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CASE REPORTS

Sleep-Related Rhythmic Movements and Sleep Terrors: A Possible Common Neurophysiological Background in a Preschool Boy

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We report the case of a 3-year-old boy with a history of frequent and injurious sleep-related rhythmic movements and sleep terrors. We documented six episodes of body rocking and head banging via video polysomnography. No epileptic seizures were observed. In addition to the association between a sleep movement disorder and a disorder of arousal, our case shows that sleep-related rhythmic movements can arise not only during relaxed wakefulness or during a stable sleep stage, but also during a less clearly defined sleep stage during which it is difficult to further subtype non-rapid eye movement sleep. On the contrary, the portion of sleep without rhythmic movement episodes were clearly depicted with their physiological features. These findings might be of relevance for understanding the pathophysiology of both sleep-related rhythmic movements and sleep terrors and emphasize the importance to assess sleep using polysomnography, especially when episodes are frequent and injurious. The neurophysiological information obtained from this assessment might be helpful and guide an eventual treatment option.

Keywords: polysomnography, sleep-related rhythmic movements, sleep terrors

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INTRODUCTION

Sleep-related rhythmic movement disorders (SRMDs) consist of stereotyped and rhythmic movements involving large muscle groups that result in significant sleep disturbance or daytime symptoms.[1] According to polysomnography (PSG), they are characterized by a minimum cluster of four movements occurring at a frequency of 0.5 to 2 Hz, for periods of variable duration.[2] Although SRMDs typically occur in normal infants and children, it can persist into adulthood and has an increased prevalence in children with autism spectrum or developmental delay.[3,4] Sleep terrors are disorders of arousal that occur during non-rapid eye movement (NREM) sleep characterized by abrupt awakening, usually with a loud scream and signs of autonomic activation (tachycardia, tachypnea, mydriasis, and diaphoresis).[2] Disorders of arousal are usually exacerbated by the presence of a comorbid sleep disorder.

PSG is the gold standard tool to diagnose sleep disorders in children. It is indicated when sleep-disordered breathing is suspected but also in cases of atypical or injurious parasomnias.[5]

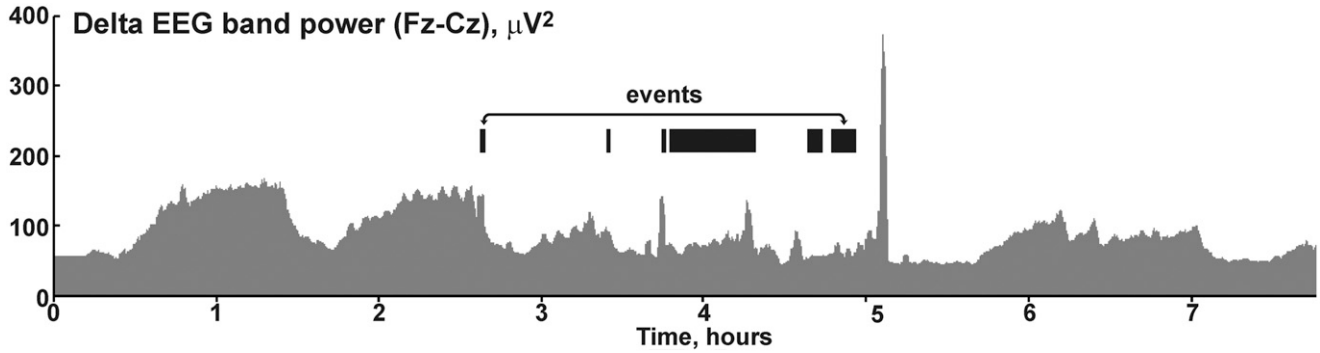
Few full-night video PSG recordings, mostly in adults, have documented rhythmic movements (RMs) during wakefulness or in stable sleep.[4]

We report the case of a 3-year-old boy with frequent episodes of RMs during sleep as well as sleep terrors. In addition to the interest connected with the association between a sleep movement disorder and a disorder of arousal, a nocturnal video PSG disclosed six episodes arising also during sleep with mixed NREM electroencephalography (EEG) features.

REPORT OF CASE

A 3-year-old boy, born from an uneventful full-term pregnancy, with typical neurodevelopment, was referred to us for evaluation of frequent sleep-related RMs. The parents stated that since he was 9 months old, the patient has had recurrent night terrors during which the child suddenly sat in bed, seemingly agitated, with a fearful facial expression, crying and sweating profusely, and not aware of his surrounding environment. Night terrors occurred weekly, several times on the night when they occurred. The parents were not too concerned about these episodes; however, the parents expressed concern about the nightly multiple episodes of body rocking and head banging that were also present since the same age as the onset of the night terrors. These episodes, which usually occurred several times during the night and every night, 1 to 2 hours after sleep onset and lasting up to 20 to 30 minutes, had been associated with bruises and injuries on the patient's head due to the aggressive head banging sometimes against the side of the bed. His bedtime routine was consistent with a bedtime of approximately 9:00 PM and a wakeup time of approximately 7:00 AM during the week and 10:00 AM on the weekend. The patient was sleepy during the day and had usually a "planned" 40- to 60-minute nap in the afternoon while at preschool. During this nap, the patient

Figure 1—Distribution of RM episodes during the night.



The bars represent RM duration. RM episodes appear in the middle part of the night, after a long period of deep sleep. EEG = electroencephalography, RM = rhythmic movements.

exhibited rhythmic movements. There was no snoring, no recollection of the events, and no suspicion of restless legs syndrome or nocturnal leg kicks. The child did not take any medications and did not have any prior medical or surgical history. Physical examination revealed normal vital signs and growth. The child was interactive without positive physical findings. Family history was negative for sleep disorders. He had a 2-year-old sibling without any sleep concerns.

Laboratory Findings

The patient underwent video PSG with an extensive EEG montage (Fp2-F4, F4-C4, C4-P4, P4-O2, Fp2-F8, F8-T4, Fz-Cz, Fp1-F3, F3-C3, C3-P3, P3-O1, Fp1-F7, F7-T3), electrooculography, chin, nuchal, sternocleidomastoid, deltoid and paraspinal muscles surface electromyogram, oronasal airflow, chest and abdomen respiratory effort, and electrocardiography. Awake and sleep EEG findings were normal, and no elevated periodic leg movements or sleep-disordered breathing was found. The patient slept for 489 minutes and sleep was visually scored. Sleep latency was normal (3 minutes), sleep efficiency was reduced (79%), and wake time after sleep onset increased (80 minutes). Rapid eye movement sleep was normally represented (22%) whereas light NREM sleep percentage was reduced (stage N1 sleep 5%; stage N2 sleep 13%); stage N3 sleep was 57.5% of total sleep time; finally, an unusual NREM-like sleep stage, in the form of a mixed state in which EEG features of relaxed wakefulness and NREM sleep appeared at the same time, occupied 2.5% of the total sleep time. In the middle part of the night, the child showed 6 episodes of body rocking and head banging, at a frequency of 1 to 1.5 Hz, lasting from 25 seconds to 31 minutes (**Figure 1**), with 2 episodes apparently arising from wakefulness and 4 from an NREM sleep stage with features that did not fit current scoring criteria.

Video Analysis

The video segment (**Video 1** in the supplemental material) shows the first RM episode recorded. Although the patient is sleeping on his left side, he suddenly moves his legs to lift up the trunk and then sits on the bed and begins rocking the body, gradually augmenting the movement intensity until banging his face on the mattress. The spectral analysis of the EEG recorded during the episode (**Figure 2**) shows increased slow-wave activity with respect to the wakefulness preceding sleep (but not at the levels obtained during stage N3 sleep) when the power of the frequency bands < 8 Hz was smaller and a small peak at 8 Hz was evident (alpha band). Both before and during the episode, there is a clear predominance of the delta and theta bands, making these periods different from the wakefulness preceding sleep.

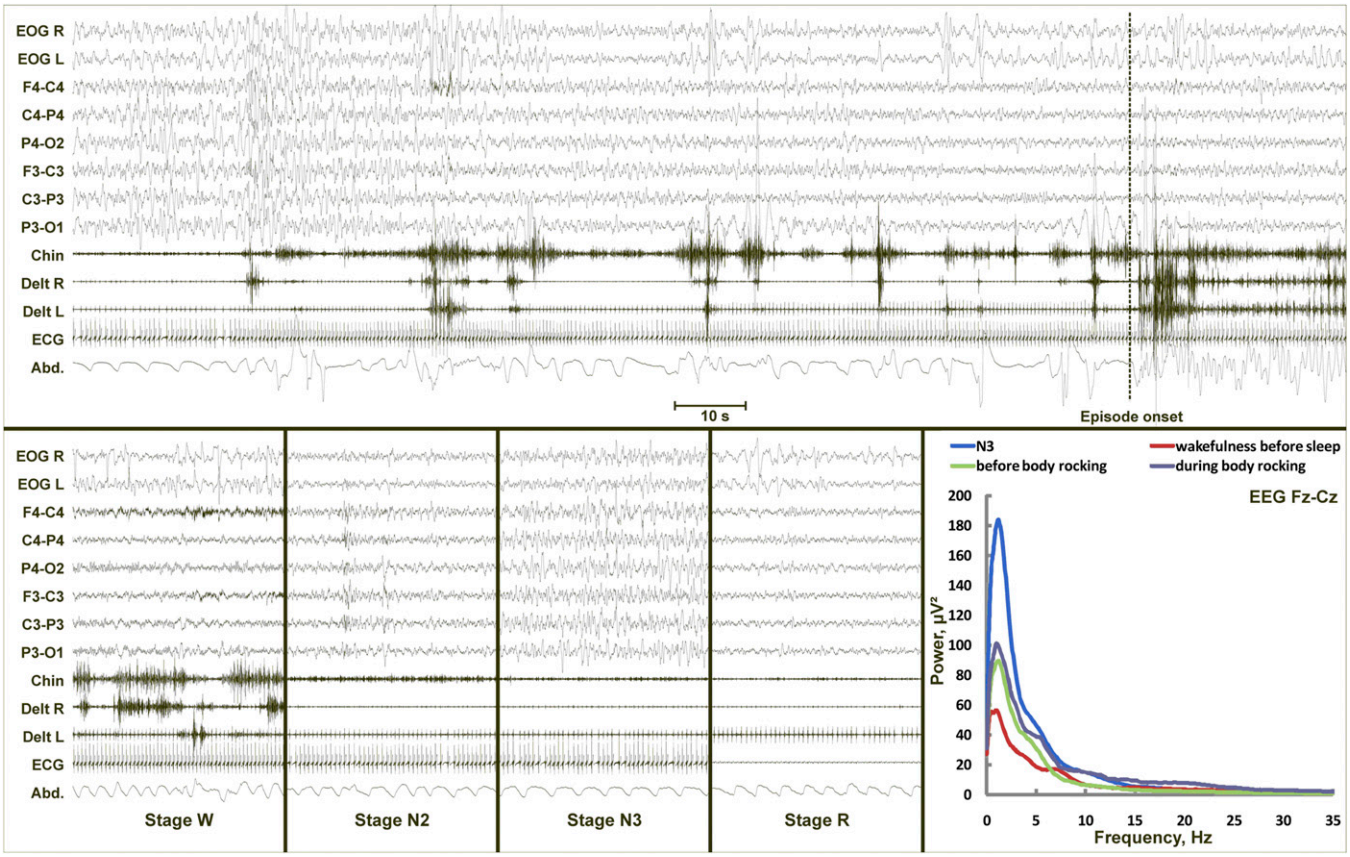
DISCUSSION

This case shows clinically persistent RM during sleep in the setting of coexisting disorder of arousal. The frequency, injurious nature, and nighttime distribution were indications for further evaluation with nocturnal video PSG. Furthermore, an extended parasomnia montage was included to characterize the movements and an extended EEG montage was added to rule out nocturnal seizures.

According to PSG, this case illustrates that nocturnal episodes of SRMDs can arise not only during relaxed wakefulness or during a stable sleep stage,[3,6] but also during a not-well-characterized sleep stage. The appearance of RM episodes in a poorly defined sleep stage might be of relevance for understanding their pathophysiology. The specific causes and physiologic mechanisms underlying SRMDs remain uncertain; however, there are some hypotheses. Some authors emphasize the close relationship between RM episodes and arousals,[7] suggesting an involvement of the brainstem and the thalamocortical reticular system.[3] In this case a vestibular self-stimulation or wake-sleep transition disorder can be a putative mechanism. Alternatively, immaturity in motor pathways could be a factor. In our case, poorly defined NREM sleep might facilitate the activation of motor subcortical central pattern generators, located in the brainstem and spinal cord.[8,9] In this case the coexistence of both night terrors and SRMDs could unify both hypotheses.

Furthermore, there is evidence that suggests that disorders of arousal consist of an NREM sleep-wake dissociated state,

Figure 2—PSG results.



Top panel: PSG results from one RM episode arising from a state with mixed non-rapid eye movement features; the vertical dashed line indicates the RM onset. Bottom left panel: Examples of PSG results obtained during normal sleep, showing the typical potentials of wakefulness and sleep stages that are clearly recognizable. Bottom right panel: Spectral analysis of the electroencephalography (Fz-Cz) during wakefulness before sleep, during the episode and just after the end of the episode, and during normal stage N3 sleep. Abd = abdomen, Delt = deltoid muscle, ECG = electrocardiography, EEG = electroencephalography, PSG = polysomnography, RM = rhythmic movements.

an admixture of wakefulness and NREM sleep: the patient produces complex behaviors that are characteristic of wakefulness (such as eye opening, retained muscle tone, and ability to move and interact with the environment) but cognitive function fitting with sleep (inappropriate actions and lack of self-related awareness, insight, and recall). Several data support this hypothesis. Scalp EEG shows some evidence of state dissociation, with posterior dominant alpha rhythm (suggestive of resting wakefulness) but anterior theta activity and sometimes vertex waves or spindles (consistent with light NREM sleep). Stereo-EEG studies in epileptic patients with co-occurrence of disorders of arousal have recently confirmed these data, identifying a dysfunctional coexistence of local cortical arousal and local cortical sleep.[10] This case is fascinating as it joins the body of literature showing that sleep may not only be characterized as NREM and REM but there could be overlapping stages, dissociated stages, and other findings still unknown and worthy of further research.

At 1-month follow-up, the patient presented with irritability, daytime somnolence, and RMs also during daytime naps. Because of the significant daytime symptoms and concerns for repetitive injuries, pharmacological treatment was offered. Treatment with clonazepam 0.3 mg at bedtime was initiated with a reduction of nocturnal episodes and disappearance of daytime symptoms at a subsequent 3-month follow-up. However, because of the subsequent increased daytime sleepiness noticed by the mother, clonazepam was substituted with niaprazine 12 mg at bedtime, a selective serotonin (5-HT2A) and α1-adrenergic receptor antagonist commonly used for pediatric insomnia in some European countries, with the aim to obtain a more stable and profound NREM sleep (which was followed by a complete disappearance of nocturnal and diurnal episodes), without residual sleepiness. Both clonazepam and niaprazine at low dosages induced clinical improvement; this might further reinforce the idea that a more stable and profound, possibly better defined, NREM sleep might underlie the efficacy of the treatment.[7]

This case emphasizes the importance to assess sleep using PSG in sleep-related RMs, especially when they are frequent and injurious. The neurophysiological information obtained from this assessment might be helpful and guide the therapeutic approach to stabilize NREM sleep.

ABBREVIATIONS

EEG, electroencephalography
NREM, non-rapid eye movement
RM, rhythmic movement
SRMD, sleep-related rhythmic movement disorder

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DISCLOSURE STATEMENT

All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work. The authors report no conflicts of interest.