



Video-Clinical Corners

Hypnagogic Foot Tremor, Alternating Leg Muscle Activation or High Frequency Leg Movements: clinical and phenomenological considerations in two cousins

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1. Introduction

Alternating Leg Muscle Activation (ALMA), Hypnagogic Foot Tremor (HFT) and High Frequency Leg Movements (HFLM) are minor motor activities during sleep with unclear significance and have overlapping features (Table 1).

In 1988, Broughton coined the term HFT and reported the phenomenon in two patients after severe head injury. He described “grouped phasic tremor potentials at varying frequencies between 0.5 and 1.5 per second, recorded in general, independently from both anterior tibialis muscles, more so on the right”, occurring during pre-sleep wakefulness, sleep stage 1 and 2 [1]. In 2001, Wichniak reported Rhythmic Feet Movements (RFM) in 7.5% of 375 subjects undergoing vPSG and used the term synonymously with HFT. RFM include the “whole foot or the toes while falling asleep with a frequency of 1–2 per second and a single movement duration between 300 and 700 ms” [2]. In 2003,

Chervin described ALMA, characterized by an “alternating pattern of anterior tibialis activation”, in a clinical series of 16 patients [3]. Yang and Winkelman investigated polysomnographies of 486 patients, reported a similar phenomenon in 37 patients and called it HFLM [4].

In the current International Classification of Sleep Disorders (ICSD 3), HFT and ALMA are represented within the category of motor disorders of sleep, among the “Isolated Symptoms and Normal Variants”, while RFM and HFLM are not [5].

We present video-polysomnography (vPSG)-clips of two female maternal cousins with suspected sleep apnea and incidental simultaneous findings of ALMA and HFT.

2. Case reports

2.1. Patient A

A 43-year-old woman treated with CPAP therapy for moderate sleep apnea (AHI 22.4/h) underwent vPSG control for one night. ALMA and HFT were found during wakefulness and in sleep stage N1 (video 1 and 2, Fig. 1a,b). Periodic leg movements in sleep (PLMS) were also present (PLMS-index 16.4/h). The patient was repeatedly asked about any Restless Legs Syndrome (RLS) symptoms which she denied.

Supplementary video related to this article can be found at <https://doi.org/10.1016/j.sleep.2018.10.024>

2.2. Patient B

A 50-year-old woman, maternal cousin of patient A, underwent vPSG for control of CPAP-treated severe obstructive sleep apnea (AHI 34.9/h). ALMA and HFT occurred in wakefulness and sleep stage N2 (video 3 and 4, Fig. 2a,b). The patient had PLMS as well (PLMS-index 39.2/h), without RLS on repeated interrogation.

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Both cousins had never been treated with antidepressants.

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Table 1
Characteristics of ALMA, HFT and HFLM (upper panel) and other motor disorders of sleep (lower panel).

| | Frequency (Movements/unit of time) | Single Movement Duration (s, ms, minutes) | Number of Muscle Activation | Sleep Stage (wakefulness, N1, N2, N3, REM) | Laterality (uni-, bilateral, alternating) | Number of patients |
|-------------------|------------------------------------|---|-----------------------------|--|---|-----------------------------|
| ALMA [3] | 0.5–3.0/s | 100–500 ms | >4 | wakefulness, N1, N2, N3 | alternating | 16 (12 male, four female) |
| HFT [6] | 0.3–4.0/s | 250–1.000 ms | >4 | wakefulness, N1, N2 | uni- and bilateral | Not determined |
| HFLM [4] | 0.3–4.0/s | 100–500 ms | ≥4 | wakefulness, all sleep stages | uni- and bilateral, alternating | 37 (19 male, 18 female) |
| EFM [7] | ≥5/60 s for at least 20 min | <150 ms | not determined | N2, N3 | asynchronously | 38 (36 male, two female) |
| Hypnic jerks/ [8] | not determined | 50–250 ms | not determined | wakefulness, N1, N2 | bilaterally/asymmetrical, unilateral | 10 (six male, four female)< |
| PLMS [6] | 1/5–90 s | 0.5–10 s | ≥4 | N1, N2, N3 | not determined | not determined |
| RMD [5,6] | 0.5–2.0/s | <15 min | ≥4 | drowsiness, all sleep stages, arousals | not applicable | not determined |

Alternating Leg Muscle Activation ... ALMA, Hypnagogic Foot Tremor ... HFT, High Frequency Leg Movements ... HFLM, Excessive Fragmentary Myoclonus ... EFM, Periodic Leg Movements in Sleep ... PLMS, Sleep Related Rhythmic Movement Disorder ... RMD, seconds ... s, sleep stage 1 ... N1, sleep stage 2 ... N2, sleep stage 3 ... N3, rapid eye movement sleep ... REM.

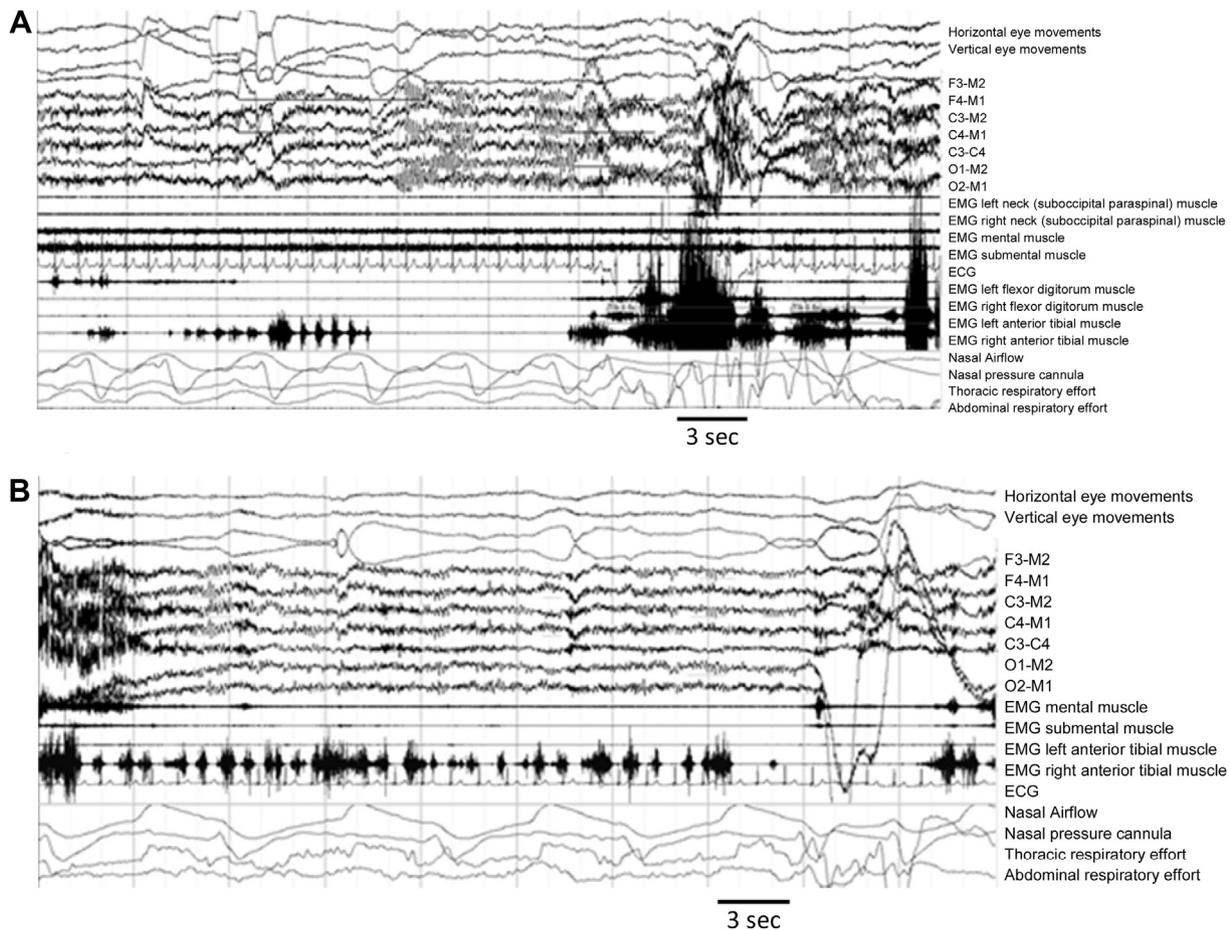


Fig. 1. a: PSG epoch with HFT during wakefulness of patient A corresponds to video-PSG clip 1. Horizontal and vertical eye-movements, horizontal and vertical electro-oculogram; F3–M2, F4–M1, C3–M2, C4–M1, C3–C4, O1–M2, O2–M1 refer to frontal, central and occipital derivations of the international 10–20 electrode placement system; EMG, surface electromyogram; ECG, electrocardiogram. **b:** PSG epoch with HFT during wakefulness of patient B corresponds to video-PSG clip 2. Horizontal and vertical eye-movements, horizontal and vertical electro-oculogram; F3–M2, F4–M1, C3–M2, C4–M1, C3–C4, O1–M2, O2–M1 refer to frontal, central and occipital derivations of the international 10–20 electrode placement system; EMG, surface electromyogram; ECG, electrocardiogram.

3. Video and polysomnography (PSG) findings

Video-PSG clips 1 and 2 show simultaneous video and PSG recordings of patient A and B, with HFT during wakefulness.

Video clip 3 shows ALMA during wakefulness in patient A.

Video clip 4 shows ALMA in sleep stage N1 in patient B.

Video-PSG clips 5 and 6 show ALMA of patient A and B.

Fig. 1a,b shows the PSG recordings of the HFT sequences (corresponding to video-PSG clips 1 and 2).

Fig. 2a,b shows PSG epochs with ALMA (corresponding to video clips 3 and 4).

4. Discussion

We present four short video clips and their representative PSG epochs of two maternal cousins in whom HFT and ALMA were accidentally detected during sleep laboratory evaluation. These motor phenomena are very subtle and not accompanied by arousals

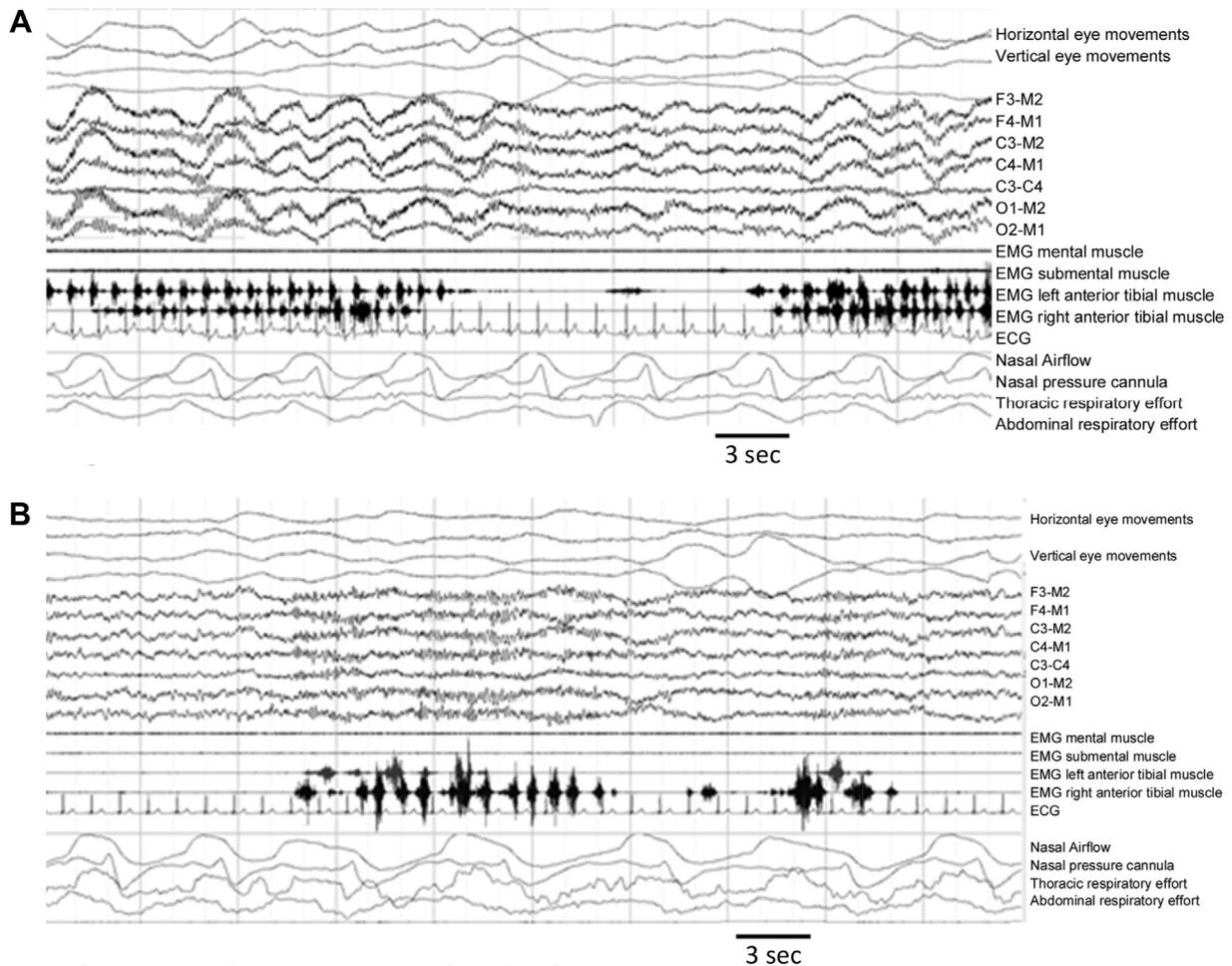


Fig. 2. a: PSG epoch with ALMA during wakefulness of patient A corresponds to video clip 3. Horizontal and vertical eye-movements, horizontal and vertical electro-oculogram; F3–M2, F4–M1, C3–M2, C4–M1, C3–C4, O1–M2, O2–M1 refer to frontal, central and occipital derivations of the international 10–20 electrode placement system; EMG, surface electromyogram; ECG, electrocardiogram. b: PSG epoch with ALMA during sleep stage N1 of patient B corresponds to video clip 4. We defined the alternating activation of the tibialis anterior muscles as ALMA even though they are not alternating in a strict 1:1 pattern. Horizontal and vertical eye-movements, horizontal and vertical electro-oculogram; F3–M2, F4–M1, C3–M2, C4–M1, C3–C4, O1–M2, O2–M1 refer to frontal, central and occipital derivations of the international 10–20 electrode placement system; EMG, surface electromyogram; ECG, electrocardiogram.

or tachycardia. Both patients had PLMS but denied RLS symptoms on repeated questioning. We suggest that ALMA and HFT in these two patients represent only incidental findings without clinical implications.

The vPSG-pattern in the tibialis anterior muscles met criteria for HFT and ALMA, occurring simultaneously in the same night in both patients. HFT, ALMA and the more recently described HFLM have overlapping features, regarding their frequency, duration and series definitions, given in the original descriptions [1,3,4]. HFT and ALMA have been included in the ICSD 3, HFLM not [5]. Whether ALMA, HFT and HFLM have the same underlying mechanism is currently unknown.

Perhaps the most comprehensive descriptive and neutral term for all these minor motor phenomena during sleep are HFLM. HFLM were defined as four or more discrete leg movements with a frequency of 0.3–4.0 per second and duration of 100–500 ms [4].

The etiological and clinical meaning of HFT and ALMA is still unclear and has been poorly investigated. Due to the overlapping features, it might be worth to discuss if all three terms should be included as synonymous under the category of “isolated symptoms and normal variants” in the ICSD [5].

Furthermore, we suggest that RLS should be excluded carefully in any patient who exhibits an incidental finding of HFLM pattern in the vPSG, and even more so if simultaneously PLMS are found,

because unconscious compensatory movements due to unpleasant sensations or unconscious urge to move caused by hitherto undiagnosed RLS or other comfort movements could produce a similar vPSG pattern. If the familial coincidence in these two cousins is accidental or true familial occurrence cannot be told in this single pair of cousins.

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Conflict of interest

None declared.

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.2018.10.024>.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.sleep.2018.10.024>.

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