Neurology Asia 2010; 15(1) : 93 – 95

Unilateral periodic limb movements during sleep as a prodromal stroke symptom: A case report

Kwang Lae Lee *MD*

Department of Rehabilitation Medicine, National Rehabilitation Hospital Seoul, Korea.

Abstract

Periodic limb movements in sleep (PLMS) is characterized by recurrent episodes of repetitive, stereotyped limb movements that occur during sleep. There have been a few reports on this condition developing after a stroke, but the movements in these reports were not prodromal stroke symptoms. We describe a case in which PLMS developed as a prodromal stroke symptom. A 78-year-old man experienced right hemiplegia and dysarthria following left striatocapsular infarct. After acute care, he was transferred to our center for rehabilitation. During rehabilitation, he complained of right-side involuntary leg movements during sleep. According to his history, it had been developed 10 days before the appearance of right hemiplegia. Polysomnography confirmed the diagnosis of PLMS. We propose that PLMS can be a prodromal stroke symptom resulting from a pyramidal tract lesion without involvement of sensory components.

INTRODUCTION

Periodic limb movement in sleep (PLMS) is characterized by recurrent episodes of repetitive and highly stereotyped limb movements.[1] It occurs in clusters while the subject is sleeping, primarily during non-rapid eye movement (non-REM) sleep.[2] It is confirmed by surface electromyographic recordings usually of bilateral tibialis anterior muscles during polysomnography following standard criteria.[3] Although various conditions are known to cause PLMS, stroke as a cause of PLMS is rare with only a few reported cases in the literature. PLMS in all of these cases developed after stroke.[2,4,5] We present a patient in whom PLMS developed as a prodromal stroke symptom before the appearance of main stroke symptoms and discuss the possible pathophysiological mechanisms.

CASE REPORT

A 78-year-old right-handed man was admitted to the Department of Neurology in a university hospital, complaining of sudden onset of right hemiplegia and dysarthria. His brain magnetic resonance imaging on the day that he developed these symptoms showed acute striatocapsular infarction (Figure 1). Carotid duplex sonography revealed multiple plaques in the left common and internal carotid arteries. After acute care, he was transferred to our center for rehabilitation. His only medical history consisted of hypertension and appendectomy. On examination, he had mild motor weakness (grade 4/5) of the right limbs without any sensory deficit. Dysarthria was improved. Deep tendon reflexes were increased in the right limbs with a positive Babinski's sign. The score on Mini-Mental State Examination was 28/30. The rest of the neurologic examination was unremarkable.

During his rehabilitation, he complained of involuntary movements of the right lower limb, appearing only when asleep, which awoke him from sleep. He and his family reported that these movements had started 10 days before the appearance of right hemiplegia and dysarthria. He denied any associated unpleasant sensation or need to move. Video monitoring during sleep revealed a mixture of repetitive dorsiflexion of the ankle, and subsequent jerking of the knee and hip. These movements occurred periodically, at about 40-second intervals. The left limbs were not involved. Nocturnal polysomnography confirmed the diagnosis of PLMS.[6]

He did not take any drugs associated with this disorder, such as fluoxetine, venlafaxine, clomipramine or lithium, and denied any family history of neurologic disease.[1] Laboratory test results, such as peripheral blood counts, serum electrolytes, erythrocyte sedimentation rate, glucose, iron, ferritin, uric acid, total iron binding capacity, liver, thyroid, and renal function were normal. Electroencephalogram, electrocardiogram, echocardiogram, and chest radiogram findings

Address correspondence to: Kwang Lae Lee MD, Department of Rehabilitation Medicine, National Rehabilitation Hospital, 111, Gaori-gil, Gangbuk-gu, Seoul, 142-884, Korea. Telephone: 82-2-901-1596, Fax: 82-2-902-3835, E-mail: 78zzz@naver.com

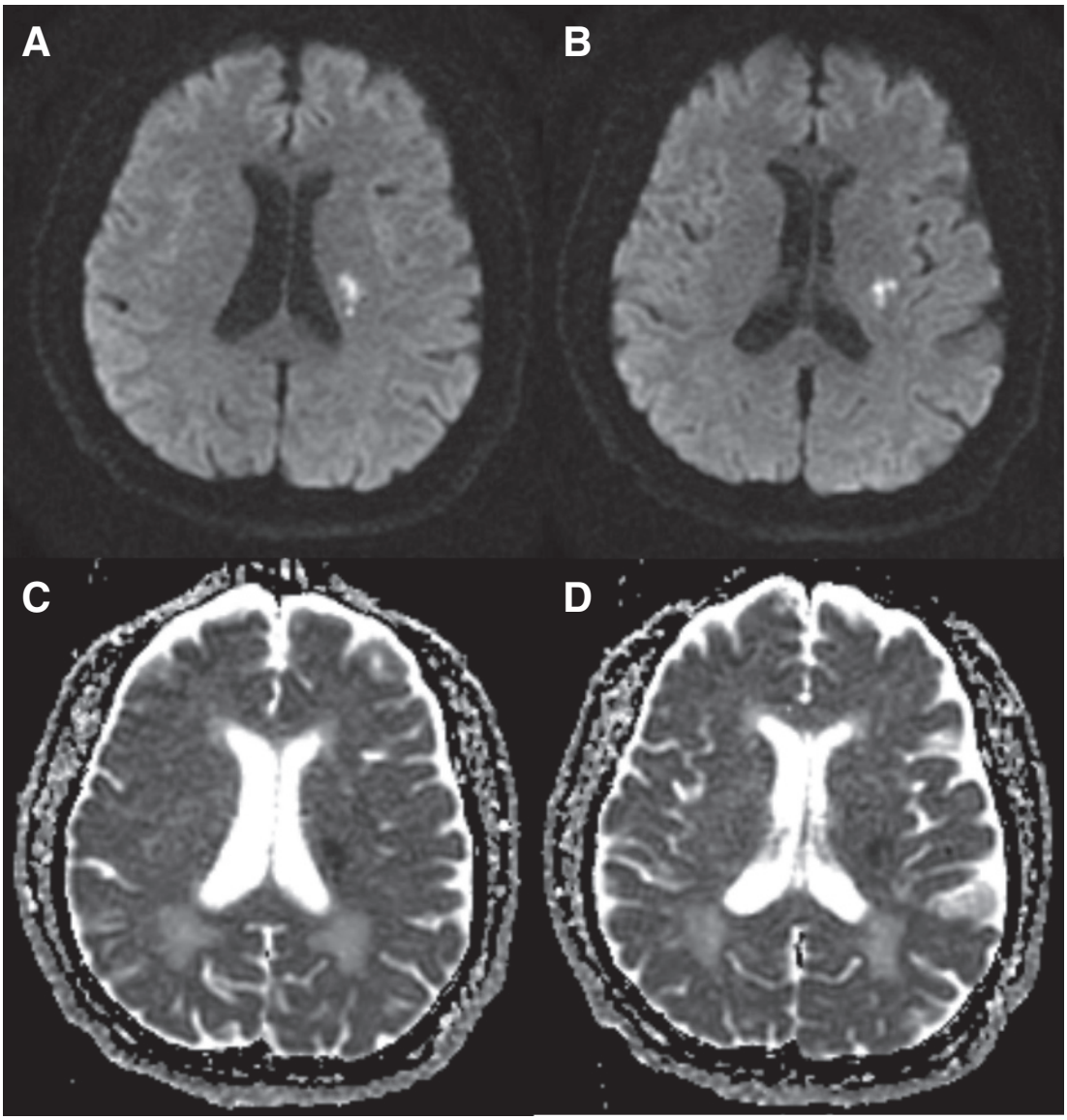

Figure 1: Diffusion-weighted images (A and B) show focal high signal intensities in left posterior basal ganglia and posterior limb of internal capsule and apparent diffusion coefficient maps (C and D) show low signal intensities in the same areas, compatible with acute cerebral infarct.

were unremarkable. Electrophysiological investigations excluded peripheral neuropathy or lumbosacral radiculopathy.

DISCUSSION

The involuntary leg movements in this patient, considering the clinical and polysomnographic findings, met the diagnostic criteria for PLMS.[3] PLMS occurs in more than 80% of patients with restless leg syndrome (RLS).[7] On account of the frequent association between PLMS and RLS, they are thought to have a common etiology, but how far they share pathogenetic mechanisms is still unclear and it is important to differentiate RLS from PLMS.[3] His symptoms did not fulfill the essential criteria for RLS because there was no unpleasant sensation and need to move, and they occurred only while he was asleep.[3]

Besides stroke, PLMS has been reported to be associated with peripheral neuropathy, uremia, anemia, gastric surgery, chronic pulmonary disease, myelopathy, various medications, and epidural or spinal anesthesia.[2,8] In this patient, none of these factors was present. The close temporal relationship between the onset of PLMS and stroke, and the occurrence of unilateral

PLMS only in the hemiplegic limb supports the contention that his PLMS was associated with the cerebral infarct. In addition, no involuntary movements were experienced by him before the onset of PLMS.

Although the anatomical structure responsible for stroke-related PLMS has not been clarified, a few reports have described an association with pontine, basal ganglia and corona radiata lesions.[2,4,8-10] Anatomical structures responsible for stroke-related RLS are similar. These lesions are in areas of the pyramidal tract and the basal ganglia-brainstem axis that are involved in motor functions and sleep-wake cycle.[11] PLMS without RLS has been reported in movement or pyramidal system disorders such as stiff-person syndrome, Huntington's disease, and amyotrophic lateral sclerosis.[12,13] It is thought that PLMS without RLS may be related to a lesion of the motor system without involvement of the sensory system. The pathogenesis of stroke-related PLMS is still under discussion and as mentioned above, how far PLMS and RLS share pathogenetic pathways is still unclear. Because of the relationship between sleep and PLMS, the generator of PLMS is presumed to be associated with sleep-related structures such as the reticular activating system. This postulation was supported by a study using functional MRI, which pointed to the reticular structure in the brainstem as the primary generator of PLMS.[14] The involuntary leg movements observed in PLMS are similar to Babinski's sign, which is usually observed in pyramidal tract lesions.[4] In addition, PLMS appears preferentially during non-REM sleep, in which the nocturnal Babinski's sign is normally observed.[15] Considering these known facts, it is proposed that PLMS without RLS may be the result of a pyramidal tract lesion without involvement of sensory components (in which there is loss of cortical or subcortical inhibition on the brainstem generator). The fact that our patient did not notice any sensory problems provides additional evidence to support this hypothesis.

In a report about prodromal stroke symptoms, episodic or prolonged prodromal symptoms preceded main stroke symptoms by a period of one to ten days, and were postulated to be caused by hypoperfusion or lacunar infarction of strategic sites.[16] Similarly, it is assumed that our patient's PLMS was caused by supraspinal disinhibition which had resulted from hypoperfusion or lacunar infarction. However, our study's main limitation is the lack of objective documentation of the PLMS prior to the onset of the patient's main stroke symptoms.

REFERENCES

1. Hornyak M, Feige B, Riemann D, Voderholzer U. Periodic leg movements in sleep and periodic limb movement disorder: prevalence, clinical significance and treatment. *Sleep Med Rev* 2006; 10:169-77.
2. Kim JS, Lee SB, Park SK, Han SR, Kim YI, Lee KS. Periodic limb movement during sleep developed after pontine lesion. *Mov Disord* 2003; 18:1403-5.
3. Stiasny K, Oertel WH, Trenkwalder C. Clinical symptomatology and treatment of restless legs syndrome and periodic limb movement disorder. *Sleep Med Rev* 2002; 6:253-65.
4. Kang SY, Sohn YH, Lee IK, Kim JS. Unilateral periodic limb movement in sleep after supratentorial cerebral infarction. *Parkinsonism Relat Disord* 2004; 10:429-31.
5. Rossetti AO, Heinzer R, Espa F, Tafti M. Unilateral periodic leg movements during wakefulness and sleep after a parietal hemorrhage. *Sleep Med* 2008; 9:465-6.
6. Allen RP, Picchietti D, Hening WA, Trenkwalder C, Walters AS, Montplaisi J. Restless legs syndrome: diagnostic criteria, special considerations, and epidemiology. A report from the restless legs syndrome diagnosis and epidemiology workshop at the National Institutes of Health. *Sleep Med* 2003; 4:101-19.
7. Montplaisir J, Boucher S, Poirier G, Lavigne G, Lapierre O, Lesperance P. Clinical, polysomnographic, and genetic characteristics of restless legs syndrome: a study of 133 patients diagnosed with new standard criteria. *Mov Disord* 1997; 12:61-5.
8. Iriarte J, Alegre M, Arbizu J, de Castro P. Unilateral periodic limb movements during sleep in corticobasal degeneration. *Mov Disord* 2001; 16:1180-3.
9. Lee JS, Lee PH, Huh K. Periodic limb movements in sleep after a small deep subcortical infarct. *Mov Disord* 2005; 20:260-1.
10. Sechi G, Agnetti V, Galistu P, *et al*. Restless legs syndrome and periodic limb movements after ischemic stroke in the right lenticulostriate region. *Parkinsonism Relat Disord* 2008; 14:157-60.
11. Lee SJ, Kim JS, Song IU, An JY, Kim YI, Lee KS. Poststroke restless legs syndrome and lesion location: anatomical considerations. *Mov Disord* 2009; 24:77-84.
12. Lugaresi E, Cirignotta F, Coccagna G, Montagna P. Nocturnal myoclonus and restless legs syndrome. *Adv Neurol* 1986; 43:295-307.
13. Martinelli P, Pazzaglia P, Montagna P, *et al*. Stiff-man syndrome associated with nocturnal myoclonus and epilepsy. *J Neurol Neurosurg Psychiatry* 1978; 41:458-62.
14. Bucher SF, Seelos KC, Oertel WH, Reiser M, Trenkwalder C. Cerebral generators involved in the pathogenesis of the restless legs syndrome. *Ann Neurol* 1997; 41:639-45.
15. Smith RC. Relationship of periodic movements in sleep (nocturnal myoclonus) and the Babinski sign. *Sleep* 1985; 8:239-43.
16. Lee H, Cho YW. Auditory disturbance as a prodrome of anterior inferior cerebellar artery infarction. *J Neurol Neurosurg Psychiatry* 2003; 74:1644-8.