A Case of Rapid Eye Movement Sleep Behavior Disorder during Continuous Positive Airway Pressure Titration in a Patient with Multiple System Atrophy

Jung-Hwan Oh, Sook-Keun Song, Ji-Hoon Kang

Department of Neurology, Jeju National University School of Medicine, Jeju, Korea

Sleep disorders are commonly observed in multiple systemic atrophy (MSA). The rapid eye movement (REM) sleep behavior disorder (RBD) is characterized by loss of normal voluntary muscle atonia during REM sleep. It usually presents during early course, and disappears over the course of disease progression. Sleep-disordered breathing (SDB) is also common sleep disorder in MSA which can be life-threatening, and continuous positive airway pressure (CPAP) treatment is useful in these patients. A 74-year-old woman with MSA presented for nocturnal respiratory disturbance. She had a five-year history of dream enacting behaviors, which had disappeared four months prior. Polysomnography revealed frequent stridor and sleep hypopnea. During the following full night CPAP titration for SDB, dream enacting behavior was observed during REM sleep stage. In MSA patients with SDB, CPAP administration may lead to increase REM sleep stage. An increase in REM sleep stage, which previously had been deprived, may have trigger RBD symptoms to reappear. The CPAP treatment should be considered with great caution in these patients.

Key Words: Multiple system atrophy, REM sleep behavior disorder, Continuous positive airway pressure.
the apnea/hypopnea index (AHI) was 47.3/h. The lowest oxyhemoglobin saturation (SpO$_2$) was 68%, and low SpO$_2$ was continuously measured (average SpO$_2$, 72.4%). Sleep architecture showed an increase in stages N1 and N2 sleep, decreased stage N3 sleep, and absent REM sleep. Six weeks after the initial PSG, CPAP titration with PSG was performed. CPAP was administered using 4 to 13 cm H$_2$O. During the titration with 11 cm H$_2$O pressure, the patient had three episodes of talking and complex motor behaviors that were not observed in the initial PSG, with increased muscle tone during REM sleep, which was compatible with RBD (Fig. 2). REM sleep (3.7% of total sleep stage) occurred after 244 minutes of latency, and was only present during the titration with 11 cm H$_2$O pressure. A fixed CPAP pressure of 11 cm H$_2$O was considered adequate for treatment of the SDB. With a pressure of 11 cm H$_2$O, the AHI decreased to 4.7/h, lowest SpO$_2$ was 83%, average SpO$_2$ was 91.8%, and stridor was nearly eliminated.

**Discussion**

Marked sleep fragmentation in patients with MSA has been reported by several polysomnographic studies and is related to motor symptoms such as urinary incontinence, rigidity, and bradykinesia with subsequent inability to turn over in the bed. Recently, two cases of RBD during CPAP titration were reported in Korean patients with OSA who developed RBD symptoms with increased REM sleep. OSA can also lead to sleep fragmentation; hence, patients with the disease show an increase in light non-REM sleep, and a decrease in REM sleep and slow-wave sleep on PSG. When CPAP is administered at the appropriate pressure level for OSA, respiratory obstructive events are eliminated; hence, the amounts of REM and slow-wave sleep increase.

Extensive research has shown that SDB is a frequent and major problem in patients with MSA. One of the most typical types of SDB in MSA is stridor caused by upper airway obstruction that frequently accompanies OSA. Such SDB can cause sleep fragmentation in patients with MSA, and CPAP is recommended as a first-line treatment for increased long-term survival. In the present case, during a therapeutic trial of CPAP, when an adequate pressure level for the treatment of SDB was reached, REM sleep occurred, and consequently, RBD developed.

Although RBD is a common manifestation in patients with MSA, it does not present in the later stages of disease, and it can disappear with worsening disease condition. These phenomena can be explained by progressive degeneration of the brain stem nuclei, which control REM sleep. Furthermore, in cases like the present, sleep fragmentation might mask RBD in MSA patients with SDB. In addition, clonaze-
pam, the first-line medication in the management of MSA patients with RBD, is known to worsen sleep apnea; this drug might exacerbate SDB, and consequently might influence sleep fragmentation in patients with MSA.

In the present report, we illustrated the case of a MSA patient with SDB who developed RBD during CPAP titration. To our knowledge, this is the first case of its kind, and hence merits reporting. Both RBD and SDB are common manifestations of MSA. Because RBD symptoms might affect CPAP adherence, physicians should carefully guide CPAP treatment in patients with MSA. Although MSA patients with SDB might have a history of RBD that has disappeared, clinicians should consider the presence of RBD symptoms during CPAP application with adequate pressure. Moreover, clonazepam treatment can exacerbate OSA and is associated with less optimal treatment outcomes in patients with RBD; hence, the choice of other classes of drugs without respiratory effects, such as melatonin, and more optimized CPAP treatment should be considered in MSA patients with SDB.

Acknowledgments
This research was supported by the 2017 scientific promotion program (Jeju National University).

REFERENCES
4. Patil SP, Schneider H, Schwartz AR, Smith PL. Adult obstructive sleep
RBD during CPAP Titration in MSA

10. Zhang J, Li SX, Lam SP, Wing YK. REM sleep behavior disorder and obstructive sleep apnea: does one “evil” make the other less or more “evil”? *Sleep Med* 2017;37:216-217.