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Video-Clinical Corners

Can body position and sleep stages influence motor patterns in periodic limb movement disorder?

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1. Introduction

Periodic limb movements disorder (PLMs) during sleep is characterized by repetitive and stereotyped limb jerks, with a typical triple flexion of lower limbs that may determine troubled sleep [1]. Periodic limb movements (PLMs) are typically more frequent during stages N1 and N2 while they occur rarely in REM sleep [2]. The putative role of body position and sleep stages in influencing motor patterns in PLMs has been suggested [3]. We describe a patient with position-modulated and sleep-stage-modulated PLMs.

2. Case description

A 67-year-old Caucasian man, with mildly overweight BMI (BMI 25) was admitted to our Sleep Medicine Center complaining of nocturnal agitation and repetitive movements during sleep, without significant daytime sleepiness (ESS = 4). His personal history was significant for virus hepatitis A, mild anxious-depressive disorder and arterial hypertension. An attended video-polysomnography showed a mild-moderate obstructive sleep apnea syndrome (AHI: 17.6/h) and a clinically relevant PLMs (PLM index 36.6/h).

3. Video-analysis

Video 1: At 1.23 a.m., lying on his left side, during stage N2 (Fig. 1), the patient presented sequences of stereotyped complex and abrupt movements. These movements were characterized by a initial extension of the left great toe followed by partial flexion of the homolateral-omolateral knee, ankle and hip, irregular flexion-extension of the left arm and hand, associated with an axial startling movement, lasting 3–5 s. In this period, inter-movements interval (IM) was 29 ± 11 s and respiratory events were absent.

Supplementary video related to this article can be found at https://doi.org/10.1016/j.sleep.2019.04.002.

Video 2: At 2.36 a.m, lying on his back, again in stage N2 (Fig. 1), the patient presented with less complex movements, with a partial flexion of the left leg, partially spreading to the right leg and the hip, without involving the upper limbs. In this period, IMs was 37 ± 9 s and obstructive hypopneas appeared associated with leg movements.

Supplementary video related to this article can be found at https://doi.org/10.1016/j.sleep.2019.04.002.

Video 3: At 4.39 a.m. lying on his back, during REM stage (Fig. 1), the patient presented obstructive hypopneas in the absence of any isolated or periodic movements.

Supplementary video related to this article can be found at https://doi.org/10.1016/j.sleep.2019.04.002.

Both motor patterns, occurring during N2, are preceded by EEG cyclic alternating pattern (CAP) sequences [4], mostly represented by A1 subtypes in the event expressed on left side, and by A2 and A3 subtypes in supine position.

4. Discussion

PLMD has been thought to be related to abnormal and involuntary activation of central pattern generators (CPG) [5]. CPG are composed of complex networks involving spinal and brainstem neurons necessary to produce coordinated and stereotyped

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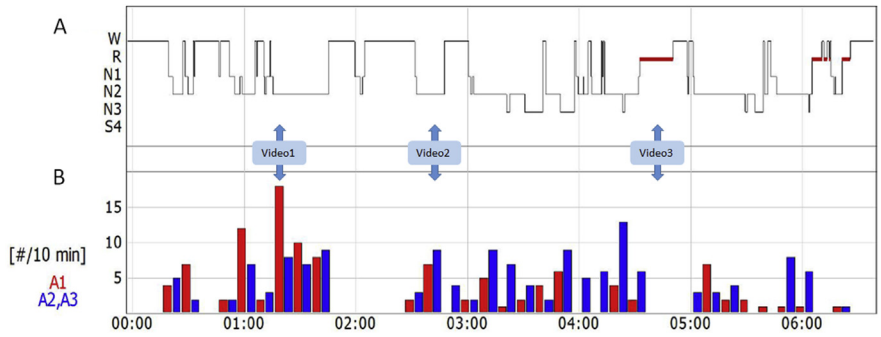

Fig. 1. (A) Hypnogram; (B) Distribution of A phases of CAP during the night. Blue arrows indicate the night-time localization of the proposed videos. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

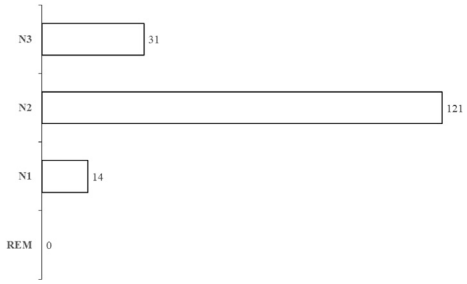

Fig. 2. PLM index across sleep stages.

locomotor movements (eg, walking or swimming) and are critical for survival [5]. Although these circuits are intrinsically able to generate rhythmic motor patterns, sensory inputs can alter their properties [3]. Changes in body position, producing different tactile superficial and proprioceptive deep sensory stimuli, may act as a complex sensory input capable of inducing CPG activation. During sleep, the transient loss of cortical control and functional rearrangement of cortico-subcortical-spinal networks may release the activation of these motor patterns, leading to parasomnias [6,7] or other sleep-disorders such as PLMD [4]. Whether differences of PLMD semiology described in our patient reflect involvement of separate CPG or of common neural circuits with different levels of activation is open to question. There is evidence that sensory stimuli influence neural activation according to sleep depth, being lighter sleep-stages more prone to be influenced by external sources than deeper stages, which appear more resilient [8,9]. Accordingly, our patient presented a clear-cut predominance of PLMs during N2 (Fig. 2).

It is known that PLMs can be associated with obstructive sleep apnea [10] and the presence of a sleep-related respiratory disturbance cannot be underrated. Nevertheless, the mild-moderate obstructive sleep apnea syndrome of our patient played a marginal role influencing motor events only in supine position during N2.

In conclusion, we suggest that body posture during sleep may influence PLMs semiology based on different levels of activation of spinal or supra-spinal CPG. Moreover, sleep stages can modulate the motor patterns of PLMs, acting as a filter in sensorial processing.

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None.

Conflict of interest

The authors declare no financial or other conflicts of interest.

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2019.04.002.

References

[1] American Academy of Sleep Medicine. The international classification of sleep disorders – Third Edition (ICSD-3). Darien, IL: American Academy of Sleep Medicine; 2014.
[2] Haba-Rubio J, Marti-Soler H, Marques-Vidal P, et al. Prevalence and determinants of periodic limb movements in the general population. Ann Neurol 2016;79(3):464–74.
[3] Dvzonic ML, Kripke DF, Klauber M, et al. Body position changes and periodic movements in sleep. Sleep 1986;9(4):484–91.
[4] Parrino L, Halasz P, Tassinari CA, et al. CAP, epilepsy and motor events during sleep: the unifying role of arousal. Sleep Med Rev 2006;10(4):267–85.
[5] Guertin P. Central pattern generator for locomotion: anatomical, physiological, and pathophysiological considerations. Front Neurol 2013;8:183. 3.
[6] Tassinari CA, Gardella E, Cantalupo G, et al. Relationship of central pattern generators with parasomnias and sleep-related epileptic seizures. Sleep Med Clin 2012;7(1).
[7] Nobili L, Sartori I, Terzaghi M, et al. Intracerebral recordings of minor motor events, paroxysmal arousals and major seizures in nocturnal frontal lobe epilepsy. Neurol Sci 2005;26(Suppl. 3):s215–9.
[8] Lavigne G, Zucconi M, Castronovo C, et al. Sleep arousal response to experimental thermal stimulation during sleep in human subjects free of pain and sleep problems. Pain 2000;84(2–3):283–90.
[9] Parrino L, Vaudano AE. The resilient brain and the guardians of sleep: new perspectives on old assumptions. Sleep Med Rev 2018;39:98–107.
[10] Manconi M, Zavalko I, Bassetti CL, et al. Respiratory-related leg movements and their relationship with periodic leg movements during sleep. Sleep 2014;37(3):497–550.