



## Original Article

# Pediatric restless legs syndrome diagnostic criteria: an update by the International Restless Legs Syndrome Study Group<sup>☆</sup>



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## ABSTRACT

**Background:** Specific diagnostic criteria for pediatric restless legs syndrome (RLS) were published in 2003 following a workshop at the National Institutes of Health. Due to substantial new research and revision of the adult RLS diagnostic criteria, a task force was chosen by the International Restless Legs Syndrome Study Group (IRLSSG) to consider updates to the pediatric diagnostic criteria.

**Methods:** A committee of seven pediatric RLS experts developed a set of 15 consensus questions to review, conducted a comprehensive literature search, and extensively discussed potential revisions. The committee recommendations were approved by the IRLSSG executive committee and reviewed by the IRLSSG membership.

**Results:** The pediatric RLS diagnostic criteria were simplified and integrated with the newly revised adult RLS criteria. Specific recommendations were developed for pediatric application of the criteria, including consideration of typical words used by children to describe their symptoms. Pediatric aspects of differential diagnosis, comorbidity, and clinical significance were then defined. In addition, the research criteria for probable and possible pediatric RLS were updated and criteria for a related condition, periodic limb movement disorder (PLMD), were clarified.

**Conclusions:** Revised diagnostic criteria for pediatric RLS have been developed, which are intended to improve clinical practice and promote further research.

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## 1. Introduction

Restless legs syndrome (RLS), also known as Willis–Ekbom disease, is a common pediatric neurologic condition affecting 2–4% of school-aged children and adolescents [1–4]. Symptoms range from mild to severe with 25–50% of pediatric cases having moderate to severe symptoms [2]. Both adult and pediatric RLS can adversely impact sleep, mood, cognition, and quality of life [1,2,5–11]. Re-

cently the pathophysiology of RLS has been defined, with genetics, the brain dopamine system, and iron found to play important roles [1,12–15].

The current pediatric RLS diagnostic criteria were published in 2003 based on the consensus of experts at a National Institutes of Health workshop (2002) and sponsored in part by the International RLS Study Group (IRLSSG) [16]. These criteria (Supplementary Table 1) were intended to conservatively but broadly define pediatric RLS, incorporating previous criteria and research up to that time, with knowledge that further refinement would be needed. To promote further research, definite, probable, and possible RLS categories were defined. Weaknesses of these criteria include their complexity and the fact that they are substantially different than the adult RLS diagnostic criteria.

Because there have been numerous pediatric RLS publications since 2003 and because the adult RLS diagnostic criteria were

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being updated [17], the IRLSSG Executive Committee appointed a pediatric RLS committee to reach consensus on possible revisions to the pediatric RLS diagnostic criteria and pediatric criteria for a related condition, periodic limb movement disorder (PLMD). In addition, the Sleep-Wake Work Group of the *Diagnostic and Statistical Manual Mental Disorders* (DSM) Task Force requested consideration that the pediatric RLS criteria be integrated with the adult RLS criteria, for inclusion of pediatric RLS in the fifth edition of the DSM (DSM-5)[18].

Our report summarizes the rationale and recommendations of the pediatric RLS committee. The IRLSSG has approved and endorsed these updates.

## 2. Methods

A panel of seven pediatric RLS experts was approved in spring 2010 by the IRLSSG Executive Committee to provide recommendations on revision of the pediatric RLS diagnostic criteria. The committee, composed of the authors of this paper, began correspondence in spring 2010 to identify key issues related to potential revisions. Fifteen consensus questions were agreed on; thirteen were discussed at a face-to-face meeting in June 2010 and two were discussed later. The questions were as follows, with responses elaborated in the next section: (1) Should the definite 1 RLS definition (Supplementary Table 1) require both urge and discomfort? (2) Should the statement, “in the child’s own words” be retained for definite RLS? (3) Should the lower age limit for definite RLS be left open or be specified? (4) Is criterion 5 (differential diagnosis) in the new IRLSSG adult criteria (Table 1) adequate for children? (5) Is the specifier for Clinical Significance in the new IRLSSG adult criteria adequate for children? (6) Does the specifier for Clinical Course in the new IRLSSG adult criteria apply to children? (7) Should the definite 2 RLS category be eliminated? (8) Do the supporting features in the IRLSSG adult criteria apply to children? (9) Should probable 1 RLS (not worse at night) be retained for children, perhaps without the supportive criteria needed? (10) Should probable 2 RLS (description of RLS symptoms by someone other than the child) be retained for children, perhaps as possible RLS? (11) Should the pediatric RLS diagnostic criteria be merged with the

adult RLS diagnostic criteria? (12) Should PLMD be retained as a pediatric diagnostic entity and what is its relationship to pediatric RLS? (13) How should clinical sleep disturbance be defined in the PLMD definition? Subsequently, two additional questions were discussed and agreed on: (14) What conditions commonly are comorbid with pediatric RLS and what is their relationship to RLS? (15) Are these criteria intended for both clinical and research applications?

To help integrate the medical literature into the recommendations, the committee conducted a formal literature search. Using the PubMed database, first in May 2010 and updated in March 2011, the search term *restless legs AND children* identified a total of 190 articles. Abstracts from these articles were reviewed to determine if the articles included information on children or adolescents and if they contained original data and any consensus statements on pediatric diagnosis. To be inclusive, no minimum number of subjects was applied, but review articles, per se, were not retained, unless diagnostic criteria consensus or discussion was included. Based on the literature search and pearling (checking of reference sections for any articles otherwise missed), 71 papers were selected for full review. Articles were divided among the seven committee members, with data pertinent to pediatric RLS diagnosis extracted and recorded on specific literature review forms. This information was then integrated into the subsequent recommendations.

After approval of the written report by all seven committee members, the recommendations were forwarded to the IRLSSG executive committee for review and approval and then to the IRLSSG membership for comment.

## 3. Revised diagnostic criteria for pediatric RLS

### 3.1. Integration with the essential RLS criteria for adults

The same diagnostic criteria for adults and children are desirable to promote accuracy, consistency, and fluency of application. In addition, it is unlikely that the basic underlying pathophysiology of RLS is different at different ages. However, there was concern at the National Institutes of Health workshop in 2002 that children

**Table 1**  
International Restless Legs Syndrome Study Group consensus diagnostic criteria for restless legs syndrome.

Restless legs syndrome (RLS), a neurological sensorimotor disorder often profoundly disturbing sleep, is diagnosed by ascertaining a syndrome that consists of all of the following features:
(1) An urge to move the legs usually but not always accompanied by or felt to be caused by uncomfortable and unpleasant sensations in the legs <sup>a,b</sup>
(2) The urge to move the legs and any accompanying unpleasant sensations begin or worsen during periods of rest or inactivity such as lying down or sitting
(3) The urge to move the legs and any accompanying unpleasant sensations are partially or totally relieved by movement, such as walking or stretching, at least as long as the activity continues <sup>c</sup>
(4) The urge to move the legs and any accompanying unpleasant sensations during rest or inactivity only occur or are worse in the evening or night than during the day <sup>d</sup>
(5) The occurrence of the above features are not solely accounted for as symptoms primary to another medical or a behavioral condition (e.g., myalgia, venous stasis, leg edema, arthritis, leg cramps, positional discomfort, habitual foot tapping). <sup>e</sup>
Specifier for clinical significance of RLS The symptoms of RLS cause significant distress or impairment in social, occupational, educational, or other important areas of functioning by the impact on sleep, energy/vitality, daily activities, behavior, cognition, or mood
Specifiers for clinical course of RLS <sup>f</sup> (A) Chronic-persistent RLS: symptoms when not treated would occur on average at least twice weekly for the past year (B) Intermittent RLS: symptoms when not treated would occur on average <2/week for the past year, with at least five lifetime events

<sup>a</sup> Sometimes the urge to move the legs is present without the uncomfortable sensations and sometimes the arms or other parts of the body are involved in addition to the legs.

<sup>b</sup> For children, the description of these symptoms should be in the child’s own words.

<sup>c</sup> When symptoms are very severe, relief by activity may not be noticeable but must have been previously present.

<sup>d</sup> When symptoms are very severe, the worsening in the evening or night may not be noticeable but must have been previously present.

<sup>e</sup> These conditions, often referred to as “RLS mimics,” have been commonly confused with RLS, particularly in surveys because they produce symptoms that meet or at least come close to meeting criteria 1–4 above. The list here gives some examples that have been noted as particularly significant in epidemiologic studies and clinical practice. However, RLS may also occur with any of these conditions, requiring a clear delineation of the RLS feelings from the other sensations.

<sup>f</sup> The clinical course criteria do not apply for pediatric cases or for some special cases of provoked RLS such as pregnancy or drug-induced RLS, in which the frequency may be high but limited to the duration of the provocative condition.

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