

# Severe Central Sleep Apnea/Hypopnea Syndrome Mimicking Rapid Eye Movement Sleep Behavior Disorder

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Rapid eye movement sleep behavior disorder (RBD) is a sleep disorder characterized by loss of muscle atonia during rapid eye movement (REM) sleep associated with dream enactment. However, these complex behaviors can also occur in other sleep disorders, a clinical situation termed as “pseudo-RBD”. Anecdotal reports of obstructive sleep apnea syndrome simulating the clinical features of RBD have been reported. It has been suggested that “pseudo-RBD” is a form of a confusional arousal caused by a combination of severe oxygen desaturations and other unknown factors. However, there have been no previous reports of RBD-like behaviors associated with central sleep apnea. Here, we describe two patients who reported complex motor and vocal behavior mimicking RBD, but REM sleep without atonia was not observed in video-polysomnography (VPSG). Furthermore, VPSG demonstrated severe central sleep apnea-hypopnea inducing abnormal motor and vocal behaviors.

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**Key Words** Central sleep apnea/hypopnea, Rapid eye movement sleep behavior disorder, Pseudo-rapid eye movement sleep behavior disorder.

## INTRODUCTION

Rapid eye movement sleep behavior disorder (RBD) is a sleep disorder characterized by loss of muscle atonia during rapid eye movement (REM) sleep associated with dream enactment.<sup>1</sup> RBD may be idiopathic or associated with neurodegenerative diseases.<sup>1</sup> Patients with RBD display complex motor behaviors such as gesturing, falling, talking, and shouting that are associated with unpleasant dreams. However, these abnormal sleep behaviors can also occur in other sleep disorders such as obstructive sleep apnea syndrome (OSAS).<sup>2</sup> This clinical situation has been termed “pseudo-RBD”. Iranzo and Santamaría<sup>2</sup> have identified patients who presented with abnormal sleep behaviors and unpleasant dreams in addition to OSAS. “Pseudo-RBD” has been reported in five cases of severe OSAS, they presented violent behaviors during OSAS-induced arousals from REM sleep.<sup>3</sup> Other anecdotal reports of OSAS simulating the clinical features of RBD also have been reported.<sup>4,5</sup>

Nevertheless, there have been no previous reports of pseudo-RBD associated with central sleep apnea-hypopnea (CSAH) syndrome. Here, we describe two patients who display complex behaviors mimicking RBD, but REM sleep without atonia was not observed in video-polysomnography (VPSG). Furthermore, VPSG demonstrated abnormal motor behaviors related to arousals induced by severe CSAH.

## CASE REPORT

### Patient 1

An 82 year old woman came into our hospital with sleep disturbance. She had abnormal behaviors during sleep, such as talking, singing and pounding on the wall, and also had unpleas-

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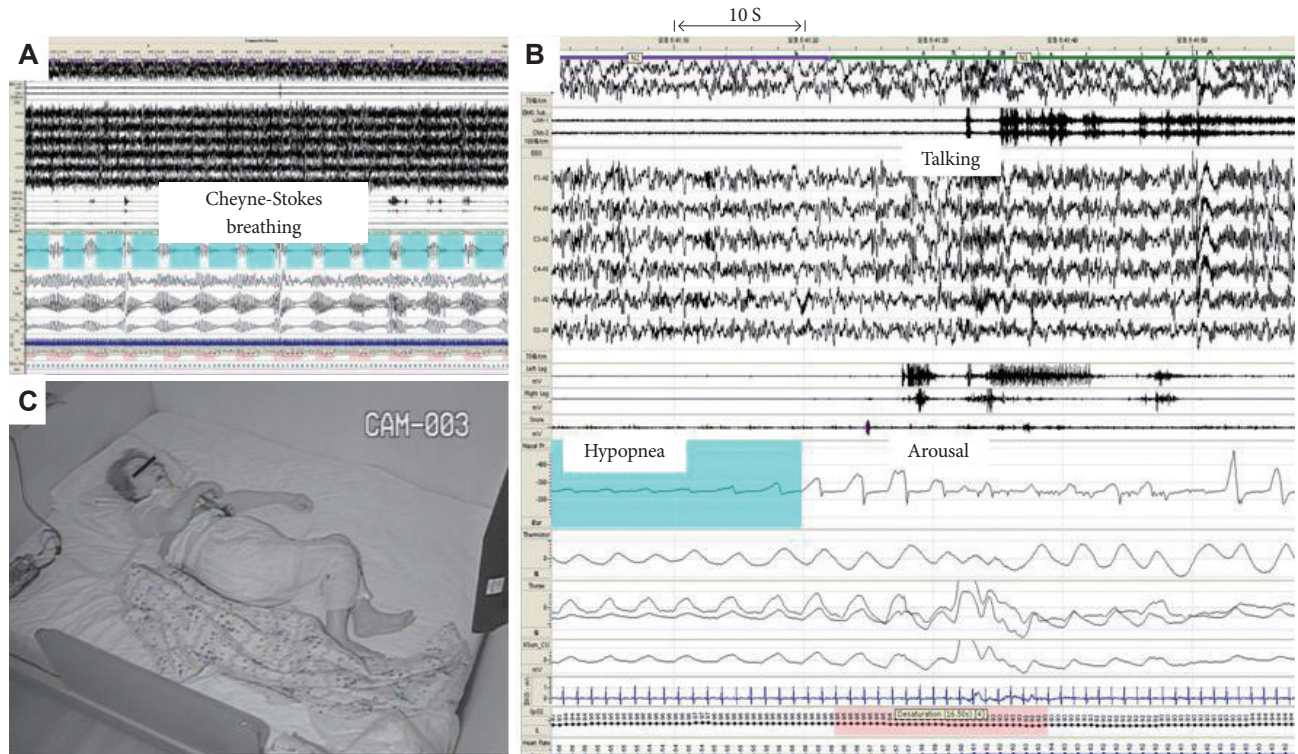
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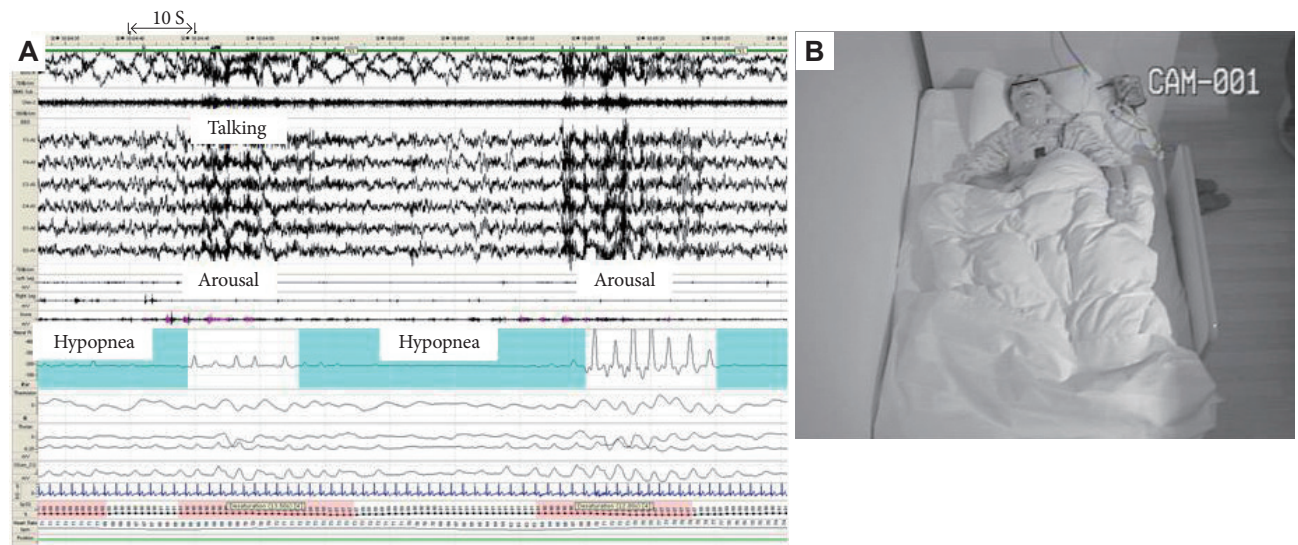
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ant dreams and daytime hypersomnolence. She had been taking antihypertensive medicine for 20 years and there was a history of intracranial hemorrhage on the right basal ganglia 15 years ago. Recently, she was diagnosed with diagnostic heart failure by a cardiologist. VPSG was done. Sleep architecture showed an increased amount of N1, and decreased N3 and R sleep. Sleep latency was 0.5 minutes. The apnea-hypopnea index (AHI) was 52.1/h, total arousal index was 23.8/h and apnea-hypopnea arousal index was 16.1/h. The lowest oxygen saturation was 88%. She

showed a waxing and waning pattern of ventilation, which is suggestive of Cheyne-Stokes respiration (Fig. 1). Talking and singing behaviors were recorded related to arousals during a hyperventilation period in N1, and no event including REM sleep without atonia (RSWA) was detected during R sleep (Fig. 1). Therefore, we diagnosed this patient as pseudo-RBD related to CSAH.



**Fig. 1.** A and B: Polysomnographic recording of patient 1 shows the evidence of typical Cheyne-Stokes respiration. C: RBD-like symptom (talking) was recorded in N1 sleep related to arousal. RBD: rapid eye movement sleep behavior disorder.



**Fig. 2.** A: Polysomnographic recording of patient 2 shows a periodic breathing pattern. B: Talking and body movements were recorded in N1 sleep related to arousal.

## Patient 2

A 60 year old man had suffered from progressive motor weakness for a year. His symptoms started with weakness in both legs, and 6 months later muscle wasting was observed, as well as dyspnea on minimal movement. The findings of an electromyography suggested anterior horn cell disease. He also had newly-developed abnormal sleep behavior such as gesturing or talking, frequent awakening during sleep, and daytime fatigue since 4 months. In VPSG, sleep architecture showed an increased amount of N1, and decreased N2, and no N3, R sleep. Sleep latency was 3.0 minutes. AHI was 120/h, total arousal index was 119.6/h and apnea-hyponea arousal index was 115.0/h. The lowest oxygen saturation was 82%. He showed a periodic breathing pattern (Fig. 2). Talking and body movements were recorded in N1 sleep at the end of a hypopnea period accompanied with arousals, and RSWA was not detected (Fig. 2). Therefore, central sleep apnea and accompanied pseudo-RBD was diagnosed.

## DISCUSSION

Here, we report two patients who presented with complex sleep behaviors in addition to CSAH syndrome. Their clinical presentations were similar to RBD, and initially we thought they may have had RBD. However, in VPSG, we identified CSAH and abnormal behaviors occurring in respiratory effort-related arousals, not in the REM sleep period.

Previous studies<sup>2-4</sup> have indicated that, in patients with OSAS, “pseudo-RBD” is only seen in those patients with a high AHI and severe oxyhemoglobin desaturations. It can be postulated that in some patients with severe OSAS syndrome, “pseudo-RBD” is a form of confusional arousal caused by a combination of severe oxyhemoglobin desaturations and other unknown factors.<sup>2</sup> However, the lowest O<sub>2</sub> saturations were 88% and 82% in our patients, which are not severe compared with that reported in the previous study<sup>2</sup> (mean minimal oxyhemoglobin saturation  $57.2 \pm 18.8\%$ ). It is due to the fact that arousals are associated more with hyperventilation than with severe desaturation in non-hypercapnic CSAH patients like ours.

Central sleep apnea-hypopnea syndrome is characterized by a lack of drive to breathe during sleep, resulting in insufficient ventilation and compromised gas exchange, repetitively.<sup>6</sup> CSAH can be broadly classified into hypercapnic and non-hypercapnic.<sup>6</sup> In our two patients, we could identify non-hypercapnic CSAH such as Cheyne-Stokes breathing and a periodic breathing pattern, which is quite different from hypercapnic CSAH that diminishes ventilatory output.<sup>6</sup> Non-hypercapnic CSAH promotes unstable breathing such as a high ventilatory drive, and vulnerability of the arousal and accompanying hyperventilation.<sup>6</sup> Therefore, it is speculated that the patient with non-hypercapnic CSAH can experience frequent arousals com-

pared to hypercapnic CSAH, and is more likely to have “pseudo-RBD” due to these frequent arousals. Additionally, arousals typically occur in the mid-cycle of Cheyne-Stokes respiration, at the peak of ventilator effort,<sup>7</sup> and at the termination of central apnea in periodic breathing.<sup>6</sup> We can also see those patterns of arousals in our patient’s polysomnography (PSG). Therefore, it is postulated that non-hypercapnic CSAH, as our patients, rather than hypercapnic CSAH, can lead to “pseudo-RBD” due to frequent arousals.

To distinguish RBD from CSAH is important because these two diseases have different clinical implications. CSAH is caused by brainstem structures or various neuromuscular disorders, and its complications are secondary polycythemia, pulmonary hypertension, and cor pulmonale.<sup>6</sup> Additionally, Cheyne-Stokes respiration is most commonly observed in patients with congestive heart failure and left ventricular systolic dysfunction.<sup>6</sup> Regarding RBD, the diagnosis in a patient with no evidence of neurodegenerative disease must be clearly identified because idiopathic RBD can frequently be a prodromal symptom of neurodegenerative diseases.

Pseudo-RBD may also occur in other conditions such as nocturnal seizures,<sup>8</sup> night terrors<sup>4</sup> and nocturnal wanderings.<sup>9</sup> In these situations, only VPSG can exclude RBD and detect these other disorders. Therefore, VPSG is essential to find concomitant sleep disorders, to distinguish true RBD and pseudo-RBD for patients suffering abnormal sleep behaviors resembling RBD.

In conclusion, we are the first to report two patients with pseudo-RBD associated with CSAH. Unlike that in OSAS, pseudo-RBD in CSAH may not present severe oxyhemoglobin desaturation because arousals in CSAH are usually related to hyperventilation. And among the two types of CSAH, non-hypercapnic CSAH rather than hypercapnic CSAH can result in pseudo-RBD due to frequent arousal during sleep. Pseudo-RBD may occur in various sleep disorders including CSAH and OSAS, and in these situations, PSG can exclude RBD and detect these other disorders.

## Conflicts of Interest

The authors have no financial conflicts of interest.

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