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CASE REPORTS

Nocturnal Hypermotor Activity during Apnea-Related Arousals

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We present a case of a 50-year-old patient who exhibits nocturnal hypermotor activity occurring exclusively during apnea-related arousals consisting of repetitive lower extremity hip-flapping. This movement is unusual and reflects a new form of lower extremity movement associated with apnea-related arousals.

Keywords: sleep apnea, apnea-related arousal, hypermotor

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INTRODUCTION

Nocturnal hypermotor movements are an unusual occurrence. We report a case of complex lower extremity movements occurring exclusively during apnea-related arousals.

REPORT OF CASE

The patient is a 50-year-old African American man with obesity, hypertension, and excessive daytime sleepiness, who presents for evaluation for witnessed snoring, and witnessed apnea. The patient has no history of seizures. He has no bed partner, and presented no complaints of unusual movements during sleep. He underwent a split night diagnostic polysomnography. The diagnostic portion of the recording showed a total sleep time apnea-hypopnea index (AHI) of 33.7, REM AHI of 80.0, and an oxyhemoglobin saturation nadir of 74%. Diagnostic portion showed total sleep time 187 minutes, sleep efficiency 78%, total REM time 22.5 minutes, and sleep stage percentages of N1 7.8%, N2 77%, N3 3.2, and REM 12%.

Repetitive lower extremity movements during arousals following apneic events were seen, consisting of repetitive hip-flapping (**Figure 1**), with these movements noted during both NREM and REM sleep. During the titration portion of the recording, patient required bilevel positive airway pressure of 24/20 to resolve the obstructive apneas. The movements resolved with resolution of the patient's sleep disordered breathing.

DISCUSSION

Nocturnal hypermotor movements are complex movements involving proximal segments of the limbs that appear inappropriate to the situation. Brief repetitive stereotyped nocturnal hypermotor movements are typically associated with nocturnal frontal lobe epilepsy with the implicated brain regions including mesial frontal cortex, orbitofrontal cortex, and insula.[1] This case demonstrates the power of nocturnal polysomnography in capturing and characterizing brief repetitive stereotyped nocturnal hypermotor movements, even in those who may not realize they exhibit such movements, and expands the differential diagnosis for such movements to include post-apnea related motor activity.

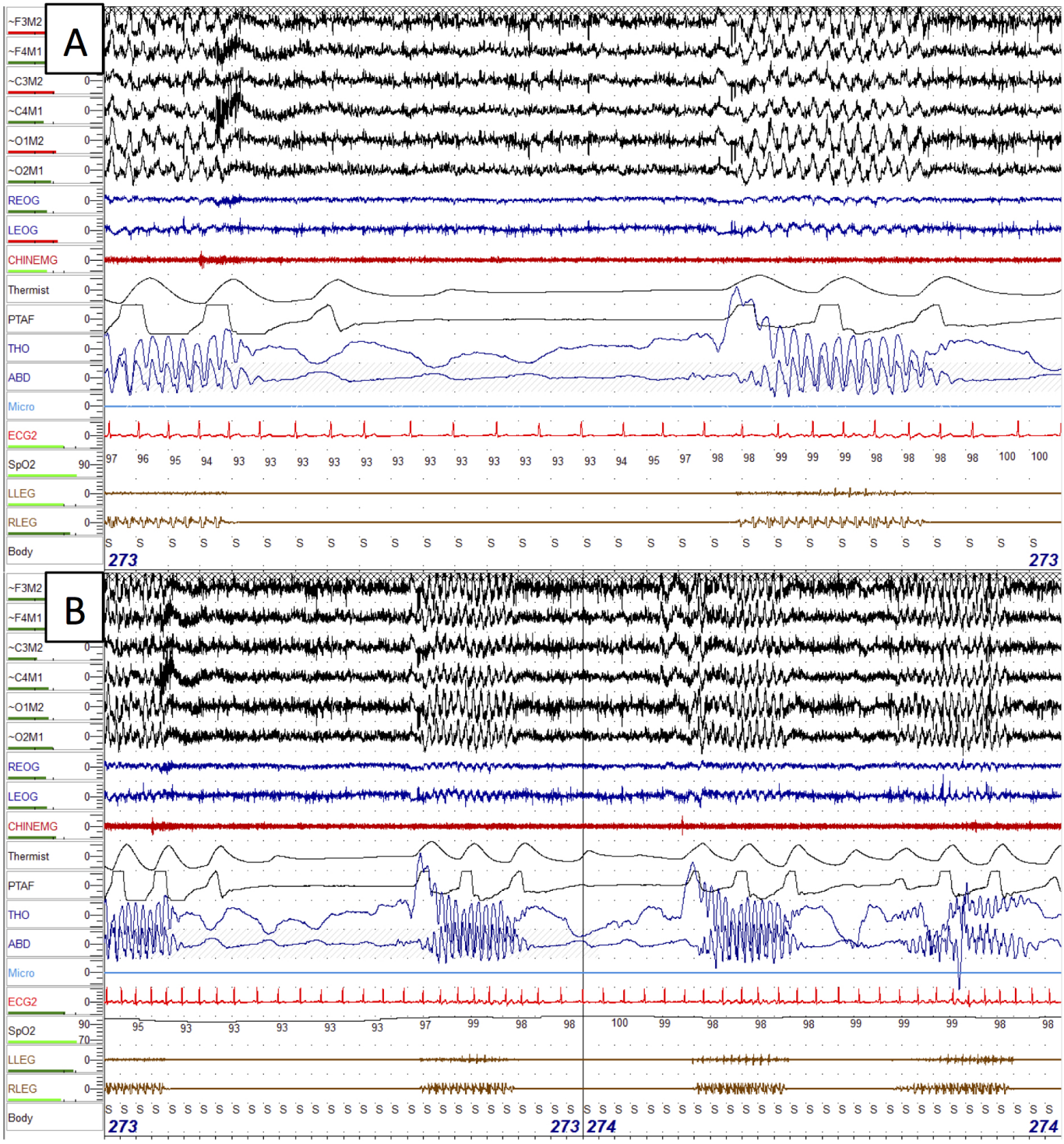
Our patient exhibited hyper-motor activity in the lower extremity, hip flapping, during arousal from sleep-disordered breathing, and this behavior resolved with positive airway pressure therapy. In patients with obstructive sleep apnea syndrome, varying degrees of arousal events occur in relation to respiratory event arousals, including lower extremity motor activation, but complex repetitive stereotyped movement are not commonly seen.

Motor response after respiratory events can be seen in both NREM and REM, as in our case, though muscle activation in REM occurs less frequently, and is likely due to active muscle inhibition during REM.[2] Muscle response to respiratory events occurs more frequently when respiratory events are associated with either electroencephalographic (EEG) micro-arousals or awakenings, vis-a-vis respiratory events with no arousal afterward. EEG arousal was difficult to assess in our case given associated movement artifact. Increased muscle sympathetic nerve activity has been demonstrated in post-apnea arousals, and may be in play a role in the motor response after respiratory events, but more research is needed.[3]

DESCRIPTION OF VIDEO 1

During arousal following a respiratory event patient exhibits repetitive hip abduction/adduction lasting a few seconds. The patient maintained sleep during this movement, and exhibited this behavior repetitively during the diagnostic portion of the recording. See **Video 1** in the supplemental material.

Figure 1—Hyper-motor lower extremity response during arousals from sleep-disordered breathing.



(A) 30-second epoch. **(B)** 1-minute segment.

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DISCLOSURE STATEMENT

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