# Severe Obstructive Sleep Apnea/Hypopnea Mimicking REM Sleep Behavior Disorder

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**Objective:** To describe the clinical and video-polysomnographic (VPSG) features of a group of subjects with severe obstructive sleep apnea/hypopnea (OSAH) mimicking the symptoms of REM sleep behavior disorder (RBD).
**Design:** Evaluation of clinical and VPSG data.
**Setting:** University hospital sleep laboratory unit.
**Participants:** Sixteen patients that were identified during routine first evaluation visits. Patients' PSG measures were compared with those of 20 healthy controls and 16 subjects with idiopathic RBD of similar age and sex distribution and apnea/hypopnea index lower than 10.
**Interventions:** NA.
**Results:** Sixteen subjects were identified presenting with dream-enacting behaviors and unpleasant dreams suggesting the diagnosis of RBD, in addition to snoring and excessive daytime sleepiness. VPSG excluded RBD showing REM sleep with atonia and without increased phasic EMG activity, and was diagnostic of severe OSAH with a mean apnea-hypopnea index of 67.5 ± 18.7 (range, 41-105) demonstrating that the reported abnormal sleep behaviors occurred only during apnea-induced arousals. Continuous positive airway pressure therapy eliminated the abnormal behaviors, unpleasant dreams, snoring and daytime hypersomnolence.
**Conclusions:** Our study shows that severe OSAH may mimick the symptoms of RBD and that VPSG is mandatory to establish the diagnosis of RBD, and identify or exclude other causes of dream-enacting behaviors.
**Key Words:** REM sleep behavior disorder, obstructive sleep apnea-hypopnea, "pseudo-RBD", video-polysomnography.
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## INTRODUCTION

RAPID EYE MOVEMENT (REM) SLEEP BEHAVIOR DISORDER (RBD) IS A PARASOMNIA CHARACTERIZED BY DREAM-ENACTING BEHAVIORS RELATED TO LOSS OF the normal generalized skeletal muscle atonia during REM sleep[1,2]. RBD may be idiopathic or associated with neurodegenerative diseases such as Parkinson's disease[3], multiple system atrophy[4], dementia with Lewy bodies[5], Machado-Joseph disease[6] and parkinsonism with *parkin* gene mutations (Park2)[7]. Patients with RBD display abnormal motor and vocal behaviors such as punching, falling out of bed and shouting that are associated to frightening dreams[1,2]. However, these dream-enacting behaviors can also occur in other sleep disorders, a clinical situation termed as "pseudo-RBD"[1]. Recently, it has been shown that 65% of the subjects initially diagnosed as having idiopathic RBD eventually developed parkinsonism and/or dementia following a mean interval of 13 years[8]. Therefore, correct diagnosis of RBD is important because if "pseudo-RBD" is misdiagnosed as true RBD, then these subjects may be misinformed that they are at risk of developing parkinsonism and/or cognitive impairment, which could carry emotional consequences and medical malpractice liability. Moreover, correct diagnosis of RBD is also important because patients with RBD may injure themselves and their bed partners, and because this parasomnia can be effectively treated with clonazepam[1,2].

Here, we describe a group of 16 subjects who reported harmful or potentially harmful dream-enacting behaviors resembling RBD, but video-polysomnography (VPSG) failed to demonstrate REM sleep without atonia and excessive phasic muscle activity. Furthermore, VPSG demonstrated severe obstructive sleep apnea-hypopnea (OSAH) inducing abnormal motor and vocal behaviors.

## PATIENTS AND METHODS.

The 16 patients described here were identified during routine first evaluation visits at our sleep unit between July, 1998, and November, 2001. Subjects were referred by their primary care physicians with complaints of abnormal sleep behaviors and daytime hypersomnolence. Sleep history was obtained from the patient and the bed partner. Excessive daytime sleepiness was estimated by the Epworth Sleepiness Scale[9].

Patients underwent a diagnostic all-night polysomnography (PSG) with continuous audiovisual recording which included electroencephalogram (C3, C4, O1 and O2, referred to the contralateral ear), electro-oculograms, submental EMG, right and left anterior tibialis surface EMG, electrocardiogram, nasal and oral air flow, thoracic and abdominal movements, and oxyhemoglobin saturation. Sleep stages were scored according to standard criteria[10]. During REM sleep, we evaluated the percentage of submental REM sleep without atonia as previously described[11], but excluding EMG increases concurrent with respiratory-induced arousals and snoring signal artifacts. We also assessed the phasic EMG activity in the chin and in the legs (right and left anterior tibialis), as the percentage of 2-second mini-epochs containing phasic twitches (bursts greater than 0.1 sec-

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onds with an amplitude exceeding four times the background EMG activity[11]). In the legs, a 2-second mini-epoch was considered "phasic" when twitches appeared in any one of the two leg channels. Apnea was defined as cessation of airflow for at least 10 seconds. In obstructive apnea respiratory effort was maintained. Hypopnea was defined as an airflow amplitude decrease greater than 50% for at least 10 seconds with either an arousal or an oxygen desaturation greater than 3%[12]. The apnea-hypopnea index (AHI) was defined as the mean number of apneas and hypopneas per hour of sleep. Arousal was defined following the scoring rules of the American Sleep Disorders Association[13], and the arousal index as the number of arousals per hour of sleep.

When VPSG detected severe OSAH, patients were offered treatment with continuous positive airway pressure (CPAP). Those who accepted underwent a second PSG where CPAP was titrated to eliminate snoring, apneic events with arousals and oxyhemoglobin desaturations in all body positions and sleep stages. During follow-up visits we assessed the effect of CPAP on the presence of abnormal sleep behaviors, dream recall content and typical symptoms of OSAH syndrome such as snoring and excessive daytime sleepiness. The mean follow-up from start of CPAP therapy to last follow-up (May 2003) was 46.1 ± 11.7 months.

Patients' baseline PSG measures were compared with those of 20 healthy controls of similar age and sex distribution who had no sleep complaints, were free of psychotropic medications and had an AHI lower than 10. Patients' baseline PSG values were also compared with those of a group of 16 unmedicated subjects with idiopathic RBD of similar age and sex distribution and an AHI lower than 10 who were diagnosed in our sleep unit during the last five years. Comparisons between patients with controls and subjects with idiopathic RBD were assessed by the Mann-Whitney U-test.

## RESULTS.

Patients were 11 men and 5 women with a mean age of 59.6 ± 7.7 years. All patients presented with a history of dream-enacting behaviors and unpleasant dreams, in addition to habitual loud snoring, witnessed apneas, unrefreshing sleep upon awakening and excessive daytime sleepiness with a mean Epworth Sleepiness Scale score of 14.1 ± 3.3 (range, 11-21). Mean duration of the abnormal sleep behaviors was 6.9 ± 5.2 years. Ten patients were unaware of their behaviors which were only noticed by their bed partners although they recalled having frequent unpleasant dreams. Abnormal sleep behaviors were noticed at any time during sleep with variable frequency ranging from once every two weeks to several times per night. Two patients assaulted their spouses, five fell out of bed and two suffered lacerations in the face and the arms. All patients reported frequent disturbing dreams such as being attacked or chased by an unknown person (Table 1). In this study, neurological examination was normal in all subjects, and none had a history of seizures, somnambulism, night terrors, and post-traumatic stress disorder. None of the patients was taking psychotropic medications three weeks before baseline VPSG studies.

In all patients, VPSG excluded RBD and was diagnostic of severe OSAH showing a mean AHI of 67.5 ± 18.7 (range, 41-105) and a mean AHI during REM sleep of 59.6 ± 18.0 (range, 32-98). During REM sleep, the longest period of obstructive apnea was 51.8 ± 16.5 seconds and the minimal oxyhemoglobin saturation was 57.2 ± 18.8. Compared to healthy controls, patients' baseline PSG showed an increase in the AHI, oxyhemoglobin desaturations, arousal index, stage II amount, and a decrease in slow wave sleep and REM sleep percentages. When compared to healthy controls, patients with OSAH had no increased REM sleep submental tonic and phasic submental and bilateral anterior tibialis EMG activity. REM sleep without submental atonia percentage, and phasic EMG activity in the chin and in the legs were significantly increased in subjects with idiopathic RBD compared to patients with OSAH (Table 2). Epileptiform activity was not detected in any subject included in this study.

In all patients, analysis of the videotapes disclosed abnormal vigorous behaviors that appeared to be acting out a dream that only occurred during arousals at the end of the majority of obstructive sleep apneic events. The most frequent behaviors observed on VPSG were gesturing, kicking, raising the arms and talking. The types and intensity of the abnormal behaviors varied among the patients but in each particular subject they were similar across the different sleep stages. In each patient, the dream-enacting behaviors displayed from arousals in REM sleep were indistinguishable clinically from those occurring in NREM sleep. Behaviors were detected in 54% of the patients in both arousals from REM sleep and NREM sleep, and in 46% of the subjects only in REM sleep.

Three patients refused to be treated with CPAP or any further therapy for OSAH, and in these subjects the dream-enacting behaviors and daytime hypersomnolence persisted. In the remaining 13, CPAP titration eliminated snoring, apneic events, arousals and oxyhemoglobin desaturations with an optimal pressure of 10.0 ± 2.1 cm of $H_20$. No other interventions or medications such as clonazepam were started. During follow-up visits these 13 subjects reported good CPAP compliance using the machine every night with complete cessation of: 1) abnormal

**Table 1**—Patients with OSAH and idiopathic RBD unpleasant dreams and abnormal sleep behaviors witnessed by their bed partners.

| | Patients with OSAH (n=16) N (%) | Patients with idiopathic RBD (n=16) N (%) |
|---|---|---|
| **Unpleasant dream content** | 100 | 100 |
| Attacked by someone | 62.5 | 93.8 |
| Chased by someone | 62.5 | 81.3 |
| Arguing with someone | 50 | 68.8 |
| Falling abruptly | 25 | 68.8 |
| Attacked by animals | 25 | 43.8 |
| **Abnormal vocalizations** | 100 | 100 |
| Talking | 100 | 100 |
| Shouting | 75 | 100 |
| Crying | 25 | 75 |
| Swearing | 6.3 | 50 |
| **Abnormal motor behaviors** | 100 | 93.8 |
| Gesturing | 75 | 75 |
| Punching | 68.8 | 87.5 |
| Falling out of bed | 31.3 | 87.5 |
| Kicking | 25 | 81.3 |
| Knocking off the nightstand | 18.8 | 68.8 |
| Assaulting the bed partner | 12.5 | 12.5 |

sleep behaviors; 2) unpleasant dreams; 3) snoring; and 4) daytime hypersomnolence.

## DISCUSSION.

We identified a group of subjects who presented with abnormal sleep behaviors and unpleasant dreams in addition to typical clinical features of OSAH syndrome. Behaviors mimicked RBD symptoms since patients had a history of limb and body movements associated with dream mentation, potentially harmful sleep behaviors, and dreams that appeared to be "acted out"[15]. The reported type of sleep behaviors (punching, gesturing, talking, etc) and dreams (being attacked and chased, etc) were the same to those occurring in RBD[1,2] (Table 1). Moreover, age and sex distribution in our patients were also similar to those in idiopathic RBD, occurring predominantly in older males[1,2]. Therefore, based on clinical history, one would believe that our patients were affected both by OSAH and RBD. VPSG, however, was diagnostic of OSAH, excluded RBD, and showed that the abnormal behaviors occurred only during respiratory-related arousals.

Anecdotal reports of OSAH simulating the clinical features of RBD have been briefly reported[15-19], but no formal clinical and audiovisual-PSG quantitative work investigating this condition has been done in a group of patients. To the best of our knowledge, here, we have demonstrated for the first time in a case series that PSG with audiovisual monitoring formally; 1) discloses that the abnormal motor and vocal sleep behaviors reported only occurred during apneic-related arousals in REM and NREM sleep; and that 2) detailed quantitative analysis of REM-sleep EMG activity excluded RBD because tonic and phasic activity was not increased when compared with a group of healthy controls and because it was decreased when compared with subjects with idiopathic RBD. In addition, we showed that specific treatment for severe OSAH syndrome with CPAP eliminated the abnormal behaviors and unpleasant dream recall, thereby proving that OSAH itself caused the abnormal sleep activity reported. It has been shown that in subjects with concomitant OSAH and RBD, CPAP therapy resolves OSAH but not RBD[20].

We showed that when subjects with untreated severe OSAH syndrome are awakened during REM sleep, they recall more violent and emotional dreams than controls[21]. We, and others[15,16], have observed that in subjects with OSAH "pseudo-RBD" is only seen in subjects with high AHI and severe oxyhemoglobin desaturations. It can be speculated that in some subjects with severe OSAH syndrome, "pseudo-RBD" is a form of a confusional arousal caused by a combination of severe oxyhemoglobin desaturations, intense chocking episodes at the end of the apneic events and other unknown factors. It remains to be elucidated why not all of the patients with severe OSAH recall disturbing dreams and display vigorous sleep behaviors.

OSAH and RBD are two different sleep disorders with different pathophysiologic substrates. While OSAH is caused by repetitive obstruction of the upper airway during sleep, RBD is thought to reflect dysfunction of the brainstem structures that modulate REM sleep[1]. To distinguish RBD from OSAH is important because these two conditions have different clinical implications. Diagnosis of RBD in a subject with no evidence of an underlying neurodegenerative disease must be accurate since idiopathic RBD frequently precedes the onset of a parkinsonian or cognitive disorder. Moreover, OSAH and RBD require very different treatments, and clonazepam, the treatment of choice for

**Table 2**—Demographic and polysomnographic findings and comparisons between patients with OSAH, healthy controls and subjects with idiopathic RBD.

| | OSAH (n=16) | Controls (n=20) | Idiopathic RBD (n=16) | *OSAH vs. controls* *p* Value | *OSAH vs. RBD* *p* Value |
|---|---|---|---|---|---|
| Age (years) | 59.6 ± 7.7 | 63.0 ± 9.8 | 64.5 ± 5.1 | NS | NS |
| Male/Female (n) | 11/5 | 16/4 | 13/3 | NS | NS |
| Total sleep time (min) | 352.1 ± 37.4 | 361.3 ± 62.0 | 316.5 ± 78.3 | NS | NS |
| Sleep efficiency (%) | 77.8 ± 8.9 | 75.5 ± 12.6 | 69.3 ± 14.7 | NS | NS |
| Sleep latency (min) | 10.5 ± 8.1 | 19.1 ± 23.0 | 29.5 ± 25.0 | NS | 0.003 |
| Arousal index | 73.9 ± 17.2 | 15.9 ± 8.5 | 21.6 ± 11.3 | <0.001 | <0.001 |
| Awakenings (n) | 21.2 ± 8.6 | 17.6 ± 7.0 | 16.6 ± 10.5 | NS | NS |
| Stage I (%) | 15.9 ± 17.6 | 13.4 ± 6.1 | 16.5 ± 13.6 | NS | NS |
| Stage II (%) | 65.9 ± 19.9 | 46.7 ± 11.5 | 40.7 ± 10.9 | 0.001 | <0.001 |
| Stage III-IV (%) | 5.1 ± 5.6 | 19.8 ± 5.9 | 25.8 ± 17.5 | <0.001 | <0.001 |
| Stage REM (%) | 12.9 ± 7.3 | 19.5 ± 6 | 20.4 ± 9.4 | 0.007 | 0.022 |
| REM sleep latency (min) | 136.5 ± 90.7 | 99.1 ± 56.7 | 110.0 ± 75.9 | NS | NS |
| Apnea-hyponea index | 67.5 ± 18.7 | 3.2 ± 2.8 | 2.5 ± 2.9 | <0.001 | <0.001 |
| CT 90(%) | 53.2 ± 28.8 | 0.6 ± 0.7 | 0.7 ± 1.5 | <0.001 | <0.001 |
| REM sleep without atonia (%) | 2.7 ± 1.5 | 4.3 ± 3.3 | 36.9 ± 35.3 | NS | 0.001 |
| REM sleep submental phasic EMG activity (%) | 3.7 ± 1.3 | 2.7 ± 1.6 | 31.4 ± 20.1 | NS | <0.001 |
| REM sleep bilateral anterior tibialis phasic EMG activity (%) | 0.4 ± 1.0 | 2.2 ± 2.5 | 17.1 ± 11.5 | 0.004 | <0.001 |
| PLMS index | 10.3 ± 22.5 | 8.6 ± 12.1 | 8.0 ± 14.7 | NS | NS |

Results are presented as means ± standard deviations. n=number of subjects, NS= non significant, CT90=percentage of sleep time with oxyhemoglobin saturation below 90%. PLMS index=number of periodic leg movements per hour of sleep.

RBD, may worsen existing OSAH.

Dream-enacting behaviors resembling RBD may also occur in the setting of other conditions such as nocturnal seizures[22], peduncular hallucinosis[23], night terrors in young-adults[15] and nocturnal wanderings[24]. In these situations, only VPSG can exclude RBD and detect these other disorders. Alternatively, RBD may be associated incidentally with OSAH, and only VPSG can detect both conditions when occurring in the same patient. In one study, PSG of 93 subjects with RBD showed an AHI >10 in 32 (34%)[2]. Therefore, our study and the available data indicate that when RBD is clinically suspected, PSG with synchronized audiovisual recording is mandatory to confirm the diagnosis.

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