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Periodic Limb Movements in Sleep After a Small Deep Subcortical Infarct

Periodic limb movements in sleep (PLMS) are characterized by episodes of repetitive, stereotyped, involuntary movements during sleep that typically recur at 5- to 90-second intervals.¹ There is no consensus on the pathogenesis of PLMS; however, their excellent response to dopaminergic drugs and worsening with dopamine receptor blockers² suggest that the dopamine system plays a key role in the pathogenesis of PLMS. We describe a patient who showed PLMS after a small deep subcortical infarct involving the pallidum.

A 58-year-old, right-handed man was admitted to our hospital with a 12-hour history of right hemiparesis. He had a 5-year history of hypertension but there was no other personal or family history of medical or neurological diseases. He and his wife denied any history of sleep problems or involuntary movements. On neurological examination, he was alert, attentive, and communicative, and had intact higher cortical functions. There was hemiparesis of the right extremity (Grade III of V by manual muscle testing) with decreased sensation of pain, temperature, and vibration. Increased deep tendon reflexes and Babinski’s sign were observed in the right extremities. Two days after admission, his caregiver reported abnormal movements of the right leg during sleep. Initially, these involuntary movements occurred intermittently, even in a daytime sleep. As time passed, they appeared only in the night, leading to severe sleep disturbance. These involuntary movements consisted of a mixture of repetitive dorsiflexion of the ankle and great toe, with subsequent flexion of the knee, which were con-

fined mostly to the right leg. He said that unpleasant sensation in the right foot sometimes preceded the involuntary movements. Motor power of the leg as well as the arm improved gradually (Grade \leq III) 5 days after admission.

Brain magnetic resonance images taken the day after admission demonstrated acute discrete infarction in the posterior limb of the left internal capsule and the adjacent pallidum (Fig. 1). Brain single photon emission computed tomography (SPECT) using $99m$ Tc-ethyl cysteinate dimer showed a markedly decreased perfusion in the left basal ganglia. Nocturnal video monitoring with surface electromyography demonstrated typical periodic involuntary movements of the right leg, consisting of flexion of the toe and the knee. These involuntary movements appeared every 14 to 60 seconds and showed periodic bursts of compound muscle action potentials lasting 1 to 2 seconds. The consecutive contraction of the left leg rarely appeared with very small compound muscle action potentials. Polysomnography revealed a total sleep time of 413.5 minutes, a sleep efficiency of 86.8% and a PLMS index of 40.3 per hour. Immediately with the introduction of levodopa (L-dopa; 100 mg/day) with carbidopa at night, the patient and his family reported a marked improvement of the involuntary leg movements during sleep. Discontinuation of L-dopa aggravated the involuntary leg movements again. Three months after L-dopa medication, the patient and his family reported that the movements were controlled well with medication.

Our patient had no history of sleep disturbance and the temporal relationship between the infarction and PLMS was obvious. The involuntary leg movements in the patient (\leq our patients) met the diagnostic criteria for PLMS^{1,3} but did not fulfill the essential criteria for restless legs syndrome (RLS).⁴ To the best of our knowledge, this is the first report of a structural lesion of the basal ganglia associated with PLMS.

As well as pharmacologic studies of dopaminergic drugs, the dopamine hypothesis in the pathogenesis of PLMS has been supported by dopamine receptor studies using SPECT or positron emission tomography (PET) images. Ruottinen and colleagues⁵ reported mild nigrostriatal presynaptic dopamine hypofunction, and Michaud and associates⁶ and Staedt and coworkers⁷ demonstrated decreased the striatal dopamine D2 receptor binding in patients with PLMS with RLS. In addition, neurophysiological studies of flexor reflexes, which resemble PLMS clinically and are known to share common mechanisms with PLMS, demonstrate that dopaminergic substances suppress the exaggerated flexor response,⁸ further strengthening the role of dopaminergic system in the pathogenesis of PLMS. The dopamine abnormality responsible for the PLMS may be mediated by dysfunction of the nigrostriatal dopaminergic pathway (either presynaptic or postsynaptic) or other descending diencephalo-spinal pathways.⁹

It has been suggested that PLMS may be the result of pyramidal tract involvement. However, motor improvement in our patient was not correlated with improvement of PLMS. A similar discrepancy between pyramidal tract dysfunction and the severity of PLMS has been found in studies of PLMS in spinal cord lesions.¹⁰ Pyramidal tract involvement may therefore give a minor contribution to the pathogenesis of PLMS.

The generator of PLMS has not been elucidated but their occurrence in patients with various spinal cord lesions indicates that spinal mechanisms are involved. Combined with previous dopamine receptor studies, it seems reasonable to hypothesize

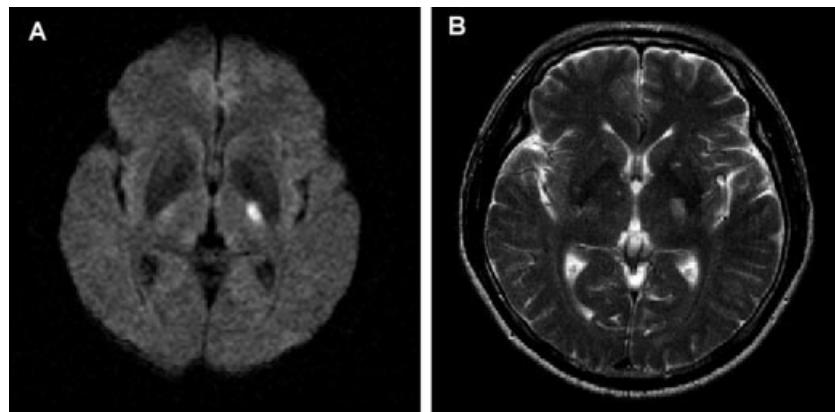


FIG. 1. Brain magnetic resonance imaging taken the day after admission reveals a small, discrete infarction involving the posterior limb of the left internal capsule and adjacent pallidum on diffusion-weighted (A) and T2-weighted imaging (B).

that PLMS result from the loss of supraspinal inhibitory influences secondary to dysfunction of the basal ganglia dopaminergic system. From a clinical viewpoint, the PLMS in our patient with basal ganglia lesion that responded to dopaminergic medication provide the additional evidence for the role of the dopaminergic system in pathogenesis of PLMS.

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Dietary Herb Extract Dai-Kenchu-To Ameliorates Constipation in Parkinsonian Patients (Parkinson's Disease and Multiple System Atrophy)

Dai-Kenchu-To (DKT) (50% ginger, 30% “Nin-jin” [ginseng], and 20% “Sansho” [Japanese pepper, *Zanthoxylum*]) is a dietary herb extract used for relieving abdominal distension in Eastern countries. Because the extract is composed of food additives generally considered safe and time-tested, we performed an open trial of DKT’s effects on constipation in parkinsonian patients, using objective measures derived from lower-gastrointestinal tract (GIT) motility tests.

We recruited 10 male patients (mean age, 72 years; range, 66–77 years, mean duration of disease 4 years; range, 3–8 years). None was taking anticholinergic agents. Six patients had Parkinson’s disease (PD; median Hoehn–Yahr grade 3; range, 2–3) and were all independent in walking, taking a mean daily levodopa dose 200 mg (range, 100–400 mg). The other 4 patients had multiple system atrophy (MSA); only 1 was independent in walking. Two MSA subjects received L-dopa treat-

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