

A patient with insomnia due to restless leg and periodic limb movement syndrome after stroke–A clinical case

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Citation: Yankova A, Georgiev K, Dimitrova D, Dimitrova-Kirilova V, Nestorova V. A patient with insomnia due to restless leg and periodic limb movement syndrome after stroke–A clinical case. *Electron J Gen Med.* 2023;20(2):em445. <https://doi.org/10.29333/ejgm/12778>

ARTICLE INFO

Received: 06 Nov. 2022

Accepted: 27 Dec. 2022

ABSTRACT

Restless legs syndrome (or Willis-Ekbom) presents with an overwhelming need for movement of the lower limbs in order to eliminate the unpleasant sensations in them, which appear during rest and sleep. The unpleasant sensations are described as “tingling, pulling or pain” in the knees, ankles, or through the entire lower limbs. Periodic limb movement syndrome presents with involuntary movements of the lower limbs or the body during sleep, for which the patient has no recollection, unlike in those in restless leg syndrome. In most cases they present with four or more successive movements of dorsal flexion of the toes and ankles with partial flexion of the knees and hips divided into five to 90 second intervals.

A 44-year-old man, a taxi driver, is admitted in a neurological clinic with complaints of a headache, dizziness, exhaustion, which he associates with difficulty falling asleep and maintaining a sleeping state after an ischemic stroke in the right middle cerebral artery four months earlier. The conducted polysomnography reveals an increased sleep latency, periodic limb movement syndrome and multiple arousals before entering REM sleep.

Keywords: insomnia, restless leg, periodic limb movement

INTRODUCTION

Restless legs syndrome (or Willis-Ekbom) presents with an overwhelming need for movement of the lower limbs in order to eliminate the unpleasant sensations in them, which appear during rest and sleep. The unpleasant sensations are described as “tingling, pulling or pain” in the knees, ankles, or through the entire lower limbs. Periodic limb movement syndrome presents with involuntary movements of the lower limbs or the body during sleep, for which the patient has no recollection, unlike in those in restless leg syndrome. In most cases they present with four or more successive movements of dorsal flexion of the toes and ankles with partial flexion of the knees and hips divided into five to 90 second intervals.

Secondary restless leg syndrome can develop after a stroke (in 12.4% of cases), mainly if there is damage to the basal ganglia, which regulate the motor functions and are part of the wake-sleep cycle. In 30% the syndrome is unilateral and in 70%-bilateral. In most cases it is accompanied by periodic limb movement syndrome. The most likely cause for the two conditions are lesions of the striatum, disinhibition of the sensory-motor cortex and the descending inhibitory pathways, with a prevailing activity of the direct extrapyramidal pathway. Both syndromes can lead to post stroke insomnia.

METHODS AND RESULTS

A 44-year-old man was admitted in a neurological clinic with complaints for two months of a dull and pulsating headache, dizziness and exhaustion. Four months earlier he had an ischemic stroke in the right medial cerebral artery, which was treated with fibrinolysis with a full clinical recovery. He has had difficulty falling asleep, maintaining a sleeping state and a lower quality of sleep ever since, with a sleep duration of four hours at most. The patient, during rest or trying to fall asleep, felt a need to move his lower limbs due to unpleasant sensations in them, which would hinder falling asleep and would lead to many nightly arousals. The patient’s comorbidities included arterial hypertension, hypertriglyceridemia, ischemic heart disease and has had an anterior myocardial infarction and a pulmonary embolism. He did not partake in any harmful habits such as smoking, drinking alcohol, drug use, unhealthy eating and exercised regularly. The patient was a taxi driver with 20 years of work experience and had a shift working schedule.

Upon admission the patient was in a satisfactory state, no pathological signs from his somatic status. The neurological status presented with a left hyperreflexia. The laboratory results revealed high triglycerides, negative results for HIV, syphilis, Ebstein-bar virus, Herpes simplex 1 and 2, Varicella-Zoster virus and *Borrelia Burgdorferi*.

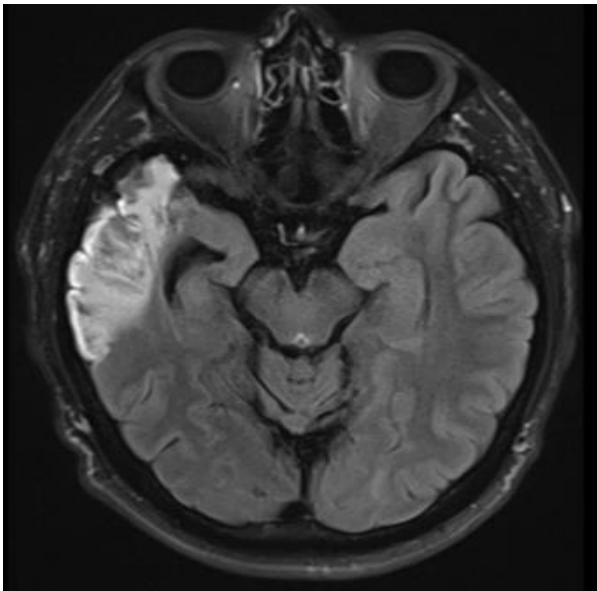


Figure 1. 1 u 2-T1 images of the cerebrum–A zone of encephalomalacia in the right temporal and frontal region with a hemorrhagic transformation-1 (Original MRI performed at the clinic, reprinted with permission of the patient)

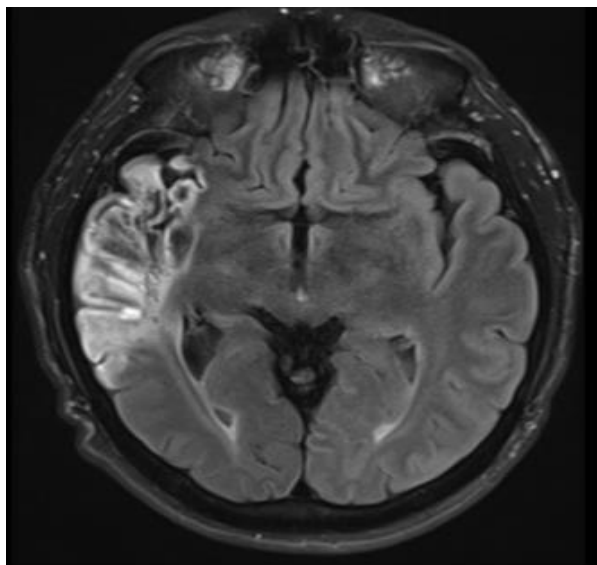


Figure 2. 1 u 2-T1 images of the cerebrum–A zone of encephalomalacia in the right temporal and frontal region with a hemorrhagic transformation-2 (Original MRI performed at the clinic, reprinted with permission of the patient)

The genetic tests for thrombophilia revealed he was heterozygous for the pathological allele for PAI-1 4G-5G, mutation in MTHFR C677T, MTHFR A1298C, factor XIII G103T, and ITGA2 C807T. The computer tomography showed a wide hypodense lesion in the right temporal region and a smaller one in the right frontal region with the characteristics of a chronic ischemic stroke. The brain MRI revealed a wide zone of encephalomalacia with perifocal gliosis in the right temporal (with partial damage to the right putamen) and frontal (mainly the inferior frontal gyrus and a small part of the medium frontal gyrus) and with signs of hemosiderin deposits–signs of a chronic ischemic stroke with a hemorrhagic transformation. The Doppler ultrasound examination of the carotid and vertebral arteries was normal (**Figure 1** and **Figure 2**).



Figure 3. EMG of the lower legs–the arrow shows the moments of movement, which are symmetrical (Original polysomnography, reprinted with permission of the patient)

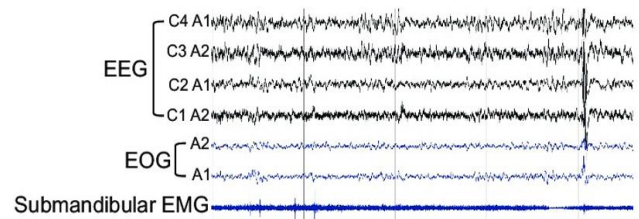


Figure 4. N2 phase, during which movements have appeared (Original polysomnography, reprinted with permission of the patient)

A polysomnography, consisting of pulse-oximetry, electrooculography, electroencephalography, electromyography, electrocardiography, was conducted. It revealed multiple arousals, a decreased total (257 min.) and REM sleep (10% of the total time), a prevailing N2 phase (80% of the total time) and multiple periodic limb movements of the lower limbs during the same phase(50/h). There were no breathing or rhythm disorders registered. The patient reported that prior to falling asleep he had unpleasant sensations in his lower limbs, forcing him to move them in order to find relief (**Figure 3** and **Figure 4**).

DISCUSSION

The sleep movement disorders consist of simple stereotypical, involuntary movements. Those can lead to sleep fragmentation and disturbance and can be primary or connected to another disease (diabetes, cardio-vascular, etc.) [1].

Restless leg syndrome (or Willis-Ekbom disease) presents with an overwhelming need for movement of the lower limbs in order to eliminate the unpleasant sensations in them, which appear during rest and sleep. The unpleasant sensations are described as “tingling, pulling or pain” in the knees, ankles or through the entire lower limbs. The patients have difficulty falling asleep. The complaints are more severe in the night and have a brief improvement in the morning. In time they can spread to the upper limbs. Between five to 15% of the whole population suffers from restless leg syndrome.

A disfunction of the dopaminergic system and lower iron concentrations in certain parts of the brain (mainly substantia nigra and the thalamus) are found in the primary restless leg syndrome. A mutation of the BTBD9 and MEIS1 genes is suspected to be the cause of the disease.

Secondary restless leg syndrome is found in different neurological diseases (diabetic polyneuropathy, Guillain-Barre syndrome, multiple sclerosis), chronic venous insufficiency, autoimmune conditions (Sjögren syndrome, rheumatoid arthritis), chronic kidney diseases, iron deficiency.

The EEG during polysomnography reveals diffuse sharp waves and K-complexes during the movements [2-5].

Periodic limb movement syndrome presents with involuntary movements of the lower limbs or the body during sleep, for which the patient has no recollection, unlike in those in restless leg syndrome. In most cases they present with four or more successive dorsal flexions of the toes and ankles with partial flexion of the knees and hips divided into five to 90 second intervals. It is present in 80 to 90% of the patients with restless leg syndrome and it is theorized that they have the same etiology.

After every movement the EEG has alpha and beta activity with a duration of five seconds and the amplitude of the EMG signal is increased [6].

Taxi drivers' work is associated with a higher stress, shift work, irregular resting hours, incorrect eating habits, usually fast food, rich in fat and salt. Frequent associated diseases are arterial hypertension, hypercholesterolemia, atherosclerosis, diabetes-risk factors for different vascular diseases. Long driving hours are also associated with higher white cell and platelet count and hematocrit, which increase the blood density and the risk for thrombosis [7, 8].

Shift workers are more likely to have different types of stroke due to the increased sympathetic activity, the presence of metabolic syndrome, arterial hypertension, hypercholesterolemia and insulin resistance. From 1998 to 2004 up to 2,226 nurses from 121,701 registered in the American society for nurse health reported the development of a stroke, from which 1,845 had an ischemic one and the rest 381-a hemorrhagic one. The data analysis showed an increase in the prevalence of stroke with 4% for every five years of shift work [9].

The patients with stroke can have a number of sleep disorders, from which the most common one is insomnia-50% of stroke cases. It can be caused by the associated depression but can be due only to the brain damage. Pontomesencephalic strokes can lead to almost total lack of sleep [10], while thalamic ones lead to loss of sleep wave patterns. Left hemisphere strokes decrease NREM, while right hemisphere ones decrease REM sleep. Some of the medication used for the treatment of stroke can affect the length and quality of sleep.

Secondary restless leg syndrome can develop after a stroke (in 12.4% of cases), mainly if there is damage to the basal ganglia, which regulate the motor functions and are part of the wake-sleep cycle. In 30% the syndrome is unilateral and in 70%-bilateral. In most cases it is accompanied by periodic limb movement syndrome. The most likely cause for the two conditions are lesions of the striatum, disinhibition of the sensory-motor cortex and the descending inhibitory pathways, with a prevailing activity of the direct extrapyramidal pathway. Both syndromes can lead to post stroke insomnia [11, 12].

CONCLUSION

Shift work by itself can lead to different vascular diseases. In the presented case the etiology of the ischemic stroke is complex- stress from the work position, shift work, hypercholesterolemia, a possible genetic thrombophilia. Considering the anamnesis of the appearance of the insomnia and restless leg syndrome after the stroke, the imaging results of damage to the right frontal and temporal cortex and putamen, the increased N2 phase, the decreased REM sleep, the multiple registered periodic limb movements, confirms an

insomnia connected to post ischemic stroke restless leg syndrome, accompanied by periodic limb movement syndrome.

Author contributions: All authors have sufficiently contributed to the study and agreed with the results and conclusions.

Funding: No funding source is reported for this study.

Ethical statement: Authors stated that the participant received written informed consent before being admitted to the clinic. Authors further stated that the study has been approved by all the authors and the clinic which treated the patient.

Declaration of interest: No conflict of interest is declared by authors.

Data sharing statement: Data supporting the findings and conclusions are available upon request from the corresponding author.

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