

## CASE REPORT

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# Vertebral artery dissection associated with Behçet's disease in elderly female

Akira Tempaku

## ABSTRACT

**Introduction:** Behçet's disease is known as an autoimmune disorder that is characterized by systemic inflammation. The disorder predominantly affects middle-aged individuals and is traditionally considered to be more prevalent in men. However, several reports point that number and ration of female patient have increased in recent years. In addition, several patient cases with significant disability due to vascular complications, such as thrombosis and aneurysms with stroke, have also been documented.

**Case Report:** An 87-year-old female patient with a medical history of Behçet's disease presented with headaches as her primary complaint. No abnormalities were identified in the central nervous system. Blood pressure was also in normal range. However, subsequent head magnetic resonance imaging studies revealed a right vertebral artery (VA) dissection, which was considered the underlying cause of the headache. The patient was subjected to conservative management, with a satisfactory recovery ensuing without complications, such as hemorrhagic or ischemic stroke.

**Conclusion:** Vertebral artery dissection is most prevalent among middle-aged individuals; however, this case is atypical in that it occurred at an older age than is typically observed for the onset of the condition. Moreover, as a vascular lesion associated with Behçet's disease, it was a rare occurrence because it manifested in the VA. Intracranial VA dissection is uncommon, which had been previously documented in few cases among middle-aged males with Behçet's disease.

**Keywords:** Behçet's disease, Dissected aneurysm, Elderly female, Intracranial vertebral artery

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## INTRODUCTION

Behçet's disease is categorized as a systemic inflammatory disorder, which is typified by the presence of oral ulcers, genital ulcers, skin rashes, and ocular symptoms [1, 2]. The disease predominantly affects individuals between the ages of 20 and 40, with males demonstrating a higher propensity to exhibit more severe manifestations [3]. While cases in men have historically been more prevalent, there has been an increasing recognition of cases in women in recent years [3]. The prevalence of the Behçet's disease patient has been documented mainly in East Asia, Middle East Asia, and Mediterranean countries [3, 4]. Patients with Behçet's usually exhibit either Human Leukocyte Antigen (HLA)-B51 or HLA-A26 antigens [5–7]. Furthermore, immune-related molecule coding genes, including the interleukin (IL)-23 receptor, IL-12 receptor  $\beta$ 2 chain, IL-10, and endoplasmic reticulum aminopeptidase (ERAP)1, have been characterized as susceptibility genes for Behçet's disease [8–11]. These factors are considered to contribute to regional and ethnic disparities in disease prevalence.

Although not prevalent, cases accompanied by gastrointestinal ulcers [12] or neurological symptoms, such as meningitis and encephalitis [13–15], tend to manifest with severe symptoms. Furthermore, vascular lesions [16, 17] involving thrombosis in major arteries or veins, and aneurysm formation with rupture, are

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reported to be more prevalent in males. Contrary to common knowledge, vertebral artery dissection, causing to aneurysm formation, was observed in an elderly female patient outside the typical age range for this condition. While vascular Behçet's disease has been predominantly reported in males, the present case involved a female patient. In addition, based on a comprehensive review of the extant literature, it is believed that this constitutes the first documented instance of intracranial VA dissected aneurysm in female.

## CASE REPORT

An 87-year-old woman presented with symptoms including persistent headache and posterior neck pain. No instances of consciousness impairment, hemiplegia, sensory disturbances, or ataxia were observed. Her general condition remained stable with blood pressure was 133/72 mmHg, and heart rate was 69/min. The patient had been undergoing medical treatment to Behçet's disease. A detailed analysis of the head and neck region was conducted using magnetic resonance imaging (MRI), which revealed a right VA dissection of the V4 segment. No acute hemorrhagic or ischemic structural abnormalities, neoplastic lesions, or white matter lesions were identified. The MRI scan revealed an acute phase mural thrombus with high T1-weighted image signal and high Black Blood signal (Figure 1). As demonstrated by the patient's symptoms, no substantial deterioration was observed at the time of the visit, which occurred on the fourth day following the onset of symptoms. Her ethnicity is Japanese. She had no family history of Behçet's disease among her known relations. Consequently, the therapeutic approach was chiefly oriented toward the regulation of blood pressure, and the patient was observed as an outpatient. In the ensuing clinical period, no adverse events have been observed.

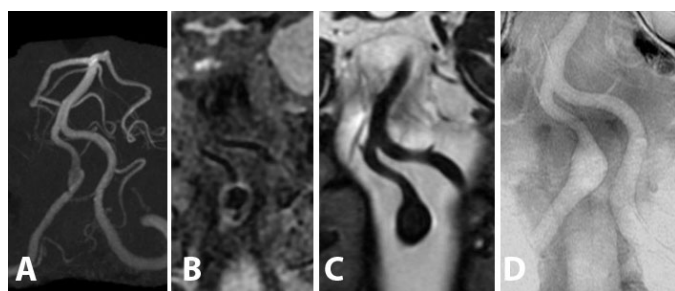


Figure 1: Head magnetic resonance imaging. (A) illustrates the time-of-flight image of the vertebral artery (VA) and the basilar artery. (B) presents the T1-weighted image of VA. The acute state of thrombus is distributed around the intima, as indicated by the high-intensity signal. A high-intensity flap in the vascular lumen reveals the dissected intima. (C) illustrates the fast imaging employs steady-state acquisition for the image of VA. Local dissection and aneurysmal deformity are observed as focal round shape. (D) illustrates the basi-parallel anatomical scanning image of VA. Focal enlargement of adventitia presents the local dissection.

## DISCUSSION

Vertebral artery (VA) dissection predominantly affects middle-aged individuals [18, 19]. Causes of VA dissection include hypertension and excessive mechanical stress by trauma to the neck [20]. This case involved an elderly patient, representing a rare age group for onset. Furthermore, no precipitating factors such as hypertension or excessive neck stress were identified.

Vascular involvement disorder is reported in 7–29% of patients with Behçet's disease. The common vascular lesions associated with Behçet's disease are arterial or venous occlusive diseases and aneurysm formation [21]. Aneurysms mainly develop in the major vessels including the abdominal aorta, femoral and pulmonary arteries. Although extracranial aneurysm is well known, intracranial aneurysm formation with Behçet's disease is rare. However, this case demonstrated dissection with aneurysm formation in the intracranial VA. The structural composition of the vascular wall in head and neck vessels is characterized by a tripartite layering. Within the intracranial area, the VA exhibits a three-layer structure: intima, media, and adventitia, with an internal elastic lamina separating the intima and media [22]. The pathogenic features of Behçet's disease are the inflammation of the vascular endothelium [23–25]. The inflammatory reactions lead to the thrombosis due to hypercoagulability [24, 25] and the enlargement of dissection cavities within the vessel wall, resulting in aneurysm formation [21, 26].

Intracranial aneurysms in Behçet's disease were reported in the several papers including case reports [26–29]. The present studies found that all patients with an intracranial VA dissected aneurysm were male. Furthermore, all patients are under 70 years of age. Inflammatory angiitis-related VA aneurysm has only been reported in one case, in a 67-year-old female patient [30]. That particular case has been linked to a condition known as eosinophilic granulomatous with polyangiitis, which is a type of disease that affects the immune system. In that report, Hayashi described two cases of fusiform aneurysm in VA among female patients as part of a literature review. However, the other cases were in 30-year-olds [30]. The present case is distinctive in that it was observed in a patient over the age of 80. Moreover, the present study documents, for the first time, the occurrence of an intracranial VA aneurysm associated with Behçet's disease in a female patient. To prevent the progression of vessel wall dissection and rupture of aneurysm, blood pressure was controlled under 140 mmHg in systolic phase with calcium channel blocker administration. Because of clinical guidelines against Behçet's disease never mention to handle with the dissected intracranial vessels, the patient was treated following to the manner of dissected intracranial aneurysm by hypertension.

In this case, it was hypothesized that inflammation of the vascular endothelium due to Behçet's disease contributed to the formation of a dissection cavity in the

intima and media. This case of vertebral artery dissection was atypical in nature, necessitating an examination that considered underlying disorders such as vasculitis or collagen diseases, in addition to hypertension.

This case includes several rare clinical features. Based on the basic pathologies of Behçet's disease, constructing the clinical treatment protocol would be needed. Furthermore, clinical evidence accumulation and characteristic analysis would remain required.

## CONCLUSION

A case of VA dissection in an elderly woman without hypertension was observed in the vascular complications of Behçet's disease. This finding indicates that VA dissection can also occur as a consequence of inflammatory diseases. Preventing the dissected aneurysm rupture, blood pressure control under 140 mmHg by calcium channel blocker might be useful. There are no obvious clinical guideline of Behçet's disease with the dissected intracranial vessels. Further case studies would be needed to construct the treatment chart.

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### Author Contributions

Akira Tempaku – Conception of the work, Design of the work, Acquisition of data, Analysis of data, Interpretation of data, Drafting the work, Revising the work critically

for important intellectual content, Final approval of the version to be published, Agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved

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### Conflict of Interest

Author declares no conflict of interest.

### Data Availability

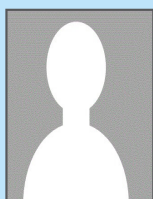
All relevant data are within the paper and its Supporting Information files.

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