



Original Article

Consensus diagnostic criteria for a newly defined pediatric sleep disorder: restless sleep disorder (RSD)



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ABSTRACT

Background: Restless sleep is a frequent complaint in clinical practice and has been reported in the medical literature since the 1970s. Most often, it has been described in association with specific sleep or medical conditions. However, more recently, publications have emerged that describe a disorder characterized by restless sleep as the core feature. To assess this further, the International Restless Legs Syndrome Study Group (IRLSSG) appointed a task force composed of international sleep experts.

Methods: A committee of 10 sleep clinicians developed a set of 16 consensus questions to review, conducted a comprehensive literature search, and extensively discussed potential diagnostic criteria. The committee recommendations were reviewed and endorsed by the IRLSSG Executive Committee.

Results: Based on the medical literature and expert clinical experience, the task force found sufficient evidence to formulate diagnostic criteria for a clinical entity designated “restless sleep disorder” (RSD). Eight essential criteria were agreed upon, which include a complaint of restless sleep, observed large body movements during sleep, video-polysomnographic documentation of 5 or more large body movements/hour, occurrence at least three times a week for at least three months, clinically significant impairment, and differentiation from other conditions that might secondarily cause restless sleep. However, the current evidence limits application to ages 6–18 years. Diagnostic coding, addition to existing diagnostic nosologies, and name selection are discussed.

Conclusions: Consensus diagnostic criteria for RSD have been developed, which are intended to improve clinical practice and promote further research.

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1. Introduction

Restless sleep has been mentioned in the medical literature since the 1970s and is a frequent complaint in the clinical setting. A

PubMed search using the search term “restless sleep” produces over 250 publications, which describe this in children ($n = 139$) and adults ($n = 112$) (MEDLINE/PubMed accessed 06/28/2020). In the first sleep disorders nosology, the *Diagnostic Classification of Sleep and Arousal Disorders* [1], “restlessness” during sleep was defined as “persistent or recurrent body movements, arousals, and brief awakenings (that occur) in the course of sleep.” It was described as a symptom or sign of several different sleep disorders, including

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sleep apnea syndrome and sleep-related myoclonus. Restless sleep has been of particular interest in the pediatric sleep literature, with questions about it in many validated pediatric sleep questionnaires, including the Pediatric Sleep Questionnaire, Child's Sleep Habits Questionnaire, and Sleep Disturbance Scale for Children. Since 2018, publications have emerged on “restless sleep disorder” (RSD), a newly described pediatric entity [2–5].

In recognition of the need to assess the accumulating evidence and clinical experience on restless sleep and RSD, and to potentially formulate diagnostic criteria for RSD, the Executive Committee of the International Restless Legs Syndrome Study Group (IRLSSG) appointed two members (LDR and DLP) to convene an RSD task force composed of international sleep experts.

This report summarizes the rationale and recommendations of this IRLSSG RSD task force.

2. Methods

A panel of nine sleep experts, all authors of the current paper, was approved in February 2019 by the IRLSSG Executive Committee to ascertain whether there was sufficient evidence to propose criteria for the diagnosis of RSD. The task force began correspondence in May 2019. Subsequently, a 10th sleep expert was added (RPA) to broaden the perspective on diagnostic criteria. Regular telephone conferences and a half-day face-to-face meeting (during World Sleep 2019 in Vancouver, Canada September 2019) took place to review the data and clinical experience on RSD and reach consensus on diagnostic criteria.

Sixteen consensus questions were agreed upon to guide the development of diagnostic criteria. These were as follows, with responses discussed in this paper: (1) Is there sufficient evidence to propose a new sleep disorder, restless sleep disorder (RSD)? (2) Is “restless sleep disorder” the most appropriate name? (3) Does each criterion clearly and concisely describe a single, essential aspect of the disorder? (4) Is each criterion unambiguous? (5) Are the criteria consistent with the ICSD-3 and DSM-5 format for diagnostic criteria? (6) Which ICD code currently best fits this disorder? (7) Is video-polysomnography (vPSG) necessary for the diagnosis? (8) What is the specific definition for vPSG movements? (9) What are appropriate frequency and duration criteria for RSD? (10) Are there supportive or associated features for RSD that are not essential criteria? (11) Which comorbidities, if any, are commonly associated with RSD? (12) What is the differential diagnosis of RSD? (13) How is RSD related to RLS? (14) Should RSD include both pediatric and adult populations? (15) Are these criteria for both clinical and research applications? (16) What are important areas for future research?

To ensure that the task force's recommendations integrated published scientific evidence on restless sleep, a formal review of the literature was performed using the broad search term “restless sleep”. The results of the systematic review will be referred to in this paper but a full report will be published elsewhere [6].

After approval of the written report by all 10 committee members, the recommendations were forwarded to the IRLSSG Executive Committee for review and endorsement.

3. Rationale for defining pediatric restless sleep disorder (RSD)

The RSD task force initially conducted meetings during which they discussed the evidence for RSD. Based on the early results of the systematic literature review and on extensive clinical experience, there was unanimous agreement to develop criteria for the diagnosis of RSD. It was determined that there was sufficient evidence for a clinical entity characterized by restless sleep that did

not meet criteria for another sleep disorder but did appear to significantly disrupt sleep and impact daytime function. However, most of the existing evidence and experience applied to school-age children (6–18 years old) and was insufficient in younger children and adults. Therefore, the task force focused on this age group in the recommendations but left open the possibility of expansion at a later date.

Several important aspects regarding RSD diagnostic criteria were agreed upon by the task force. Differentiating a sign or symptom (restless sleep) from a disorder (restless sleep disorder) was to be an essential characteristic of the criteria. Critical to that issue is defining the impact on function, rather than simply describing a characteristic of sleep. In addition, it was determined that there should be an objective measure of restless sleep included, even if that limited the applicability of the criteria in clinical settings where there might not be the resources to measure sleep objectively. At this point, it is considered more important to avoid over diagnosis than risk under diagnosis. Also, there needed to be a clear differentiation from other sleep disorders or conditions, which might have restless sleep as a secondary aspect. The goal was to have clear and concise diagnostic criteria that would evolve with subsequent research, applicable in both clinical and research settings.

4. Consensus diagnostic criteria for restless sleep disorder (RSD)

The clinical diagnosis of RSD is based on both subjective reports obtained in an interview with the patient (and parent, caregiver, or bedpartner when relevant) and the results of a single night of vPSG. Assessment of these subjective and objective findings should be guided by the eight diagnostic criteria below.

4.1. Essential diagnostic criteria (all must be met) (Table 1)

- (A) A complaint of “restless sleep” as reported by the patient's parent, caregiver, or bedpartner, or by the patient.

A complaint of “restless sleep” is the core diagnostic feature of RSD and must be present [2]. This is typically reported by a parent, caregiver, or bedpartner. Sometimes “restless sleep” is reported by the patient, based on the observations of others or the condition of the bed upon awakening. As much as possible, a first-hand description of the patient's sleep should be obtained. Table 2 lists descriptive terms that are used for RSD. Many of these convey a perceived disruption of sleep.

- (B) Restless sleep movements involve large muscle groups of the whole body, all four limbs, arms, legs, or head.

Criterion B highlights the presence of gross, visible body movements, involving large muscle groups of the whole body, all four limbs, arms, legs, or head [2,3]. The sleep movements may be characterized further as frequent repositioning, disruption of the bedsheets, or falling out of bed. Children with RSD are often found in a completely different position compared to the position in which they fell asleep and blankets are frequently found on the floor. vPSG data indicate that the movements involve the whole body (43.7%), all four limbs (28.9%), arms (20.2%), and legs (7.1%) with minimal movements of the head alone (0.1%) [3]. These large movements are to be differentiated from normal sleep movements such as phasic rapid eye movement (REM) twitches and hypnic jerks, as well as movements of other sleep disorders, such as REM sleep behavior disorder, sleep-related rhythmic movement disorder, or periodic limb movement disorder (PLMD) (also, see criteria D and H below). When the presentation is primarily

Table 1

Consensus diagnostic criteria for restless sleep disorder (RSD).

Criteria A-H must be met
A. A complaint of “restless sleep” as reported by the patient’s parent, caregiver, or bedpartner, or by the patient.
B. Restless sleep movements involve large muscle groups of the whole body, all four limbs, arms, legs, or head.
C. The movements occur during sleep or when the individual appears to be asleep.
D. Video-polysomnography shows a total movement index (by video analysis) of 5 or more per hour of sleep.
E. Restless sleep occurs at least three times per week.
F. Restless sleep has been present for at least three months.
G. Restless sleep causes clinically significant impairment in behavioral, educational, academic, social, occupational, or other important areas of functioning, as reported by the patient’s parent, caregiver, or bedpartner, or by the patient (eg, daytime sleepiness, irritability, fatigue, mood disturbance, impaired concentration, or impulsivity).
H. The condition is not better explained by another sleep disorder, medical disorder, mental disorder, behavior disorder, environmental factor, (eg, sleep-disordered breathing, restless legs syndrome, periodic limb movement disorder, sleep-related rhythmic movement disorder, insomnia disorder, atopic dermatitis, seizure disorder, etc.), or the physiological effects of a substance (eg, caffeine).

Supportive findings.

(1) Large body movements during sleep typically occur throughout the night.

(2) Delayed sleep onset is uncommon.

Note: These criteria are intended for the diagnosis of RSD in children and adolescents ages 6–18 years. Future work may support use in younger children and adults.

Table 2

Descriptive terms for restless sleep disorder (RSD).

<ul style="list-style-type: none"> • Restless sleep • Trashes bed • All over the bed • Beats you up during sleep • Sleep disturbance • Abnormal sleep • Poor sleep quality • Large body movements • Sleep movements • Jerking during sleep • Like a helicopter during sleep
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with head movements during sleep, then motor tics associated with Tourette syndrome [7–9] or sleep-related rhythmic movement disorder, head rolling type [10], should be considered. Since the movements are of large muscle groups, they are typically quite visible to direct observation or video monitoring, even under bedsheets or blankets [3]. Fig. 1 shows examples of the body movements seen in children with RSD.

(C) The movements occur during sleep or when the individual appears to be asleep.

Criterion C emphasizes that the large muscle movements occur during sleep or when the individual appears to be asleep, characterizing RSD as a disorder of sleep. The presence or absence of movements while awake should not be taken into consideration. There are currently no published data on awake motor activity in RSD, compared to controls or other disorders.

(D) Video-polysomnography shows a total movement index (by video analysis) of 5 or more per hour of sleep.

This criterion highlights two essential points: that vPSG is essential for the diagnosis of RSD and that a total movement index cut-off of 5 movements per hour has been established. A diagnosis of RSD cannot be made on clinical history alone. vPSG is essential to identify these large body movements and provides objective data on the movements, as well as other disorders that might mimic RSD, such as obstructive sleep apnea (OSA) and PLMD. PSG without video is insufficient as large body movements are easily overlooked using only electroencephalogram (EEG) and limited electromyogram (EMG) channels.

Scoring of movements is by visual analysis of the vPSG [3]. Movements are counted that: 1) begin during sleep; 2) are clearly visible; 3) last for at least 1 s; 4) involve the whole body, arms, legs,

or head; and 5) are NOT associated with respiratory events or seizure activity, or meet the criteria for periodic limb movements of sleep (PLMS), sleep-related rhythmic movements, hypnagogic foot tremor, alternating leg muscle activation (ALMA), motor tics during sleep, or REM sleep behavior disorder [7,11]. The total movement index is calculated by adding together all scored movements and dividing by the total sleep time (in hours). Movements may be, but are not necessarily, associated with cortical arousals or awakenings.

A total movement index of 5 or more per hour accurately separates RSD from controls with 100% accuracy (sensitivity 1, specificity 1) and from children with RLS with 90% accuracy (sensitivity 1, specificity 0.833) [3]. There are no data available on the night-to-night variability of this parameter. However, restless sleep is reported to occur clinically almost every night or every night in affected children, suggesting that the night-to-night variability is relatively low. Some movements typically scored as non-periodic leg movements by leg EMG may meet criteria for RSD movements on video analysis. However, if a patient meets criteria for RLS or PLMD, a diagnosis of RSD should not be made, whether there are frequent non-periodic movements or not (criterion H).

A standard video sampling rate of “at least 1 frame per second” will be sufficient for detection of most restless movements [11] but a higher sampling rate of at least 4 frames per second is ideal.

The task force is aware that a requirement for vPSG will make it more difficult to diagnose RSD. However, this more conservative approach is considered warranted until other methods of objective measurement, such as home video monitoring or wearable devices, are found to be accurate. Objective measurement, instead of reliance on history alone, will help prevent overdiagnosis.

(E) Restless sleep occurs at least three times per week.

The current literature and consensus among task force members indicate that restless sleep in pediatric RSD is typically described on a nightly basis; however, for the purposes of diagnosis, the task force agreed that “at least three times a week” is an appropriate determinant of frequency. It is considered likely that this frequency criterion will help differentiate clinically significant cases that might need treatment from individuals with restless sleep as an occasional symptom.

(F) Restless sleep has been present for at least three months.

The duration of symptoms was unanimously agreed upon as being an important criterion for the diagnostic process. “At least three months” is a typical determinant of chronic persistence and, as such, will help exclude restless sleep occurring due to transient events such as acute otitis media or acute pain.

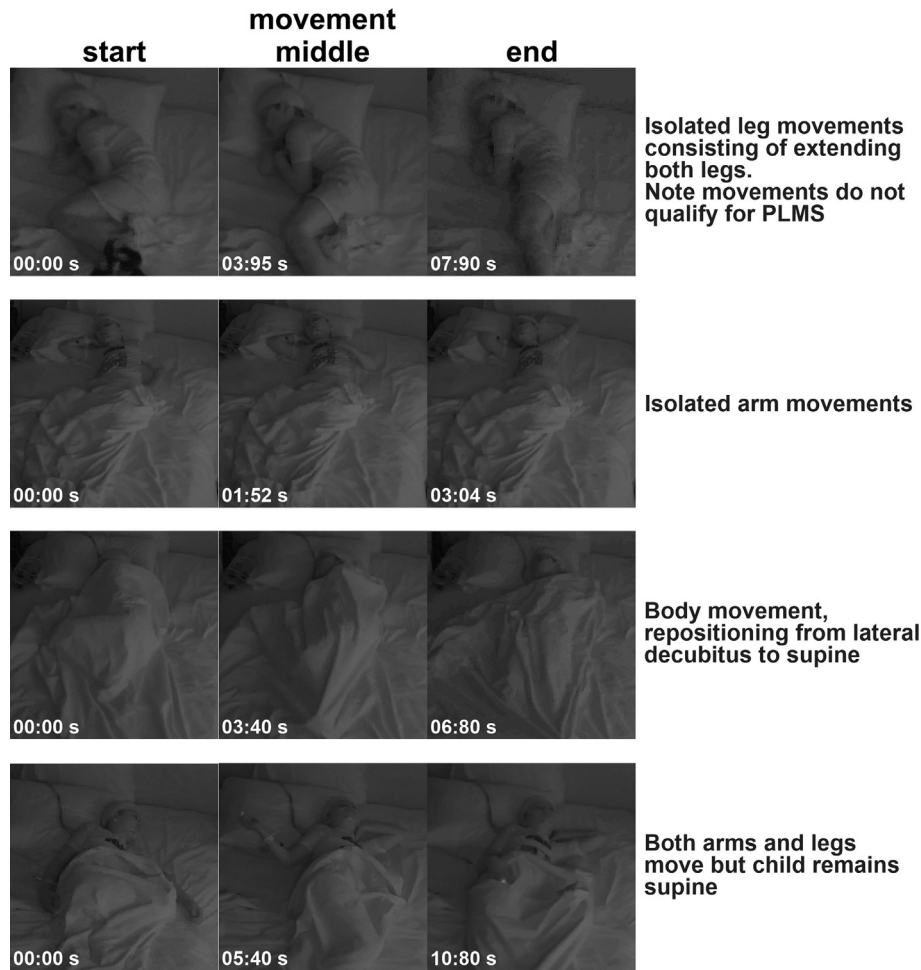


Fig. 1. Typical body movement types in children with RSD. Images taken during video-polysomnography demonstrating body movements in a child with RSD. Photos in the same row show the initial position during sleep, mid-movement position, and final position with the time spent in the movement (seconds). Images are taken with an infrared camera synchronized to the polysomnogram at a sampling rate of 30 frames per second.

- (G) Restless sleep causes clinically significant impairment in behavioral, educational, academic, social, occupational, or other important areas of functioning, as reported by the patient's parent, caregiver, or bedpartner, or by the patient (eg, daytime sleepiness, irritability, fatigue, mood disturbance, impaired concentration, or impulsivity).

The task force agreed that based on the current literature and clinical experience, the specifier for causing clinically significant impairment was appropriate for the diagnostic criteria of RSD. This identifies restless sleep severe enough to affect function, a key issue in treatment decisions. The current evidence shows an impact on daytime alertness as well as cognitive, emotional, and behavioral domains manifest by poor school performance, daytime sleepiness, behavioral concerns, fatigue, irritability, and/or hyperactivity [2]. These are impacts similar to those seen in children and adolescents with OSA, RLS, insufficient sleep, and chronic insomnia [10,12,13].

- (H) The condition is not better explained by another sleep disorder, medical disorder, mental disorder, behavior disorder, environmental factor, (eg, sleep-disordered breathing, restless legs syndrome, periodic limb movement disorder, sleep-related rhythmic movement disorder, insomnia disorder, atopic dermatitis, seizure disorder, etc.), or the physiological effects of a substance (eg, caffeine).

The differentiation of RSD from other disorders is an essential aspect of the diagnostic process. Important common and less common mimics of RSD are presented in Table 3. More detailed information can be found in a recent publication that provides a systematic review of conditions associated with restless sleep in children [6].

Sleep disorders to be differentiated from RSD are sleep-disordered breathing (SDB) [14], RLS [15], PLMD [16], sleep-related rhythmic movement disorder [10], sleep-related bruxism [17], and chronic insomnia disorder [10]. Restless sleep associated with SDB typically improves with adenotonsillectomy indicating that restlessness can be secondary to SDB events [18,19]. RLS and PLMD have other diagnostic features that can be determined by history and/or polysomnography [3]. An “urge to move” the legs is an essential aspect of RLS. Also, PLMS and a family history of RLS are common with RLS but not RSD [3]. If a child does not have sufficient language skills to describe the urge to move and does not meet criteria for PLMD, it is possible that an initial diagnosis of RSD could change to RLS over time [20]. Although restlessness while awake can be part of insomnia disorder, restless sleep is not typically described [10]. In addition, sleep onset or maintenance problems are not prominent in RSD [2].

Medical conditions that may result in restless sleep include asthma [21], acute otitis media [22], atopic dermatitis (eczema)

Table 3
Differential diagnosis of restless sleep disorder (RSD).

Common mimics
• Sleep-disordered breathing
• Restless legs syndrome
• Periodic limb movement disorder
• Sleep-related rhythmic movement disorder
• Sleep-related bruxism
• Chronic insomnia disorder
• Asthma (especially if poorly controlled)
• Acute otitis media
• Atopic dermatitis (eczema)
• Pain
• Physiologic effect of a substance (eg, caffeine)
Less common mimics
• Sleep-related seizures
• Motor tics during sleep
• Hypnagogic foot tremor
• Alternating leg muscle activation (ALMA)
• REM sleep behavior disorder

[23], and pain [24]. In addition, the physiologic effect of a substance, especially caffeine, may cause restless sleep [25].

Less common conditions to be differentiated from RSD include sleep-related seizures, motor tics during sleep [7], hypnagogic foot tremor, alternating leg muscle activation (ALMA), and REM sleep behavior disorder [10].

There are other conditions best considered as comorbid with RSD, ie, in addition to rather than instead of RSD. Restless sleep is often described in attention-deficit/hyperactivity disorder (ADHD), autism, and iron deficiency anemia but not as a consistent feature [6,26–28]. Interestingly, iron deficiency has been identified in children with RSD [2,29] and might be a common underlying pathophysiologic features of ADHD, autism, and RSD.

4.2. Features supporting the diagnosis of restless sleep disorder

In addition to the essential criteria, the task force identified two supportive features that can help confirm a diagnosis of RSD. The current evidence is not sufficient to require these features to be present for an RSD diagnosis.

- (1) Large body movements during sleep typically occur throughout the night.

RSD movements were found to be evenly distributed throughout the night by time of night distribution analysis on vPSG, occurring in both non-REM and REM sleep [3]. This is in contrast to the clustering of PLMS in RLS during the first half of the night, as well as the occurrence of most non-REM parasomnias during the first half.

- (2) Delayed sleep onset is uncommon.

Children with RSD typically do not experience a delay in sleep onset, by history or PSG [2]. No significant difference in sleep latency was found on PSG between RSD and controls.

The task force concluded that there is not enough evidence about family history of restless sleep to consider it a supportive feature.

5. Diagnostic coding for restless sleep disorder

The task force examined the International Statistical Classification of Diseases and Related Health Problems, 10th revision (ICD-

10-CM) codes and, since there is no specific code for RSD, agreed that the most appropriate code in the current classification system is G47.69 – “Other sleep-related movement disorder”. This congruously classifies RSD under “Diseases of the nervous system” (G00-G99), “Sleep disorders” (G47), and “Sleep-related movement disorders” (G47.6). “Other sleep disorder” (G47.8) and “Sleep disorder, unspecified” (G47.9) were not considered specific enough. G47.69 is a billable code. Sleep-related rhythmic movement disorder, benign sleep myoclonus, and propriospinal myoclonus at sleep onset are under this same code. It is recommended by the task force that a specific code for RSD be developed with future ICD code updates.

6. Compatibility with international classification of sleep disorders (ICSD) and diagnostic and statistical manual (DSM) formats

There was careful attention to adherence to ICSD and DSM guidelines for the construction of diagnostic criteria, including clarity and conciseness of each criterion. The task force was fortunate to have members with extensive experience on committees for the ICSD (RPA, DLP, SK, JAO) and DSM (DLP, JAO).

7. Discussion of name selection

The task force considered potential names for this clinical entity, including restless sleep disorder (RSD), restless sleep syndrome (RSS), restless sleep disease (RSD), disruptive sleep disorder (DSD), and DelRosso-Ferri syndrome (DRFS). Restless sleep disorder (RSD) was chosen because it is appropriately descriptive of the core feature and identifies this as a clinical concern. “Primary restless sleep disorder” was also discussed. However, “secondary restless sleep disorder” should not be considered a disorder. Rather, restless sleep as a symptom or sign should be differentiated from restless sleep disorder, which is now clearly defined by the RSD diagnostic criteria. “Pediatric restless sleep disorder” was also considered but both ICSD and DSM formats discourage diagnostic titles restricted by age or different diagnostic criteria by age, even if the disorder is seen predominantly in children or adults. Examples include sleep-related rhythmic movement disorder (children), sleep-related leg cramps (adults), attention-deficit/hyperactivity disorder (children), and oppositional defiant disorder (children). However, an important note in the RSD diagnostic criteria identifies the current intended use for children and adolescents, ages 6–18 years (Table 1), but allows flexibility for other age groups should future research warrant this. The name restless sleep disorder (RSD) is to be differentiated from reflex sympathetic dystrophy (RSD), now known as complex regional pain syndrome (CRPS), and restless legs syndrome (RLS).

8. Search term in PubMed

A review of U.S. National Library of Medicine, MEDLINE/PubMed Medical Subject Headings (MeSH) found that there is no MeSH search term specific for “restless sleep” or “restless sleep disorder”. “Restless” maps to “restless legs syndrome”. Using the search term “restless AND sleep AND disorder” produced 3478 results; “restless sleep” produced 251 results; and “restless sleep disorder” produced two results, this last search missing 2 of the 4 recent papers that included children with RSD (MEDLINE/PubMed accessed 06/20/2020). The task force submitted a request for an update in MeSH search terms to include “restless sleep disorder” as a specific search term.

9. Conclusions

Diagnostic criteria for RSD have been developed and are intended for use in both clinical and research settings.

9.1. Directions for future research

It is hoped that these criteria will facilitate future research in the field and that future studies will bring greater understanding of the pathogenesis and treatment of this disorder. These criteria could also be a basis for possibly expanding the diagnosis of RSD to children younger than 6 years of age and adults. Other recommended areas for future research include the natural course of RSD at different development stages throughout the life span, population prevalence of RSD, diagnosis of RSD using portable devices, clinical impairments related to RSD, comorbidity with other disorders, and response to iron supplementation or other treatment options.

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CRedit authorship contribution statement

Lourdes M. DelRosso: Supervision, Conceptualization, Data curation, Project administration, Writing - original draft. **Raffaele Ferri:** Visualization, Conceptualization, Methodology, Data curation, Resources, Writing - original draft. **Richard P. Allen:** Conceptualization, Methodology. **Oliviero Bruni:** Conceptualization, Methodology. **Diego Garcia-Borreguero:** Conceptualization, Methodology. **Suresh Kotagal:** Conceptualization, Methodology. **Judith A. Owens:** Conceptualization, Methodology. **Patricio Peirano:** Conceptualization, Methodology. **Narong Simakajornboon:** Conceptualization, Methodology. **Daniel L. Picchietti:** Supervision, Conceptualization, Methodology, Project administration, Writing - review & editing.

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Conflict of interest

The authors have no competing interests to declare.

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.2020.08.011>.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.sleep.2020.08.011>.

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