

Short communication

Two cases of dural arteriovenous fistula presenting with parkinsonism and progressive cognitive dysfunction

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ABSTRACT

Dural arteriovenous fistula (DAVF) is a rare type of cerebral arteriovenous malformation. The occurrence of parkinsonism together with progressive cognitive dysfunction caused by DAVF has been rarely reported. The probable underlying pathophysiology could be due to venous hypertension caused by DAVF which leads to basal ganglia and cortical dysfunction. Here, two DAVF cases were reported presenting with parkinsonism and progressive cognitive dysfunction. A 54-year-old man (case 1) and a 75-year-old man (case 2) presented with bradykinesia, gait disturbances and cognitive dysfunction who were initially misdiagnosed as having Parkinson's disease. Case 1 exhibited a characteristic type of subcortical calcification on CT scan, while case 2 had specific resting tremor and intracranial hypertension. Both cases showed transient response to the dopaminergic treatment. After 3D time-of-flight (TOF) magnetic resonance arteriography (MRA) or digital subtraction arteriography (DSA), they were diagnosed as having DAVF. This report suggests the possibility of DAVF in patients presenting with parkinsonism and progressive cognitive dysfunction, which requires further attention to be paid, especially in those with transient response to levodopa. Early 3D TOF MRA or DSA is recommended for the diagnosis of DAVF.

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1. Introduction

Dural arteriovenous fistula (DAVF) is a rare type of cerebral vasculopathy, which accounts for 10–15% of all intracranial arteriovenous malformations [1]. DAVFs are abnormal connections of the arterial and venous systems involving dural sinuses and branches of the external and internal carotid arteries, or vertebral arteries. Typically, DAVF exhibits various symptoms including pulsatile tinnitus, ophthalmoplegia, proptosis, headache, and cognitive dysfunction. DAVF with the onset of parkinsonian symptoms and cognitive dysfunction is rarely reported [2–6]. Here, two DAVF cases were reported presenting with parkinsonism and progressive cognitive dysfunction. These cases' diagnosis was eventually confirmed by 3D time-of-flight (TOF) magnetic resonance angiography (MRA) or digital subtraction arteriography (DSA). Although angiography is the gold standard for the diagnosis of DAVF, 3D TOF MRA as a noninvasive diagnostic tool we employed had provided a satisfactory delineation of DAVF. The causes of these two symptoms may be due to venous hypertension of deep cerebral vein and cortical

venous congestion caused by DAVF, which eventually leads to basal ganglia and cortical dysfunction [3,7].

2. Case reports

2.1. Case 1

A 54-year-old man was admitted to our hospital with a 10-month history of slow-movement and slowed thinking. He was initially diagnosed as having Parkinson's disease by the outpatient doctor at the 7th month after the onset of the symptoms. Madopar, at the dose of 62.5 mg Qid was effective at the beginning. Later the symptoms gradually aggravated although madopar was added up to the maximum dose of 187.5 mg Qid. Slowness of movement, cognitive dysfunction and urinary incontinence finally made him lose the ability to take care of himself. The patient had a mild impact on the right occiput 5 years before the disease and was diagnosed of incurring a brain concussion. He recovered satisfactorily without any neurological sequelae. On neurological examination, his facial expressions were significantly diminished and muscle tone increased in all four of his limbs. The patient had a slow movement while walking with wide-based gait and was very unstable when turning back. Muscle strength and sensation were normal. There was no pathological reflex or signs of cerebellar ataxia. The patient was of primary school level education but his mini-mental state examination (MMSE) score was 14/30. Blood tests had excluded some

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reversible causes of cognitive dysfunction, such as hypothyroidism, vitamin deficiencies and HIV. EEG showed as low as 60 μV 8.5–9 Hz α waves and small amount of scattered θ waves which was at times observed on the frontal head in short ranges. Subsequent non-enhanced CT scan showed curvilinear calcification in the cortico-medullary junction at the bottom of the cerebral sulcus bilaterally (Fig. 1A). T2-weighted magnetic resonance imaging (MRI) revealed flow void clusters at the inner part of the left temporal lobe (Fig. 1B). Source imaging of MRA showed findings of multiple high-intensity curvilinear adjacent to the right transverse sinus wall. Hyperintense signals coming from feeders of the tentorial artery were well visualized (Fig. 1C). 3D TOF MRA showed flow-related enhancement of a dural arteriovenous fistula located in the right transverse-sigmoid sinus. High-intensity areas in the dilated straight sinus and the right transverse sinus suggested venous hypertension due to the DAVF (Fig. 1D). DSA finally confirmed the diagnosis of DAVF of the right transverse sinus. Unfortunately, the medical file-keeping department lost the DSA information of this patient for some special reason, so we could not provide this patient's DSA imaging for reference. Considering the risk and the expenses of endovascular embolization treatment, his family chose to give up. After being discharged, his situation continued to get worse. Unfortunately, we eventually lost contact with this patient.

2.2. Case 2

A 75-year-old man came with the complaint of slow-movement for 3 years. 2 years after the onset, the patient started experiencing difficulties to start walking and the gait became shuffling with short steps.

Resting tremor started from his right hand insidiously, and then to his left hand. Simultaneously, his recent memory and arithmetic ability suffered tremendous damage. He had a 10-year history of hypertension and no history of head trauma. On the first visit to our department, the patient had apparent bradykinesia, gait disturbances and resting tremor of upper extremities. He had a mask-like face with mouth open and diminished blinking. His muscle tone increased moderately. No abnormality of muscle strength and sensation was found, and there were also no pathological reflexes. He had a MMSE score of 8/30. Hypothyroidism, vitamin deficiencies and HIV were also excluded by blood tests. Initially the patient was misdiagnosed as having Parkinson's disease and was given a small dose of madopar treatment. The tremor of his hands improved and he was able to walk faster than before. However, this improvement lasted only for three months. Increasing the dose of madopar did not achieve a better effect to his condition. A moderate headache was complained when the patient was lying down. In order to confirm a possible intracranial hypertension and differentiate other secondary diseases which could cause headache we carried out lumbar puncture and his cerebrospinal fluid pressure exceeded 300 mmH₂O. α waves completely disappeared and diffuse slow waves could be seen in his EEG. Further, the MRI revealed that there was a significantly dilated vein on the left temporal cortex on T2-weighted imaging. There were also apparently increased flow void signals indicating deep dilated vessels of the brain (Fig. 2A). The left dilated cortical veins could also be found on the same level of the source MRA imaging (Fig. 2B). 3D TOF MRA revealed the apparently inflated left middle meningeal vein connected with the left transverse-sigmoid sinus. The amount of dilated intracranial

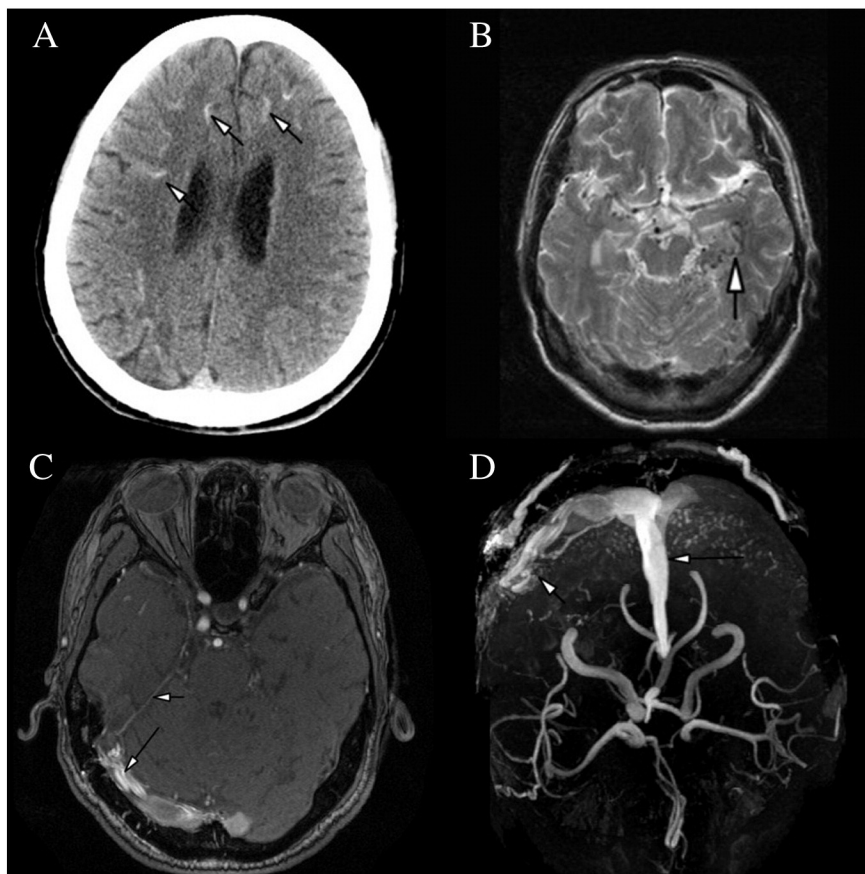


Fig. 1. A dural arteriovenous fistula (DAVF) in a 54-year-old man with slow-movement and slowed thinking. (1A) Non-enhanced CT scan shows multi-curvilinear calcification (arrows) in subcortical region of the bilateral frontal and parietal lobe. (1B) T2-weighted MRI imaging shows flow void clusters at the left medial temporal lobe (arrow). (1C) Source imaging of MRA shows findings of multiple high-intensity curvilinear (long arrow) adjacent to the right transverse sinus wall. Hyperintense signals coming from feeders of the lateral tentorial artery (short arrow) are well visualized. (1D) 3D TOF MRA shows flow-related enhancement of a DAVF located in the right transverse-sigmoid sinus (short arrow). High-intensity areas in the dilated straight sinus (long arrow) and the right transverse sinus suggest venous hypertension due to the DAVF.

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