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Case report

Churg-Strauss syndrome presenting with subarachnoid hemorrhage from ruptured dissecting aneurysm of the intracranial vertebral artery

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Abstract

Churg-Strauss syndrome (CSS) represents a rare systemic vasculitis that is almost invariably accompanied by bronchial asthma and eosinophilia. We report a case of a 36-year-old woman with previously diagnosed CSS presented with subarachnoid hemorrhage (SAH) from dissecting aneurysm in a vertebral artery (VA). Two months before onset of SAH, the patient had presented with numbness on her right lower leg due to peripheral neuropathy. On admission, angiography revealed dissecting aneurysm of the right intracranial VA and stenosis of the basilar artery. Hematological examination revealed an increased percentage of eosinophils. Ruptured dissecting aneurysm of the intracranial VA was diagnosed. Emergent coil embolization of the dissecting aneurysm and occlusion of the parent artery was performed to prevent repeated hemorrhage from the dissecting aneurysm. Then pharmacotherapy with predonisone was initiated for CSS. The patient recovered well and was discharged without any neurological deficit. As far as we know, this is the first reported case of CSS presented with SAH from dissecting aneurysm on posterior circulation.

Keywords: Churg-Strauss syndrome; Dissecting aneurysm; Subarachnoid hemorrhage; Vertebral artery

1. Introduction

Churg-Strauss syndrome (CSS) represents systemic small-vessel vasculitis characterized by the presence of asthma and eosinophilia [1]. Organ involvement of the central nervous system (CNS) associated with CSS appears uncommon [1–3], and CSS presenting with subarachnoid hemorrhage (SAH) is particularly rare [4–6]. We report the case of a 36-year-old woman with CSS presenting with SAH caused by rupture of intracranial dissecting aneurysm in the vertebral artery (VA).

2. Case report

A 36-year-old woman was admitted to Hiroshima University Hospital complaining of severe headache and vomiting on March 28, 2003. The patient had suffered from severe bronchial asthma and eosinophilia for 8 years. Furthermore, the patient sometimes suffered from nausea due to eosinophilic gastroenteritis and had been clinically diagnosed with CSS and received treatment with betamethasone (1.5 mg/day) for two years. No clear history was present for risk factors of cerebrovascular diseases, including hypertension, diabetes mellitus, hyperlipidemia, smoking, and trauma.

Over the preceding 2 months, the patient had noticed numbness and a few erythema on the dorsal aspect of the right lower leg. Sensory nerve conduction study had revealed low amplitude in proximal side of the right sural nerve. There-

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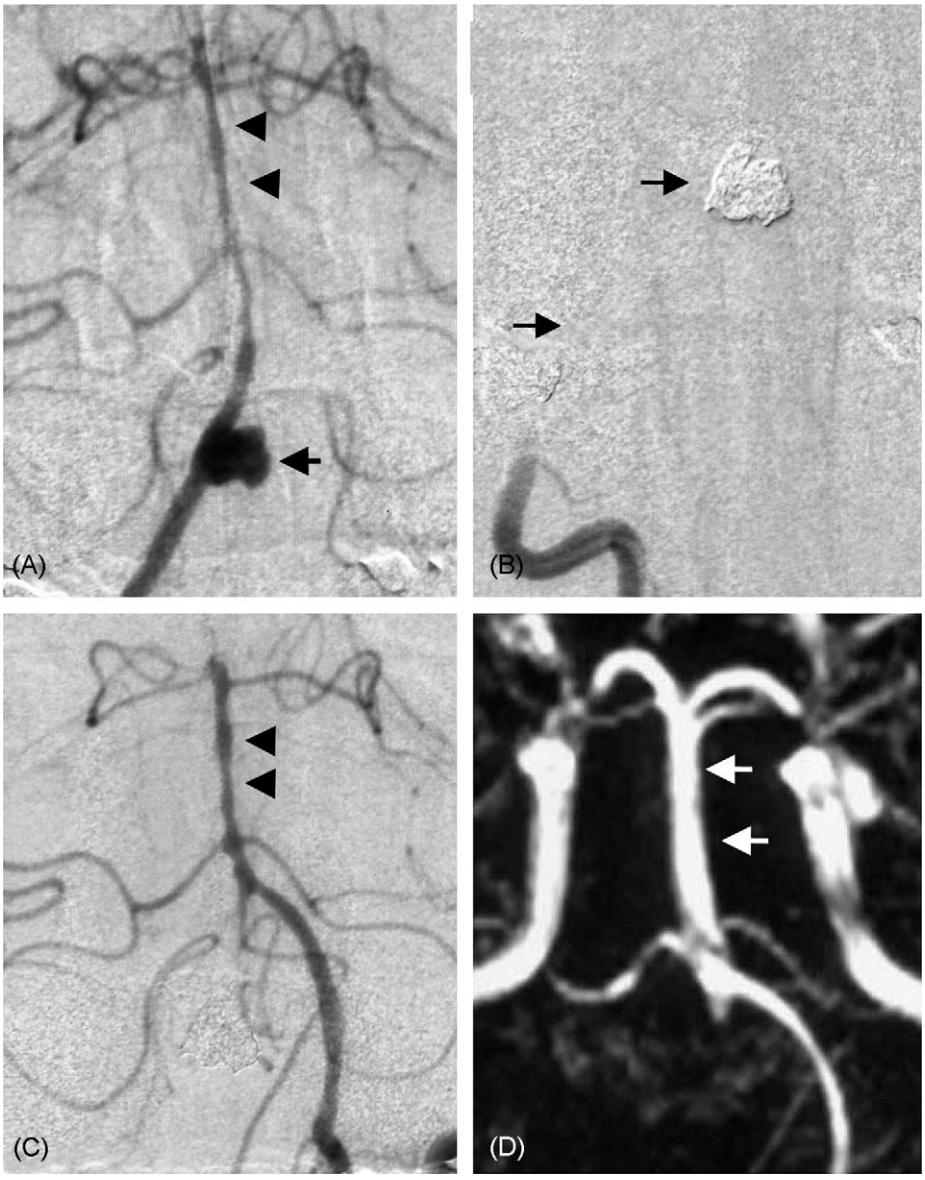

Fig. 1. (A) Right vertebrobasilar angiography on admission showing dissecting aneurysm of the intracranial vertebral artery (arrow) and irregular stenosis of the basilar artery (arrowheads). (B) Right vertebrobasilar angiography after endovascular treatment showing excellent occlusion of dissecting aneurysm and parent artery (arrows). (C) Left vertebrobasilar angiography showing irregular stenosis of basilar artery (arrowheads) and no retrograde filling of contrast medium into dissecting aneurysm. (D) Magnetic resonance angiography one month after embolization, showing complete obliteration of dissecting aneurysm of the right vertebral artery and improvement of stenosis of the basilar artery (arrows).

fore, it had been diagnosed that numbness had been caused by peripheral neuropathy of the right sural nerve. On admission, Glasgow Coma Scale score was 14. Computed tomography on admission revealed SAH and hydrocephalus. Hunt and Hess grade for SAH was 1. Hematological and laboratory examinations revealed: white cells, 24,780/mm^3 (38.4% eosinophils, 48.4% neutrophils); platelets, 6.89×10^5/mm^3; IgE, 1000 IU/ml (normal < 321.4 IU/ml); and C-reactive protein, 1.2 mg/dl (normal < 0.4 mg/dl). Positive results were obtained for rheumatoid factor. Negative results were obtained for myeloperoxidase antineutrophil cytoplasmic antibody. Digital subtraction angiography on admission revealed dissecting aneurysm of the right intracranial VA and irregular stenosis of the basilar artery presumably caused by vasculitis (Fig. 1A), and intracranial dissecting aneurysm associated with CSS was diagnosed. Endovascular occlusion of the right VA was performed on the day after admission to prevent repetitive hemorrhagic event. Balloon occlusion test (BOT) and coil occlusion procedures were performed with local anesthesia to observe neurological examination. A 6Fr guiding catheter was placed in the proximal right VA via a right femoral artery. A 5Fr diagnostic catheter via a left femoral artery was placed in the proximal left VA to verify collateral flow during BOT. The patient was anticoagulated by the intravenous injection of 5 mg argatroban. The balloon microcatheter was introduced just distal to the portion