

Unilateral Periodic Limb Movement in Sleep after Ischemic Brain Stroke: A Case Report

Hamed Amirifard, Arezu Najafi, Elham Afzalinejad*

Occupational Sleep Research Center, Baharloo Hospital, Tehran University of Medical Sciences, Tehran, Iran

Received: 23 Nov. 2018 Accepted: 20 Dec. 2018

Abstract

Background and Objective: Various types of abnormal movements such as pain, cramp, jerk, creeping, or itching may occur during sleep, many of which often involve the legs. In this study, we reported a case of periodic limb movement in sleep (PLMS) in the setting of a neurological disease.

Case Report: We report a patient with involuntary left leg movements during sleep. The patient developed this problem after an ischemic brain stroke that involved right temporal and basal ganglia. The patient underwent an overnight polysomnography (PSG) for the diagnosis of PLMS.

Conclusion: Although we do not know the exact pathogenesis of PLMS, it has been proposed that the brain lesions might cause PLMS. The present case provided evidence to support that brain lesions could be considered as a cause of unilateral PLMS.

© 2019 Tehran University of Medical Sciences. All rights reserved.

Keywords: Excessive sleep-related periodic leg movements; Stroke; Polysomnography; Restless legs syndrome

Citation: Amirifard H, Najafi A, Afzalinejad E. **Unilateral Periodic Limb Movement in Sleep after Ischemic Brain Stroke: A Case Report.** *J Sleep Sci* 2019; 4(1-2): 48-51.

Introduction

Various types of abnormal movements such as pain, cramps, jerks, creeping, or itching may occur during sleep, many of which often involve legs. Periodic limb movement disorder (PLMD), which was originally named nocturnal myoclonus by Symonds (1), is characterized by periodic episodes of repetitive limb movements caused by contractions of muscles during sleep. Restless legs syndrome (RLS), which was initially reported by Ekbom (2), is characterized by strong urge to move legs. There is often an unpleasant feeling in legs that improves partially by moving them at rest circumstances. This is often described as aching, tingling, or crawling in nature. Moving the affected part of the body modulates the sensations and provides temporary relief (2).

Periodic limb movements during sleep (PLMS) are repetitive leg movements occurring in a series

with an interval between 5 to 90 seconds (3). The real prevalence of PLMD is still unknown (4). In 2016, however, a population-based study on 2162 adult subjects from the HypnoLaus Sleep Cohort Study found that the prevalence of PLMS more than 15/hour was 28.6% (3).

PLMS even occurs in healthy individuals. It is known that up to 28% of individuals have PLM index > 15/hour. PLMS can activate autonomic system and increase heart rate (HR), which is considered as a risk factor for cerebrovascular and cardiovascular diseases (CVDs). Patients with PLMS are likely to have over hundred events at each night, potentially for many years (5).

Although PLMS was related to the occurrence of stroke, the role of stroke on the pathogenesis of secondary PLMS is remained underestimated (6). In patients with RLS or PLMS secondary to stroke (6, 7), associations between lesions of the pons or corona radiata (CR) and post-stroke RLS or post-stroke PLMS are reported. However, only a small number of cases of post-stroke RLS and post-stroke PLMS is reported. Some authors have

* **Corresponding author:** E. Afzalinejad, Occupational Sleep Research Center, Baharloo Hospital, Tehran University of Medical Sciences, Tehran, Iran
Tel: +98 21 55460184, Fax: +98 21 55648189
Email: e.afzalinejad94@gmail.com

suggested that disinhibition plays a role in the mechanism of the both conditions (8).

Case Report

The patient was a 71 years old woman, who was admitted in our sleep clinic complaining of limb movement during sleep. She reported severe symptoms that awakened her during sleep. These movements started from the precedent two years after a neurologically confirmed ischemic brain stroke. Episodes repeated nearly every night with more than 30 times per night. Her children reported that these episodes were more frequent at first half of the sleep. Unilateral dorsiflexion of left foot and hip flexion was frequently occurred while she was sleeping. After a careful enquiry, her husband also reported that movements were more severe in left side.

The movements disappeared after awakening. During some movements, she awakened and then resumed to sleep. That led to fragmented sleep. No history of snoring, witnessed apnea, sleep talking or sleep walking, rapid eye movement (REM) sleep behavior disorder, or other parasomnias was detected.

She had a 10-year history of diabetes mellitus (DM) and hypertension (HTN). She had a history of ischemic heart disease (IHD) and atrial fibrillation (AF) that were under medical treatment. She had a history of ischemic cerebrovascular accident (CVA) 2 years ago with history of hospitalization in a neurologic ward for 12 days.

She was taking 100 mg metoprolol, 20 mg rivaroxaban, 0.25 mg digoxin, 10 mg atorvastatin, and 10 mg amlodipine daily. General clinical examination and pulse rate were normal at the visit time.

The Epworth Sleepiness Scale (ESS) and Beck Depression Inventory (BDI) scores were estimated as 12 and 17, respectively.

Neurologic examination showed a normal mental status. Cranial nerves were normal except mild left central facial palsy. In motor examination, left upper and lower limbs forces were 4/5 and right side was 5/5. Deep tendon reflexes (DTRs) in left side increased (3+) and left plantar reflex was upward (Babinski +). Mild spasticity in left lower limb was detected. Sensory examinations were normal and no cerebellar symptoms were detected. Her gait was paretic; however, she could walk without any aid.

She showed infarction of right deep temporal region with involvement of internal capsule and

basal ganglia in brain magnetic resonance imaging (MRI) (Figure 1). In recent imaging, gliosis in this region was occurred.

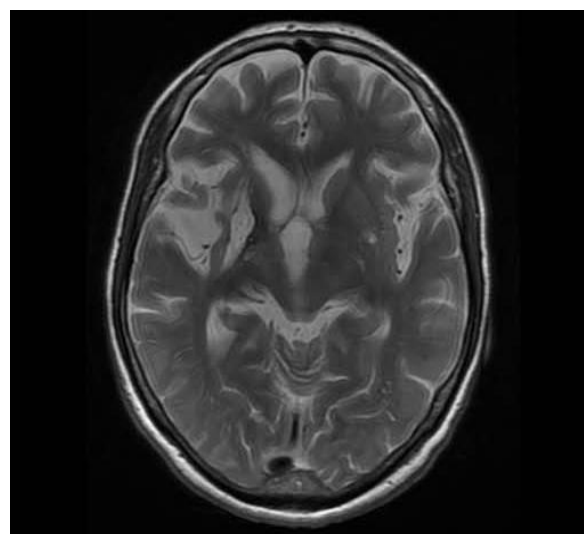


Figure 1. Axial T2 magnetic resonance imaging (MRI) of the patient depicting infarction of right deep temporal region with involvement of internal capsule and basal ganglia

One-night full polysomnography (PSG) with Embla N7000 was performed to evaluate sleep phenomena. The results of PSG were analyzed by two trained sleep specialists.

Total sleep time was measured 112 minutes with 15 awakening episodes. Sleep latency was estimated as 118 minutes. She spent 367 minutes in bed and had sleep efficiency of 32.1%.

In this regard, stage 1, 2, 3, and REM were 25%, 48%, 16%, and 11%, respectively. She had one central apnea and five obstructive apneas and hypopneas, which gave a total respiratory disturbance index (RDI) of 3.2. The mean oxygen saturation (SpO₂) during sleep time was 91.6% and the minimum SpO₂ was 80%. SpO₂ was between 90-100 percent for 91% of the times.

She had 112 leg movements during sleep. All of them were in left side. Minimum period of each movement was 2-9 seconds and was separated by an interval of 20-40 seconds. These movements were stereotypic extension of great toe and triple flexion of knee and hip.

The total arousals consisted of spontaneous electroencephalography (EEG), respiratory, and limb movement arousal events. From total arousals, 30 were of spontaneous nature (3.9 events/hour). A total of one arousal was associated with respiratory events. There was no evidence of epileptic dis-

charge during analysis and all episodes were carefully noticed for possibility of any type of epilepsy. The possibility of focal motor seizure, i.e., cortical myoclonus was also excluded.

Discussion

Although the main pathogenesis of the RLS and PLMS has not yet been determined, the relationship between PLMS and disruption of brainstem, anemia, uremia, peripheral neuropathy, radiculopathy, and spinal cord lesion has been reported in several studies. Several medications, epidural and spinal anesthesia, gastric surgery, and chronic pulmonary diseases are reported to probably be concomitant with unilateral PLMS (6, 9). Some authors reported that the cerebellum had a pivot role in PLMS (9). Others have reported that ischemic lesions in right lenticulostriate region of brain had a potential role in the pathophysiology of PLMS and RLS (10).

In some studies, unilateral PLMS without RLS symptoms were diagnosed in patients with cerebral supratentorial infarction, cerebellar lesion, and some types of limb affecting corticobasal degeneration (CBD) (11). Kang *et al.* reported a patient with unilateral right leg movements during sleep after an acute ischemic stroke in the CR. Overnight PSG confirmed the diagnosis of PLMS. Although the pathogenesis of PLMS is still controversial, Kang *et al.* indicated that the loss of cortical or subcortical inhibition because of the pyramidal tract lesion might cause PLMS (7).

The involuntary leg movement in our patient, when considering its clinical presentation and PSG findings, was consistent with PLMS. But only left lower extremity was affected.

Patients usually do not notice their PLMs in pure PLMS, because it occurs during sleep. In our case, however, the symptoms were severe and led to sleep fragmentation and excessive sleepiness. A similar association has been shown by previous studies (12).

The possibility of focal motor seizure, i.e., cortical myoclonus, was excluded by careful EEG evaluation during the leg movements and matching of leg electromyography (EMG) activity.

PLMS is estimated to affect one third of general population (3); therefore, the development of PLMS in this patient might be coincidental. However, the close temporal association between PLMS onset and stroke, and unilateral PLMS in

the paretic leg supported the idea that PLMS in this patient may be due to the brain ischemia in the CR. In addition, she and her family did not notice this leg movement prior to the stroke.

The pathogenesis of PLMS has not yet been fully clarified. The association of PLMS with wide range of clinical conditions makes it difficult to establish a single mechanism for its effect. PLMS may be associated with sleep-related structures such as the ascending reticular activating system (RAS). This correlation was confirmed by a functional MRI study that indicated the reticular structure in the brainstem as the primary generator of PLMS (13). Unilateral PLMS associated with lesions near the ascending reticular formation provides additional evidence to support the role of this structure in generating PLMS (6). Unilateral PLMS has been observed in neurodegenerative diseases. Unilateral PLMS is seen in the symptomatic limbs of the patients with CBD (14). Accordingly, it has been proposed that PLMS is the result of a pyramidal tract lesion. The loss of cortical or subcortical inhibition exerting on the brainstem generator might cause PLMS (6, 14).

In this patient, PLMS persisted after recovery of leg weakness. However, recovery of motor weakness does not directly indicate that pyramidal tract is functionally intact. Various upper motor neuron signs including Babinski's sign and spasticity often persist after recovery of motor weakness. It is unclear why PLMS sometimes occurs in patients with CVA. Various lesions in the brainstem, spinal cord, and cerebrum can cause secondary PLMS or unilateral PLMS and should be considered in sleep clinic settings.

Conclusion

Any lesion in the corticospinal tract or brainstem generator can cause PLMS. In patients with unilateral PLMS, the lesions of these brain areas should be considered in clinical evaluations.

Conflict of Interests

Authors have no conflict of interests.

Acknowledgments

Authors would like to thank the patient for her kind contribution. We also are grateful to the staff of Imam Sleep Lab.

References

1. Symonds CP. Nocturnal myoclonus. *J Neurol Neurosurg Psychiatry* 1953; 16: 166-71.
2. Walters AS. Toward a better definition of the restless legs syndrome. The International Restless Legs Syndrome Study Group. *Mov Disord* 1995; 10: 634-42.
3. Heinzer R, Haba-Rubio J, Tafti M. HypnoLaus sleep cohort study. *Rev Med Suisse* 2011; 7: 2137-1. [In French].
4. Hameed S, Tan EK. Restless legs syndrome: Diagnostic criteria and differential diagnosis. In: Ondo WG, editor. *Restless legs syndrome: Diagnosis and treatment*. Boca Raton, FL: CRC Press 2016. p. 173-84.
5. Manconi M, Fanfulla F, Ferri R, et al. Periodic limb movements during sleep in stroke/TIA: Prevalence, course, and cardiovascular burden. *Neurology* 2018; 90: e1663-e1672.
6. Kim JS, Lee SB, Park SK, et al. Periodic limb movement during sleep developed after pontine lesion. *Mov Disord* 2003; 18: 1403-5.
7. Kang SY, Sohn YH, Lee IK, et al. Unilateral periodic limb movement in sleep after supratentorial cerebral infarction. *Parkinsonism Relat Disord* 2004; 10: 429-31.
8. Woo HG, Lee D, Hwang KJ, et al. Post-stroke restless leg syndrome and periodic limb movements in sleep. *Acta Neurol Scand* 2017; 135: 204-10.
9. Lee KL. Unilateral periodic limb movements during sleep as a prodromal stroke symptom: A case report. *Neurol Asia* 2010; 15: 93-5.
10. Kizilay F, Ozkaynak S, Hatipoglu E, et al. Periodic limb movement during sleep following cerebellar infarct. *Acta Neurol Belg* 2010; 110: 284-6.
11. Valko PO, Siccoli MM, Bassetti CL. Unilateral RLS with predominantly ipsilateral PLMS and variable response to dopaminergic drugs: a variant of idiopathic RLS? *Eur J Neurol* 2009; 16: 430-2.
12. Leary EB, Moore HE, Schneider LD, et al. Periodic limb movements in sleep: Prevalence and associated sleepiness in the Wisconsin Sleep Cohort. *Clin Neurophysiol* 2018; 129: 2306-14.
13. Bucher SF, Seelos KC, Oertel WH, et al. Cerebral generators involved in the pathogenesis of the restless legs syndrome. *Ann Neurol* 1997; 41: 639-45.
14. Iriarte J, Alegre M, Arbizu J, et al. Unilateral periodic limb movements during sleep in corticobasal degeneration. *Mov Disord* 2001; 16: 1180-3.