

CASE REPORT

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Transverse-Sigmoid Sinus Dural Arteriovenous Fistula Presenting With Parkinsonism

—Case Report—

Tomoji HATTORI,¹ Taku TAKEUCHI,¹ Ryusuke KABEYA,¹
Kazuhiko ANDO,¹ and Fujio TOSAKI¹

¹Department of Neurosurgery, Ichinomiya Municipal Hospital, Ichinomiya, Aichi

Abstract

Dural arteriovenous fistula (DAVF) is rarely associated with parkinsonism. A 52-year-old woman presented with a rare case of DAVF manifesting as parkinsonism and subsequently akinetic mutism. She showed dramatic recovery after endovascular treatment. We also review 10 published reports of DAVF presenting with parkinsonism. The clinical features of these cases at presentation was more closely compatible with lower body parkinsonism or vascular parkinsonism rather than Parkinson's disease. Most lesions are located at the transverse-sigmoid sinus (TSS) with venous reflux into the straight sinus with probable venous congestion of the basal ganglia. Most importantly, parkinsonism due to TSS DAVF is reversible if embolization is achieved successfully.

Key words: cognition disorder, dural arteriovenous fistula, embolization, parkinsonian disorder

Introduction

Dural arteriovenous fistula (DAVF) accounts for only 10–15% of all intracranial vascular malformations.³⁾ The incidence is 0.29 persons/100,000/year in Japan,³⁾ higher than 0.16 persons/100,000/year in Western countries.¹⁾ DAVFs may present incidentally or with symptoms relat-

ed to the location and pattern of venous drainage. Chemosis, proptosis, and ophthalmoplegia are symptoms of cavernous sinus lesions, and pulsatile tinnitus is common for transverse-sigmoid sinus (TSS) DAVF. However, DAVF is rarely associated with parkinsonism.^{2,4–7,9,10)} We report a case of TSS DAVF presenting with parkinsonism, and review 10 similar cases.

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Case Report

A 52-year-old woman had complaints of general fatigue, stumbling, urinary incontinence, and disorientation of 3 months duration. She consulted the Neurology Department because of emesis and gait disturbance. Masked face, hypophonia, short-stepped gait, and mental slowness were observed. Recent onset of depression and psychotropic medication led to a presumptive diagnosis of drug-induced parkinsonism. Computed tomography (CT) showed tight brain, and diffusion-weighted and T₂-weighted magnetic resonance (MR) imaging revealed diffuse high intensity lesions in the deep white matter and basal ganglia (Fig. 1A). CT with contrast medium revealed multiple vermiform enlarged cortical vessels, and she was referred to the Neurosurgery Department. Angiography showed right TSS DAVF with reflux to the straight sinus and cortical veins (Fig. 1B). The right sigmoid sinus had a proximal stenosis; the contralateral transverse sinus was hypoplastic; the posterior third of superior sagittal sinus (SSS) had a stenosis; and the cerebral veins were severely

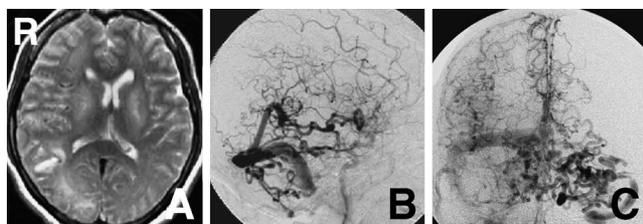


Fig. 1 A: T₂-weighted magnetic resonance image showing hyperintense lesions in the basal ganglia and deep white matter of the right occipital lobe. B: Right common carotid angiogram showing right transverse-sigmoid sinus dural arteriovenous fistula with retrograde venous reflux into the straight sinus and cortical veins. C: Anteroposterior view of right common carotid angiogram showing diffuse venous congestion and drainage into the contralateral sigmoid sinus. The left transverse sinus is hypoplastic.

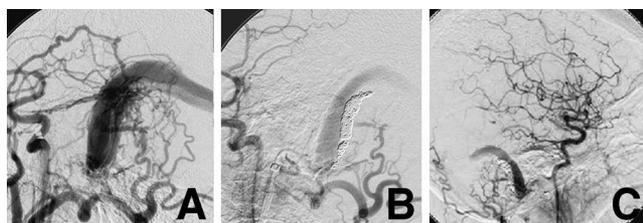


Fig. 2 A: Left anterior oblique view of right common carotid angiogram showing feeders from the middle meningeal, posterior auricular, occipital, and tentorial arteries, converging to a shunt point at the posteroinferior corner of the transverse-sigmoid sinus junction. B: Left anterior oblique view of right common carotid angiogram showing the coil mass in the parallel venous channel. C: Right common carotid angiogram after selective transvenous embolization showing significantly decreased shunt and stagnant but still patent right transverse-sigmoid sinus.

congested. Feeding arteries were the middle meningeal, posterior auricular, occipital, and tentorial arteries, which converged to the shunt point at the posteroinferior corner of the transverse-sigmoid junction (Fig. 2A). Right cerebral blood flow (CBF) mainly drained to the contralateral sigmoid sinus through the cortical veins (Fig. 1C).

One week later, on admission to the Neurosurgery Department, akinetic mutism had developed. She was bedridden and occasionally whispered. She showed marked rigidity with forced neck and forced gaze to the right. Both elbows were flexed as in the decorticate posture, with eyes open. Her husband revealed that she had complained of pulsatile tinnitus for the last 8 months. CBF study showed hypoperfusion in the basal ganglia on both sides and diffuse cortex prominent in the right occipital cortex. Repeat MR imaging revealed asymptomatic small subcortical bleedings without subarachnoid hemorrhage.

Five days later, selective transvenous coil embolization of a parallel venous channel was achieved (Fig. 2B) with preserved patency of the right sigmoid sinus (Fig. 2C). Venous drainage to the contralateral hemisphere diminished immediately. Her condition improved remarkably. She regained speech the next day, and normal mentation and eating in a few weeks. The first follow-up angiography 1 month later showed disappearance of the right TSS DAVF, but a de novo DAVF at the left transverse sinus. The shunt showed retrograde cortical reflux but was very small, and the left transverse sinus was hypoplastic, so transarterial embolization was regarded to be safe and sufficient to occlude the lesion. Coil and particle transarterial embolization through the external carotid artery branches diminished the lesion. She still had parkinsonian gait and was transferred to a rehabilitation hospital. Three months later, she returned to normal life at home, despite moderate parkinsonian gait and micrographia under medication.

Three years later, MR imaging revealed diminished high intensity in basal ganglia (Fig. 3A). Follow-up angiography showed a newly developed DAVF at the SSS (Fig. 3B). The shunt was small and showed no cortical reflux and asymptomatic, so we continued observation. Antegrade venous flow had been preserved in the right



Fig. 3 A: Follow-up T₂-weighted magnetic resonance image revealing diminished hyperintense lesions in the basal ganglia. B: Follow-up right common carotid angiogram 3 years after embolization showing a tiny sagittal sinus dural arteriovenous fistula. C: Venous phase of the right common carotid angiogram showing preserved venous flow in the right transverse-sigmoid sinus.

Table 1 Summary of cases of DAVF presenting with parkinsonism

Case No.	Author (Year)	Age (yrs)/ Sex	Tinnitus/ Bruit	Gait difficulty*/ Tremor	Angiography location/ Drainage	MR imaging change	SPECT hypoperfusion	Time from onset to treatment (mos)**	Treatment	Outcome
1	Okuzumi et al. (1998) ¹⁰⁾	81/M	ND/+	++/+	rt TS/SSS	ND	bil WM	5/5/3	TAE, surg/TVE	improved
2	Matsuda et al. (1999) ⁵⁾	55/M	ND/-	+/-	rt SigS/SSS, StS	WM, BG	lt thalamus	NA/6/8	TAE × 2	improved
3		78/M	ND/-	+/NA	rt SigS/SSS, StS	absent	diffuse cortex	NA/0/NA	TAE × 2	improved
4		69/F	ND/-	++/-	lt SigS/TS, SSS, StS	WM	ND	NA/2/<2	TAE	unimproved
5	Lee et al. (2005) ⁴⁾	60/F	-/ND	++	lt TSS/SSS, StS	WM	lt FT, BG	18/12/NA	TAE	improved
6	Kajitani et al. (2007) ²⁾	75/M	ND/ND	++	rt TSS/SSS	absent	frontal, BG	6/3/3	TAE × 2, feeder coagulation	improved
7	Nogueira et al. (2009) ⁹⁾	79/M	+/+	++	lt TS/SSS, StS	absent	ND	<4/4/NA	TAE × 2, surg/TVE	normalized
8	Miura et al. (2009) ⁶⁾	65/M	+/ND	+/NA	lt TSS/SSS, StS	WM, BG	lt frontal, BG	NA/4/4	TAE × 3	improved
9	Netravathi et al. (2011) ⁷⁾	54/M	+/+	++/+	torcula/StS	BG (thalamus)	ND	NA/ND/12	TAE	minimal change
10		40/M	ND/ND	+/NA	SSS/NA	ND	ND	NA/3/3	TVE/TAE	unchanged
11	Present case	52/F	+/-	++/-	rt TSS/SSS, StS	WM, BG	diffuse cortex, BG	NA/3/3	TVE	improved

*Gait difficulty + indicates some gait difficulty but able to walk; ++, unable to walk. **Tremor/Gait difficulty/Dementia. BG: basal ganglia, F: female, FT: frontotemporal, M: male, MR: magnetic resonance, NA: not applicable, ND: not described, SigS: sigmoid sinus, SPECT: single photon emission computed tomography, SSS: superior sagittal sinus, StS: straight sinus, Surg/TVE: combined surgical and transvenous endovascular approach, TAE: transarterial embolization, TS: transverse sinus, TSS: transverse-sigmoid sinus, TVE: transvenous embolization, WM: white matter.

TSS despite the proximal and distal stenoses (Fig. 3C).

Discussion

DAVF cases with parkinsonism were reviewed (Table 1). The pathophysiology of parkinsonism has been discussed based on MR imaging and single photon emission computed tomography findings. Frontal white matter hypoperfusion was speculated to be responsible for parkinsonism in DAVF,⁹⁾ and this condition may be associated with basal ganglia dysfunction, rather than white matter lesions.⁴⁾ Concomitant frontal lobe and basal ganglia dysfunction may be important.²⁾ In previous cases, MR imaging detected white matter changes in 5 cases, and intensity changes in the basal ganglia in 4 cases, including ours. Review suggests that the pathogenesis of parkinsonism due to DAVF is heterogeneous. However, the clinical features of these cases at presentation were more closely compatible with lower body parkinsonism or vascular parkinsonism¹¹⁾ rather than Parkinson's disease. History of tinnitus or bruit is suggestive of DAVF-related parkinsonism. Typical angiographical features of this condition include the location of the DAVF at the TSS, reflux into the straight sinus, and probable venous congestion of the basal ganglia. Three unsuccessful treatments were reported in a case of transarterial embolization without attempted transvenous embolization in a comatose patient (Case 4),⁵⁾ in a case of delayed diagnosis with diffuse cerebral atrophy with

thrombosed straight sinus (Case 9),⁷⁾ and in a case of multifocal distal shunt points at the SSS (Case 10).⁷⁾ On the other hand, reversibility seems possible even after 12 months from onset (Case 5).⁴⁾ Therefore, it is important to recognize that parkinsonism and dementia due to DAVF is reversible if embolization is achieved successfully.

In our case, sinus stenoses were multiple and future de novo DAVF was possible, so selective transvenous coil embolization of the venous pouch preserving the normal dural sinus was performed, and that was advantageous for improving venous congestion, restoring normal venous drainage, and for preserving the access route of transvenous embolization for future de novo lesions. In summary, although rare, DAVF is a potential cause of parkinsonism and severe akinetic mutism, which can be reversed by appropriate and timely endovascular treatment.

Conflicts of Interest Disclosure

The authors have no personal financial or institutional interest in any of the drugs, materials, or devices in the article. All authors who are members of The Japan Neurosurgical Society (JNS) have registered online Self-reported COI Disclosure Statement Forms through the website for JNS members.

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Address reprint requests to: Tomoji Hattori, MD, Department of Neurosurgery, Ichinomiya Municipal Hospital, 2-2-22 Bunkyo, Ichinomiya, Aichi 491-8558, Japan.
e-mail: hatket@nifty.com