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CASE REPORTS

# Sleep-Related Rhythmic Movement Disorder and Obstructive Sleep Apnea in Five Adult Patients

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Sleep-related rhythmic movements (SRRMs) are typical in infancy and childhood, where they usually occur at the wake-to-sleep transition. However, they have rarely been observed in adults, where they can be idiopathic or associated with other sleep disorders including sleep apnea. We report a case series of 5 adults with sleep-related rhythmic movement disorder, 4 of whom had a previous history of SRRMs in childhood. SRRMs mostly occurred in consolidated sleep, in association with pathological respiratory events, predominantly longer ones, especially during stage R sleep, and recovered in 1 patient with continuous positive airway pressure therapy. We hypothesize that sleep apneas may act as a trigger of rhythmic motor events through a respiratory-related arousal mechanism in genetically predisposed subjects.

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## INTRODUCTION

Sleep-related rhythmic movement disorder (SRRMD) is characterized by repetitive, stereotyped, and rhythmic motor behaviors occurring predominantly during drowsiness or stage N1 sleep, involving large muscle groups, with possible significant consequences such as sleep disruption and injuries. Based on the anatomical district and the type of rhythmic movement events (RMEs), SRRMD is classified in body or head rocking, banging, rolling, or combinations thereof.[1] SRRMD is common in infants and children, and usually disappears spontaneously before adolescence. Only severe cases require a first-line treatment with benzodiazepines, such as clonazepam.[1] In childhood, SRRMD is usually idiopathic; however, other sleep disorders can trigger or exacerbate motor events. Rarely, adult cases have been described; herein the relationship with a previous SRRMD in infancy or with other sleep disturbances is unclear.[2–7] We report a detailed video-polysomnographic documentation of 5 adults with SRRMD.

## REPORT OF CASES

Five adult patients, referred either to the Sleep Center of Lugano or to the Center for Investigation and Research in Sleep of Lausanne (Switzerland), received a diagnosis of SRRMD according to the International Classification of Sleep Disorders, Third Edition criteria. Nocturnal video-polysomnography (VPSG) studies were carried out as previously described.[8] Polysomnographic results for all patients are available in **Table 1**, as well as detailed data of RMEs and the associated respiratory events. Results are presented as mean ± standard deviation (SD).

A two-tailed Student *t* test was used to compare the duration of breathing events associated with SRRMD to the duration of those unassociated with SRRMD.

### Case 1

A 63-year-old woman, admitted to our stroke unit for an ischemic lesion of the left paramedian pons, was screened for sleep apnea in the context of the SAS-CARE study.[9] VPSG performed 3 days after the stroke showed obstructive sleep apnea (OSA), periodic limb movements, and a significant sleep fragmentation. A total of 20 rhythmic rolling/banging episodes were recorded in this patient, 6 of which involved the whole body, 6 the pelvis, 4 the head, and 4 both the head and the trunk. All RMEs were preceded by a pathological breathing event. Breathing events were followed by RMEs in 5.5% of the cases, and their duration was significantly longer in comparison to those unassociated with motor events (mean ± SD duration 40.58 ± 17.64 versus 29.15 ± 15.87 seconds; $P = .0141$). CPAP treatment was refused by the patient.

Three months later, after a complete recovery from the stroke, VPSG was repeated. Nineteen episodes of body rolling and 7 of head rolling, for a total of 26 events, were recorded. All RMEs occurred at the end of an apnea or hypopnea. Apneas/hypopneas were associated with motor events in 10.9% of the cases and they were significantly longer than those not associated with motor events (mean ± SD duration 60.77 ± 33.81 versus 33.16 ± 22.63 seconds; $P = .0004$) (**Figure 1**; **Video 1** in the supplemental material). The patient and her relatives were already aware of the SRRMD, which had been observed since

**Table 1**—Polysomnographic findings, rhythmic movement events, and their association with respiratory events.

| | Case 1 VPSG 1 | Case 1 VPSG 2 | Case 2 | Case 3 | Case 4 | Case 5 pre-CPAP | Case 5 with CPAP |
|---|---|---|---|---|---|---|---|
| TST (min) | 374.0 | 430.5 | – | 355.0 | 454.6 | 158.5 | 228.7 |
| WASO (min) | 228.0 | 125.9 | – | 88.1 | 26.0 | 54.7 | 22.5 |
| SE (%) | 54.6 | 75.6 | – | 70.9 | 93.3 | 70.0 | 90.2 |
| Sleep latency (min) | 83.4 | 13.3 | – | 57.6 | 6.5 | 12.9 | 2.4* |
| REM latency (min) | 162.0 | 77.0 | – | 56.0 | 156.0 | * | 3.0* |
| Stage N1 sleep (% of TST) | 19.4 | 8.6 | – | 13.8 | 10.9 | 6.3* | 2.0* |
| Stage N2 sleep (% of TST) | 39.7 | 26.5 | – | 40.0 | 54.9 | 47.4* | 28.7* |
| Stage N3 sleep (% of TST) | 21.8 | 45.8 | – | 23.0 | 17.3 | 22.3 | 15.8* |
| Stage R sleep (% of TST) | 19.1 | 19.2 | – | 23.2 | 16.9 | * | 44.6* |
| AHI (events/h) | 47.0 | 29.3 | 48.5 | 4.6 | 5.3 | 54.3 | 8.5 |
| RDI (events/h) | 47.2 | 29.3 | 48.5 | 6.5 | 9.0 | 54.3 | 9.7 |
| RERA index (events/h) | 0.3 | 0.1 | – | 1.9 | 3.4 | 0.0 | 1.2 |
| ODI ≥ 3% (events/h) | 34.5 | 32.8 | 130.7 | 4.1 | 6.3 | 73.3 | 8.7 |
| Arousal index (events/h) | 39.3 | 13.5 | 0.0 | 21.8 | 31.0 | 59.4 | 5.6 |
| PLMS index (events/h) | 29.4 | 29.4 | 0.0 | 2.6 | 19.8 | 16.0 | 23.5 |
| Known comorbidities | Minor brainstem stroke | | Obesity | Epilepsy | None | OSA | |

| Case Number | | RMEs (n) | RMEs Duration Range (seconds) | Onset of RMEs by Sleep Stage, n (%) | | | RMEs related to REs (%) | REs related to RMEs (%) |
|---|---|---|---|---|---|---|---|---|
| | | | | W / N1 | N2 / N3 | R | | |
| 1 | *VPSG 1* | 20 | 3.7–18.8 | 5 (25.0) | 2 (10.0) | 13 (65.0) | 100.0 | 5.5 |
| | *VPSG 2* | 26 | 3.5–16.3 | 10 (38.4) | 6 (23.0) | 10 (38.4) | 100.0 | 10.9 |
| 2 | | 779 | 2.4–260.8 | – | – | – | 100.0 | 100.0 |
| 3 | | 36 | 3.5–3,402.9 | 26 (72.2) | 6 (16.6) | 4 (11.1) | 69.4 | 64.1 |
| 4 | | 262 | 1.4–174.1 | 57 (21.7) | 163 (62.2) | 42 (16.0) | 27.8 | 77.6 |
| 5 | Split-night | 144 | 4.0–363.0 | 31 (21.5) | 99 (68.7) | 14 (9.7) | 91.6 | 55.6 |
| | *Baseline* | 128 | 4.0–363.0 | 29 (22.6) | 99 (77.3) | 0 (0.0) | 97.0 | 98.0 |
| | *Under CPAP* | 16 | 5.0–32.0 | 2 (12.5) | 0 (0.0) | 14 (87.5) | 88.0 | 45.0 |

* = split-night. AHI = apnea-hypopnea index, CPAP = continuous positive airway pressure, ODI = oxygen desaturation index, OSA = obstructive sleep apnea, PLMS = periodic limb movements in sleep, RDI = respiratory disturbance index, REs = respiratory events, RERA = respiratory effort-related arousal, RMEs = rhythmic movement events, SE = sleep efficiency, TST = total sleep time, VPSG = video-polysomnography, WASO = wake after sleep onset.

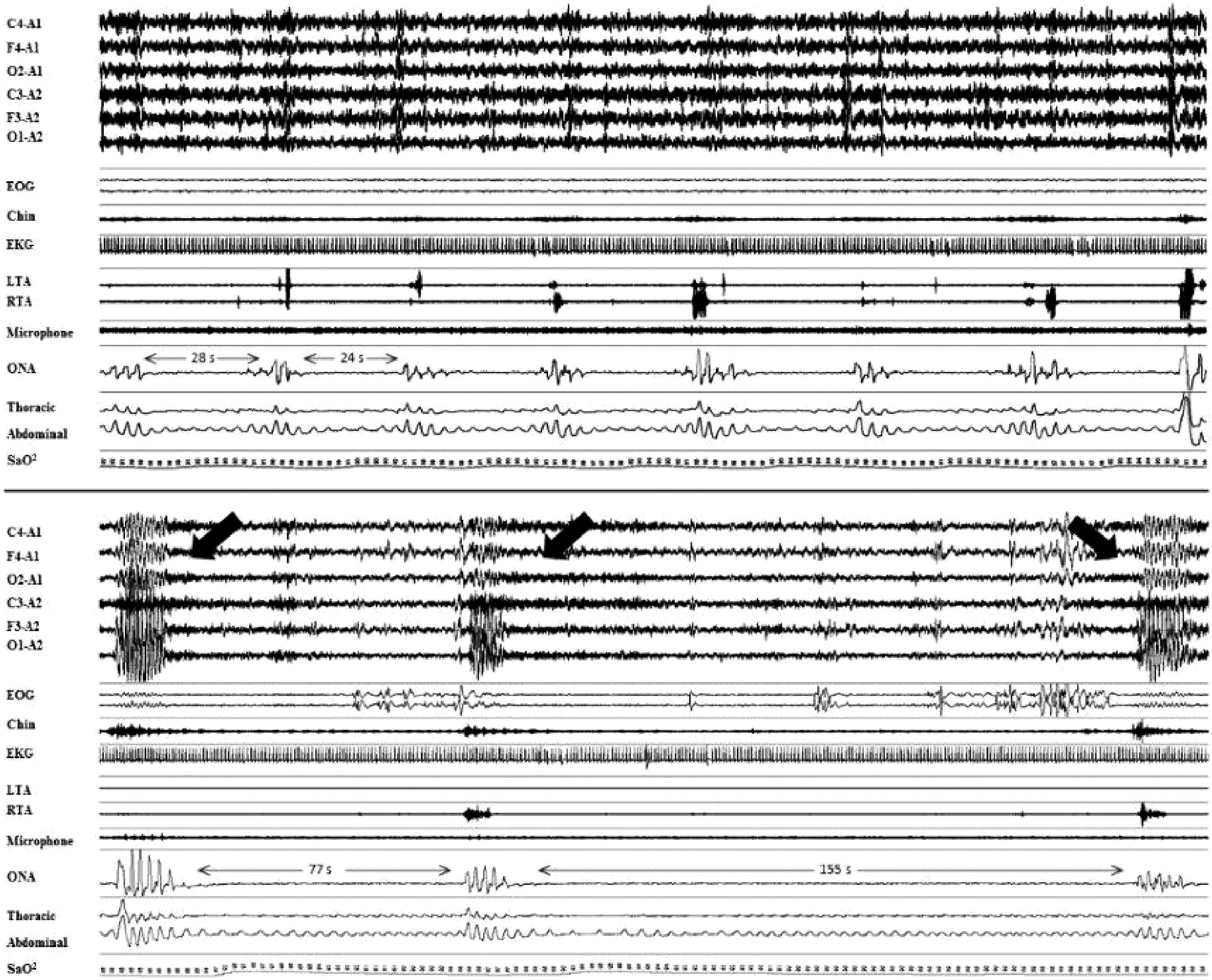


childhood, without any significant period of remission. She currently lives abroad and has recently contacted a local sleep center to seek alternative therapies for sleep-disordered breathing. Rhythmic movements still persist.

## Case 2

A 30-year-old obese male (body mass index = 45 kg/m$^2$) scheduled for bariatric surgery was investigated for suspected OSA in the presence of severe daytime sleepiness (Epworth Sleepiness Scale score 18/24), snoring, frequent nocturnal awakenings, and nocturnal restlessness. Because of the continuous movement artifacts, sleep stages could not be scored, but the VPSG recording revealed subcontinuous obstructive respiratory events occurring every 10 to 15 seconds and often lasting less than 10 seconds (apnea-hypopnea index [AHI] 48.5), resulting in a very severe oxygen desaturation index (≥ 3%) of 130.7. Arousals and awakenings following respiratory events were always associated with stereotyped body rolling movements. A total of 779 RMEs were recorded, widely ranging in duration and often occurring in clusters lasting up to 65 minutes (**Video 1**). In this patient, SRRMD was not known during childhood, it had started about 5 years before, in parallel with weight gain. CPAP was started before bariatric surgery and was continued for another 6 months afterward. After losing about 70 kg, the patient discontinued ventilotherapy because of a reported drastic reduction in OSA. Rhythmic movements have disappeared, based on telephone interview.

**Figure 1**




**Top:** Polysomnographic recordings (5 minutes) of a sequence of short obstructive apneas during non-rapid eye movement sleep, without rhythmic movement episodes. **Bottom:** Polysomnographic recordings (5 minutes) of a sequence of long obstructive apneas (duration specified in the picture) during REM sleep, associated with rhythmic movement episodes (see black arrows). Abdominal = abdominal movements, Chin = mylohyiod muscle electromyogram, EKG = electrocardiogram, EOG = electrooculograms, LTA = left tibialis anterior, ONA = oronasal airflow, RTA = right tibialis anterior muscle, $SaO_2$ = pulse oximetry, Thoracic = thoracic movements.

## Case 3

A 39-year-old man with a history of cryptogenic epilepsy had 3 generalized epileptic seizures in 2004, was treated with valproate, and underwent VPSG because of excessive daytime sleepiness. Although recorded with a reduced montage, and under antiepileptic drugs, no specific critical electroencephalographic activity was documented and no seizures were observed. A total of 36 head-rolling events were obtained. Longer sequences of head rolling occurred at the sleep-to-wake transition. Twenty-five RMEs (69.4%) were associated with apneic and hypopneic events, which were not significantly longer than those unrelated to rhythmic movements (mean ± SD duration 25.86 ± 15.35 versus 22.92 ± 10.98 seconds; $P$ = .4932). Although AHI was low, RMEs were preceded by respiratory effort-related arousals in 22% of the cases.

The patient had a history of whole-body rocking movements at sleep onset since childhood, which persisted into adulthood, without any changes in frequency or intensity over time. He did not receive any specific treatment for nocturnal respiratory anomalies, because these were minimal on VPSG recording.

## Case 4

A 26-year-old man with a history of sleep-onset rhythmic movements during childhood, which had somewhat diminished during adolescence, was evaluated because of a recent relapse of RMEs within the night, associated with sleep disruption due to numerous head banging events with injuries.

He also complained of snoring and nonrestorative sleep. We recorded 262 RMEs, characterized either by whole-body or pelvic rocking, leg or head rolling, or a combination thereof. Respiratory events associated with RMEs (77.6% of the cases) were longer than those unrelated to motor events (mean ± SD duration 24.96 ± 27.35 versus 15.10 ± 7.12 seconds; $P = .0067$). He did not receive any specific treatment for nocturnal respiratory anomalies, because these were minimal on VPSG recording.

### Case 5

A 50-year-old man was referred to our sleep center because of snoring, witnessed apneas, and excessive daytime somnolence. He, as well as his two brothers, was known for sleep-onset rhythmic movements during childhood, which had diminished during adolescence and had recently relapsed and shifted across the whole night.

The patient underwent a split-night VPSG, which consists of a first half of the night of baseline sleep study recording, followed by a CPAP titration study in the second half. During the entire night, we recorded a total of 144 RMEs, characterized either by leg or body rolling. Respiratory events associated with RMEs (55.6%) were longer than those unrelated to motor events (mean ± SD duration 31.22 ± 9.79 versus 27.48 ± 10.95 seconds; $P = .0068$).

During baseline recording, a total of 128 SRRMs were noted, with 98% of the respiratory events associated with RMEs, and 97% of RMEs occurring at the end of a respiratory event. During CPAP ventilation, RMEs reduced to 16 events, which were preceded by respiratory events in 45% of the cases. The patient still benefits from CPAP treatment regularly and reports a beneficial effect on sleep continuity and daytime somnolence. Rhythmic movements have also remitted, based on telephone follow-up.

## DISCUSSION

To the best of our knowledge, 39 cases of adult SRRMD have been reported in the literature.[6,7,10–12] Of these, 24 were assumed to be idiopathic, 7 associated with sleep apnea, 5 with REM sleep behavior disorder, and 3 with RLS or PLMS. Few cases were reported to improve with CPAP.

In this case series, all but 1 patient reported a previous history of SRRMD during their childhood, with events mostly located at the wake-to-sleep transition. In the latter four cases, RMEs persisted in an attenuated form into adulthood, assuming different characteristics; in particular, motor events tended to shift from the wake-to-sleep transition into deeper sleep, persisting across the entire night (**Figure S1** in the supplemental material). Considering the 488 RMEs recorded in those 4 subjects whose sleep stages could be identified, 56.5% of RMEs occurred during consolidated stage N2 and N3 sleep, and 17% in stage R sleep.

In total, 81.4% of RMEs were triggered by a preceding pathological breathing event. The association between RMEs and respiratory events reached almost 100%, when RMEs occurring at the wake-sleep transition were not taken into consideration. Even in case 3, in which RMEs were apparently poorly associated with breathing events, RMEs were indeed triggered by respiratory effort-related arousals.

The link between RMEs and breathing events was reinforced by their positive relationship with the event duration, which seemed to be an important factor implied in triggering the associated rhythmic movements in 4 patients. Eventually, RMEs and pathological breathing events retreated proportionally after CPAP ventilation in 1 case.

Because of the presence of SRRMD since childhood, we hypothesized that respiratory-related arousals drive the occurrence of motor events in genetically predisposed subjects, as previously suggested.[7] However, 1 of the 5 subjects had no history of SRRMD, which appeared in adulthood after weight gain. In another 2 subjects, attenuation and a subsequent re-exacerbation of SRRMD occurred across age.

## CONCLUSIONS

In conclusion, we recommend the investigation of sleep-related breathing patterns by means of a full VPSG in all adults with SRRMD, particularly in those with a new-onset disorder or a recent aggravation, as well as in those whose RMEs tend to occur also in consolidated sleep. SRRMD in adult patients may imply sleep-disordered breathing, possibly associated with longer respiratory events. This may speak against an unsafe treatment with benzodiazepines, which are usually the first-line therapy for idiopathic SRRMD, but which can aggravate sleep apnea. When sleep apnea is confirmed, treatment with CPAP might be beneficial also for RMEs.

## ABBREVIATIONS

AHI, apnea-hypopnea index
CPAP, continuous positive airway pressure
ODI, oxygen desaturation index
OSA, obstructive sleep apnea
PLM, periodic limb movements
RDI, respiratory disturbance index
RERA, respiratory-effort related arousal
RLS, restless legs syndrome
RME, rhythmic movement event
SD, standard deviation
SRRMD, sleep-related rhythmic movement disorder
VPSG, video-polysomnography

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