Sleep Medicine 33 (2017) 43–46

Contents lists available at ScienceDirect

Sleep Medicine

journal homepage: www.elsevier.com/locate/sleep

Video-Clinical Corners

Video-polysomnographic aspects of painful legs and moving toes syndrome

Francesco Zellini [a], Raffaele Ferri [b], Manuel Abbafati [a], Claudio Staedler [a], Massimiliano Tiberti [a], Claudio Gobbi [a], Mauro Manconi [a, *]

[a] Sleep and EEG Center, Neurocenter of the Southern Switzerland, Civic Hospital of Lugano, Via Tesserete 46, 6900 Lugano, Switzerland
[b] Sleep Research Centre, Department of Neurology I.C., Oasi Research Institute (IRCCS), Via Conte Ruggero 73, 94018 Troina (EN), Italy

ARTICLE INFO

Article history:
Received 11 October 2016
Received in revised form
2 January 2017
Accepted 3 January 2017
Available online 27 January 2017

Keywords:
Painful legs syndrome
Video-polysomnography
Restless legs syndrome
Alternating leg muscle activation

1. Introduction

Painful legs and moving toes (PLMT) syndrome is a rare chronic disorder [1] characterized by pain and repetitive stereotyped toe movements that may resemble restless legs syndrome (RLS) [2]. PLMT is idiopathic or associated with other conditions [3]. No effective therapy is available for PLMT. A video-polysomnography of PLMT has never been carried out.

2. Case description

A 69-year-old woman was referred to us with a diagnosis of RLS, which started five years earlier and refractory to dopamine agonists. Her medical history was positive for hypertension, diabetes, and retinal melanoma. She complained of a sub-continuous pain in the anterior part of both feet, mainly on the left foot. She also reported permanent involuntary movements of the left and, rarely, of the right foot that she could stop by volition for only a few seconds. These symptoms caused insomnia, which was treated with zopiclone (7.5 mg daily) for three years. A previous ineffective attempt with pramipexole (1 mg/day) and then ropinirole (2 mg/day) was performed.

The neurological examination was unremarkable except for continuous flexion and extension digit movements of the left and sporadically the right foot (Video). A cerebro-spinal MR and an electro-neuro-myography were unremarkable. On this basis, a diagnosis of PLMT was established, and a full night video-polysomnography was performed to rule out comorbid RLS/PLMS or other causes of insomnia.

Supplementary video related to this article can be found at doi:10.1016/j.sleep.2017.01.008.

Sleep was characterized by a normal sleep latency (15 min), increased wakefulness after sleep onset (290 min), severe reduction in sleep efficiency (35.3%) and total sleep time (167 min), with a representation of all sleep stages: N1 1.5%, N2 27.5%, N3 45.2%, R 25.7%. Neither breathing abnormalities (AHI 0.8/h) nor PLMS (index 1.2/h) were noticed. Prolonged night awakenings were motivated by the patient with pain and insomnia, without restlessness. Continuous flexion-extension movements of feet were recorded only during wake time, with immediate disappearance at sleep onset and an immediate reappearance with transition from sleep to wakefulness (Fig. 1).

Continuous movements occurred on the left side (Fig. 1b), and occasionally on the right side. When both sides were affected, movements always occurred with a regular alternation between left and right (Fig. 1a). From a total of 951 limb movements we selected a series of regular rhythmic movements for frequency analysis (620 movements, 421 on the right and 199 on the left), considering the onset of consecutive movements. The mean frequency of bursts on one limb was 0.36 ± 0.11 Hz (both limbs 0.18 Hz), while the mean burst duration was 2.1 ± 0.93 s. No cortical pre-motor potentials were detected at back-averaging EEG analysis triggered by the onset of well-defined leg movements (Fig. 2).

* Corresponding author. Sleep and Epilepsy Center, Neurocenter of the Southern Switzerland, Civic Hospital of Lugano, Via Tesserete 46, Lugano, Switzerland. Fax: +41 091 811 6915.

http://dx.doi.org/10.1016/j.sleep.2017.01.008

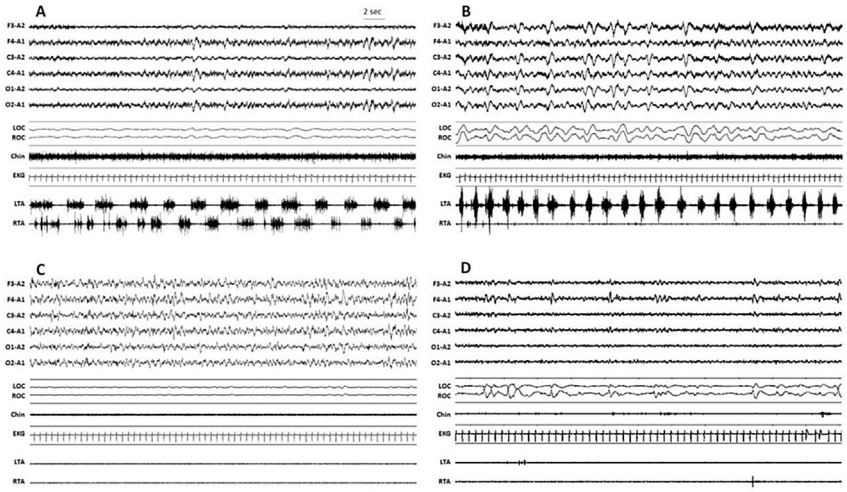

Fig. 1. Four 45-s long polysomnographic fragments in the patient with PLMT performed in the sleep laboratory with the following signals: electroencephalogram; electrooculogram; electrocardiogram; electromyogram of the submentalis; both tibialis anterior muscles; oronasal airflow; thoracic and abdominal movements; oxyhemoglobin saturation. A) alternating leg movement activity during wakefulness; B) monolateral left leg movement activity during wakefulness; C) absence of leg movement activity during NREM sleep; D) absence of leg movement activity during stage R. LOC and ROC left and right ocular cantus; LTA and RTA left and right anterior tibialis muscles. A1 left mastoid, A2 right mastoid, EEG: F frontal, C central, O occipital.

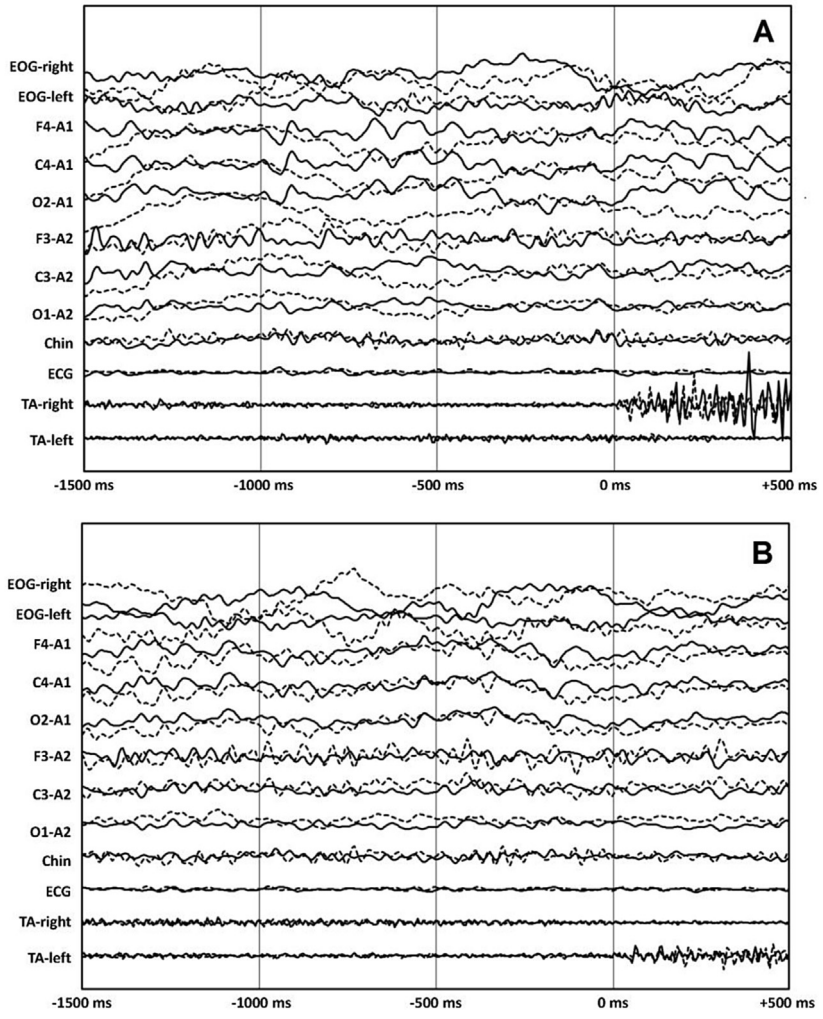

Fig. 2. Back-averaging of EEG signals with time 0 at the beginning of EMG bursts over the right leg (top panel A) and of the left leg (bottom panel B). The epoch acquisition length was 2000 ms and included 1500 ms before the beginning of EMG bursts and 500 ms after. Signals were band-pass filtered at 05–30 Hz (EEG signal) and at 15–90 Hz (EMG signal) and sampled at 200 Hz. Signals were averaged separately for each side and the 0 μV baseline was determined as the mean amplitude of the first 200 ms of the epoch. Two averages are shown (continuous and dotted lines) for each leg, obtained from two separate series of 100 leg movements each, which do not disclose any evident EMG time locked potential in any of the other channels recorded. Only the increase in the tibialis anterior activity is evident after the time 0, in agreement with the method used and the side considered (right or left).