Disturbed nighttime sleep in children and adults with rhythmic movement disorder

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ABSTRACT

Background

Rhythmic movements during sleep are frequent and often considered as benign in children. Disabling forms are diagnosed as rhythmic movement disorder and may persist in adulthood. Whether rhythmic movements severely impact sleep continuity in patients with rhythmic movement disorder remains unclear.

Objectives

To describe the clinical and polysomnographic patterns of children and adults diagnosed with rhythmic movement disorder in comparison with healthy controls and to assess the relationships between the rhythmic movements and sleep architecture.

Methods

All consecutive patients (n=50; 27 children, 35 males) with rhythmic movement disorder from a single sleep clinic (from 2006 to 2019) underwent a comprehensive clinical evaluation and a polysomnographic recording to assess the sleep continuity in comparison to 75 controls (42 children, 53 males).

Results

Disturbed nighttime sleep were reported by 82.0% of children and adult patients. Comorbid neurodevelopmental, affective or sleep disorders were found in 92%. Rhythmic movement sequences defined by videopolysomnography were found in 82.0% of patients, in wakefulness and in all sleep stages, all along the night, without any similar sequence in controls. Patients had altered sleep continuity, with low sleep efficiency, increased wake time after sleep onset, frequent periodic leg movements and apnea events. The severity of rhythmic movements was associated with disrupted sleep, after controlling for comorbid motor and respiratory events.

Conclusions

Rhythmic movement disorder is a rare, highly comorbid and disabling condition in children and adults, with frequent objective disturbed nightime sleepthat may contribute to the burden of the disease.

Keywords: Rhythmic movement disorder, bodyrocking, headbanging, polysomnography

INTRODUCTION

Rhythmic movement disorder (RMD) is a sleep-related motor disorder characterized by repetitive, stereotyped and rhythmic oscillations involving large groups of muscles predominately at sleep onset that can persist during sleep, associated with daytime or nighttime disturbances or physical injury(1). Rhythmic movements (RMs) encompasse a large spectrum of manifestations: body rocking, body rolling, head rolling or head banging. The prevalence of RMs is thought to be high in infants and to decrease with age (1-4), but persistence in adulthood is not uncommon with a male predominance (1, 5-9). In contrast, RMD is less frequent, probably around 1-3% in children, often considered as a benign phenomenon; however severe forms exist also in adults with risk of injuries (9-15).

RMD remains a poorly studied sleep disorder, and little is known regarding sleep quality in those patients, with only few case series reporting associated disturbed nighttime sleep (5, 7, 14-23). RMs frequently occur at bed time leading to sleep onset insomnia, but also during sleep that may impair sleep continuity and quality (6, 9, 14, 16, 24-27). RMD may also be associated with other sleep disorders, disorders of arousal (DOA), restless legs syndrome (RLS), periodic limb movement (PLM) or sleep-related breathing disorders (25, 28-32). All these conditions shared an excessive nighttime sleep fragmentation. Whether disturbed nighttime sleep quality in patients with RMD is associated with RM severity or with frequent comorbid sleep disorders remains unknown.

To our knowledge, no case-control studies assessing both clinic and polysomnographic characteristics of patients with RMD were reported. We thus conducted a study aiming to 1) provide a clinical and polysomnographic evaluation of children and adult patients with RMD, 2) compare sleep parameters assessed by polysomnography in patients with RMD and controls, and 3) to study the relationships between RMs characteristics and polysomnographic parameters of disturbed nighttime sleep in patients with RMD.

METHODS

Population

From 2006 to 2019, 50 patients (35 males, median age 14.3, range 3.4 – 60.7) were diagnosed with RMD in our sleep clinic. The population included 27 children (aged ≤16 years, 24 males) and 23 adults (>16 years old, 11 males). The diagnosis of RMD was made according to the Third Revision of the International Classification of Sleep Disorders (1). It required 1) a clinical history of repetitive, stereotyped and rhythmic movements involving large muscle groups; 2) predominately related to sleep; 3) resulting in a significant complaint such as interference with normal sleep,

impairment in daytime functioning or injuries; and 4) being not better explained by epilepsy or another movement disorder.

We also recruited 75 community-dwelling healthy participants (53 males, median age 15.3 years, range 3.9 – 62.4, including 33 > 16 y.o.), without past or present history of RM or other sleep disorder, through advertisements and local association networks from the general population.
All participants or their parents signed informed consent, and this study was conducted in accordance with the Declaration of Helsinki and French Good Clinical Practices.

Clinical evaluation

RM phenotype (body rocking, body rolling, head rolling or head banging), age at onset, and frequency of RM episodes were systematically recorded by a clinical interview. Different domains of impairment potentially associated with RM were assessed: RM-related injuries; sleep disruption; fatigue, excessive daytime sleepiness and interferences with parent/ bedpartner's sleep. Information on positive family history of RM (first and second degrees) was detailed for 37 patients. Current comorbid neurodevelopmental (i.e. attention deficit hyperactivity disorder - ADHD, autism spectrum disorder, intellectual and learning disabilities), affective (i.e. mood and anxiety disorders), and sleep disorders including RLS and DOA were also reported.

Polysomnographic assessment

All patients underwent one-night PSG recording in the sleep laboratory. This included electroencephalogram (EEG; leads at C3/A2, Fp1/T1, T1/O1, O1/C3, C4/A1, Fp2/T2, T2/O2, O2/C4), electro-oculogram, chin electromyogram (EMG), and electrocardiogram. Respiration was monitored with a nasal canula/pressure transducer system, mouth thermistor, chest and abdominal bands, and pulse oximeter. Leg movements were assessed with surface EMG electrodes placed on the right and left anterior tibialis muscles. Sleep stages, microarousals and respiratory events were manually scored according to standard criteria(33). Sleep-stage scoring during RMs was unreliable because of pervasive motor artifacts in all channels **(Figure 1)**. The epochs occupied by more than 15 seconds of RMs were scored as "movement time" (34). The remaining epochs containing RM sequences were scored according to the underlying sleep stage, and RMs were scored as microarousal.
Nighttime sleep was assessed by total sleep time (TST), sleep efficiency (SE), sleep latency (SOL), wake after sleep onset (WASO) duration and microarousal index. The epochs scored as "movement time" were not used for the calculation of SE and WASO. Moderate sleep apnea syndrome (SAS)

was defined as having an AHI >15/h in adults and >1/h in children (>30/h and >5/h for severe SAS, respectively). Periodic leg movements (PLMs) were scored following the criteria set by the International Restless Legs Syndrome Study Group (35). None of the participants were treated by a psychotropic medication during the two weeks prior to the PSG recording.

Rhythmic Movements scoring

Twenty PSGs synchronized with video recording were re-analyzed to identify RMs by a first rater (C.L.). A RM sequence was defined by a cluster of at least four body and/or head movements, with a frequency of 0.5 to 2Hz.

RMs were systematically associated with artifacts on EMG, EEG, respiratory or EKG leads **(Figure 1)**. We estimated the diagnostic value of those artifacts identification in re-assessing 11 representative PSGs involving body and/or head RMs (ranging from 21 to 108 sequences per PSG) by a second rater (R.L.) blinded of the video recording. Among the 539 RM sequences identified by the video-PSG, 518 (96.1%) were correctly identified by the PSG only (second rater). Only 19 (3.4%) artifacts scored as RMs on the PSG were not confirmed on the video recording. Overall, the agreement between the two scoring methods (video-PSG and PSG alone) was 92.8%. However, the PSG scoring alone could not identify the muscle group involved in the RM sequence (e.g. body or head movements), nor their subtype (e.g. rolling, banging, rocking movements).

We further applied this scoring method to analyze the 30 remaining PSG recordings without the full video recording saved. For these patients, details on the muscle group involved in RM was retrieved from the sleep technician's PSG report and from selected saved video captures of RM. Various RM-related parameters were detailed: RM subtype (i.e. body or head RM), total number of sequences, duration, total time spent in RM, occurrence from sleep stages and wakefulness, and distribution (i.e. before sleep onset, in the first, second and third parts of the night). We calculated the RM index defined by the number of RM sequences/h of time in bed, as well as the RM duration defined as the percentage of time in bed occupied by RMs, according to a recent work (36).

Statistical analyses

Clinical and RM characteristics were described using median and range for continuous variables, and percentages for categorical variables. Independent t-tests were used to assess case-control differences for continuous PSG variables and chi-squares for dichotomous variables. Comparisons between cases and controls were made according to the two age groups (≤16 and >16 y.o) and the full population. Partial Pearson's correlations were used to assess association between RM characteristics and other PSG parameters while controlling for age and comorbid sleep disorders.

All statistical analyses were made using SPSS version 24 for Windows. Statistical significance was set at $p<0.05$.

RESULTS

Clinical characteristics of RMD patients

Thirty-six (72%) patients reported body RMs (29 with body rolling, 6 with body rocking and one with unspecified body RM), 26 (52%) head RMs (11 with head rolling, 10 with head banging, four with both phenotypes, and one with unspecified head RM), and 12 patients (24%) both head and body RMs. Thirty-two patients reported a daily occurrence of RM. A complaint of disturbed nighttime sleep was found in 82%, 81% in children, 83% in adults, altered daytime functioning in 80%, and parent's or siblings's sleep disruption in 72%. Fifteen patients had a past history of injurious RMs. All patients reported an age at onset before 12 year-old, with 32 (67%) before 3 year-old, except for one patient with an adult onset. The clinical RM phenotype was comparable between children and adults, except for a higher sleep parent disruption and a younger age at onset in children **(Table 1)**.

At time of study, 23 patients (46%, 17 children) had a comorbid neurodevelopmental disorder, 13 (26%) with ADHD, two (4%) with autism spectrum disorder, five (10%) with intellectual disabilities, and 15 (30%) with specific learning disorders. Fourteen patients (28%, five children) had an affective disorder, 13 (26%) with anxiety and six (12%) with depressive disorder. Sixteen patients (32%, four children) had RLS at least twice a week, with a median age at RLS onset at 17 years (i.e. after RMD onset in 13 patients, 81.2%). Disorder of arousal (i.e. sleepwalking or sleep terrors) was found in 13 patients (26%, nine children). Finally, only four (8%, three children and one adult) patients with RMD had no comorbid neurodevelopmental, affective nor sleep disorder **(Figure 2)**.

Sixteen patients among 37 (43%) reported a positive family history of RMD, 12 with 1st degree and four with 2nd degree relatives, including three RMD families with a at least 3 affected patients . No between sporadic and familial RM differences were found for demographic characteristics, or comorbid neurodevelopmental, affective and other sleep disorders.

Rhythmic movements during the polysomnography assessment

During the polysomnograohy, 41 patients (82%) had at least one RM sequence, 32 (78.0%) with more than 10 and nine (18%) more than 100 episodes. No RMs were seen in the controls. Altogether, 2709 RM sequences were analyzed, with a mean duration of 60.75 seconds per episode, ranging

from 3 seconds to 29.6 minutes. RM sequences often emerged from wakefulness (67.8% of sequences), and seven (14.0%) patients had only wakefulness-related episodes. The remaining RM sequences emerged from sleep: 33.9% in N1, 38.8% in N2, 2.6% in N3, and 24.6% in REM sleep. Two adult patients had RM sequences only in REM sleep, patients with neither comordid condition nor positive RMD family history. The episodes were regularly distributed throughout the night, 30% during the first part (including 12.3% before sleep onset), 39% during the 2nd part, and 31% during the last part of the night (including 3.1% of episodes after sleep offset). The mean RM index was 6.6 per hour of PSG recording, the mean total time spent in RM 43.8 minutes that corresponds to 8.8% of time in bed. We found no differences between adults and children for the RM PSG parameters **(Table 1)**.

Polysomnographic findings in patients and controls

Compared to adult controls, adult patients with RMD had reduced TST and SE, and increased WASO duration, SOL and microarousal index **(Table 2)**. PLMS index was higher in patients compared to controls (PLMS index >5/h :40% vs 11%, χ^2=14.350, p<0.001 ; PLMS index >15/h: 22% vs 1% , χ^2=14.765, p<0.001). Patients with moderate OSAS were comparable between the groups ; however patients were more likely to have severe OSAS (22% vs 2% χ^2=7.098, p=0.008).

Comparing the children groups, patients with RMD had a lower SE, an increased WASO duration, and higher AHI, oxygen desaturation, PLMS and microarousal indices **(Table 2)**. Children with RMD also had higher PLM (>5/h, χ2=16.833, p<0.001; >15/h , χ2=8.385, p=0.004) and AHI indexes (> 5/h, χ^2=4.548, p=0.033).

Considering all patients (children and adults), similar differences were observed for TST, SE, WASO, and micro-arousal index between patients and controls. An increased percentage of N1 sleep was also found in patients compared to controls together with higher AHI, PLMS and oxygen desaturation indexes.

Disturbed nighttime sleep and its relations with RM phenotype

Considering all patients, RMs index and duration correlated to TST, SE and WASO (i.e. parameters of sleep continuity), after controlling for age, AHI, PLMS and RLS (**Table 3**).The number of RM sequences in N2 and N3 sleep correlated with the microarousal index, and the sequences occurring before sleep onset with a longer SOL.

DISCUSSION

Here, we performed a case-control study and showed that objective disturbed nighttime sleep was more frequent in children and adult patients with RMD. RMD is highly often associated with neurodevelopmental, affective and sleep disorders; however RM characteristics were independently associated with markers of sleep fragmentation .

Despite a well-known high prevalence of RMs in the general population in children, (3, 4) we diagnosed only 50 patients with RMD over 13 years in a single reference national sleep center with half of our population being adults. This observation may confirm that RMs in children is often a benign condition that rarely requires an evaluation in a specialized sleep disorder clinic in contrast to a most severe and debilitating form of RMs, named RMD, a less frequent and understudied condition. Symptoms of RMD were often comparable between children and adults suggesting a similar underlying pathology. While RMs in children tend to resolve spontaneously, without consequences, our sample of children and adults had persistent RMs with nighttime and or daytime impairment.(37) We observed a male RMD preponderance, but exclusively in children (89%) that may reflect a bias selection as most of epidemiological studies reported no association between RM/RMD and gender (4, 38).

The diagnosis of RMs should rely on visual analysis of video synchronized to PSG or home videosomnography (2, 36, 39, 40). Our procedure of assessing RMs in first video-PSG, and second in PSG only (blinded of the video) - confirmed an excellent concordance (92.8%) between both methods, with highly stereotyped motor artifacts on most of channels during RM sequences. None of the controls and 41 patients (82.0%) displayed RMs during the PSG assessment, suggesting that some patients suppressed such movements in the laboratory setting,(41) or that RMs may have a night-to-night variability as described for PLMs (42). Most of RMs emerged from wakefulness, with a majority of sequences being observed after sleep onset. The repartition of the RM sequences was spread out across the night, not predominantly in the first part of the night that further challenge the historical view of RMD as a pre-dormitum sleep-wake transition phenomenon (19). In NREM sleep, RMs often occurred during light N1 and N2 sleep, with rare episodes emerging from N3 sleep. The decrease in RMs frequency with NREM stages proportionally to sleep profundity has recently been reported (41). In two patients, RMs occurred only in REM sleep episodes, without any comorbidities as being already reported (6, 9, 18, 19, 27). We found that both children and adults with RMD had an excessive sleep fragmentation, characterized by a reduced SE, and higher

micro-arousal index, in comparison to controls. Our results revealed that these differences were larger in the adult subsample, with almost 20% reduced SE and a twice-longer WASO duration, compared to adult controls. A shorter TST and an increased SOL were also found in this age group. These results were associated with the RM index and duration although RMs were not scored as wakefulness but as “movement time”.

RMD often occurred with comorbid affective, neurodevelopmental or sleep disorder. Comorbid neurodevelopmental disorders were found especially in children with RMs that confirms earlier studies(8, 25, 27, 41). In the general population, RMs and RMD decrease as function of age, as for neurodevelopmental disorders. These findings would suggest that RMs reflect a delayed maturation of motor control during sleep, with possible implication of the vestibular system and the central pattern generator network, as hypothesized (8, 43). We also found a high frequency of affective disorders, namely depression and anxiety disorders in both adults and children with RMD. These psychiatric conditions are known to interfere with sleep initiation and maintenance. In this context, RMD could be considered as a learned behavior to facilitate sleep onset and return to sleep following awakenings, by reproducing parental rocking, known to sooth infants and decrease arousal in experimental paradigms. Whether RMs directly disrupt sleep continuity or are soothing behaviors that partially prevent from more severe sleep fragmentation remains to be determined.`We found a high frequency of RLS, PLMS and sleep-disordered breathing, also underlined by previous studies, that may aggravate the disrupted nighttime sleep reported by patients with RMD (29, 32, 44, 45). We found that RM severity was associated with poorer SE, increased WASO duration, longer SOL, and increased microarousal index even controlling for RLS, PLMS and AHI. Altogether, these results suggest that RMD severity impacts PSG continuity and architecture parameters, independently of age and comorbid sleep disorders.

We found finally that only 8% of our patients with RMD were without any comorbidities, suggesting that primary or idiopathic RMD would be a rare condition. In contrast, we found a high frequency of a positive family history of RM (43%), without differences in phenotyping between sporadic and familial RMD. Familial occurrence of RM has been rarely reported in isolated cases (7, 9, 26, 46, 47), and three case series reported a lower frequency of 8.0% to 20.0% (8, 25, 41). The frequent familial occurrence we observed does not necessarily mean that RMD has a genetic origin, given that RM could be a learned behavior from the parents or siblings. Attarian, Ward (47) reported on a multigenerational family with comorbid RMD and insomnia and proposed that there might be

a common genetic predisposition for both disorders. However, genetic investigations have not been yet performed in RMD.

Some limitations need to be acknowledged. Our clinical population sample diagnosed in a tertiary sleep clinic was well-characterized, relatively small and probably biased and most severe than the one from the general population that prevent the generalization of our main results. Our study did not provide definitive explanations of mechanisms underlying associations between poor sleep quality and RMD severity. Finally, we did not use standardized psychometric tools to quantify the complaint of the disturbed nighttime sleep, daytime functionning and quality of life.

In conclusion, we confirmed that RMD is a rare and potentially disabling condition with objective disturbed nighttime sleep. The severity of RMD and the frequent associated sleep-related motor and respiratory disorders may contribute to the global altered sleep continuity. Combined therapeutic approaches that target both RMs and comorbid sleep disorders are mandatory to improve nighttime sleep, and potentially daytime functioning and quality of life in adults and children with RMD.

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Table 1. Clinical and polysomnographic RM phenotype in children and adults with RMD

	Total (n=50)	***Adults (n=23)***	**C*hildren (n=27)***	*p-value*
	Mean (SD) or %	Mean (SD) or %	Mean (SD) or %	
Clinical characteristics (patient report)				
Body RMs	72%	70%	74%	0.723
Head RMs	52%	52%	52%	0.982
Body and head RMs	24%	22%	26%	0.730
Familial history (yes)	43%	44% *	43% *	0.957
Daily occurrence of RM (yes)	74%	68% *	79% *	0.423
Age at onset (>3 years old)	33%	52% *	19%	**0.014**
History of RM-related injuries (yes)	30%	22%	37%	0.239
Sleep disruption (yes)	82%	83%	81%	0.918
Altered daytime functioning (yes)	80%	87%	74%	0.256
Family impairment (yes)	72%	48%	93%	**<0.001**
Comorbidities				
RLS	32%	52%	15%	**0.005**
DOA	26%	17%	33%	0.200
Neurodevelopmental disorder	46%	26%	63%	**0.009**
Affective disorder	26%	39%	19%	0.106
Polysomnographic RMs phenotype				
RM sequences (yes)	82%	83%	81%	0.918
RM duration (min)	43.82 (61.70)	54.65 (74.94)	34.59 (47.22)	0.256
RM duration Index (%)	8.79 (12.60)	11.58 (15.69)	6.40 (8.83)	0.169
RM sequences (TIB)	54.18 (84.09)	68.52 (94.57)	41.96 (73.64)	0.270
RM Index (/h)	6.61 (10.72)	8.90 (12.79)	4.65 (8.33)	0.164
RM sequences (wakefulness)	36.72 (63.15)	45.35 (70.77)	29.37 (56.18)	0.378
RM sequences (N1)	5.92 (12.63)	7.26 (14.72)	4.78 (10.69)	0.494
RM sequences (N2)	6.78 (14.02)	8.48 (15.55)	5.33 (12.69)	0.435
RM sequences (N3)	0.46 (1.55)	0.70 (2.10)	0.26 (0.86)	0.328
RM sequences (REM)	4.3 (13.50)	6.74 (17.73)	2.22 (8.24)	0.242
RM sequences before sleep onset	6.66 (13.36)	8.65 (16.48)	4.96 (10.05)	0.336
RM sequences first third	16.24 (25.06)	21.30 (27.48)	11.93 (22.42)	0.190
RM sequences second third	21.14 (41.23)	24.39 (39.11)	18.37 (43.49)	0.612
RM sequences last third	16.80 (29.51)	22.83 (38.09)	11.67 (18.84)	0.185

- ***Note.*** * data not reported for all patients (familial history: adults n=16, children n=21; daily occurrence: adults n=19, children n=24; late onset: adults n=21)
- Abbreviations:

Table 2. Comparison of polysomnographic characteristics in children and adults with RMD and controls

	Children					***Adults***				
	Controls (n=42)		RMD (n=27)			Controls (n=33)		RMD (n=23)		
	Mean	SD	Mean	SD	p-value	Mean	SD	Mean	SD	p-value
TST (min)	454.31	66.58	436.78	76.64	0.318	418.51	45.11	324.13	85.78	**<0.001**
Sleep efficiency (%)	86.27	8.52	80.92	11.97	**0.033**	85.30	6.85	67.62	16.44	**<0.001**
Sleep onset latency (min)	29.88	68.56	27.22	21.60	0.846	17.18	11.14	38.30	41.59	**0.026**
WASO (min)	46.69	34.05	75.00	66.70	**0.049**	44.54	26.91	114.83	68.85	**<0.001**
N1 (%)	3.89	2.57	5.17	3.66	0.093	4.19	2.50	8.31	9.50	0.053
N2 (%)	45.54	8.33	45.44	10.05	0.965	57.34	5.70	53.53	11.32	0.103
N3 (%)	31.08	8.94	28.55	8.17	0.240	19.20	5.81	18.17	7.00	0.552
REM (%)	19.47	5.08	20.82	6.06	0.322	19.08	4.46	19.98	6.36	0.537
Micro-arousal index (/h)	6.31	3.38	12.87	6.35	**<0.001**	11.90	4.91	19.70	9.93	**0.002**
AHI (/h)	1.34	1.56	3.75	7.24	**0.041**	3.18	4.33	7.40	12.58	0.079
Desaturation index (/h)	1.35	1.99	3.77	4.90	**0.020**	2.82	4.26	9.20	15.20	0.061
PLMS index (/h)	1.02	1.36	6.52	8.02	**0.002**	2.34	3.66	12.17	18.64	**0.020**

Abbreviations:

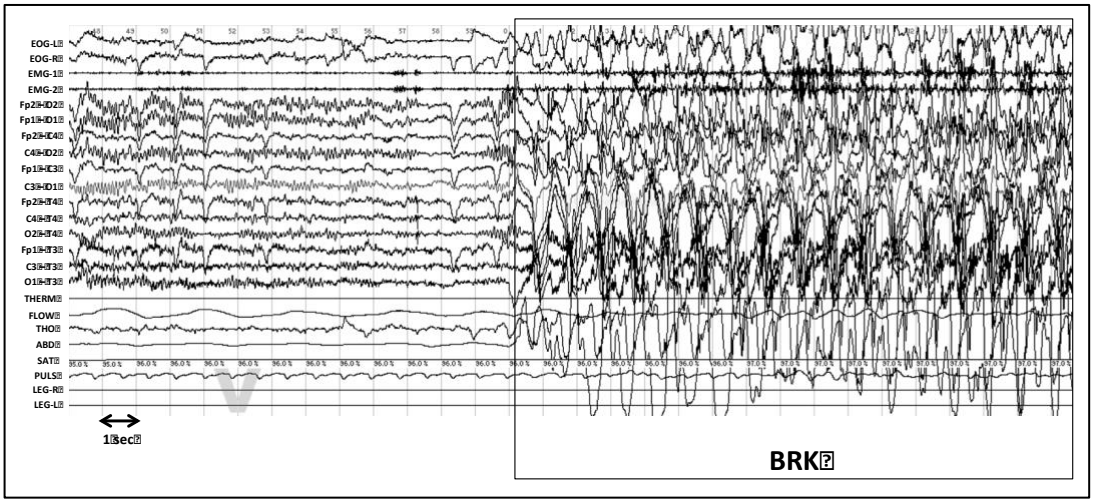
Table 3. Correlations between general sleep architecture parameters and RM phenotype in adults and children with RMD

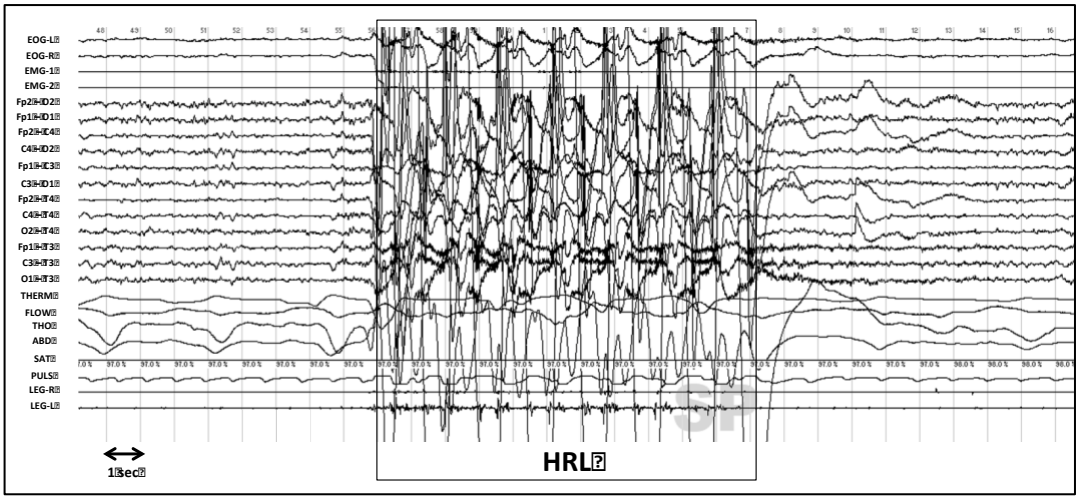
	Total Sleep Time		***Sleep efficiency***		***Sleep onset latency***		***Wake after sleep onset***		***Microarousal index***	
	Pearson r	p-value	Pearson r	p-value	Pearson r	p-value	Pearson r	p-value	Pearson r	p-value
RM Index (/h)	-0.400	**0.006**	-0.439	**0.002**	0.053	0.726	0.400	**0.006**	0.065	0.667
RM duration Index (%)	-0.332	**0.024**	-0.412	**0.004**	0.039	0.799	0.416	**0.004**	0.152	0.314
RM in wake	-0.489	**0.001**	-0.540	**<0.001**	0.104	0.492	0.476	**0.001**	-0.028	0.855
RM in NREM1	-0.226	0.132	-0.263	0.078	0.019	0.899	0.226	0.130	0.043	0.779
RM in NREM2	-0.099	0.513	-0.167	0.267	-0.044	0.770	0.258	0.083	0.385	**0.008**
RM in NREM3	0.257	0.085	0.210	0.161	-0.084	0.580	-0.096	0.524	0.387	**0.008**
RM in REM	0.145	0.337	0.167	0.266	-0.163	0.280	-0.052	0.732	0.255	0.087
RM before sleep onset	-0.256	0.085	-0.231	0.123	0.365	**0.013**	0.019	0.898	-0.137	0.365
Note. Controlling for age, AHI, PLMS, and RLS										

Abbreviations:

Figure 1. Polysomnographic aspects of RMs sequences.

Bodyrocking episode emerging from wakefulness (Fig. 1.A), Headrolling episode emerging from N1 sleep (Fig 1.B) and 4 successive bodyrolling episodes emerging from REM sleep (Fig 1.C).





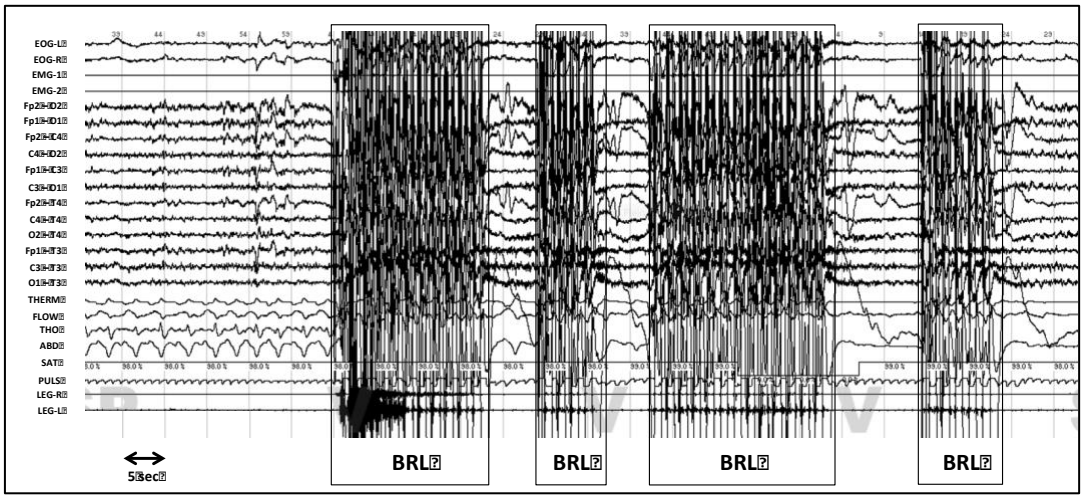


Figure 2. Comorbidities in patients with RMD