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Special Section

The official World Association of Sleep Medicine (WASM) standards for recording and scoring periodic leg movements in sleep (PLMS) and wakefulness (PLMW) developed in collaboration with a task force from the International Restless Legs Syndrome Study Group (IRLSSG)

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1. Prologue

The definition and scoring criteria for periodic leg movements (PLM) during sleep have not changed since 1990 [1] and 1993 [2] and are substantially based on the work carried out by Coleman et al. in 1982 [3]. The aim of the Atlas and Scoring Rules [2] was to provide recommendations for the correct recording of motor events, the use of standard terminology and the definition of some common rules to quantify PLM.

However, the current standard for recording sleep is based on computerized technology, and new pathologies have been recognized to be associated with PLM during sleep (PLMS), different motor patterns have also been detected and new sophisticated methods of signal analysis have changed our understanding of the impact of PLM on sleep patterns. In particular, the analysis of the electromyogram (EMG) signal and of the periodicity of the phenomenon provides different methods for the evaluation of muscle activity in normal and pathological sleep.

Even small changes in the criteria of detection of the single PLM may have a significant impact on the final results of the analysis by altering the range of the PLM included. This could significantly modify the numerous characteristics derived from analyses of the PLM. Moreover, changes in the classification criteria of events, inter-movement intervals and periodicity can influence significantly the indexes related to PLM, thereby impacting the evaluation of sleep disorders.

The Task Force of PLM Scoring of the International Restless Legs Syndrome Study Group (IRLSSG) wishes to define innovative strategies to evaluate, count and score leg movements during sleep and wakefulness, which respond both to the new requirements of computerized sleep recording and also to the developing understanding of the different pathologies presenting with PLM. These new guidelines apply to adults; they are expected to be valid also for children, but are recommended for cautious use with children pending further pediatric evaluations of PLM.

2. Recommended standards for recording and scoring of periodic leg movements (PLM)

2.1. Introduction

The differing needs of research and clinical studies lead to two recommended standards: (1) clinical, a minimally acceptable standard for clinical recordings of PLM, and (2) research, a standard adequate for a complete analysis of the leg movements, as they are currently understood. The characteristics that differ between these standards are indicated when they occur; otherwise, in the absence of any specific indication, the standard applies to both applications. In these standards the electromyographic (EMG) activation

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of leg muscles, and in particular the anterior tibialis muscle, defines the 'leg movement' event. This may not be identical to an actual visually noticeable movement of the leg. The 'leg movement' events defined by the EMG activation are considered in themselves to be the primary phenomena.

2.2. Electromyographic (EMG) recording techniques

Surface electrodes will be placed at 2–3 cm apart or 1/3 of the length of the anterior tibialis muscle, whichever is shorter. Electrodes should be placed longitudinally on the muscles, symmetrically around the middle. Impedance should be $\leq 10\,K\Omega$ for clinical studies, but $\leq 5\,K\Omega$ is recommended for research studies. Bilateral recordings are required. Two channels, one for each leg, are strongly recommended for all studies and required for research. Clinical applications may, however, combine the electrodes from both legs into one recorded channel, although this practice is discouraged.

Recording PLM activity from other muscles besides the tibialis anterior is recommended only for research purposes or for special clinical conditions (e.g. arm restlessness, bruxism, other movement disorders).

Filters should be no more restrictive than 10–100 Hz for clinical use and 10–200 Hz for research studies. Digital sampling rates should be no less than 200 Hz for clinical studies and 400 Hz for research studies.

Baseline resting EMG amplitude from these electrode placements with a relaxed muscle should be ± 2–3 μV (or 4–6 μV peak-to-peak), for a non-rectified signal. When using rectified signal, filtering should be applied as needed to remove DC levels; consequently, the amplitude can be measured by the voltage increase above zero. In this case, amplitude can be measured as an increase of 2–3 μV from the baseline.

2.3. Calibration

Calibration used in the past [2], based on definitions of maximum dorsiflexion at the ankle without resistance, was considered too vague and susceptible to technician differences to be useful. It was, therefore, unanimously decided to use the absolute increase in microvolts for detecting a significant event, as described below. Aside from meeting the criteria as defined below there are no magnitude requirements.

Since the change in microvolts defines the event, the calibration should document the microvolts for a baseline recording of the relaxed anterior tibialis lasting 5–10 s and this should be for a non-rectified signal no greater than ± 5 μV (or 10 μV peak-to-peak) for clinical purposes and ± 3 μV (or 6 μV peak-to-peak) for research; these values should be 5 and 3 μV, respectively, for rectified signals.

2.4. Definition of a candidate leg movement event

2.4.1. Onset criteria

An event starts when the EMG increases to ≥ 8 μV above the resting baseline (e.g. 10 μV for a baseline of 2 μV, for a rectified signal).

2.4.2. Offset criteria

An event ends when the EMG decreases to < 2 μV above the resting level and remains below that value for 0.5 s. Note that an event may have one or more periods where the EMG drops below the offset criteria for less than 0.5 s (Fig. 1).

2.4.3. Duration criteria

Duration is defined as the time between onset and offset of the candidate event. The duration must be at least 0.5 s and no longer than 10 s (Fig. 2).

For research studies, particularly on characteristics of PLM, it is recommended that a wider range of events could be recorded with durations up to 15 s. The minimum duration for research is that required to obtain a valid stable measure of the EMG increase. The maximum duration is that where the activity would possibly be considered an awakening. Recording all movements with this wider range of duration is recommended where possible, but the candidate events are only those that meet the duration criteria defined above.

2.4.4. Special criteria for events during wake time

During wake time there may be periods when the subject tenses the leg muscles, changing the baseline. Events may occur during these periods. If the EMG does not return below the threshold set for the detection of the end of the movement (see above) before 15 s, then a new increased baseline should be defined as the average amplitude of the EMG during such a period. The above criteria apply using this new baseline as the resting EMG value. These criteria end when the EMG fits the criteria for end of movement using the original baseline.

2.4.5. Morphology of PLM

The morphology of PLM varies considerably across multiple movements and different states and stages (such as myoclonic, polymyoclonic, rhythmic, multiple and tonic discharges, etc.). Some of these activities, such as polymyoclonus as an example, will still be detected applying the above criteria; however, further studies are needed.

2.5. Definition of PLM events

2.5.1. Definition of period for events

The period is measured from onset to onset of consecutive candidate events. Note that events that are not candidate events are not considered; thus, very short (< 0.5 s) or very

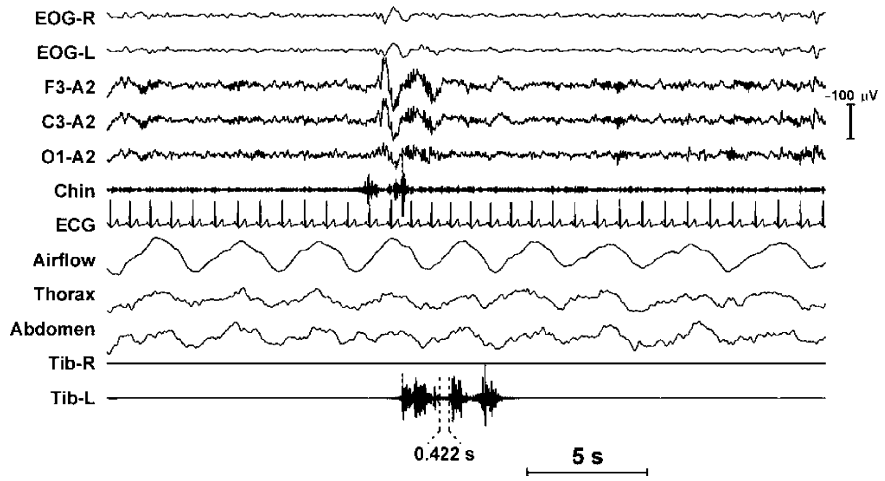

Fig. 1. Single leg movement during which the EMG drops below the offset criteria for less than 0.5 s. Abbreviations: EOG-R, EOG-L, right and left electrooculogram, respectively; F3-A2, C3-A2, O1-A2, electroencephalogram from 3 different left scalp locations referred to the right earlobe; Chin, electromyogram of the submentalis muscle; ECG, electrocardiogram; Airflow, oronasal airflow by thermistors; Thorax and Abdomen, chest wall and abdominal movements by strain gauges, respectively; Tib-R and Tib-L, right and left anterior tibialis muscle electromyogram, respectively.

long (> 10 s) EMG events that occur between two candidate events do not affect the period duration measured from onset to onset of the candidate events.

2.5.2. Period criteria

The period length for two consecutive events to be considered as PLM must be at least 5 and no more than 90 s. The distribution of periods outside this range may inform about the distribution of events and would be useful for research evaluations. Therefore, all periods between candidate events should be measured and recorded, if possible. The minimum period between candidate events as defined above is 1.0 s (minimum leg movement duration of 0.5 s plus minimum offset-to-onset interval of 0.5 s) and the overall distribution of events in the lower range of period lengths (1–5 s) deserves further research evaluation. For clinical scoring, if the interval between two consecutive candidate leg movement events is less than 5 s, then the latter candidate movement is ignored and the period is calculated from the onset of the earlier movement to the onset of the next candidate movement. For research, the period should be measured for all candidate events including a full analysis of the distribution of the periods over the full range (up to at least 100 s).

2.5.3. Number of events for PLM

The number of consecutive candidate events meeting the period criteria must be four or more. Candidate events can occur in any stage of sleep or wake and the sequence of consecutive events continues across changes in sleep–wake state. Thus, a PLM sequence starting in sleep can continue in waking and vice versa. Similarly, an arousal occurring before or during a candidate event does not alter the assessment of that event.

2.5.4. Combined versus separate leg analysis

Periodicity can be evaluated either in separate channels, one for each side, or combining the events detected in both sides. The choice should be clearly stated. For diseases involving predominantly one side, the former is the only acceptable method. Detections from the two sides can be combined, and bilateral movements are counted as one. Movements occurring on the two sides are considered bilateral when they are overlapping or

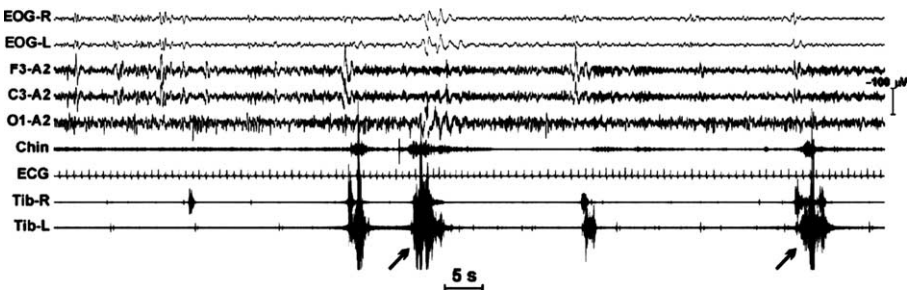

Fig. 2. Sequence of PLM, two of which show a duration longer than 5 s (arrows). Abbreviations as in Fig. 1.

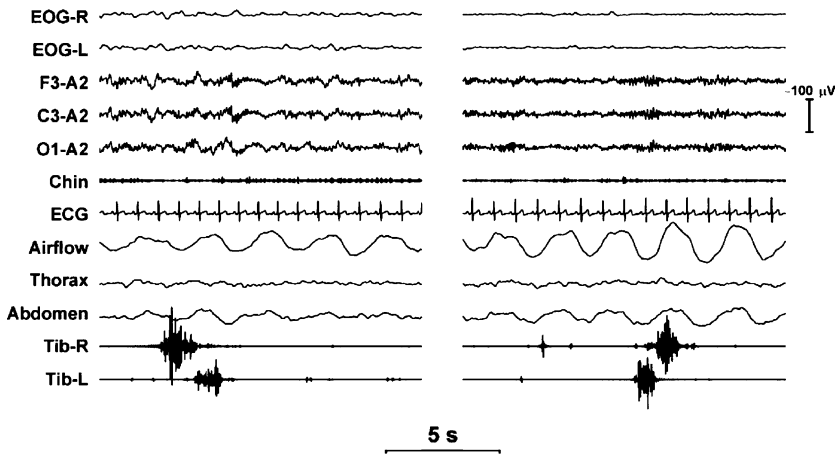

Fig. 3. Two examples of movements occurring on the two sides considered as bilateral because they are overlapping or separated (offset-to-onset) by less than 0.5 s. Abbreviations as in Fig. 1.

separated (offset-to-onset) by less than 0.5 s (Fig. 3). Bilateral movements can include two or more monolateral movements separated (offset-to-onset) by less than 0.5 s from each other.

Fig. 4 provides an overview of all the parameters described above.

2.6. Definition of a leg movement arousal event

2.6.1. Definition of an arousal

This follows scoring rules specified in the 1992 Atlas Task Force publication by the American Sleep Disorders Association (ASDA) [4]; however, some additional considerations on arousal events are reported in the Appendix. The 3 s minimum arousal duration criteria used in this standard can be relaxed when computer analysis is used. Recommended duration with computer scoring could be 2 s, but 2 s is a best-guess estimate not based on research data. Further research should help define this more precisely in the future.

2.6.2. Definition of temporal association of events

An arousal event and movement event are considered associated with each other when there is less than 0.5 s between the end of one event and the onset of the other event, regardless of which is first.

2.7. Quality control

Visual inspection of the recording should be maintained throughout the recording period. If the resting baseline increases during sleep, then consideration should be given to replace the electrodes, depending on the clinical or research demands.

Visual recording of leg movement events can be useful to evaluate characteristics of the movement.

2.8. Basic measurements for sleep period recordings

It was felt that the following measurements should be reported for sleep period recordings:

2.8.1. Required

- *PLMS/h of sleep*. Number of leg movement events occurring during sleep that meet PLM criteria divided by the number of hours of sleep with leg movement recording (PLMS/h).

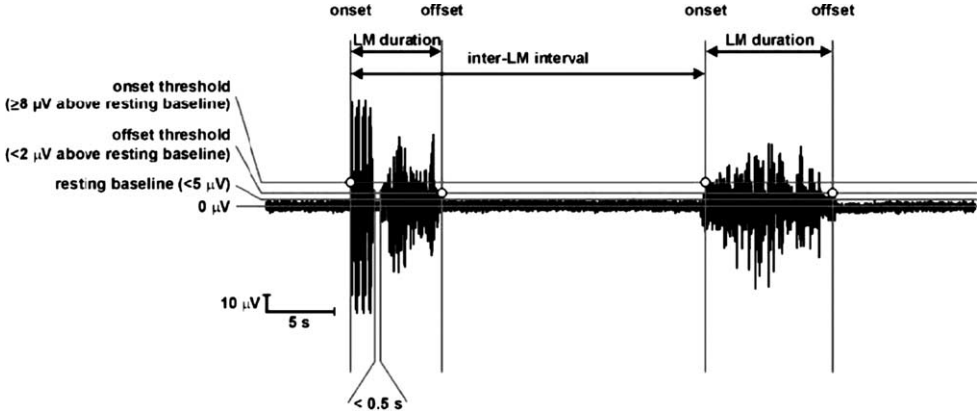

Fig. 4. Overview of the detection parameters for candidate PLM.

- *PLMS with arousals/h (PLMA/h)*. Number of leg movement events occurring during sleep that meet criteria for PLM and having an associated arousal divided by the number of hours of sleep with leg movement and arousal recording (PLMA/h).
- *PLM during wakefulness (PLMW)/h of waking*. Number of leg movement events occurring during wakefulness during time in bed that meet PLM criteria divided by the number of hours of wake with leg movement recording (PLMW/h); time in bed is defined as all time between lights off and lights on, while the legs are horizontal (i.e. while the patient is in bed and not out of bed or sitting on the side of the bed).

2.8.2. Recommended

- PLMS/h for non-rapid eye movement (NREM) sleep only
- PLMS/h for REM sleep only
- Duration of each of the PLM occurring during sleep (PLMS) and wakefulness (PLMW)—average, standard deviation (separate PLMS for REM and NREM sleep)
- Inter-movement interval (onset-to-onset of movements) of PLMS and PLMW (separate PLMS for REM and NREM sleep)

2.8.3. Optional

- PLMS by sleep stages (include duration and inter-movement intervals)
- Isolated leg movements: leg movements with onset–onset intervals >90 s or PLM in sequences formed by less than 4, number/h of sleep.

2.9. Basic measurements for suggested immobilization test (SIT) [5]

It was felt that the following measurements should be reported for wake period recordings during the SIT:

2.9.1. Required

PLMW/h of waking. Number of the leg movement events occurring during wake time that meet PLM criteria divided by the number of hours of wake time with PLM recording (PLMW/h); wake time is defined as all time between start and stop of the SIT, with waking EEG and while the legs are horizontal.

3. Respiration and PLM special recording rules

Respiration must be recorded to accurately assess if leg movements are associated with respiratory events and therefore are not pathophysiologic markers for other neurological disorders, such as restless legs syndrome (RLS) or PLM disorder (PLMD). Thus, a leg movement is associated with the breath ending an apnea/hypopnea event or introducing a significant transitory improvement in respiration during apnea/hypopnea when the temporal occurrence of the critical breath event and the leg movement event overlap or the offset of the earlier event precedes the onset of the other by less than 0.5 s, regardless of which is first (Fig. 5); these leg movements should not be included in PLM-related parameters. It should be noted that PLM and obstructive sleep apnea might coexist. Therefore, the clinician must attempt to treat the sleep apnea first to unmask the possible independent leg movements, if present. Respiration recordings should include measures of flow and effort [6]. Flow can be recorded with thermistor, flow-sensitive cannula, or capnograph and effort with rib cage and abdominal movements and/or intercostal EMG. Subtle increases in inspiratory resistance can be detected with nasal pressure, paradoxical motion of chest wall and abdomen, increasing intercostal EMG or esophageal pressure.

4. Cyclic alternating pattern (CAP) recording standards

PLM is often associated with periodic electroencephalographic (EEG) events including arousals, k-alpha complexes and sequences of k-complexes. This periodicity suggests that a central nervous system oscillator may be involved. The cyclic alternating pattern (CAP) [7] is an EEG

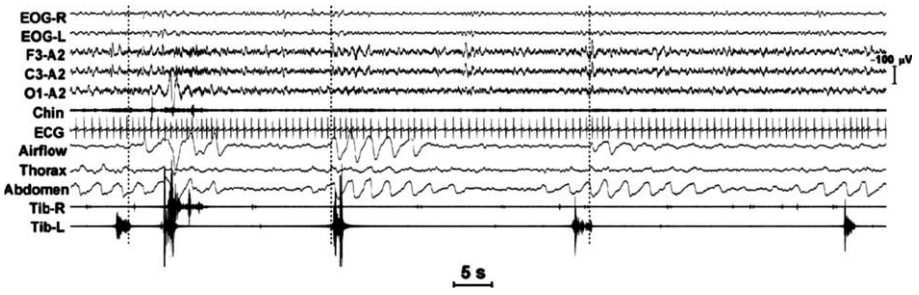

Fig. 5. Sequence of leg movements overlapping with or occurring within ± 0.5 s of breathing resumption of sleep apnea events (vertical dashed lines). Abbreviations as in Fig. 1.

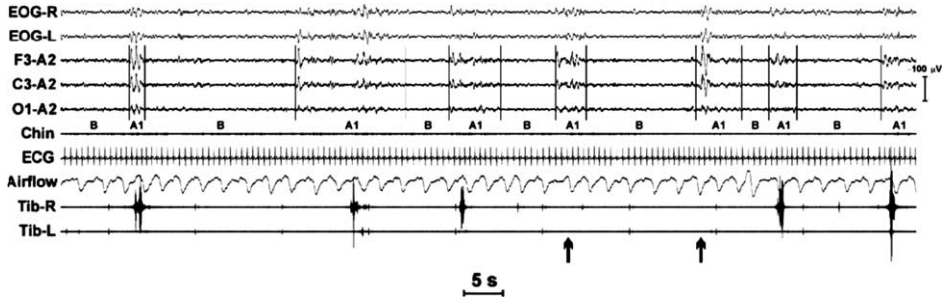

Fig. 6. Example of missing PLM (arrows). Note the presence of short bursts of delta waves (A1 CAP subtypes) approximately where the missing PLM were expected to occur. Abbreviations as in Fig. 1.

oscillator composed of these periodic EEG phenomena that underlies many periodic phenomena occurring during sleep, including PLM (see Appendix).

CAP associated with PLM may represent an upstream phenomenon; however, CAP and PLM are likely controlled by different mechanisms. For example, drugs that decrease RLS and PLM may increase CAP. From a neurophysiological perspective, the presence of CAP may provoke, facilitate, or elicit leg movements. A classic illustration of this occurs in the case of the so-called missing leg movement (Fig. 6). In such cases, CAP and PLM occur simultaneously; however, after several leg movements, a leg movement fails to occur in the expected temporal location. Nonetheless, the concurrent CAP includes an A phase in the expected temporal location. CAP phase A1 reflects a synchronization pattern which may represent unstable sleep; nonetheless, it may also help to preserve sleep continuity. Thus, it is not surprising to find phase A1 reduced in patients with PLM, whereas A2 and A3 phases are increased [8]. Phases A2 and A3 represent an arousal phenomenon that likely leads to clinical correlates of disturbed sleep. The PLM/CAP association more strongly maps onto A2 and A3 phases. PLM seldom occur during non-CAP periods. Recommendations for simple non-computerized clinical recording include awareness of CAP to help appreciate the EEG and arousal events. For those clinicians or researchers who wish to apply a more systematic analysis we recommend quantification of CAP rate, CAP time, CAP distribution, and PLM/CAP phase tabulation [9].

5. Leg activity monitoring standards

Activity monitoring of leg or foot movements by a motion detector system provides another measure of leg movements in sleep (Fig. 7). These devices provide the possibility of multiple nights of recording in a home environment, reducing somewhat the vexing problems caused by the relatively large night-to-night within-subject variation reported to occur for PLM [10,11]. These devices should be able to meet approximately the same scoring criteria for the EMG signal. The following are considered

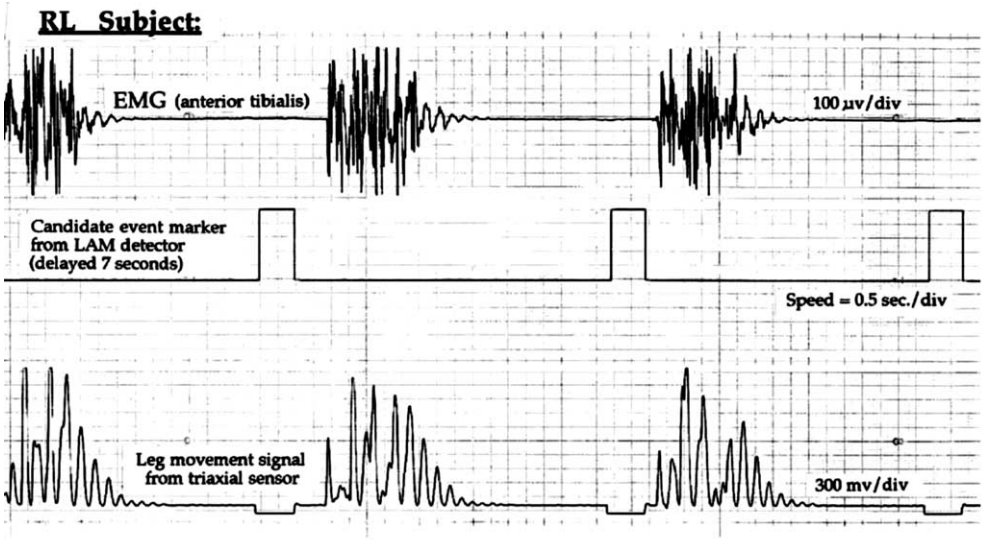

Fig. 7. Anterior tibialis EMG and rectified leg activity data for PLM events. The middle line is a marker showing the detection of the prior event as meeting criteria for PLM. Note the remarkable similarity in duration and morphology of the activity meter and the EMG.

minimal criteria for a satisfactory actigraphic leg activity measurement:

- Movement sensor can detect a wide range of accelerations (0.01–8 g) and have a reasonably linear relationship between output and acceleration over a frequency range of 0.8–14 Hz. Documentation of the calibration of the sensor in each unit should be available.
- Sampling rates of acceleration measures should be $\geq$10 Hz in order to reliably measure durations of 0.5 s.
- Data are stored, analyzed and presented for data review showing the sampling rate of $\geq$10 Hz.
- Some record of body or leg position needs to be obtained from a position indicator time synched to the leg monitor or from a patient hourly log during the sleep period. PLM should only be counted for times the patient's legs are horizontal. This becomes important for unattended recordings in a home environment where the patient may get up one or more times during the night. This also permits assessment of time in bed for determining the PLM/h. Note that in addition to a diary, the button press on a recording device could also be used to determine time lying or sitting in bed with legs horizontal during the night. These methods need to be specified in any report of actigraphic leg activity monitoring during the sleep period.
- The maximum recording time for the recorder must be at least 10 consecutive hours for a night. Preferably multiple nights are recorded
- At least three nights of recording are recommended (but more are preferred, if possible). Further research may provide guidelines for the number of nights in relation to stability of the measurement.
- Leg activity should be measured on each leg separately and recorded accordingly. Recording of activity from one leg only is not recommended.
- The leg activity scoring system for identification of PLM events should specify the thresholds for onset and offset of movement events and the duration parameters used in the measurement. Ideally, these parameters could be adjusted for special conditions such as pediatric studies.
- The analyses of the leg activity measurements must include the total number of PLM and PLM/h lying in bed. In addition, the analyses should provide information on the duration of the intervals between onsets of successive movements, the durations of the movements themselves and the time of occurrence of the movements. Similar information could ideally be available for isolated leg movements.
- Validation of leg activity measurements should be made by simultaneous comparison with blind scoring of a standard EMG assessment of the events. Event-by-event assessment for accuracy of detection is preferred but correlation with overall rates for a full night or test condition recording may be considered minimally acceptable for a validation. Comparisons with all 1–1 night EMG recordings should be done against the combined PLMS and PLMW from the night. Comparisons with only the PLMS suffices for conditions where, for the recording, the total PLMW is less than 10% of the total PLMS + PLMW, but when this occurs it should be clearly noted.

Appendix

In the state progression of different NREM stages the balance between the arousal and the anti-arousal system is regulated by long period infraslow oscillation (0.002–0.02 Hz) expressed in the scalp electrocortical activities by the cyclic alternating pattern (CAP). When they appear in NREM sleep, these oscillations tend to recur in sequences with a periodicity of 20–40 s. This regular distribution determines the organization of serial frequency/amplitude EEG modifications of CAP cycles, composed of a phase A (specific, repetitive and transient EEG pattern) and a phase B (interval between 2 consecutive A phases). Phase A of CAP is the EEG marker of cerebral activation, including cortical arousal, and for this reason it is a potential trigger of somatomotor activities. Phase B is an EEG indicator of rebound deactivation which induces somatomotor inhibition and becomes a potential limiting factor for the duration of any body movement during the NREM sleep period.

CAP is the EEG translation of unstable sleep which coordinates responses in disparate brain regions and accompanies the dynamic evolution of the sleep process such as falling asleep, stage shifts, NREM/REM transition and intra-sleep awakenings, which are the crucial points for motor event generation during sleep. The absence of CAP for more than 60 s is scored as non-CAP and reflects a condition of stable consolidated sleep in which body movements have less chance to appear due to the overall multi-system stability.

The A phases of CAP can be classified in subtypes A1, A2 and A3. In particular, subtypes A1 are composed almost entirely by slow waves (0.5–2.5 Hz, with a significant portion of $<$1 Hz rhythms), subtypes A2 contain approximately 50% of slow oscillations and 50% of low-voltage fast activities, while subtypes A3 include only a minimal percentage of slow oscillation with a predominance of rapid activities. CAP subtypes, A2 and A3 in particular, include ASDA arousal events [4]; however, special attention should be paid to the eventual slow-wave content of these events because some differences between phase A subtypes and ASDA arousals exist. According to the ASDA criteria, slow-wave activity is included in the arousal definition only when occurring within the EEG fast-frequency shift; in the view of CAP, slow waves can occur not only within but also before (more frequently) or at the end of the fast-frequency shift (Fig. 8).

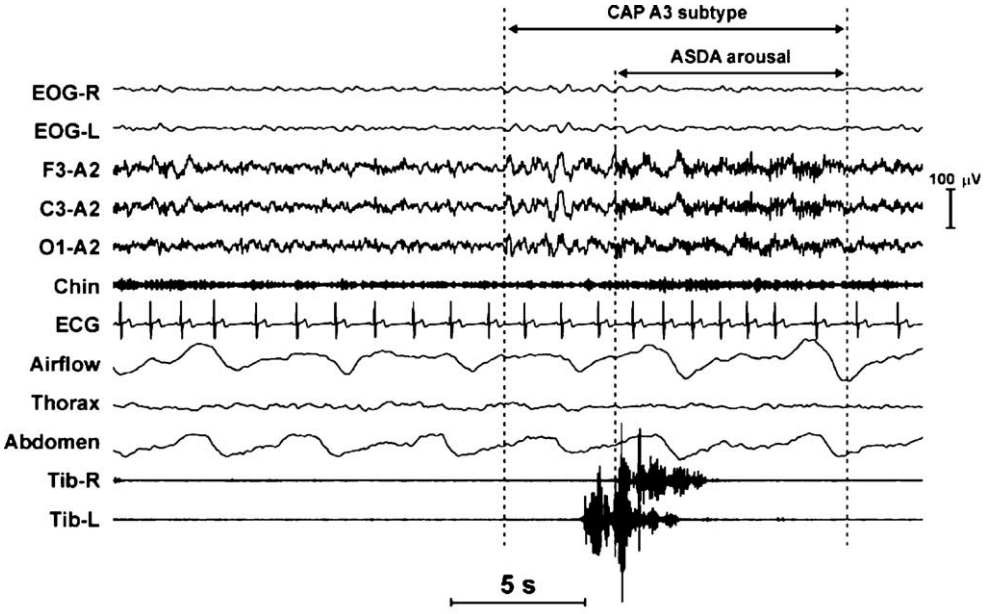

Fig. 8. Example of association of PLM with arousal. Note the different extension of the same EEG event if considered as an ASDA arousal or CAP A3 subtype. Abbreviations as in Fig. 1.

With regard to motor and autonomic functions, subtypes A1 are associated with a mild activation, subtypes A2 with a moderate activation and subtypes A3 with a powerful activation. Subtypes A1 are essentially expressions of transient activation restricted to the frontal lobe; on the contrary, subtypes A3 are projected into the parieto-occipital regions and subtypes A2, with mixed slow-rapid components, span from frontal to occipital lobes. These differences might have physiologic and pathologic implications for motor pattern activation during the sleep period. The motor pattern generated in the brain might be either directly activated by cortical arousal or indirectly activated by transient suppression of cortical or subcortical inhibition. Most of them, such as physiologic body movements, bruxism, generalized tonic–clonic seizures, major episodes of nocturnal frontal lobe epilepsy (NFLE) and most parasomnia events, seem to be inserted only in specific points of the sleep structure. On the contrary, PLM and minor seizures of NFLE seem to be related to a resonant association with the cyclicity of the brain arousability and the origin of the central pattern generator (cortical, subcortical and spinal) is not clearly determined. In particular, PLM is a disorder characterized by a pattern of motor phenomena and EEG changes, both recurring at intervals of 20–40 s. This periodicity parallels the recurrence of CAP cycles. Therefore, it is not surprising that the two polysomnographic manifestations (PLM and CAP) are mutually connected. CAP and PLM also appear to be related to the cyclic alternation of autonomic activation and quiescence, as indicated by the increase in electrocardiogram (ECG)-established heart rate that accompanies both the A phase and PLM. In the sequence of events, cardiac and EEG changes take place before the onset of the motor phenomenon. The temporal relationship between delta activity, cardiac activation and PLM onset suggest that these phenomena as a preparatory condition, involve both central and autonomic nervous systems, exerting a permissive function on the activity of spinal motor neurons.

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