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CASE REPORTS

Violent Parasomnia With Recurrent Biting and Surgical Interventions: Case Report and Differential Diagnosis

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A case is reported of recurrent, injurious self-biting during sleep, requiring surgical interventions, in a 55-year-old obese man with a 20-year history of violent complex parasomnia, with greatly increased frequency and severity of episodes induced by work stress during the preceding 3 years. After clinical evaluation and overnight, hospital-based video-polysomnography, the cause of the chronic injurious parasomnia was deemed to be a non-rapid eye movement (NREM) sleep parasomnia comorbid with severe obstructive sleep apnea. Therapy with bedtime clonazepam and bilevel positive airway pressure was effective, with injurious parasomnia relapse occurring with cessation of either or both of these therapies. The differential diagnosis of sleep-related biting should now include NREM sleep parasomnia (with or without comorbid obstructive sleep apnea), besides previously reported cases of REM sleep behavior disorder (RBD), sleep-related dissociative disorder, sleep-related rhythmic movement disorder and anticipated cases of parasomnia overlap disorder (RBD + NREM sleep parasomnia), sleep-related biting seizures, and sleep-related eating disorder.

Keywords: biting injury, clonazepam, CPAP, NREM parasomnia, obstructive sleep apnea, polysomnography, REM sleep behavior disorder, sleep-related rhythmic movement disorder, sleep-related dissociative disorder

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INTRODUCTION

Parasomnias comprise the group of sleep disorders characterized by abnormal behaviors, experiences, and autonomic activity during entry into sleep, during any stage of non-rapid eye movement (NREM) or REM sleep, and during sleep-wake transitions.[1] We now present a unique case of a chronic, violent parasomnia, associated with severe obstructive sleep apnea (OSA), which included recurrent biting of the same index finger that caused major injury requiring surgical interventions.

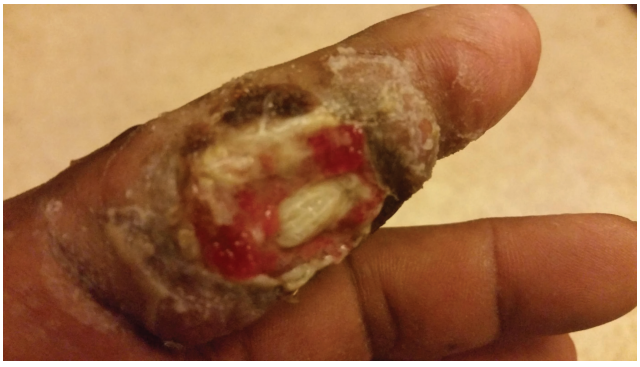
REPORT OF CASE

A 55-year-old, single, obese black male, with a body mass index of 41 kg/m^2, who worked on night shifts at a manufacturing company, presented with a 20-year parasomnia history in which he would wake up in the first 2 to 3 hours of the night with a sense of fear while "running" away from snakes and sometimes "fighting" back at wild animals attacking him. He would find himself "wrestling" with pillows, falling off the bed, running out of the bedroom to the living room, and running into tables. On one occasion, he stepped on a glass table and broke it. He had sustained injuries during these episodes, but never had any witnessed events because he has lived alone and slept alone. He once had a bed partner for several years more than 20 years ago, but she had not witnessed any events. He never had more than one episode nightly, and the initial frequency was once or twice a year.

He reported that in relation to stress at work his "night terrors" became more frequent and aggressive since 2014, and started occurring once every 2 to 3 weeks. On one occasion in 2015, he woke up pounding the floor with his fists. In an episode that he described as his "worst one," he woke up while running and found himself biting his index finger, which was bitten down to the tendon, for which he needed surgical intervention for tendon repair (**Figure 1**). He was referred to a psychiatrist who diagnosed REM sleep behavior disorder (RBD) and prescribed clonazepam 1 mg at bedtime, which stopped the nocturnal events. He was then referred to a sleep physician, and underwent polysomnography (PSG) while on clonazepam, which revealed severe OSA, with an apnea-hypopnea index (AHI) of 38.9 events/h. He was titrated on bilevel positive airway pressure (BPAP) at 16/12 cm H_2O, and had a residual AHI of 4.4 events/h and a minimum SaO_2 of 91%. No behavioral sleep-related episodes were observed. At first, he was not reliably BPAP compliant and complained of frequent awakenings. He had another parasomnia episode in October 2016, when he woke up biting his right wrist, sustaining a superficial bite mark that did not require any wound care. He later became BPAP compliant.

In July 2017, he ran out of clonazepam and subsequently experienced the prompt re-emergence of abnormal sleep behaviors, including a recurrent episode of severe biting of the same index (left) finger that caused a deep puncture wound (**Figure 2**). He was not using BPAP on that night. He consulted the plastic surgery department for wound care. He then

Figure 1—Self-biting during sleep in 2015.



Avulsion injury to left index finger resulting in exposure of flexor digitorum tendon. Skin flap was repaired and later he underwent rehabilitation exercises to prevent contractures.

Table 1—Differential diagnosis of sleep-related biting.

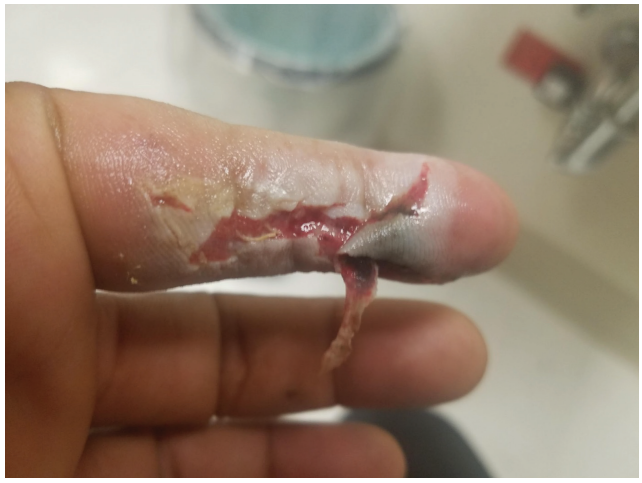
1. NREM sleep parasomnia
2. Obstructive sleep apnea
3. NREM sleep parasomnia + OSA
4. REM sleep behavior disorder
5. Parasomnia overlap disorder (RBD + NREM parasomnia)
6. Sleep-related dissociative disorder
7. Sleep-related rhythmic movement disorder
8. Sleep-related seizures
9. Sleep-related eating disorder

NREM = non-rapid eye movement, OSA = obstructive sleep apnea, RBD = REM sleep behavior disorder, REM = rapid eye movement.

presented to an author's (IK) sleep clinic the following month after his third sleep-related biting episode. He was restarted on clonazepam, 1 mg at bedtime, with immediate and sustained benefit. A video-PSG with seizure montage took place in November 2017. He was taken off the clonazepam 2 days prior to the video-PSG. Total sleep time was less than 5 hours (285.5 minutes), sleep latency was 0.3 minutes, and REM sleep latency was 55.5 minutes. Sleep efficiency was 90.8%. Percentages of sleep stages were as follows: N1: 9.6%, N2: 69.7%, N3: 1.8%, R: 18.9%. There was no electroencephalogram (EEG) epileptiform activity, nor any confusional arousals from NREM sleep. Periodic leg movement index was 7.1 events/h. Nasal continuous positive airway pressure (CPAP) with 12 cm H_2O, which was used throughout his video-PSG, was effective in eliminating sleep-disordered breathing, with an overall AHI of 2.5 events/h. During some epochs of stage R sleep, there was clinically insignificant REM without atonia (RWA), and no abnormal movements or behaviors were observed during REM sleep.

There was a childhood history of generalized tonic-clonic seizures, but he had not been prescribed any anticonvulsant since the age of 14 years, without seizure recurrence. He denied any family history of parasomnias or seizures. Medical history included asthma, hypertension, and hypercholesterolemia. Physical and neurologic examinations were negative. Psychiatric history was positive for a brief depression in 2005

Figure 2—Self-biting during sleep in 2017.



Later the patient presented to the emergency department for wound care, and was treated with wound débridement and antibiotics.

for which he took an antidepressant medication for several months, without subsequent depressive recurrence.

DISCUSSION

This is a complex case of a violent parasomnia that included severe recurrent biting of the same index finger, requiring surgical interventions. The key diagnostic elements of the case will now be considered, along with the differential diagnosis of sleep-related biting.

The most likely parasomnia diagnosis is combined NREM parasomnia and severe OSA. First, in regard to an NREM parasomnia, there was an increased frequency and severity of the patient's parasomnia related to work stress, with stress being a well-known aggravator in NREM parasomnias[1]; also, response to bedtime benzodiazepine therapy (clonazepam) is common in NREM parasomnias[2]; running out of the bedroom is far more common in NREM parasomnias than in RBD[1]; brief episodes of dream-enacting behavior in adults is not uncommon in NREM parasomnias.[3] Second, in regard to severe OSA triggering parasomnia, the patient's parasomnia relapse in October 2016, when he bit his wrist, occurred when he was taking bedtime clonazepam, but not using his BPAP therapy for severe OSA, and then his relapse with a deep finger bite in July 2017 occurred when he was not using BPAP (nor taking clonazepam); also, OSA is known to trigger an NREM parasomnia,[4] including severe assaultive parasomnia (a pet cat chopped to death),[5] and aggressive dream-enacting behaviors (punching, kicking),[6] with prompt and full resolution being achieved with successful CPAP or other OSA therapy.[4–6] Presumably OSA triggers confusional arousals with complex and assaultive behaviors. RBD is a far less likely diagnosis in this case because insufficient RWA and no REM sleep behaviors were documented during the second video-PSG study (and no information was available about RWA or RBD behaviors in the first PSG study done elsewhere).

To our knowledge, this is the first reported case of biting during sleep related to a NREM parasomnia, with or without associated (severe) OSA. **Table 1** contains the differential diagnosis of sleep-related biting. There are three reports on RBD associated with biting. In a series of 203 consecutive patients with idiopathic RBD, the prevalence of biting in RBD was 8.4%, which usually involved bed partners.[7] In a case report on RBD, a man was divorced four times and had three relationship breakups because of repeatedly biting his partners during dream enactment.[8] In a case of duloxetine-induced RBD, a 62-year-old woman dreamed of biting something, but she was actually biting the hand of her grandson.[9] Parasomnia overlap disorder (ie, combined RBD and NREM parasomnia,[1] with or without associated OSA) should also be considered, given the separately documented cases of sleep-related biting with RBD and NREM parasomnias.

A case of sleep-related dissociative disorder with recurrent biting has been reported.[10] A 19-year-old male presented with a 4-year history of exclusively animalistic episodes arising during the sleep period 1 to 2 nights weekly, when he would act like a large jungle cat. He would leave his bed and crawl and leap about, and bite the furniture and various objects (leaving imprints), for up to an hour. He had injured his lips and gums on numerous occasions from biting sharp objects. During video-PSG, two of his characteristic episodes were recorded, and each arose from clear-cut EEG wakefulness, with the first episode occurring 53 minutes after sleep onset, several minutes after an epoch of stage N1 sleep had terminated in sustained EEG wakefulness. There was a 2-minute prodrome of intermittent growling, and then he left his bed and crawled around the room, hissing, growling, loudly grinding his teeth, and pulling the mattress with his jaws. He also chewed and swallowed portions of the airflow monitoring device. The second episode emerged 75 minutes after the first episode (having slept during NREM and REM sleep). During EEG wakefulness, he growled for 15 seconds before crawling out of bed, knocking a lamp over, biting that lamp, chewing and swallowing portions of another airflow monitoring device, and repeatedly banging his head against a wall.

A case of recurrent sleep-related injurious tongue biting from rhythmic movements of the jaw associated with body rocking in NREM sleep was reported in a 2-year-old girl.[11] Therefore, she suffered from two sleep-related rhythmic movement disorders (SRRMD), and SRRMD is another category of sleep disorder that can manifest with sleep-related biting.

Finally, sleep-related epileptic seizures with biting should be considered, as biting seizures are well documented in wakefulness,[12] and because NREM sleep is a seizure-promoting state. Sleep-related eating disorder[1] should also be considered in the differential diagnosis, even though non-food biting has not yet been reported, because repetitive masticatory movements during stage N2 sleep have been documented by electromyography in these patients.[13]

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DISCLOSURE STATEMENT

The authors report no conflicts of interest. Dr. Schenck reports working as a consultant for Axovant Sciences, Inc. in a capacity not related to this case report