

# Dural Arteriovenous Fistula With Sinus Thrombosis and Venous Reflux Presenting as Parkinsonism

## A Case Report

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**Abstract:** The occurrence of dural arteriovenous fistula (DAVF) is rare. The clinical manifestation varies and depends on the location and venous drainage pattern. We present a case of a 57-year-old man with a left transverse sinus DAVF along with sigmoid thrombosis, cortical venous reflux, and congestion, that initially presented as parkinsonism. The patient was alert and fully oriented; however, decreased facial expressions were noted. His left forearm showed rigidity and bradykinesia, and it was difficult for the patient to smoothly perform rapid alternating movement testing. His complaints about hearing a rhythmic bruit above the left ear, particularly when lying down, alerted the physician of the presence of vascular lesions. Magnetic resonance imaging and angiography confirmed the diagnosis of DAVF. The patient received a combined surgical and endovascular approach to permanently block the fistula blood flow. The motion and movement of his left upper limb improved after fistula embolization. Images at the 1-month follow-up showed a decrease in the volume of tortuous vessels, and the fistula was completely occluded.

**Key Words:** angiography, brain, etiology

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The term “parkinsonism” usually refers to a group of diseases, including movement disorders. It is a syndrome characterized by bradykinesia, rigidity, and tremors. Parkinsonism is easily linked to Parkinson disease, particularly in the elderly.<sup>1</sup> However, some other factors and differential diagnoses should be considered. Although the direct cause and mechanisms of Parkinson disease remain unknown, it is now thought to be related to genetic and environmental factors and responsive to dopaminergic medications.<sup>2</sup> In contrast, for the patients with parkinsonism, the prompt and accurate diagnosis of the reversible underlying conditions might be critical. Differential diagnosis of Parkinson disease and parkinsonism is complicated and difficult. The spectrum of parkinsonism includes neurodegenerative disorders, infectious diseases, cerebrovascular diseases, metabolic disorders, exposure to drugs and toxins, and structural brain lesions, etc.<sup>3</sup>

Dural arteriovenous fistula (DAVF) is a vascular malformation. A dural fistula is an abnormal crossover connection between 2 sides of the dural layer. Penetrating the dura mater, arachnoid mater, and pia mater, DAVF directly drains blood

flow from the meningeal and extracranial arteries into the meningeal veins, pia veins, and/or dural sinuses.<sup>4</sup> The true incidence and prevalence of DAVF is not known because patients with DAVF might completely be asymptomatic.<sup>5</sup> In addition, DAVF might mimic other neurological conditions because of its location and the associated effects of space occupation and brain tissue perfusion.

We present a case of a patient with left transverse sinus DAVF with sigmoid thrombosis, cortical venous reflux, and congestion, that initially presented as parkinsonism.

### CASE REPORT

A 57-year-old man without any history of head trauma or central nervous system infection visited the clinic because of his clumsy left hand. He was healthy, was not taking any medication, and had no known risk factors for cerebrovascular diseases. He complained that he currently could not perfectly play a Chinese violin as he used to.

During the examination, the patient was alert and fully oriented; however, decreased facial expressions were noted. His left forearm showed rigidity and bradykinesia, and it was difficult for the patient to smoothly perform rapid alternating movement testing. Parkinsonism and Parkinson disease were then suspected. Although his muscle strength of the bilateral limbs was symmetric and full, his complaints during the following visit about hearing a rhythmic bruit above his left ear, particularly when lying down, alerted the physician about the presence of vascular lesions.

Electroencephalography displayed slow waves over the left temporal area, and magnetic resonance imaging and angiography (MRI and MRA, respectively) showed engorged and tortuous vessels above the cerebellum, the deep gray matter, and the left temporal lobe (Fig. 1). Digital subtraction angiography (DSA) revealed early opacification of the left transverse sinus with occlusion of the sigmoid sinus. Reverse blood flow to the straight sinus and right transverse sinus and many engorged cortical veins were also noted (Fig. 2). The diagnosis of left transverse sinus DAVF with sigmoid thrombosis and cortical venous reflux was confirmed.

Transvenous embolization through the occluded sigmoid sinus was attempted but failed because the sigmoid thrombosis was too long to fully advance the catheter. Intervention from the opposite transverse sinus also failed because of a tortuous confluence of sinuses. Finally, the patient received a combined surgical and endovascular approach. The neurosurgeon punctured the left transverse sinus directly with an 18-G needle sheath through a burr hole in the left occipital bone. A microcatheter was inserted into the left transverse sinus via the sheath, and detachable coils were deployed in the distal sigmoid sinus posterior to the distal left transverse sinus. After embolization, only minimal residual fistula with slow flow remained.

One month later, the patient mentioned that the noise above his left ear had almost disappeared, and the movement of his left forearm and hand improved. The patient underwent a follow-up MRA and DSA, which showed a decreased volume of tortuous vessels. The previous fistula was completely occluded.

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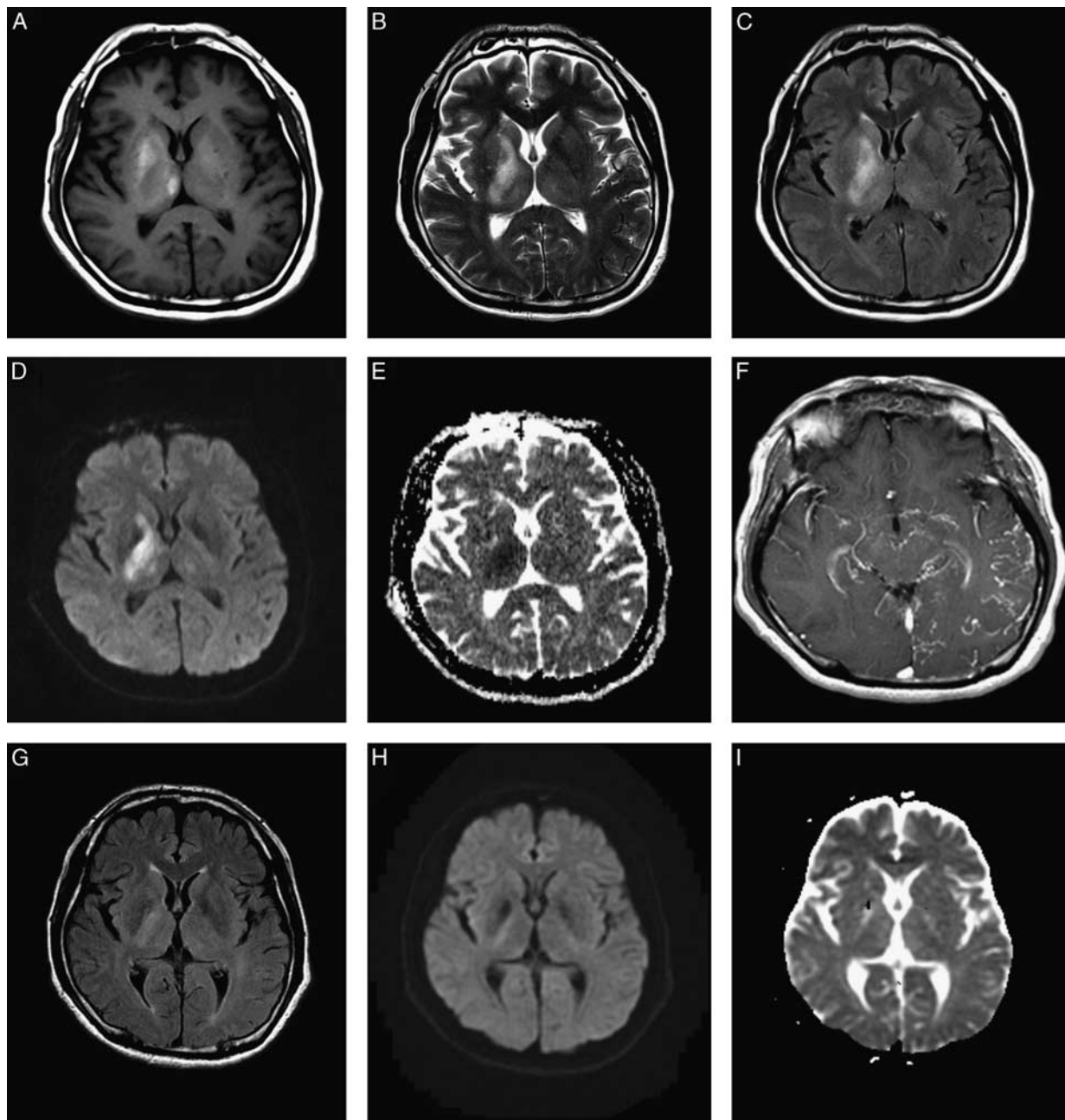
The authors declare no conflict of interest.

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**FIGURE 1.** Brain magnetic resonance imaging of a 57-year-old man with dural arteriovenous fistula, preembolization (A–F) and post-embolization (G–I). A, T1WI showed high signal intensity above the right thalamus and basal ganglion; T2-weighted image (B–E), fluid-attenuated inversion recovery, and diffusion-weighted image showed high signal intensity above the right basal ganglion, internal capsule, and thalamus. Apparent diffusion coefficient showed low signal intensity in the same area. F, T1WI with contrast revealed tortuous and engorged vessels above the left temporal area and deep gray matter. G–I, Images a month after embolization revealed improvement of these abnormal signals. T1WI indicates T1-weighted image.

### DISCUSSION

In this case, we demonstrated that DAVF might be listed as one of the differential diagnoses in patients with secondary parkinsonism. The timely diagnosis and definite treatment prevented prolonged useless treatment in this patient, as well as potential critical complications such as intracranial hemorrhage.<sup>5</sup>

In 1999, Matsuda et al<sup>6</sup> reported on 3 patients (2 women and 1 man) with DAVF who had major symptoms of progressive dementia and parkinsonism, which may be the first case reported

on the association of DAVF and parkinsonism. However, when searching on PubMed for DAVF and parkinsonism, only sporadic cases were found reporting the link between DAVF and parkinsonism in the last 2 decades.<sup>6–14</sup> Almost all the reports were in Asia (6, 3, 1, 1, and 1 in Japan, China, South Korea, India, and Israel, respectively), and only one 79-year-old man was reported in North America,<sup>13</sup> with DAVF in the traditional occidental area. These patients were predominantly male (only 3 female patients), with an age distribution from 40 to 79 years.



**FIGURE 2.** Digital subtraction angiography of a 57-year-old man with DAVF. A, Angiography of the left internal carotid artery revealed a feeding artery to the left transverse sinus (arrow head). B and C, Angiography of the left external carotid artery revealed early opacification of the left transverse sinus with occlusion of the left sigmoid sinus and reverse flow to the straight sinus, right transverse sinus, and multiple cortical veins. D, The microcatheter was placed into the left transverse sinus through a transcranial sheath (white arrow), and the coils were deployed within it. E and F, Angiography 1 month later confirmed the complete occlusion of DAVF. DAVF indicates dural arteriovenous fistula.

Previously, DAVF was thought to be congenital but is now considered an acquired disease associated with venous hypertension, thrombosis, and factors triggering angiogenesis.<sup>4</sup> The presented symptoms of DAVF range from asymptomatic to emergent critical and might depend on the location, pattern of venous drainage, and vascular status at diagnosis. The symptoms might be varied and nonspecific. With the increased accessibility to computed tomography, MRI, and MRA, the diagnosis of DAVF may become more accurate and efficient.<sup>15,16</sup>

In this case, DAVF with retrograde flow caused venous congestion and ischemia above the right basal ganglia, internal capsule, thalamus, left cerebellum, and left temporal lobe, as well as a hemorrhage above the right basal ganglia and thalamus. These are likely the cause of the patient's clumsy hand, reduced left arm movement, masked face, and lethargy. Similar to what was seen in previous reports,<sup>7,13</sup> the patient's parkinsonism dramatically improved after successful embolization. This made our diagnosis of DAVF-induced parkinsonism even more certain. Interestingly, progressive cognitive dysfunction or dementia, except for parkinsonism, was frequently

mentioned in previous reported DAVF cases. Although Lee and colleagues reported an isolated parkinsonism-related DAVF case of a 60-year-old woman without clinically noticeable cognitive dysfunction; the patient's mini-mental status examination score was only 15.<sup>9</sup> In contrast, our patient showed perfect performance in his mini-mental status examination. This might be because his frontal lobes were not involved in this pathologic venous abnormality, as shown in Figure 1. For DAVF patients presenting with parkinsonism, bruit and pulsating tinnitus is an important clue.

When considering Parkinson disease and parkinsonism, the clinicians should always keep DAVF in mind for differential diagnosis. Early diagnosis of DAVF not only reverses the clinical course of parkinsonism but also decreases the risk of possible intracranial hemorrhage.

## REFERENCES

1. Savica R, Grossardt BR, Bower JH, et al. Time trends in the incidence of parkinson disease. *JAMA Neurol.* 2016;73:981-989.

2. Ascherio A, Schwarzschild MA. The epidemiology of Parkinson's disease: risk factors and prevention. *Lancet Neurol*. 2016;15:1257–1272.
3. Frank C, Pari G, Rossiter JP. Approach to diagnosis of Parkinson disease. *Can Fam Physician*. 2006;52:862–868.
4. Wanke I, Rufenacht DA. The dural AV-fistula (DAVF), the most frequent acquired vascular malformation of the central nervous system (CNS). *Clin Neuroradiol*. 2015;25(suppl 2):325–332.
5. Hiramoto M, Sugi K, Hishikawa T, et al. Epidemiology of dural arteriovenous fistula in Japan: analysis of Japanese Registry of Neuroendovascular Therapy (JR-NET2). *Neurol Med Chir*. 2014;54:63–71.
6. Matsuda S, Waragai M, Shinotoh H, et al. Intracranial dural arteriovenous fistula (DAVF) presenting progressive dementia and parkinsonism. *J Neurol Sci*. 1999;165:43–47.
7. Fujii H, Nagano Y, Hosomi N, et al. Dural arteriovenous fistula presenting with progressive dementia and parkinsonism. *BMJ Case Rep*. 2014;2014:pil:bcr2014203921.
8. Hattori T, Takeuchi T, Kabeya R, et al. Transverse-sigmoid sinus dural arteriovenous fistula presenting with parkinsonism. *Neurol Med Chir*. 2013;53:224–227.
9. Lee PH, Lee JS, Shin DH, et al. Parkinsonism as an initial manifestation of dural arteriovenous fistula. *Eur J Neurol*. 2005;12:403–406.
10. Luo Y, Qi J, Cen Z, et al. Two cases of dural arteriovenous fistula presenting with parkinsonism and progressive cognitive dysfunction. *J Neurol Sci*. 2014;343:211–214.
11. Ma C, Lu Q, Shi W, et al. Diagnosis and treatment of a dural arteriovenous fistula presenting with progressive parkinsonism and dementia: a case report and literature review. *Exp Ther Med*. 2015;9:523–526.
12. Netravathi M, Pal PK, Bharath RD, et al. Intracranial dural arteriovenous fistula presenting as parkinsonism and cognitive dysfunction. *J Clin Neurosci*. 2011;18:138–140.
13. Nogueira RG, Baccin CE, Rabinov JD, et al. Reversible parkinsonism after treatment of dural arteriovenous fistula. *J Neuroimaging*. 2009;19:183–184.
14. Shahar T, Gadoth A, Nossek E, et al. Reversible freezing of gait caused by dural arteriovenous fistula and congestion of the globus pallidus. *Mov Disord*. 2012;27:1690–1693.
15. Lin YH, Lin HH, Liu HM, et al. Diagnostic performance of CT and MRI on the detection of symptomatic intracranial dural arteriovenous fistula: a meta-analysis with indirect comparison. *Neuroradiology*. 2016;58:753–763.
16. Al-Radaideh AM, Rababah EM. The role of magnetic resonance imaging in the diagnosis of Parkinson's disease: a review. *Clin Imaging*. 2016;40:987–996.