were not contributory.

Magnetic resonance (MR) imaging showed bilateral inferior cerebellar infarctions (right < left), possibly due to an embolic mechanism based on the multiplicity of infarcts (Fig. 1A). Laboratory data including hematological tests were all within normal limits. Electrocardiography and echocardiography were normal. Digital subtraction angiography (DSA) 1.5 months after the ictus was initially interpreted as normal (Fig. 1B, C). The left VA originated directly from the aorta and entered the transverse foramen of the fifth cervical vertebra. However, retrospective analysis of the initial DSA revealed dilative change of the V_3 segment and stagnant flow in this segment. The left PICA, which had dual origins from the left VA, was supplied by both antegrade flow from the left VA and retrograde flow from the right VA. The dilative change of the V_3 segment did not extend to the origin of the proximal leg of the PICA.

The patient was treated with low dose aspirin for half a year, which was then discontinued. She had no recurrence of ischemic episodes for 2 years. Fol-

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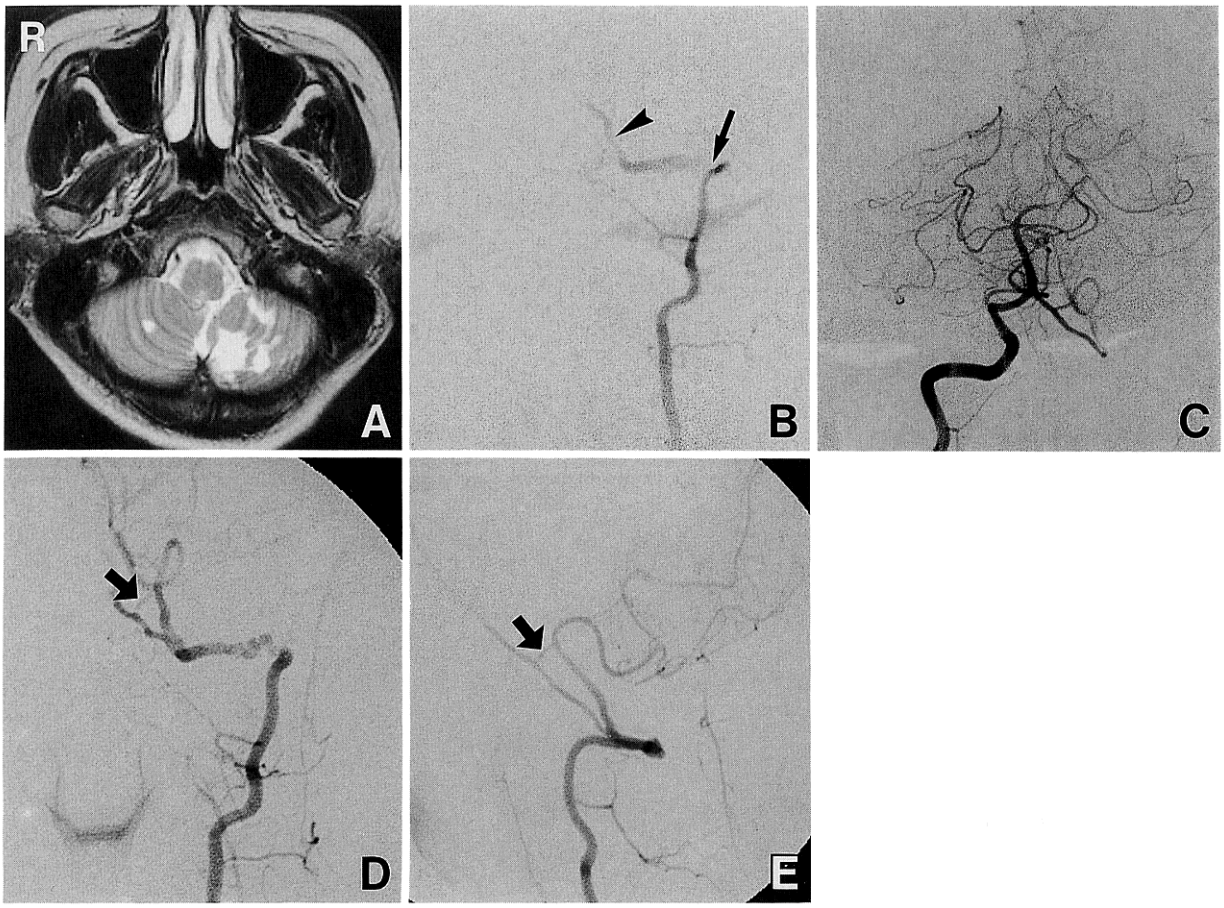

Fig. 1 Case 1. Magnetic resonance image taken 1.5 months after ictus (A) showing bilateral inferior cerebellar infarctions (left > right). Left vertebral angiogram at the same time (B: frontal view) showing dilative change in the V_3 segment (*arrow*) of the left extracranial vertebral artery and proximal leg (*arrowhead*) of the dual origin of the left posterior inferior cerebellar artery (PICA). Right vertebral angiogram at the same time (C: frontal view) showing retrograde filling of the dual origins of the PICA. Follow-up angiograms 2 years later (D: frontal view, E: lateral view) showing wall irregularity in the same V_3 segment and the dual origins of the left PICA. *Thick arrow* indicates the distal leg of the dual origin of the PICA.

low-up DSA at 2 years after ictus revealed persistent wall irregularity at the V_3 segment of the left VA (Fig. 1D, E).

Case 2: A 38-year-old male suddenly developed vertigo, nausea, and gait disturbance. He was admitted to a local hospital 2 days later. MR imaging showed bilateral inferior cerebellar infarctions (left > right) (Fig. 2A). DSA on day 3 revealed incomplete occlusion of the left PICA with capillary blush suggestive of partial recanalization. The left PICA supplied the bilateral inferior portions of the cerebellum (Fig. 2B, C). The right PICA was not observed (Fig. 2D). The left VA was initially interpreted as normal in that hospital. The patient was transferred to us 1 month later. Neurological examination found no abnormalities. His past and family history were not contributory.

Laboratory data including hematological tests were all within normal limits. Cardiac examinations were also normal. Repeat DSA on day 38 after the ictus was normal with recanalization of the left PICA (Fig. 2E, F). Retrospective analysis of the initial DSA revealed stenotic change at the V_3 segment, which had resolved on the follow-up DSA.

The patient was treated with warfarin for half a year, which was then discontinued. He had no recurrent stroke during the 11-month follow-up period.

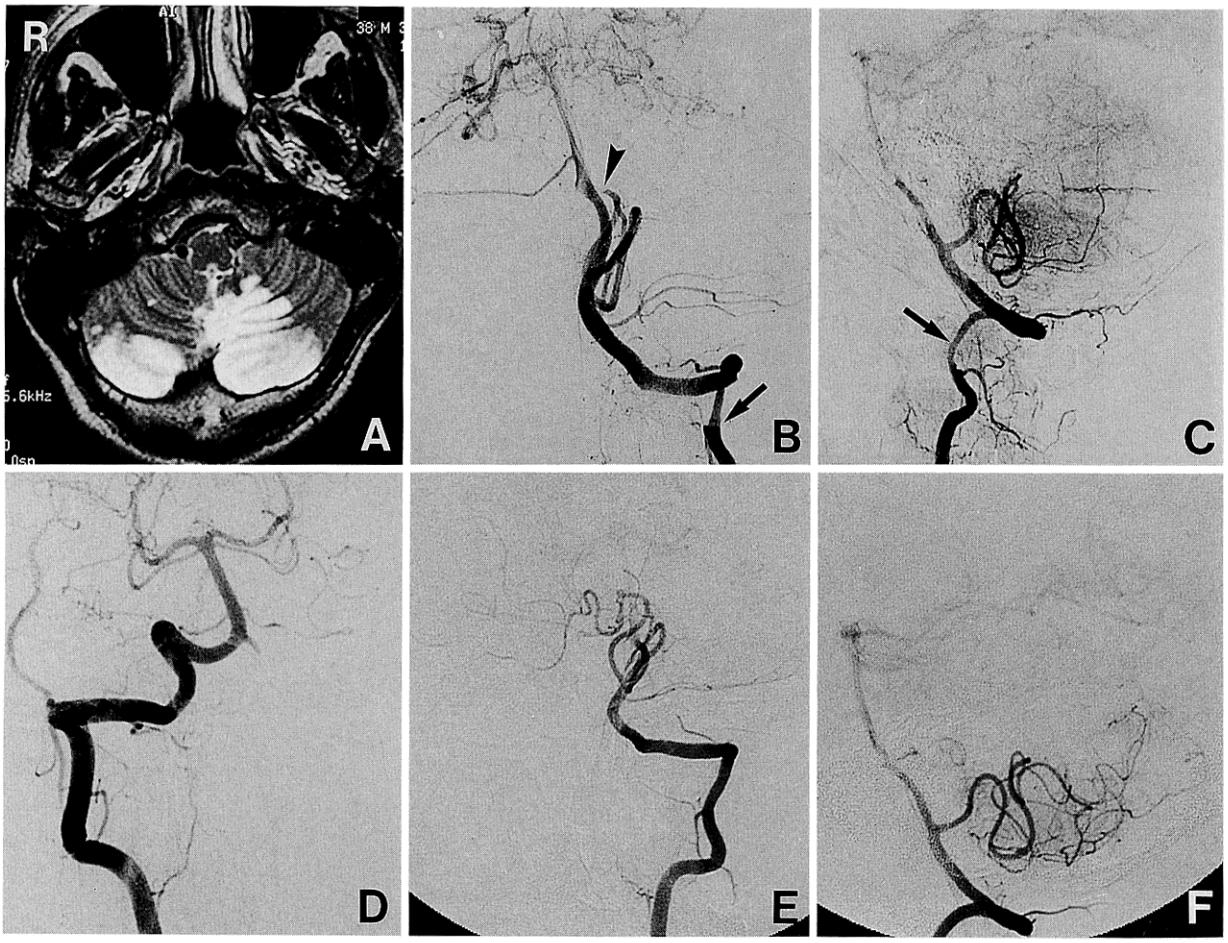

Fig. 2 Case 2. Magnetic resonance image on day 2 (A) showing bilateral inferior cerebellar infarctions (left > right). Left vertebral angiograms on day 3 (B: frontal view, C: lateral view) showing stenotic change in the V_3 segment of the left vertebral artery (*arrow*), and capillary blush and occlusion of the left posterior inferior cerebellar artery (PICA) (*arrowhead*). Right vertebral angiogram on the same day (D: frontal view) showing no apparent PICA on the right side. Follow-up angiograms on day 38 (E: frontal view, F: lateral view) showing resolution of the stenosis in the V_3 segment and recanalization of the left PICA.

Discussion

Extracranial VA dissection can be classified into spontaneous and traumatic causes based on the obvious presence of trauma history. Underlying etiologies of spontaneous VA dissection include minor trauma, hypertension, syphilis, fibromuscular dysplasia, migraine, oral contraceptives, and pregnancy.[2,4,8,9,11,14] In particular, minor trauma due to unusual neck torsion is emphasized.[7] Our two patients had no predisposing risk factors including history of minor trauma.

Infarction associated with VA dissection can result from development of intraluminal thrombi or arterial occlusion due to intramural hematoma. Ischemic stroke due to intracranial VA dissection is generally considered to be caused by direct occlusion of the VA, PICA, or their perforating arteries.[5] Distal embolism from extracranial VA dissection is less frequently reported. We highly suspected the embolic mechanism in our two cases. Extracranial VA dissection causing distal embolism is difficult to diagnose *in the acute stage* when there are not enough laboratory or cardiac data, and when serial angiographic information is not available, as in the present two cases.

Intracranial VA dissection occurs predominantly in the VA segment immediately distal to the origin of the PICA. VA dissection involving the PICA origin or VA dissection in the segment proximal to the PICA origin is less common.[5,10,12] Spontaneous extracranial VA dissection is less frequent than in-

tracranial VA dissection. Cerebellar infarction was caused by intracranial VA dissection in eight cases and extracranial VA dissection in two.[1] Irregular stenosis usually began in the V_3 at the exit from the transverse foramen of the axis and commonly extended to the horizontal portion during the course over the posterior arch of the atlas.[3] Extracranial VA dissection is rare in the V_1 portion,[11] but VA dissection occurred in the V_1 segments in two cases, the V_1 and V_2 segments in two, the V_2 segments in three, the V_2 and V_3 segments in 10, the V_3 segments in two, and the V_3–V_4 segments in one.[4] In our two cases, arterial dissection occurred in the V_3 segments, located in the C-1 space (atlanto-occipital space) and in the C-2 space (atlanto-axial space).

The angiographic appearance of VA dissection includes stenosis, occlusion, dilation of the VA, double lumen, and retention of the contrast material. Extracranial VA dissection appears as either stenosis or occlusion in most cases because the extracranial VA is enclosed in an inextensible osseous canal except for the V_1 and V_3 segments.[4] Double lumen sign has long been believed to be a definite diagnostic sign of VA dissection, but is rarely observed because there is usually no blood flow in the pseudolumen.

Vascular abnormalities associated with subarachnoid hemorrhage are not so difficult to identify on angiograms, because of the extensive knowledge of the etiologies of subarachnoid hemorrhage. However, ischemic stroke caused by VA dissection is not so easy to evaluate. Theoretically, extracranial VA dissection should not cause subarachnoid hemorrhage unless the dissection extends to the intracranial VA. Patients with ischemic stroke due to VA dissection are younger than those with atherosclerotic stroke.[7] When otherwise healthy young or the middle-aged subjects present with ischemic stroke, arterial dissection including VA dissection (either intracranial or extracranial) should be considered in addition to cardioembolic stroke and thromboembolic stroke associated with anti-phospholipid antibody syndrome or other systemic autoimmune diseases. Angiographic normalization or improvement of the VA dissection is frequently observed,[3,6,11,13] so initial angiography should be performed as early as possible. Follow-up angiography is also necessary to observe the chronological change of the vascular abnormalities.

The primary treatment for extracranial VA dissection is anticoagulant or antiplatelet drugs for secondary prevention of thromboembolic complications.[7,9,11] Surgical intervention is reserved only for patients with progressive dissection after conservative treatment or repeated ischemic symptoms.

Extracranial VA dissection is not uncommon, but can be difficult to identify. Spontaneous extracranial VA dissection should be suspected in young patients presenting with ischemic stroke but without predisposing risk factors or associated trauma. Careful angiographic evaluation in the early stage is necessary together with follow-up angiography for definite diagnosis.

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