Sleep Medicine 64 (2019) 112–115

Contents lists available at ScienceDirect

Sleep Medicine

journal homepage: www.elsevier.com/locate/sleep

Brief Communication

Sleep-related rhythmic movements and rhythmic movement disorder beyond early childhood

Iva Prihodova [a], Jelena Skibova [b], Sona Nevsimalova [a, *]

[a] Department of Neurology and Center for Clinical Neurosciences, 1st Faculty of Medicine, Charles University and General University Hospital, Prague, Czech Republic
[b] Unit of Statistics, Institute of Clinical and Experimental Medicine, Prague, Czech Republic

ARTICLE INFO

Article history:
Received 20 February 2019
Received in revised form
14 May 2019
Accepted 16 May 2019
Available online 29 June 2019

Keywords:
Rhythmic movement disorder
Rhythmic movement type
Clinical comorbidities
Childhood and young adulthood
Polysomnography

ABSTRACT

Introduction: Sleep-related rhythmic movements (SRRMs) are common in young children and become less prevalent with increasing age. When SRRMs significantly interfere with sleep and/or affect daytime functioning, potentially resulting in injury, rhythmic movement disorder (SRRMD) is diagnosed.
Objective: The aim of our study was to assess clinical comorbidities, types of SRRMs, sleep stage/wakefulness distribution during night, and age-dependence of these parameters.
Material and methods: In sum, 45 patients (age range 1–26 years, mean age 10.56 ± 6.4 years, 29 men) were clinically examined for SRRMs or SRRMD. Nocturnal polysomnography (PSG) was recorded in 38 patients. To evaluate clinical and sleep comorbidity, the cohort of 38 patients was divided according to age into four groups: (1) younger than 5 years (N = 7), (2) 5–9 years (N = 12), (3) 10–14 years (N = 11), and (4) ≥ 15 years (N = 8).
Results: A clear relationship between perinatal risk factors and developmental disorders (attention deficit hyperactivity disorder - ADHD, specific learning disability) was found which extended population prevalence at least five times. A total of 62 recordings were evaluated in 38 patients; SRRMs were found in PSG in 31 of 38 patients (82%). No age-dependent correlation between type of SRRMs and sleep stage/wakefulness distribution during the night was observed. However, when all recordings were correlated together, rolling stereotypes occurred more frequently in REM sleep, and rocking stereotypes in superficial NREM sleep.
Conclusion: Developmental disorders and perinatal risk factors were connected with SRRMs and SRRMD in children and young adults. Rolling movements were significantly associated with REM stage and rocking stereotypes with superficial NREM sleep, independent of age.

1. Introduction

Sleep-related rhythmic movements (SRRMs) are common in normal infants and toddlers. Up to 60% of infants has been reported to exhibit one or more following rhythmic movements – body rocking, head banging or head rolling [1]. Vestibular stimulation and/or learned behavior which soothes the child at sleep onset and following night awakenings have been proposed as the initiating factors in infants and toddlers [2]. Movements generally resolve during childhood and rarely persist until adolescence or adulthood [3–6].

Sleep-related rhythmic movement disorder (SRRMD) is reserved for cases in which SRRMs involve large muscle groups while drowsy or asleep, and interfere with normal sleep and/or significantly impair the daytime function, potentially resulting in bodily injury. In addition to the three main sleep-related rhythmic movement subtypes mentioned above, the ICDS-3 [1] also includes body rolling, leg rolling, and leg banging. A combination of two or more individual stereotypes may be observed.

Polysomnographic recordings (PSG) show that rhythmic movements occur mostly during drowsiness and in non-rapid eye movement (NREM) sleep, but in some cases occur or even prevail in rapid eye movement (REM) sleep, likely more frequently in adults [1]. SRRMD has also been described in association with obstructive sleep apnea (OSA) [7–9].

The aim of our study was to characterize the type and sleep stage/wakefulness distribution of SRRMs, age-dependence of these parameters and possible association with perinatal risk factors and

* Corresponding author. Katerinska 30, 128 00 Prague 2, Czech Republic.
E-mail address: snevsi@lf1.cuni.cz (S. Nevsimalova).

https://doi.org/10.1016/j.sleep.2019.05.021
1389-9457/

developmental disorders (attention deficit hyperactivity disorder –ADHD, specific learning disabilities, others). We further aimed to evaluate the co-existence of other sleep disorders (eg, parasomnias, insomnias, sleep-related breathing disorders, restless legs syndrome), as well as other comorbidities.

2. Methods

2.1. Participants and procedures

We performed a retrospective analysis of 45 patients (age range 1–26 years, 29 men, mean age 10.56 ± 6.4 years) evaluated within the past 20 years at our Neurological Department and Sleep Center. All patients and/or their legal guardians provided signed informed consent to the examination.

A detailed interview with the patients and/or their guardians and neurological examination was done in all cases. Psychological tests were performed in only subset of patients. SSRMs were found incidentally in three patients during PSG monitoring. In all other cases repetitive movements interfered with normal sleep, in some cases resulted in impaired daytime function and exceptionally in injury, and therefore, were considered as SRRMD. In one case permanent nocturnal body rolling led to scoliosis.

Thirty-eight patients older than three years underwent video-PSG examination in our Sleep lab. However, the seven youngest patients were excluded from the monitoring. A total of 62 recordings were evaluated (some patients had two nights of PSG), and rhythmic movements were found in 43 recordings. Each PSG recording was manually scored according to the AASM rules [10], and SRRMs were subjoined to each sleep stage/wakefulness. Five types of SRRMs were distinguished: head banging, head rolling, body rocking, body rolling, and leg banging. A combination of up to three subtypes was observed in some patients. Video-PSG data were additionally correlated with each age group.

2.2. Analyses

Statistical analyses were performed using Microsoft Excel 2010 and MedCalc software. Pearson χ^2-test for contingency tables was used to determine a statistical significance with respect to possible association between sleep stages and character of movements, comorbidities and other covariates. Whenever necessary, we merged categories to meet test assumptions (adequate expected cell counts $\geq$5).

3. Results

3.1. Clinical findings

Out of 45 total patients (35.5%), 16 had prenatal and perinatal risk factors in their history: prematurity, surgical delivery for intrauterine hypoxia, and neonatal jaundice requiring phototherapy. Moreover, six (13.3%) had mildly delayed psychomotor development. The most obvious clinical comorbidity was ADHD which was present in 16 of 45 patients (35.6%). When only school-aged children (5–15 years) were considered, the number of ADHD cases increased to 54.1%. Specific learning disabilities (most often dyslexia) were found in 17.8% of cases, and similarly, if only the above-mentioned school-aged children were considered, the percentage of affected patients increased up to 33.3%. Furthermore, 12 of 45 patients (26.7%) suffered from some type of allergy. Increased anxiety was reported by 13% of patients and/or their guardians. Two children had chronic tics disorder.

Notably, three patients (8.9%) had a positive family history of SRRMs/SRRMD in their close relatives. Three siblings and a father with SRRMs/SRRMD were reported in one case. Regarding sleep disorder comorbidities (reported by patients/guardians), ronchopathy was reported most frequently (eight cases), followed by disorders of arousal (five cases). Insomnia was reported by two responders as was sleep delay and excessive daytime sleepiness.

3.2. Video- PSG results

SRRMs were detected in 31 of 38 patients (81.6%) in at least one video-PSG recording. Table 1A illustrates standard PSG parameters and indices. Insufficient sleep of youngest group was influenced by poor accommodation in the sleep lab. A survey of sleep stages including any rhythmic movement is shown in Table 1B. Rhythmic movements were found most frequently in wakefulness (23 of 38 patients, 60.5%), their frequency decreased in NREM stages proportionally to sleep profundity (from 47.4 to 22.2%); however, rhythmic movements also occurred during REM sleep (14 of 38 patients, 38.9%). It is noteworthy that women had more rhythmic movements in REM sleep ($p = 0.028$), and patients exhibiting rhythmic movements in REM sleep had a higher level of self-reported anxiety ($p = 0.042$).

The occurrence of rhythmic movements in different PSG stages in four age groups is illustrated in Table 1C. No association with age, subtypes of rhythmic movement and sleep stages/wakefulness was found. However, when statistical correlation was evaluated in all recordings together, REM-associated rhythmic movements were frequently correlated with movements during wakefulness ($p = 0.049$) and NREM 2 sleep ($p = 0.005$). REM sleep-related rhythmic movements had a tendency to head rolling ($p = 0.057$) and especially to body rolling ($p = 0.028$) compared with other sleep stages/wakefulness. Conversely, body rocking was observed most frequently in NREM 1 ($p = 0.046$). The association of rhythmic movements with different sleep stages is illustrated in Fig. 1. Leg banging was omitted due to low amount of observations ($N = 3$). Video-PSG monitoring revealed ronchopathy in eight patients (21.8%) and OSA in three children (7.9%). Disorders of arousal was present in eight cases (21%). In four cases (10.5%), bruxism was present, and periodic leg movements were verified in another four patients, and two children manifested groaning.

4. Discussion

SRRMs are common in young children and become less prevalent with increasing age. The etiology of persistent SRRMs/SRRMD until late childhood and adulthood is unclear and likely multifactorial. The persistence of SRRMs is associated with developmental

Table 1A
Sleep variables in different age groups.

Years	<5	5–9	10–14	≥15
Number of patients	N = 7	N = 12	N = 11	N = 8
Sleep latency (min)	24,3 ± 33,9	23,8 ± 23,2	35,4 ± 33,6	17,6 ± 9,1
REM latency (min)	64,9 ± 40,1	102,8 ± 26,7	90,5 ± 26,6	103,9 ± 37,3
Total sleep time (min)	354 ± 70,3	478 ± 72	455 ± 57	455 ± 38,8
Sleep efficiency (%)	65,2 ± 14,5	88,3 ± 10	88,2 ± 10	88,9 ± 5,1
Wakefulness (%)	8,7 ± 13,4	4,5 ± 3,6	6,1 ± 5,4	4,9 ± 3,2
NREM 1 (%)	4,2 ± 2,3	2,9 ± 1,3	4,2 ± 3,9	5,0 ± 5,1
NREM 2 (%)	34,9 ± 11,3	40,8 ± 5,1	38,2 ± 6,3	45,6 ± 4,6
NREM 3 (%)	16,7 ± 3,6	31,7 ± 5,2	29,0 ± 5,1	21,8 ± 5,1
REM (%)	15,0 ± 7,2	19,1 ± 4,2	22,0 ± 4,1	22,7 ± 4,1
AHI	0,5 ± 0,4	2,0 ± 2,5	1,4 ± 1,4	1,2 ± 0,9
PLMI	1,1 ± 2,9	1,5 ± 1,8	1,5 ± 3,0	1,6 ± 2,3

REM rapid eye movement sleep, NREM = non-rapid eye movement sleep, AHI – apnoea/hypopnoea index, PLMI – periodic limb movements index.

Table 1B
Survey of rhythmic movements related to different sleep stages/wakefulness in video-PSG recordings of 38 patients.

PSG recording stages	Wakefulness	NREM 1	NREM 2	NREM 3	REM
Occurrence of RMs	N = 23 60.5%	N = 18 47.4%	N = 15 39.5%	N = 8 22.2%	N = 14 38.9%

PSG = polysomnography, NREM = non-rapid eye movement sleep, REM rapid eye movement sleep, RMs = rhythmic movements.
Note: patients may have RMs in different sleep stages/wakefulness in the same recording.

Table 1C
Occurrence of rhythmic movements in different PSG stages in 38 patients divided according age.

Age (years)	<5	5–9	10–14	≥15	Significance
Number of patients	N = 7	N = 12	N = 11	N = 8	
Wakefulness	2 (28.6%)	8 (66.7%)	9 (81.8%)	4 (50.0%)	NS
NREM 1	4 (57.1%)	7 (58.3%)	4 (36.4%)	3 (37.5%)	NS
NREM 2	4 (57.1%)	5 (41.7%)	3 (27.3%)	3 (37.5%)	NS
NREM 3	0 (0.0%)	4 (33.3%)	3 (27.3%)	1 (12.5%)	NS
REM	3 (42.9%)	3 (25.0%)	5 (41.6%)	3 (37.5%)	NS

PSG = polysomnography, NREM = non rapid eye movement sleep, REM rapid eye movement sleep, NS = not significant.
Note: patients may have RMs in different sleep stages/wakefulness in the same recording.

disabilities or intellectual disorders [11]. Dyken et al. [12], first reported the association between ADHD and SRRMD, and we confirmed this association previously [4]. The relationship can be bi-directional: SRRMD appears to be more prevalent in the ADHD population [13], and children with rhythmic movements may be predisposed to develop ADHD. ADHD may thus predispose to increased motor activity not only during the day, but during nighttime as well [14].

The present study also highlights the possible role of developmental factors. More than 1/3 of our patients had positive prenatal and perinatal risk factors, more than half of patients aged 5–15 years were diagnosed with ADHD, and almost one third of children of the same age had some manifestation of specific learning disability. Our observations highly exceeded the prevalence of these disorders in childhood population [15]. Haywood and Hill [11] noted that SRRMD is not only associated with learning difficulties, but also with autism spectrum disorder, and other developmental disorders such as Tourette and/or Rett syndrome. An alteration of central motor pattern generator neural networks has been proposed as being responsible for rhythmic movements [16], and developmental disorders likely predispose to their dysfunction.

The etiology of RMD is much more complex and may involve psychological factors, namely high levels of anxiety [17,18]. A higher arousal threshold as a predisposing factor for SRRMD has been suggested in adult males with OSA [9]. SRRMD also frequently appears in association with other sleep disorders, as we have shown. Two interesting cases of head rolling movements in REM sleep behavior disorder without family history of SRRMD were published by Manni et al., [19]. Genetic predisposition to SRRMs/SRRMD has been proposed in some cases [20], however no molecular analyses have been done. Nine percent of cases in our cohort had one or more (up to four) close relatives affected by SRRMs/SRRMD. The etiology of many adult cases remains to be clarified [5].

SRRMs manifest through the whole sleep cycle [1,2], and this finding supports our study. Regarding the different age of our patients, no correlation between rhythmic movements and sleep stages/wakefulness and/or dependence on their type was detected. However, when evaluating pooled recordings of the entire cohort, REM rhythmic movements showed a tendency toward head rolling and particularly toward body rolling, while body rocking was seen most frequently in NREM 1. Shah et al. [9], speculated about loss of physiological muscle atonia transiently associated with the release of central pattern generators in the brainstem from cerebral cortical inhibition triggering rhythmic movements during REM sleep. The tendency toward rolling movements in REM sleep and rocking movements in superficial NREM sleep needs to be verified and clarified by further studies.

We evaluated SRRMs/SRRMD in a large cohort from childhood until adulthood, from clinical and PSG perspectives, and focused on association between the type of SRRMs, PSG stages and on clinical/sleep comorbidities with increasing age.

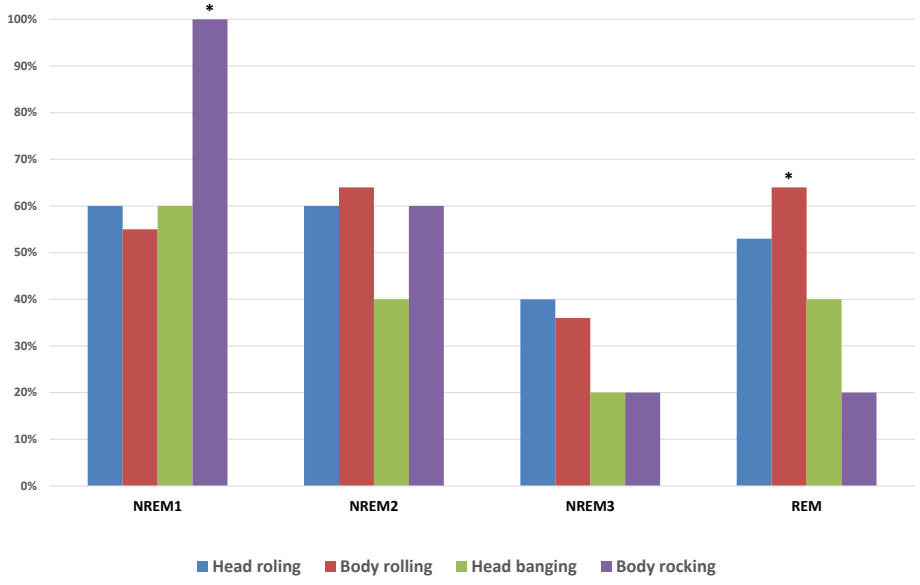

Fig. 1. Percentage of different rhythmic movement types in PSG stages. H Rol = head rolling, B Rol = body rolling, HB = head banging, B Roc = body rocking.

A limitation is the retrospective character of study and the enrollment of the patients from neurological department, in which developmental disorders are concentrated.

Acknowledgement

Supported by Progres Q27/LF1.

Conflict of interest

Nothing to be 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.2019.05.021.

References

[1] American Academy of Sleep Medicine. International classification of sleep disorders. 3rd ed. Darien, IL: American Academy of Sleep Medicine; 2014.
[2] Gwyther ARM, Walters AS, Hill CM. Rhythmic movement disorder in childhood: an integrative review. Sleep Med Rev 2017;25:62–75.
[3] Kohyma J, Matsukura F, Kimura K, et al. Rhythmic movement disorder: polysomnographic study and summary of reported cases. Brain Dev 2002;24:33–8.
[4] Stepanova I, Nevsimalova S, Hanusova J. Rhythmic movement disorder in sleep persisting into childhood and adulthood. Sleep 2005;28:851–7.
[5] Mayer G, Wilde-Frenz J, Kurella B. Sleep-related rhythmic movement disorder revisited. J Sleep Res 2007;16:110–6.
[6] Chokroverty S, Bhat S, Allen RP. Motor control and dyscontrol in sleep. In: Chokroverety S, editor. Sleep disorders medicine. Basic science, technical considerations and clinical aspects. 4th ed. New York: Springer; 2017. p. 713–57.
[7] Urrestarazu E, Escobar F, Manzanilla O, et al. Body rolling in adults associated to obstructive sleep apnea. J Sleep Med Disord 2016;3:1049.
[8] Chiaro G, Maestri M, Riccardi S, et al. Sleep-related rhythmic movement disorder and obstructive sleep apnea in five adult patients. J Clin Sleep Med 2017;13:1213–7.
[9] Shah B, Bhat S, Polos P, et al. Co-occurrence of respiratory event-related and spontaneous rhythmic movements in REM sleep. Sleep Med 2018;51:115–7.
[10] Iber C, Anconi-Israel S, Chesson AL, et al. The AASM manual for the scoring of sleep and associated events. Rules, terminology and technical specifications. 1st ed. Westchester, IL: American Academy of Sleep Medicine; 2007.
[11] Hywood PM, Hill CM. Rhythmic movement disorder: managing the child who head-bangs to get to sleep. Pediatrics Child Health 2012;22:207–10.
[12] Dyken ME, Lin-Dyken DC, Yamada T. Diagnosing rhythmic movement disorder with video-polysomnography. Pediatric Neurol 1977;16:37–41.
[13] Silvestri R, Gagliano A, Aricó I, et al. Sleep disorders in children with Attention-Deficit/Hyperactivity Disorder recorded overnight by video-polysomnography. Sleep Med 2009;10:1132–8.
[14] Walters AS, Silvestri R, Zucconi M, et al. Review of the possible relationship and hypothetical links between attention deficit hyperactivity disorder (ADHD) and the simple sleep related movement disorders, parasomnias, hypersomnias, and circadian rhythm disorders. J Clin Sleep Med 2008;4:591–600.
[15] American Psychiatric Association. Diagnostic and statistical manual of mental disorders. 5th ed. Arlington, VA: American Psychiatric Publishing; 2013.
[16] Manni R, Terzaghi M. Rhythmic movements during sleep: a physiological and pathological profile. Neurol Sci 2005;26(Suppl. 3):181–5.
[17] Laberge T, Tremblay RE, Vitaro F, et al. Development of parasomnias from childhood to early adolescence. Pediatrics 2000;106(1 Pt 1):67–74.
[18] Blunden S, Nair D. An unusual clinical phenomenon: a case of bedtime ritual with apparent sexual overtones. Clin Child Psychol Psychiatry 2010;15:55–64.
[19] Manni R, Terzaghi M. Rhythmic movements in idiopathic REM sleep behavior disorder. Mov Disord 2007;22:1797–800.
[20] Attarian H, Ward N, Schuman C. A multigenerational family with persistent sleep-related movement disorder (RMD) and insomnia. J Clin Sleep Med 2009;5:571–2.