

[CASE REPORT]

The Sudden Onset of Pure Parkinsonism Caused by Intracranial Dural Arteriovenous Fistulas

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Abstract:

We herein report a case of sudden-onset parkinsonism, with no other symptoms, caused by intracranial dural arteriovenous fistulas (DAVFs). Diffusion-weighted magnetic resonance imaging (MRI) revealed an increased signal intensity in the bilateral lenticular nucleus. Endovascular embolization improved the patient's parkinsonism and MRI findings. DAVF should be suspected in cases of sudden-onset parkinsonism.

Key words: intracranial dural arteriovenous fistula, parkinsonism, striatal presynaptic dopaminergic function

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Introduction

Common initial symptoms of intracranial dural arteriovenous fistula (DAVF) are pulsatile tinnitus, cranial nerve palsy, headache, convulsion, and cognitive deficits. Parkinsonism associated with DAVF is rare. To our knowledge, there have been four reported cases of isolated parkinsonism, unaccompanied by other symptoms, that were caused by DAVF (1, 2). Of these, only one case was examined using ^{18}F -DOPA positron emission tomography/computed tomography (CT) to evaluate the striatal dopaminergic presynaptic function (3). This small number of case reports of DAVF with parkinsonism is therefore insufficient to elucidate its pathophysiology.

Case Report

We recently encountered a case of DAVF presenting with sudden-onset parkinsonism only. A 67-year-old man developed sudden-onset bradykinesia and was admitted to a local hospital at the beginning of November, 20XX-1. He had a history of mild renal failure but no history of head trauma.

A neurological examination indicated parkinsonism. Brain magnetic resonance imaging (MRI) showed very faint high-signal intensities of the bilateral globus pallidus on diffusion-weighted imaging and a normal signal on T2-weighted imaging (Figure A, B). Magnetic resonance (MR)

angiography and three-dimensional (3D)-CT angiography revealed two DAVFs: one at the right transverse sinus and one at the right sigmoid sinus (Figure C, D). Because the patient's renal failure worsened after the 3D-CT angiography, he was transferred to another hospital for treatment. He was discharged from the hospital three months later, with improvement of renal failure.

He visited our hospital at the beginning of March, 20XX. He was alert, and his cognitive function was well-preserved. He showed isolated parkinsonism, including a masked face, small voice, rigidity of all four limbs with left-side predominance, short-stepping gait with gait freezing, and postural instability. His Movement Disorders Society Unified Parkinson's Disease Rating Scale (MDS-UPDRS) motor scale score was 48 out of 108. His Parkinsonism suddenly developed with short-stepping gait which gradually worsened over the next month. After that, his Parkinsonism did not change. Diffusion-weighted imaging and T2-weighted MRI revealed an abnormal high signal intensity in the bilateral lenticular nucleus (Figure E, F). ^{123}I -FP-CIT single-photon emission computed tomography (SPECT) revealed a decreased uptake in the bilateral striatum with left-side predominance (Figure G). The specific binding ratio was 4.49 on the right side and 3.93 on the left side (Figure I). The specific binding ratio of this case was below the 95% predicted interval calculated from the data of Japanese healthy subjects (4).

MR angiography revealed two DAVFs, as had been ob-

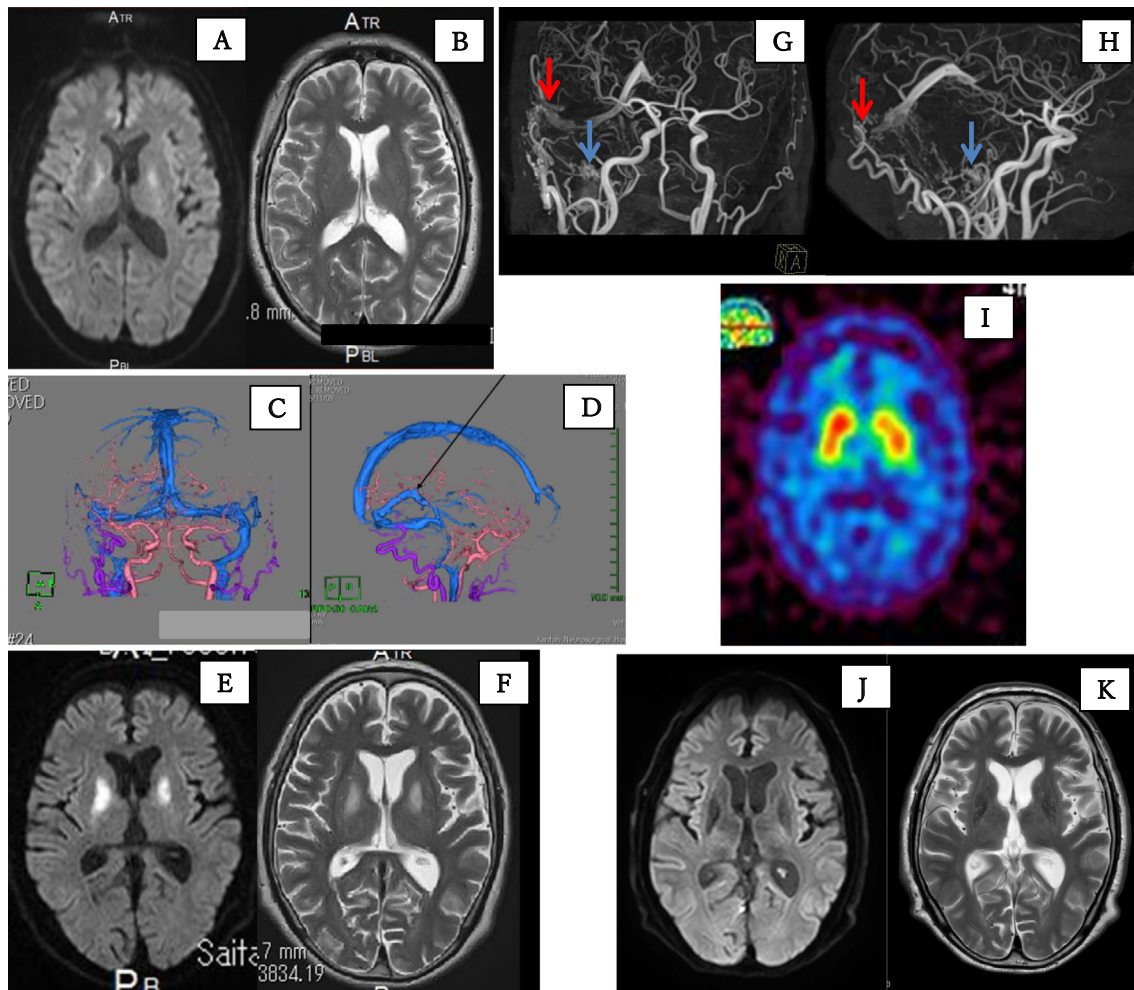


Figure. Representative brain images of the patient. A, B: Magnetic resonance (MR) brain images (2 days after onset). Faint high-signal intensities were observed in the bilateral globus pallidus on diffusion-weighted imaging (DWI), and normal signals were observed on T2-weighted imaging (T2WI). C, D: 3D computed tomography angiography (4 days after onset). The blood vessels are color-coded. Pink indicates the internal carotid and vertebral arteries, blue indicates the cerebral venous sinus, and violet indicates the external carotid artery. E, F: MRI brain images (154 days after onset). Bilateral high signal intensities were observed in the globus pallidus on DWI and T2WI. G, H: MR angiography (183 days after onset). There were two DAVFs. One was in the right transverse-sigmoid sinus fed by the right occipital artery (red arrow), and the other was in the sigmoid sinus fed by the right ascending pharyngeal artery (blue arrow). I: ^{123}I -FP-CIT single-photon emission computed tomography (2 days after onset). The specific binding ratios were 4.49 on the right side and 3.93 on the left side. These values demonstrate a decline in dopamine transporter binding in the bilateral striatum. J, K: MR brain images after the endovascular embolization of the fistula. Bilateral low signal intensities were observed in the globus pallidus on DWI and T2WI.

served previously (Figure G, H). One was in the right transverse-sigmoid sinus fed by the right occipital artery, and the other was in the right sigmoid sinus fed by the right ascending pharyngeal artery. MR angiography also showed abnormal signals in the straight sinus and vein of Galen, which are not usually depicted. It was hypothesized that the abnormal blood flow of the straight sinus and vein of Galen was reflux from the fistulas. His parkinsonism did not respond to levodopa (400 mg/day). Intravascular embolization of the fistula was effective for treating his Parkinsonism, and his MDS-UPDRS motor scale score improved to 19. The

MRI findings also improved (Figure J, K).

Discussion

We herein report a case of isolated parkinsonism caused by DAVFs. Based on the radiographic findings, the pathophysiological mechanisms of this patient's parkinsonism can be considered as follows: according to 3D-CT angiography, the right transverse sinus had communication with the straight sinus but no communication with the left transverse sinus or superior sagittal sinus. This type of anatomical variation of

the torcular Herophili reportedly occurs in 24.5% of adults. It is therefore speculated that the blood regurgitated from fistulas of the right transverse sinus mainly flows into the straight sinus (5). There was no congestion of the superior sagittal sinus in our patient, which may explain why he showed no cerebral cortical symptoms, such as cognitive decline. The reversed blood flow toward the straight sinus caused venous congestion of the basal ganglia, leading to his parkinsonism. The patient's contrast medium-induced nephropathy meant that it took about four months for him to start receiving treatment for DAVF. However, both his MRI findings and his parkinsonism were improved (especially his MRI findings, which improved significantly) by endovascular embolization of the fistulas. These findings indicate that the basal ganglia were damaged not by cytotoxic edema but by vasogenic edema. The patient's specific binding ratio was bilaterally decreased on presynaptic dopamine transporter SPECT, although this was inconsistent with the laterality of his parkinsonism. The parkinsonian symptoms in this patient did not improve with L-dopa, suggesting that venous congestion caused by DAVF might impair the postsynaptic function in the striatum. We speculated that there was dominant dysfunction of the postsynaptic nerves in the right striatum, resulting in left-handed parkinsonism.

DAVF-induced parkinsonism has a generally good prognosis with early treatment. Parkinsonism reportedly becomes irreversible if treatment is delayed considerably, so early treatment is required (6).

In conclusion, we should consider the possibility of DAVF-associated parkinsonism when we see cases of

sudden-onset parkinsonism. Because few case reports of parkinsonism associated with DAVF exist, further studies are needed to elucidate its pathophysiology.

The authors state that they have no Conflict of Interest (COI).

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