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SCIENTIFIC INVESTIGATIONS

Parental Symptom Report and Periodic Limb Movements of Sleep in Children

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Study Objectives: To examine the prevalence of raised periodic limb movements of sleep (PLMS) index in children referred for polysomnography (PSG) and whether parental report of symptoms correlates with objective measurement during PSG.

Methods: Records of children undergoing PSG from January 2006 to July 2006 were retrospectively reviewed. At their initial sleep clinic visit, parents had been asked whether their child was restless or moved their legs excessively during sleep. Their response to these questions was compared to the child's PLMS index (number of periodic limb movements per hour) during a full PSG. PLMS were scored according to internationally accepted criteria.

Results: Data were examined for 101 children (60 male) with mean age 6.5 years (range 1.2 to 17.6 years). Excessive leg movements were reported by parents in 50% and restlessness in 73%. A raised PLMS index (defined as ≥5 per hour) was noted in 10 cases (prevalence 10%). Asking parents about whether their child kicks their legs excessively in sleep had sensitivity 50%, specificity 51%, positive predictive value (PPV) 10%, negative predictive value (NPV) 90% and positive likelihood ratio (LR^+) 1.02 when compared to objective analysis. Asking parents about whether their child is restless in sleep had sensitivity 70%, specificity 26%, PPV 9%, NPV 89% and LR^+ 0.95.

Conclusions: Asking parents about their child's symptoms is not an accurate predictor of raised PLMS index. We recommend that leg electromyography be used in all pediatric sleep studies to record PLMS.

Keywords: Periodic limb movements of sleep; child; questionnaires; polysomnography; sensitivity and specificity; likelihood ratio.

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Periodic limb movements of sleep (PLMS) are recognized as stereotyped, repetitive movements of the limbs during sleep. Movements described include extension of the great toe (similar to the Babinski response), flexion of the foot and lower leg or abrupt extension of the lower leg.[1] Movements of the upper limbs may also occur. PLMS occur most commonly in light sleep (stage 1 and 2) and are scored according to internationally accepted criteria originally developed by Coleman[2] and recently updated by the American Academy of Sleep Medicine.[3] An index of 5 or more PLMS per hour is considered abnormal, although data supporting this figure are limited.[4] PLMS in children have been associated with iron deficiency,[5-7] symptoms of attention deficit hyperactivity disorder (ADHD)[8-12] and "growing pains."[1,13]

In periodic limb movement disorder (PLMD), a rate of 5 or more PLMS per hour is accompanied by clinical sleep disturbance with a further diagnostic criterion that the leg movements cannot be accounted for by sleep disordered breathing (SDB) or medication such as antidepressants.[4] The condition may be due to underactive dopaminergic pathways in the central nervous system.[14-16] PLMD is a well-recognized but controversial phenomenon in adults but is thought to be under-recognized in children.[17] PLMS index ≥5 has been noted to be uncommon in children and adolescents compared to adults aged >40 years.[18] A large European survey reported a prevalence of 3.9% in the general population including 3.2% in adolescents aged 15 to 19 years, but polysomnography (PSG) data did not form part of the study.[19] Studies including PSG have quoted prevalence in children ranging from 5.6% to 26% in several general and referred populations.[10,17,20,21]

Restless legs syndrome (RLS), which can be considered a separate but related condition to PLMD, is diagnosed clinically by the presence of uncomfortable sensations in the legs which tend to be worse in the evening or night and are relieved by movement. Most people with PLMS do not have clinical symptoms of RLS but up to 80% of RLS sufferers have significant PLMS noted during PSG.[22] PLMS do not form part of the essential diagnostic criteria for RLS but they do provide supportive evidence.[4]

The prevalence of PLMS in children is not well defined[23] and symptoms are often not well reported by children or their par-

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ents. Furthermore, PLMS are not routinely recorded and scored in all pediatric sleep laboratories. We sought to examine the prevalence of raised PLMS index in a population of children referred for PSG. We also examined whether parental report of symptoms predicted presence of PLMS on objective measurement. To limit confusion among PLMS, PLMD, and RLS, which often arises in the literature, we primarily examined PLMS as a PSG finding rather than PLMD or RLS as clinical disorders.

METHODS

Subjects

Records of children who underwent PSG at Sydney Children's Hospital, Randwick, NSW, Australia between January and July 2006 were examined. At their initial clinic appointment with one of the authors (AYT), a standard questionnaire had been administered. This included asking parents whether their child was restless or moved their legs excessively during sleep. From the patient records, data were obtained for the following parameters: age; sex; indication for PSG; parental report of restlessness in sleep; parental report of excessive leg movements in sleep; PLMS index (number of PLMS per hour); and mixed/obstructive apnea/hypopnea index (MOAHI: number of mixed or obstructive apneas and hypopneas per hour of sleep). Children were excluded if they were younger than one year of age, did not have a complete diagnostic PSG, or had a neuromuscular or severe neurodevelopmental condition which could interfere with estimation of PLMS. We included studies supervised by a single physician to ensure consistency in questioning about symptoms.

Polysomnography

PSG was performed using Compumedics S or E Series (Compumedics, Melbourne, Australia). Studies were scored by the authors according to the criteria of Rechtschaffen and Kales.[24] Sleep stages were scored manually using electroencephalography, electro-oculography, and submental electromyography (EMG). Audio and video were digitally recorded. Respiratory efforts were measured by respiratory bands as well as EMG of diaphragm and abdominal muscles. Nasal airflow was recorded using nasal cannulae with a pressure transducer and oronasal flow was measured by thermistor. Oxygen saturation (SpO_2) was measured by pulse oximetry (Nellcor 595, Nellcor Puritan Bennett, Pleasanton, CA, USA) and electrocardiogram was recorded continuously. Carbon dioxide was recorded continuously by a transcutaneous electrode (Radiometer, Copenhagen, Denmark). Bilateral anterior tibialis EMG signals were calibrated at commencement of recording by voluntary movement in older children and as part of general calibration in younger children.

Respiratory Event Scoring

Central apnea was defined as cessation of airflow and effort for two or more respiratory cycles with SpO_2 decrease of >3% from baseline. Obstructive apnea was defined as cessation of oronasal airflow with continuing respiratory effort regardless of SpO_2. Hypopnea was defined as airflow 20% to 50% of baseline amplitude and as central or obstructive according to associated absence or presence of respiratory effort. Events with both central and obstructive components were scored as mixed apneas. MOAHI was calculated as the total number of mixed apneas, obstructive apneas and obstructive hypopneas per hour of sleep. MOAHI ≥1 is considered abnormal in children.[25]

PLMS Scoring

Periodic limb movements were scored according to accepted criteria as follows: (1) movements 0.5 s to 5 s in duration; (2) interval between movements 5 s to 90 s; (3) ≥4 movements in sequence; (4) movement at least 25% of calibrated magnitude.[3] Movements following arousals due to respiratory events were not scored.

Statistical Analysis

Data were compiled in Microsoft Excel (Microsoft Corporation, Redmond, WA, USA) and analysed using SPSS Version 12.0 (SPSS Inc, Chicago, IL, USA). PLMS data were dichotomised into categories of <5 and ≥5 with tables constructed to calculate sensitivity, specificity, positive predictive value (PPV), negative predictive value (NPV) and positive likelihood ratio (LR^+) for each of the symptoms investigated. McNemar chi-squared tests were used to compare parental symptom report with objective measurement of PLMS.

Ethical approval for the study was obtained from the Human Research Ethics Committee of the South Eastern Sydney Area Health Service.

RESULTS

Subjects

There were 235 PSGs undertaken in our unit between January and July 2006 of which 101 were included in the analysis. Reasons for exclusion were age less than one year (n = 26), study supervised by another physician (n = 82), neuromuscular disease (n = 5), severe neurodevelopmental problems (n = 7), and repeat or incomplete studies (n = 14). In the included studies, there were 60 males and 41 females with mean age 6.5 years (range 1.2 to 17.6 years). Indications for PSG were snoring (n = 89), restless sleep (n = 4), CPAP/VPAP adjustment (n = 4), excessive daytime sleepiness (n = 2), oxygen titration (n = 1), and previous obstructive sleep apnea (n = 1). Characteristics of subjects are summarised in Table 1.

Prevalence Data

Of the 101 included subjects, PLMS index was 0 for 73 (72%), 0.1 to 4.9 for 18 (18%) and ≥5 for 10 subjects (10%). Overall range of PLMS index was 0 to 96.7. Obstructive sleep apnea (OSA), defined as MOAHI ≥1, was noted in 43 subjects (43%). Of the 10 subjects with PLMS index ≥5, 8 (80%) were male and 6 (60%) showed concurrent evidence of OSA. Hence, 4 (4%)

Table 1—Characteristics of All Subjects, Subjects with PLMS Index ≥5 and Subjects with PLMS Index <5

	All Subjects (n=101)	PLMS Index ≥ 5 (n=10)	PLMS Index < 5 (n=91)
Age (y)	6.5 (4.2)	9.0 (5.7)	6.2 (3.9)
Male	60 (59.4%)	8 (80%)	52 (57.1%)
MOAHI ≥ 1	44 (43.6%)	6 (60%)	38 (41.8%)
PLMS Index	0 (0 – 96.7)	10.7 (5 – 96.7)	0 (0 – 4)
MOAHI	0.6 (0 – 64.9)	2.2 (0 – 6.1)	0.6 (0 – 64.9)

Data are mean (SD) for age; N (% of n listed at top of column) for sex and MOAHI ≥ 1; median (range) for PLMS Index and MOAHI due to skewed distributions.
PLMS, periodic limb movements of sleep; MOAHI, mixed obstructive apnea/hypopnea index.

of our subjects could be considered to have primary PLMD. One subject with PLMS index ≥5 was taking fluoxetine, which may have exacerbated the condition.[26] Parents of this child had reported both restlessness and excessive leg movements in sleep. This child was also the only one of the 10 with raised PLMS index to have reported "growing pains."

Association of PLMS with Symptoms

Excessive leg movements in sleep were reported by 50 parents (50%) and restlessness was reported by 74 parents (73%). Parental report rate was significantly higher than the rate obtained by objective recording for excessive leg movements (McNemar $\chi^2_{1df} = 30.4$, $p < 0.001$) and for restlessness (McNemar $\chi^2_{1df} = 56.7$, $p < 0.001$).

Asking parents about whether their child kicks their legs in sleep had sensitivity 50%, specificity 51%, PPV 10%, NPV 90% and LR^+ 1.02 when compared to objective analysis of sleep studies for PLMS (Table 2). Asking parents about whether their child is restless in sleep had sensitivity 70%, specificity 26%, PPV 9%, NPV 89%, and LR^+ 0.95 (Table 3).

DISCUSSION

Our study of children referred to a specialist pediatric sleep clinic demonstrates that asking parents about their child's leg movements or restlessness during sleep is not predictive of the presence of abnormal PLMS index on objective measurement. Negative report of these symptoms was fairly accurate in excluding significant PLMS but overall the questions performed poorly as diagnostic tools. The positive likelihood ratios of 1.02 for excessive leg movements and 0.95 for restlessness indicate that the presence of these symptoms does not alter the probability of significant PLMS to an important degree.

The prevalence of PLMS index ≥5 of 10% in our referred population is similar to that reported by Crabtree et al of 11.9% in a community sample ($\chi^2_{1df} = 0.33$; $p = 0.57$) and 8.4% in a referred population ($\chi^2_{1df} = 0.24$; $p = 0.63$).[17] It is also not statistically different from the rate of 5.6% reported by Kirk and Bohn in a referred population ($\chi^2_{1df} = 2.76$; $p = 0.10$).[20] Traegar et al performed PSG on a group of children specifically excluding those likely to have OSA and reported a similar rate to ours of 8% ($\chi^2_{1df} = 0.26$; $p = 0.61$).[21] Our rate, however, was lower than the 26% reported by Chervin and Archbold in children referred for suspected sleep disordered breathing ($\chi^2_{1df} = 8.89$; $p = 0.003$).[10] Chervin and Archbold used slightly broader criteria for scoring PLMS (periodicity of 5 s to 120 s and sequence ≥3 movements), which may have resulted in more children being classified with raised PLMS index. It is important to note, however, that study populations and referral patterns are likely to have differed between the above studies so it is difficult to draw firm conclusions from these comparisons.

Chervin et al examined symptoms useful in predicting PLMD in order to compile a questionnaire.[13] Asking about restlessness of the legs, growing pains in bed, insomnia, and morning headache were moderately predictive; but questions about leg movements during sleep, daytime sleepiness, and daytime behavior were not. Overall sensitivity and specificity of the final questionnaire based on symptoms associated with PLMS ≥5 per hour were only 79% and 56% respectively. Receiver-operator analysis suggested only moderate utility of their PLMS score as a diagnostic tool. This implies, as does our study, that clinical suspicion cannot be relied upon in the diagnosis of PLMS.

Martinez and Guilleminault noted that the report of leg pains at morning waking was associated with PLMS of any degree but that other symptoms such as sleep initiation problems, sleep maintenance problems, and daytime tiredness were not specific.[1] Reporting of leg pains had sensitivity 80% and specific-

Table 2—Comparison of Parental Report of Excessive Leg Movements in Sleep with PLMS Index Measured on PSG

		PLMS Index ≥ 5		
		Yes	No	
Parental Report	**Yes**	5	45	50
	No	5	46	51
		10	91	101

Sensitivity = 50% PPV = 10%
Specificity = 51% NPV = 90%
$LR^+ = 1.02$
PLMS, periodic limb movements of sleep

Table 3—Comparison of Parental Report of Restlessness in Sleep with PLMS Index Measured on PSG

		PLMS Index ≥ 5		
		Yes	No	
Parental Report	**Yes**	7	67	74
	No	3	24	27
		10	91	101

Sensitivity = 70% PPV = 9%
Specificity = 26% NPV = 89%
$LR^+ = 0.95$
PLMS, periodic limb movements of sleep

ity 82% and may be a more useful symptom about which to enquire than leg movements in sleep or restlessness, although only one of the 10 subjects with raised PLMS index in our study reported such pain.

Picchietti and Walters, in a study of 129 children with PLMS index >5, reported that parents had noted the presence of PLMS in only 25 cases (19%), and even after being asked to look for leg movements did not notice them in 39 cases (30%).[27] Furthermore, symptoms were only reported in 3 of 16 children considered severe cases with PLMS index >25. Another study noted substantial disparity between parental symptom report and PLMS index in a study of children with ADHD.[9] Crabtree also noted no significant difference in reported symptoms by parents of children with and without PLMD,[17] again highlighting the difficulty in using parental reports for diagnosis.

PLMD is an important diagnosis to consider as patients may be misdiagnosed with conditions such as ADHD and seizures if the diagnosis is not considered.[26] Normal phenomena such as body shifts, hypnic jerks and phasic REM twitches may also be misinterpreted as PLMS.[9] The condition is also potentially treatable, particularly if associated with SDB or low ferritin levels.[1] Unfortunately, we were not able to perform repeat PSG on all of our patients to examine the effect of treatments such as adenotonsillectomy, CPAP, or iron therapy on PLMS index.

Six of the 10 subjects with raised PLMS index in our study also showed evidence of SDB. As leg movements directly following arousal due to respiratory events were not scored, we feel that PLMS can be considered a separate but perhaps related phenomenon in these children. Kirk and Bohn[20] (60%), and Martinez and Guilleminault[1] (50%) have reported similar comorbidity rates. Martinez and Guilleminault noted similar rates of SDB in children with and without PLMs, again implying independence of the conditions. They also noted that in 15 of 29 comorbid children treated for SDB, PLMs also resolved, but PLMs persisted or increased in the others.[1] Chervin and Archbold found that PLMS index was related to hyperactivity scores in children with SDB—but not those without—and hypothesized that SDB may act as an effect modifier of PLMS.[10] These and other studies highlight the difficulty in assessing the relationship between PLMS and SDB and the necessity to exclude movements associated with SDB-related arousals from scoring of PLMS. Regarding other comorbidities, none of our subjects was diagnosed with narcolepsy or REM sleep behavior disorder; these disorders are also commonly associated with PLMS. As data were not available for specific symptoms of RLS, we were not able to assess accurately the presence of this associated condition in our study sample.

PSG is widely recognized as the gold standard for diagnosis of PLMS as well as many other sleep disorders, but its complexity and expense limit its use in many environments. Actigraphy has been assessed as a more economical, less labor-intensive alternative method of diagnosis, but results have been conflicting.[28-30] PSG, therefore, remains essential in the recording of PLMS.

As this was a retrospective study, PSGs had been reported in a clinical context and hence the reporter was not necessarily blinded to the symptom reports of the parents. We feel, however, that the use of standard, validated criteria to score PLMS counterbalanced this potential bias. Our sample size was based on pragmatic considerations, but we feel it compares well with other studies on this topic. Our use of a population referred for investigation of sleep problems, primarily snoring, also means that our results cannot be extrapolated to the general population but should be relevant for the practice of sleep medicine in most pediatric units.

CONCLUSION

Parental report of excessive leg movements or restlessness in sleep is not an accurate predictor of raised PLMS index compared with objective measurement during PSG. A negative report of symptoms is fairly likely to exclude significant PLMS but overall these symptoms perform poorly as diagnostic tools. As PLMS represent a potentially treatable condition in children which is associated with significant comorbidities, we recommend objective scoring of PLMS during PSG for all children over the age of one year.

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ABBREVIATIONS

ADHD = attention deficit hyperactivity disorder
CPAP = continuous positive airway pressure
EMG = electromyography
LR^+ = positive likelihood ratio
MOAHI = mixed obstructive apnea/hypopnea index
NPV = negative predictive value
OSA = obstructive sleep apnea
PLMs = periodic limb movements
PLMD = periodic limb movement disorder
PLMS = periodic limb movements of sleep
PPV = positive predictive value
PSG = polysomnography
REM = rapid eye movement
RLS = restless legs syndrome
SD = standard deviation
SDB = sleep disordered breathing
VPAP = variable positive airway pressure

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