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The spectrum of disorders causing violence during sleep



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Abstract

Violent behavior during sleep is a common problem, affecting > 2% of the population > 15 years old as found in two large epidemiologic studies. The differential diagnosis of sleep related injury and violence includes: REM sleep behavior disorder (RBD); NREM sleep parasomnias (sleepwalking, sleep terrors); parasomnia overlap disorder (RBD + NREM sleep parasomnias); obstructive sleep apnea; sexsomnia (sleep related abnormal sexual behaviors); sleep related dissociative disorder; trauma-associated sleep disorder/post-traumatic stress disorder; periodic limb movement disorder; rhythmic movement disorder; nocturnal scratching disorder; nocturnal seizures; and miscellaneous/mixed conditions. Careful clinical interviews, preferably with bed partners participating, and extensive, hospital-based, technologist-attended, overnight video-polysomnography (for one or more nights), is crucial for determining the accurate diagnosis that will guide proper therapy. Most of the disorders mentioned above are classified as parasomnias in the International Classification of Sleep Disorders, 3rd Edition, with parasomnias defined as abnormal behavioral, experiential and/or autonomic nervous system activity during entry into sleep, during any stage of sleep, and during emergence from any stage of sleep. Parasomnias are often manifestations of "state dissociation" in which components of one state of being (wake, REM sleep, NREM sleep) intrude into, and become admixed, with other states of being, with clinical consequences. There are forensic implications related to the parasomnias, with inadvertent and unintentional assaults, murder, and "pseudo-suicide" resulting from aggressive and violent behaviors arising from sleep with suspended awareness and judgement.

Keywords: REM sleep behavior disorder, NREM sleep parasomnias, Parasomnia overlap disorder, Obstructive sleep apnea, Sexsomnia, Sleep related dissociative disorder, Trauma-associated sleep disorder/post-traumatic stress disorder, Periodic limb movement disorder, Rhythmic movement disorder, Nocturnal seizures

Violent behavior during sleep (VBS): epidemiology

Two major studies have been conducted in this area (Ohayon et al. 1997; Ohayon and Schenck 2010). The first study was conducted in the United Kingdom in which a representative sample of nearly 5000 males and females (> 15 years old) participated in a telephone interview directed by the Sleep-EVAL expert system (Ohayon et al. 1997). The prevalence of current VBS was 2.1%, with a significantly higher rate in males (2.6%) than in females (1.7%). A complex set of medical, substance use, psychological and hereditary factors affecting those with VBS was identified. The second study explored the

prevalence, comorbidity and consequences of VBS in a much larger sample from five other European countries and in Finland (Ohayon and Schenck 2010). A random stratified sample of nearly 20,000 participants, > 15 years old, from the general population were interviewed by telephone using the Sleep-EVAL Expert System. VBS was reported by 1.6%. VBS was higher in subjects younger than 35 years. During VBS episodes, 79% of VBS subjects reported vivid dreams and 31% had hurt themselves or someone else. Only 12.3% had consulted a physician for these behaviors. In 73% of cases, VBS was associated with other parasomnias (highest odds of VBS for sleepwalking [SW] and sleep terrors [ST]). Family history of VBS, SW and ST was reported more frequently in VBS than in non-VBS subjects with odds of 9.3, 2.0 and 4.2, respectively. Therefore, both these studies indicate that VBS is frequent in the general population and often associated

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with dream-enactment. The high frequency of VBS, SW, and ST in the families of VBS subjects indicates that some families have greater vulnerability to sleep disorders involving motor dyscontrol.

An epidemiological study of sleep-related injury among the elderly in Hong Kong has been reported, with 0.8% of 1034 subjects > 70 years old interviewed at home answering positive to the screening question "have you ever inflicted injuries to yourself or other people during your sleep?" (Wing et al. 2000). Those subjects who answered in the affirmative then underwent extensive clinical interviews and sleep laboratory monitoring, and the estimated prevalence of REM sleep behavior disorder (RBD) was 0.38%.

The critical role of an accredited, experienced sleep disorders center in evaluating cases of VBS has been comprehensively addressed (Mahowald et al. 1992), and guidelines for assisting in the determination of the putative role of an underlying sleep disorder in a specific violent act have been formulated (Mahowald et al. 1990).

Differential diagnosis of sleep-related injury and violence

The first systematic video-polysomnographic (vPSG) and clinical study on sleep-related injury (usually caused by violent behavior) reported on 100 consecutive patients from an established multi-disciplinary sleep disorders center (Schenck et al. 1989a, 1989b). Five disorders were identified as the basis for sleep-related injury: i) NREM sleep parasomnias (SW/ST), n = 54; REM sleep behavior disorder (RBD), n = 36; sleep related dissociative disorders, n = 7; nocturnal seizures, n = 2; and obstructive sleep apnea (OSA)/periodic limb movement disorder (PLMD), n = 1. Among NREM parasomnia patients, one-third developed their parasomnia after the age of 16 years (extending up to age 58 years). Ninety-five patients had sustained recurrent ecchymoses, 30 had lacerations, and 9 had fractures.

The following vignette was provided by the wife of a man with a NREM sleep parasomnia: "He seems to have the strength of 10 men and shoots straight up from bed onto his feet in one motion. He's landed clear across the room on many occasions and has pulled down curtains, upset lamps, and so forth. He's grabbed me and pulled on me, hurting my arms...He's landed on the floor so hard that he's injured his own body. The description 'vaults explosively' describes it most clearly. There are low windows right beside our bed and I'm afraid he'll go through them some night." Another vignette involving a 67 year old man with RBD focused on how one night he was awakened "by his wife's yelling as he was choking her. He was dreaming of breaking the neck of a deer he had just knocked down. This patient had tied himself to his bed with a rope at night for 6 years as a protective measure, owing to repeated episodes of jumping from bed and colliding with furniture and walls." Since the time of this report, the differential diagnosis of sleep-related injury and violence has expanded, as listed in Table 1.

Furthermore, a report has been published on a series of 20 patients with injurious parasomnias who were admitted to intensive care units (ICUs) while their parasomnias had not yet been diagnosed or treated (Schenck and Mahowald 1991). In this series, the mean age during ICU confinement was 63 years, and 85% were males. Subsequent comprehensive sleep center evaluation with extensive vPSG monitoring was diagnostic for RBD in 85% of patients and for NREM parasomnia (SW/ST) in 15% of patients. Three types of parasomnia-ICU relationships were identified: i) Parasomnia originating in ICUs, stroke-induced (n = 3); ii) Admission to ICUs resulting from parasomnia-induced injuries (n = 2): C2 odontoid process fracture; and C3 spinous process fracture with severe concussion; iii) Parasomnias in patients admitted to ICUs for various other medical problems (n = 15). This report called attention to the possibility of injurious, and treatable, parasomnias affecting patients on various ICU units. One patient in this series was presented as a case vignette: an 81 year-old female developed RBD acutely at the time of a stroke. In the ICU she jumped out of bed and fractured a hip while enacting a dream in which she was escaping from "grotesque men with hands like claws and swarms of bees who were attacking me." After discharge, her RBD worsened with her frequently jumping out of bed. One night, 3 days after cataract surgery, she jumped from bed and punched her cheeks repeatedly, which caused profuse gingival and palatal bleeding.

Disorders causing sleep-related injury and violence

NREM sleep parasomnias

Two major concepts will first be presented to provide the pertinent clinical-scientific background for helping

Table 1 Differential diagnosis of sleep-related injury and violence

- 1. REM Sleep Behavior Disorder (RBD)
- 2. NREM Sleep Parasomnias (Sleepwalking, Sleep Terrors)
- 3. Parasomnia Overlap Disorder (RBD + NREM Sleep Parasomnias)
- 4. Obstructive Sleep Apnea
- 5. Sexsomnia (Sleep Related Abnormal Sexual Behaviors)
- 6. Sleep Related Dissociative Disorder
- 7. Trauma-Associated Sleep Disorder/Post-Traumatic Stress Disorder
- 8. Periodic Limb Movement Disorder
- 9. Rhythmic Movement Disorder
- 10. Nocturnal Scratching Disorder
- 11. Sleep Related Eating Disorder
- 12. Nocturnal Seizures
- 13. Miscellaneous/Mixed Disorders

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understand the subsequent case series and case reports illustrating aggression and violence associated with NREM parasomnias.

Factors that predispose, prime and precipitate SW episodes

Factors that predispose, prime and precipitate episodes of SW, including violent SW have been carefully considered (Pressman 2007a, 2007b). Predisposition to SW is based on genetic susceptibility and has a familial pattern. Priming factors include conditions and substances that increase slow wave sleep or make arousal from sleep more difficult. These factors include sleep deprivation, alcohol, medications, situational stress and fever, etc. A patient with a genetic predisposition to SW and with priming factors usually also needs a precipitating factor to trigger a SW episode, such as noise, touch, sleep disordered breathing, periodic leg movements, etc.

NREM sleep parasomnias as disorders of sleep-state dissociation

Patients affected by NREM parasomnias exhibit waking behaviors arising abruptly out of NREM sleep. Although these individuals remain largely unresponsive to the external environment, their EEG shows both typical sleep-like and wake-like features, and they can report dreaming afterwards. Therefore, these disorders offer a unique natural model to explore the abnormal coexistence of local sleep and wake brain activity and the dissociation between behavior and various aspects of consciousness, as discussed in two major critical review articles. The first review article stated that the "traditional idea of somnambulism as a disorder of arousal might be too restrictive and a comprehensive view should include the idea of simultaneous interplay between states of sleep and wakefulness. Abnormal sleep physiology, state dissociation, and genetic factors might explain the pathophysiology of the disorder" (Zadra et al. 2013). The second more recent review article provided major findings and updates on NREM sleep parasomnias, focusing on neurophysiological studies, and offering an overview of new clinical frontiers and promising future research areas (Castelnovo et al. 2018). Furthermore, a common genetic background has been found for all NREM parasomnias involving HLA DQB1*05:01 (Heidbreder et al. 2016). Additionally, the neuroanatomical substrate for the simultaneous co-existence of arousal originating from the motor and cingulate cortices and persistent sleep in associative cortical regions has been proposed, based on gray matter volume decline in the dorsal posterior and posterior midcingulate cortex reported in a 3 Tesla MRI study of 14 drug-free, PSG-confirmed adults with NREM parasomnia and 14 healthy controls matched for age and gender (Heidbreder et al. 2017). Finally, not only NREM parasomnias, but virtually all parasomnias provide compelling examples of the pervasive clinical phenomenon of dissociated states,

with multiple combinations of admixed states of being encompassing NREM sleep, REM sleep and wakefulness ((Mahowald and Schenck 1991; Mahowald and Schenck 1992; Mahowald et al. 2011a; Mahowald et al. 2011b).

Selected case series

In a study of 64 consecutive adult SW/ST patients, with mean age of 30 years, the patients were categorized by clinical history into three groups: serious violence during sleep to other people, or to property, or to self (n = 26); harmful, but not destructive behavior (n = 12); and nonviolent behavior (n = 26) (Moldofsky et al. 1995). Log linear analysis showed that a predisposing factor (childhood parasomnia and/or family history of parasomnia) and a stressor (psychologic distress, substance abuse, or sleep-wake schedule disorder) predicted the presence of SW, ST. Serious violent acts were more likely to occur with males (p < 0.004) who had a sleep-wake schedule disorder (p < 0.03). Both harmful and serious violent sleep behavior occurred with drug abuse (p < 0.009). In comparison to all other groups, those who were violent to other people were males who experienced more stressors (p < 0.02), drank excessive caffeinated beverages, abused drugs (p < 0.03), showed less N4 sleep (p < 0.02), and less alpha EEG activity in NREM sleep (p < 0.02). Being male and having < 2%N4 sleep had 89% sensitivity, 80% specificity and 81% diagnostic accuracy for individuals who were violent to others.

In another study on adults with SW that included data on sleep violence, among 63 SW adults, 45% had ≥1 episodes/month, 54% had partial recall of the episodes and 36% reported trigger factors for SW (Bargiotas et al. 2017). In this series, 73% reported childhood-onset SW and 27% adult-onset SW. Violence during SW episodes was more frequent in males and in subjects with childhood-onset SW (45% for self-injury and 44% for violent behavior towards others). Thus, adult SW was demonstrated to represent a complex and potentially dangerous condition.

Aggression and violent behaviors were found in a study on dream-enactment behavior during SW/ST (Uguccioni et al. 2013). The subjects in this study completed aggression, depression, and anxiety questionnaires. The dreaming (mentations) associated with SW/ST and RBD behaviors were collected over their lifetime and on the morning after vPSG. The dream reports were analyzed for complexity, length, content, setting, bizarreness, and threat. Of the 32 subjects with SW/ST, 91% recalled an enacted dream, virtually identical to 87% of 24 subjects with RBD who recalled an enacted dream, including 41 dreams recalled on the morning after the vPSG studies. The dreams of RBD subjects were more complex and less bizarre, and with a higher level of aggression, compared to the dreams of SW/ST subjects.

Another notable finding was the low levels of aggression, anxiety, and depression scores during the daytime in both the SW/ST and RBD groups, thus underscoring how aggression and violence during sleep can be solely sleep-state dependent (and generated) behaviors, and not an extension of waking mental status and aggressive predispositions. As many as 70% of enacted dreams in SW/ST and 60% in RBD involved a threat, but there were more misfortunes and disasters in the SW/ST dreams and more human and animal aggressions in the RBD dreams. The response to these threats differed, as the SW/ST subjects mostly fled from a disaster. However, 25% of the SW/ST subjects fought back when attacked and engaged in aggressive and at times violent behaviors. In contrast, 75% of RBD subjects counterattacked when assaulted. Finally, the dream setting included the bedroom in 42% of SW/ST dreams, with this setting being exceptionally rare in the RBD dreams.

Selected case reports

Various reported cases will now be summarized to illustrate some of the diverse clinical (and forensic) settings associated with SW/ST violence. In addition, a book of transcribed parasomnia patient interviews contains 25 NREM patient stories illustrating aggressive and violent behaviors in most of these cases, as reported by the patients and the spouses (Schenck 2005).

The most dramatic case, with forensic consequences that made Canadian legal history for being the first time the Sleepwalking Defense was successfully used in a murder trial, was the Ken Parks case in Toronto on the night of May 24, 1987 (Broughton et al. 1994). Parks drove 23 km to the home of his in-laws, where he proceeded to the bedroom of his sleeping in-laws and strangled his father-in-law to unconsciousness (he survived), and he beat his mother-in-law with a tire iron. Parks also stabbed them both with a large knife that he had taken from their kitchen. His mother-in-law was killed. Parks had the "perfect storm" of predisposing, priming and precipitating risk factors that resulted in the tragic episode. He had a prior history of SW, and during childhood he once nearly went out of a 6th floor window while asleep. He had always been a very deep sleeper. There was a very strong family history of NREM parasomnias, with 20 parasomnias, across 4 categories, affecting 10 first and second degree family members, including a grandfather with 4 persistent, active parasomnias (Schenck 2005). Parks had been severely sleep-deprived leading up to the tragic night, and he had major physical stress (playing rugby on a hot, humid afternoon hours before his episode), and he had major psychological stress (gambling addiction with major debt incurred that he had just revealed to his wife). Nevertheless, Parks had been referred to as "a gentle giant" by family and friends (he was 6'5" tall and weighed 280 pounds [127 kg]), as testified in court. Furthermore, the police from their observations described "dissociative analgesia" after the murder, as Parks showed no signs of pain despite having severed most of the tendons of both hands while bludgeoning his in-laws with the large kitchen knife. Dissociative analgesia is a common finding in SW. In one study, of 47 SW patients with at least one previous violent parasomnia episode, 79% had perceived no pain during episodes, allowing them to remain asleep despite injury (Lopez et al. 2015). Parks in a forensic setting clearly demonstrated this clinically finding. The not-guilty verdict was appealed by the Crown, without success. The Parks case has been extensively analyzed, with sleep medicine experts in agreement with the not-guilty verdict (Broughton et al. 1994; Schenck 2005).

Another case of NREM parasomnia with "sleep driving", a recognized rare complex SW behavior (Pressman 2011), associated with sleep violence has been reported as a strictly clinical case (Schenck and Mahowald 1995). A 43 year-old man with childhood-onset SW presented for sleep center evaluation on account of repeated sleep-related injuries incurred during violent nocturnal activity, which included frenzied running, throwing punches and wielding knives. He had also driven an automobile a long distance (8 km) during an apparent somnambulistic state and arrived at his parents' home (without any weapon) and pounded on their door, and they found him clad only in his underwear. He had bolted from bed, grabbed the car keys and smashed through a screen door at home on his way to the car. His wife had observed the following frequent SW behaviors that he demonstrated on many nights for years: stabbing at the furniture or the air with knives; swinging and throwing baseball bats; punching furniture and sustaining lacerations and fractures of his digits; running into doorways and furniture and sustaining ankle sprains and knee injuries; and various injuries from falling down the staircase. His wife had been repeatedly injured from being punched and elbowed, and she felt that her life was threatened by his nocturnal violence 2-3 times yearly, especially when she had to struggle to avoid strangulation. Most of these episodes, according to the wife, began with his "flying out of bed in a highly energized state." vPSG documented multiple episodes of complex and violent behaviors arising exclusively from N3 sleep, thus confirming the diagnosis of NREM parasomnia. Other causes of sleep-related violence were excluded, such as sleep-disordered breathing. The patient responded promptly to treatment with bedtime clonazepam, and benefit was maintained at 5-year follow-up.

Two cases of premenstrual sleep terrors and injurious SW have been reported (Schenck and Mahowald 1995a). A 17 year-old presented with a 6 year history of exclusively premenstrual ST and injurious SW that began one

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year after menarche. During the four nights before each menses, the following parasomnia sequence took place, beginning 30-120 min after sleep onset: on the first night, she would talk and shout; on the second night she would scream loudly; on the third and fourth nights, she would scream while engaged in agitated activity, such as frenzied SW, running, knocking furniture over, and attempting to go through windows, often sustaining ecchymoses and lacerations. She underwent two consecutive vPSG studies, beginning four nights before the predicted onset of her menses. On the second night, there were two episodes of complex behaviors arising abruptly from N3 sleep, thus confirming the diagnosis of NREM parasomnia. There was no psychiatric history. Bedtime pharmacotherapy was ineffective or could not be tolerated. However, bedtime self-hypnosis (after an instructional appointment) was beneficial, and at 2.5 year follow-up the patient reported only mild premenstrual ST occurring about three times yearly.

The second patient was 46 years old without psychiatric disorder (nor history of sexual or physical abuse) who presented with a 5 year history of ST and injurious SW that initially was not menses-related, but starting 8 months before referral had become exclusively a premenstrual parasomnia, occurring each month beginning 5-6 days before each menses. The episodes were witnessed by her husband who reported the following: she would scream loudly and bolt from bed, sustaining ecchymoses and lacerations. She recalled feeling the walls collapsing around her, or a sense of a threatening presence, at the onset of each episode. A vPSG study a week before her expected menses did not detect any episode, but there was an excessive amount of abrupt NREM sleep arousals (24/h) that were consistent with a NREM parasomnia. Treatment with bedtime self-hypnosis and 0.25 mg clonazepam was effective, with minor episodes recurring every few months.

Another case of sleep-related injury with NREM parasomnia resulted in acute PTSD affecting the spouse (Baran et al. 2003). Her 29 year-old husband had a severe ST episode when he suddenly aroused from sleep and jumped through a closed second-story window of their bedroom, sustaining major lacerations to his arms. He hung onto the roof as his wife screamed at him from the window, and he eventually climbed back inside. vPSG: confirmed the diagnosis of NREM parasomnia. His STs were effectively treated with behavioral and pharmacologic interventions. However, at a routine follow-up appointment, the wife's PTSD symptoms came to clinical attention, and she was referred for treatment, with marked improvement in her PTSD after an 8 week course of cognitive-behavioral therapy. This case is a striking example of how family members of patients with parasomnias can suffer psychological trauma even if they are not physically injured.

An unusual case of injurious NREM parasomnia associated with lancinating throat pain has been reported in a healthy 30 year-old married woman with childhood-onset, persistent, non-problematic SW (Buskova and Sonka 2014). Within two hours after sleep onset she would sit up in bed dreaming that she was swallowing indigestible objects, such as scissors, needles, worms, etc., which were accompanied by very unpleasant feelings and lancinating throat pain. She would then vigorously try to remove these objects from her throat with her hands, and in the process injured her throat, with pain lasting for weeks afterwards. She would be confused for several minutes and ask her husband to remove the objects from her throat. One episode posed a risk of harming her two year-old daughter who was sleeping with her parents that night. The patient forcibly opened the mouth of her daughter and tried to insert her fingers into the daughter's mouth, but her husband quickly intervened. She reported dreaming of her daughter swallowing a coin. vPSG documented seven spontaneous arousals from N3 sleep, without any behaviors. Medical, neurological and psychologic testing was normal. However, during seven sessions of psychotherapy, she revealed prior trauma involving sexual assault at age 7 years, with an object being forced into her mouth to prevent her from screaming during the assault. Psychotherapy was successful, as she had no parasomnia episode at 6 month follow-up.

REM sleep behavior disorder (RBD)

The typical clinical profile of chronic RBD consists of middle-aged or older men with aggressive dream-enacting behaviors that cause repeated injury to themselves and/or their wives. This profile was demonstrated in the first two large published series on RBD, involving 96 and 93 patients, respectively (Schenck et al. 1993; Olson et al. 2000). In these two series, male predominance was 87.5 and 87%, mean age at RBD onset was 52 years and 61 years, dream-enacting behaviors were reported in 87 and 93% of patients, and sleep-related injury as the chief complaint was reported in 79 and 97% of patients, respectively. Injuries included ecchymoses, subdural hematomas, lacerations (arteries, nerves, tendons), fractures (including high cervical-C2), dislocations, abrasions/rug burns, tooth chipping, and hair pulling. RBD causing subdural hematomas has been reported in 5 additional cases (Gross 1992; Dyken et al. 1995; McCarter et al. 2014; Ramos-Campoy et al. 2017).

A review of the published cases of RBD that were associated with potentially lethal behaviors identified choking/headlock in 22–24 patients, diving from bed in 10 patients, defenestration/near-defenestration in 7 patients, and punching a pregnant bed partner in 2 patients (Schenck et al. 2009). The concept of "victim vulnerability factors" for increasing the risk morbidity and mortality from vigorous RBD behaviors was discussed extensively. A

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"spectrum of vulnerability" was formulated for RBD (and other parasomnias) whereby at one end of the spectrum is the degree of vigor and violence of the RBD behavior, and at the other end of the spectrum is the degree of medical vulnerability of the victim (patient or spouse). Furthermore, the fact that the patient or bed partner is asleep, and in which sleep stage (e.g., REM sleep with generalized muscle paralysis in the bed partner, or slow-wave NREM sleep in the bed partner predisposing to an agitated and violent confusional arousal induced by a RBD episode), or if the bed partner suffers from a sleep disorder predisposing to abnormal and potentially violent arousals (e.g., OSA; sleep inertia; confusional arousals, ST, SW) would add an additional sleep-related vulnerability risk factor. The circumstances of the sleeping environment may also confer additional vulnerability. Some of the medical factors that can increase the morbidity and mortality risk from RBD behaviors include: pregnancy; deafness; blindness; osteopenia, osteoporosis; bleeding disorder, anticoagulant therapy, status postsurgical procedure, spinal-vertebral disorder; and various advanced age-related vulnerabilities.

A study was reported on factors associated with injury in RBD (McCarter et al. 2014). Among 53 patients, median age was 69 years, 73% were men, and 55% had idiopathic RBD (iRBD). Injuries were reported by 55%, including 38% to self, 17% to the bed partner, and 11% had severe injuries requiring medical intervention or hospitalization, including two (4%) with subdural hematomas. iRBD diagnosis and dream recall were significantly associated with injury; and iRBD diagnosis was independently associated with injury and injury severity. Falls were also significantly associated with injury severity. The frequency of dream-enacting episodes did not predict RBD-related injuries, calling attention to the importance of prompt initiation of treatment for RBD in patients having even rare episodes of dream-enactment. Therefore, injuries appear to be a frequent complication of RBD, with one in nine patients suffered injury requiring medical intervention.

The experiences of the initial series of RBD patients and their spouses presenting to the Minnesota Regional Sleep Disorders Center, where RBD was formally identified (Schenck et al. 1986; Schenck et al. 1987), were captured by audiotaped interviews (with signed permissions) that were transcribed and edited, and then published in a book (Schenck 2005). These patients with RBD had been married for decades before the onset of RBD, and so the spouses knew that the onset in later life of sleep violence did not reflect any waking personality disturbance with propensity for irritability and violence. This is probably the main reason for having only two published cases of divorce (Ingravallo et al. 2010; Zhou et al. 2017) and one published case of marital discord (Yeh and Schenck 2004) related to RBD. Patient and spouse dialogues, with

Table 2 Sample dialogues of men with rbd and their wives^a

57 Year Old Man with RBD and Wife:

"It seems like I am extra strong when I sleep."-man

"It almost seems like a force picks him up."-wife

"He is sleeping and his body is in motion."-wife.

"I don't think he ever could hit as hard while awake as he hits during sleep. A year ago he punched right through a wall board in our bedroom at our lake cabin."—wife.

"Oh yes, there were always bloody sheets." wife

67 Year Old Man with RBD and Wife:

"It's amazing. You should see the energy behind that activity. Oh, it's so unreal."—wife.

"He pounded my head one night and my head still hurt for another 2 weeks"—wife

"His legs go fast, just like he's running."—wife.

"We've put as much distance between us in bed as we can."—wife.

"I didn't really sleep soundly until he got up in the morning."—wife.

65 Year Old Man with RBD and Wife:

"I was wrestling someone and I had her by the head. What scares me is what a catastrophe that would be to wake up and find that I had broken her by the neck."—man

"This went on for 3 years, and then I retired—but nothing changed afterwards whatsoever."---man

"What happens to people like my husband who don't get diagnosed? Do they kill their wives in these experiences? Do we know?"—wife.

70 Year Old Man with RBD and Wife:

"I didn't remember the dream because I knocked myself out."--man

"The next morning I asked her what I had done, and she told me I had beat her."—man

"It was hard for me to sleep, because I never knew when I was going to get hit."—wife.

"When all this started, I figured it was part of getting old, part of being normal, I guess."--wife

75 Year Old Man with RBD and Wife:

"I just started kicking—the big, faceless, shapeless figures were still there. And my wife was afraid for herself, the dog, and for me."--man

"I told him I'd have a Devil of a time explaining how I got a broken arm in bed with both of us asleep."—wife

"When a man his size comes down on that floor, honestly, it's a miracle he has not broken a hip or a shoulder."—wife.

^aAdapted from Schenck 2005

comments on the imminent dangers posed by RBD, are contained in Tables 2, 3.

Violent RBD carries an increased forensic risk, including both inadvertent death from "Parasomnia Pseudo-Suicide" (Mahowald et al. 2003), and inadvertent homicide (Mahowald et al. 1990).

Parasomnia overlap disorder (POD)

POD was formally described in 1997 with a series of 33 cases of RBD combined with a disorder of arousal from NREM sleep (confusional arousals, SW, STs) that

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Table 3 Comments by patients and spouses on RBD behaviors causing imminent danger^a

1. Comments by RBD Patients:

"I ran right smack into the wall, an animal was chasing me. I think it was a big black dog." (p. 157)

"I thought I was wrestling someone and I had her by the head." (p. 136)

"Pounding through the curlers into her head." (p. 157)

"What scares me is what a catastrophe that would be to wake up and find that I had broken her neck." (p. 137)

"I have hit her in the back too, and she has had a couple of (vertebral) disc operations." (p. 143)

"One night I woke up as I was beating the hell out of her pillow... that's when I realized that I had a problem." (p. 106)

"Just recently, I rammed into her pelvis with my head...during a dream." (p. 93)

2. Comments by the Wives

"It's amazing. You should see the energy behind that activity, oh, it's unreal."(p. 107)

"He literally just kind of flew out of bed and landed on the floor with tremendous strength" (p. 53)

"It almost seems like a force picks him up." (p. 130)

"His legs go so fast, just like he's running" (p. 155)

"It is his kicking, violent kicking, his feet are just like giant hammers when they hit you over and over again." (p. 73)

"I felt that kick on the ankle for two months afterwards." (p. 82)

"That's the reason we got the waterbed—because he was wrecking his hands on the wooden bed." (p. 111)

"Oh, yes, there were always bloody sheets." (p. 105)

"Roaring like a wounded wild animal: he roared, he crouched, he punched." (p. 75)

emerged idiopathically or symptomatically with neurological and other disorders (Schenck et al. 1997). The presenting complaint was sleep-related injury; mean age was 34 + 14 years, and mean age of parasomnia onset was 15 + 16 years (range 1-66); 70% were males. An idiopathic subgroup (n = 22) had a significantly earlier mean age of parasomnia onset (9 + 7 years) than a symptomatic subgroup (n = 11) (27 + 23 years). The clinical diagnosis was confirmed by vPSG that documented both RBD and a NREM parasomnia. Treatment outcome was available for 20 patients, and 90% (n = 18) reported substantial control with bedtime clonazepam (n = 13), other medications (n = 4), or hypnosis (n = 1). Thus, POD was found to be a treatable condition that emerges either idiopathically or with various clinical disorders. Although POD is classified as a subtype of RBD in the International Classification of Sleep Disorders, 3rd edition (American Academy of Sleep Medicine 2014), diagnostic criteria for both RBD and a NREM parasomnia must be met in order to diagnose POD. In the first reported series on POD (Schenck et al. 1997), a literature review had also identified two prior cases of violent POD (1): (i) a 51 year-old man with Machado-Joseph disease (spinal-cerebellar-ataxia type-3 [SCA-3]), who had episodes of prolonged nocturnal wandering that were often violent, which had begun 15 years prior to the diagnosis of SCA-3. vPSG demonstrated multiple behavioral events from NREM sleep and during REM sleep together with REM-without-atonia (Kushida et al. 1995). (ii) a 49 year-old man with a 23 year history of nightly violent sleep-related episodes had vPSG confirmation of POD; bedtime clonazepam therapy was immediately effective in controlling the parasomnia, as confirmed by the patient's wife and by follow-up vPSG two months later (Bokey 1993). Since 1997 the literature on POD has grown substantially, with additional categories of NREM parasomnias linked with RBD, and with additional types and central nervous system locations of symptomatic cases being identified, and with additional cases involving 5 or 6 total parasomnias affecting individual patients. Many of these additional reports have included cases of aggressive and violent POD, as reviewed (Schenck and Howell 2013; Schenck and Howell 2018). One notable case involved a 60 year-old woman who presented with VBS that were documented to be caused by RBD, but also during her vPSG she had an episode of sleep masturbation arising from N3 sleep, and so her POD involved aggressive/violent (RBD) behavior along with appetitive behaviors (sexsomnia, and sleep related eating disorder) (Cicolin et al. 2011).

In the evolution of POD over the life cycle, it appears that the NREM parasomnia component may predominate in the earlier stages, with a transition (of variable duration) to more predominant RBD in the later stages. However, this topic needs further investigation. Nevertheless, POD is a prime example of generalized sleep motor dyscontrol, associated with disturbed dreaming.

Obstructive sleep apnea (OSA)

The associations between OSA and violent parasomnias include "OSA Pseudo-RBD" and OSA-induced confusional arousals and SW, i.e. NREM parasomnias.

"OSA Pseudo-RBD"

In a carefully conducted study on "OSA Pseudo-RBD", 16 patients (11 men), with mean age of 59.6 (± SD 7.7) years had presented with complaints of snoring, excessive daytime sleepiness (EDS), and abnormal nocturnal sleep behaviors that were often injurious dream-enacting behaviors associated with disturbed dreams that strongly suggested RBD (Iranzo and Santamaria 2005a, 2005b). Two control groups consisted of 16 patients with idiopathic RBD with an apnea/hypopnea index < 10, and 20 healthy controls. vPSG findings were diagnostic for severe OSA/hypopnea, with a mean apnea-hypnopnea index of 67.5 (range, 41–105). Parasomnia behaviors,

^aAdapted from Schenck 2005

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including dream-enacting behaviors, occurred only during apnea/hypopnea-induced arousals from REM and NREM sleep. REM sleep electromyography (EMG) was normal, with absence of increased muscle tone and increased phasic twitching, as found in RBD. The most frequent behaviors observed on vPSG included kicking, gesturing, raising the arms, and talking. In 54% of patients the OSA-induced arousal parasomnia behaviors occurred from both REM and NREM sleep, and in 46% of patients the OSA-induced arousal parasomnia behaviors occurred only from REM sleep. Nasal CPAP therapy in 13 of 16 treated patients eliminated snoring, daytime somnolence, unpleasant dreams, and parasomnia behaviors, according to the patients and their spouses. Repeat vPSG with nCPAP therapy confirmed that the apneas and hypopneas had been eliminated, and normal oxygen hemoglobin saturation levels were documented. The EMG during REM sleep remained normal, once again excluding the presence of RBD. Therefore, severe OSAH may mimic the symptoms of RBD, and so vPSG monitoring is mandatory to establish the diagnosis of RBD, and to either identify or exclude other causes of dream-enacting behaviors.

OSA-induced NREM parasomnias

A case of violent parasomnia triggered by OSA was reported in a 54 year old female with no history of parasomnia up until 5 years previously when she engaged in complex behaviors during both nocturnal sleep and daytime naps, including episodes of "sleep-driving" from naps approximately 5 times monthly (Lateef et al. 2005). She would go SW barefoot in the snow. Once she was found by police wandering in a nearby town. The most disturbing incident was when she chopped up her cat on a cutting board in the kitchen, and then awakened at 6 a.m. with her hands covered in blood and found the cat's remains next to the trash can. During this 5 year time period, her sleep history was positive for loud snoring, non-restorative sleep, daytime somnolence, and weight gain. Overnight vPSG documented severe OSA with marked oxygen desaturation that was controlled with nasal CPAP. At 4 month follow-up the patient reported no parasomnia recurrence.

In another case, a 55 year-old morbidly obese man with documented OSA was experiencing progressive cognitive and psychological deterioration due to suboptimal treatment of his OSA with nasal CPAP (Baron and Auckley 2005). One night, the patient reached for his bilevel positive airway pressure mask, but accidentally picked up his pistol (kept by his bedside for self-protection) along with the mask and straps. In his confused state, he was unaware that he had a gun in his hand, and attempted to pull the straps of the mask over his head. In the process, he

accidentally fired the pistol, inflicting a tangential gunshot wound to his parietal scalp.

A forensic case involved a 37-year-old man with severe OSA (AHI, 124/h; SaO2 low 80% range, and nadir of 63%) who one night fatally shot his wife (Nofzinger and Wettstein 1995). At the trial he invoked an "OSA defense", but was found guilty of first-degree murder, despite two (of three) expert witnesses testifying that the OSA was severe enough to have induced a confusional arousal with violent behavior during the night in question. Pertinent medical-legal aspects of OSA, confusional arousals, and other clinical information were discussed in this report.

NREM parasomnia/severe OSA violent parasomnia with biting

A unique case was recently reported of a chronic, violent NREM parasomnia that was associated with severe OSA aggravating the parasomnia, which included recurrent biting of the same index finger that caused major injury requiring surgical interventions (Danish et al. 2018). Combined bedtime clonazepam therapy and control of the severe OSA with nCPAP was needed to control the violent parasomnia. The case involved a 55-year-old, single, obese male, with a BMI of 41 who had presented with a 20-year parasomnia history in which he would wake up in the first 2 to 3 h 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 and once stepped on a glass table and broke it. He had sustained injuries during these episodes. 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. He was referred to a psychiatrist who prescribed clonazepam 1 mg at bedtime, which stopped the nocturnal events. He was then referred to a sleep physician, and underwent vPSG while on clonazepam, which revealed severe OSA, with an AHI of 39/h. He was titrated on bilevel positive airway pressure (BPAP) at 16/12 cm H₂O, and had a residual AHI of 4.4 events/hr. and a minimum SaO2 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,

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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. He was not using BPAP on that night. He consulted the plastic surgery department for wound care. He then presented to an author's 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 vPSG with seizure montage took place in November 2017. He was taken off the clonazepam 2 days prior to the video-PSG. There was no EEG epileptiform activity, nor any confusional arousals from NREM sleep. PLM index was 7/h. Nasal CPAP with 12 cm H2O, which was used throughout the vPSG, was effective in eliminating sleep-disordered breathing, with an overall AHI of 2.5 events/hr. Psychiatric history was positive for a brief depression in 2005.

In this report (Danish et al. 2018), the differential diagnosis of sleep-related biting was presented and discussed, with citations from the pertinent literature. Table 4 lists the differential diagnosis of sleep-related biting. In regards to RBD, in a series of 203 idiopathic RBD patients, the prevalence of biting in RBD was 8.4%, which usually involved bed partners (Fernández-Arcos et al. 2016). There have been two published cases involving biting during RBD episodes, with the first case being an extraordinary case of a 63 year old man whose four consecutive wives had divorced him because of his aggressive and violent dream-enacting behaviors, including repeated biting (Zhou et al. 2017). With his first wife, one night he dreamed that he was eating an apple, but instead he was biting her ear. On subsequent nights, during similar dreams he would bite her ears, nose and face, which culminated with his wife divorcing him after four years of marriage. His 3 next marriages were also terminated by the wives on account of his repeated RBD-related sleep violence, including aggressive biting

Table 4 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

during dreams. The second case of RBD with biting involved duloxetine-induced RBD in a 62 year-old woman who one night dreamed of biting something, but she was actually biting the hand of her grandson (Tan et al. 2017).

Severe OSA/PLMD violent parasomnia

A case has been reported of a 75 year-old man firing a loaded gun during sleep who had previously undiagnosed severe OSA and PLMD, and other multiple risk factors for sleep violence (Ingravallo et al. 2018). Although this was a clinical case, there were forensic implications. This man was employed and was a hunter/firearms collector, married for 32 years, with no prior criminal or psychiatric history, who presented to a sleep center reporting to have fired a shot in his bedroom during sleep while his wife was away, without memory of hearing the gunshot. The patient had a normal day before the event, apart from major concern about recent nearby burglaries that prompted his sleeping with a loaded gun placed behind his bed. Since on the night of the episode, his wife was not at home, he decided to put a loaded gun on the shelf behind the bed for security for when he was asleep. The next morning, he found a dark gunpowder stain around a bullet hole on the bed where his wife usually slept. He had no recall of causing the gunshot nor of the noise from the explosion. Also, the gun had been placed back behind the bed after discharge.

The patient and his wife had been unaware of any sleep problems, apart from his mild daytime sleepiness and rare episodes of minor motor activity (sleeptalking and kicking). There was no alcohol or drug abuse history. He drank a half-bottle of beer at 8 p.m. on the evening of firing the gunshot during sleep. He had had his usual one expresso the prior morning. The patient had an implantable cardioverter defibrillator since 2011, and hypertension was treated with a calcium channel antagonist. He had mild obesity (BMI 31.6). Family history was negative for sleep disorders. Medical and neurological exams were normal, as was a CT scan of the brain.

At vPSG, sleep structure was markedly disrupted, with 96 brief awakenings, an elevated arousal index of 25/h, and poor sleep efficiency of 67%. Sleep structure was markedly disrupted with only one clear sleep cycle with REM sleep that had preserved REM-atonia. Severe OSA and PLMs were documented (AHI = 60/h, with average SaO2 = 93; PLM index = 38.7/h). Brief abnormal movements from REM sleep without apparent precipitant were recorded. CPAP therapy was effective, with a residual AHI of 4.5. The patient and his wife did not report any subsequent abnormal behaviors during sleep at 6-month follow-up. The patient had been immediately advised to keep all weapons out of his bedroom.

This was the first reported case of a near-miss gunshot discharge during sleep, with potential dramatic clinical Schenck Sleep Science and Practice (2019) 3:2 Page 10 of 14

consequences (he could have inadvertently killed his wife if she were sleeping with him that night), and with forensic implications. This patient had a "perfect storm" of sleep and psychological risk factors that converged to strongly promote precipitous arousals with sleep-related violence. He was mentally primed (and perhaps even expecting) to arouse from sleep on the night of the episode. At the point of falling asleep, his mind (and then presumably his sleeping mind) knew about the potential danger surrounding him, because of recent nearby burglaries, while he slept alone; his mind knew the location of his loaded gun behind the bed, and his mind knew about the perhaps likely need for rapid use of the gun for self-protection upon suddenly arousing from sleep in response to immediate danger. So, the patient was psychologically hyper-primed to arouse from sleep to use a loaded gun for self-protection. Superimposed on this sleep-related mental set was previously undiagnosed severe organic sleep pathology with multiple precipitous arousals from high frequency obstructive apneas and high-frequency PLMs, which have been documented in the literature to be associated with complex parasomnia episodes (Iranzo and Santamaria 2005a, 2005b; Gaig et al. 2017).

The patient's heightened mental and physiological state for abrupt arousals could have resulted in either a spontaneous arousal or an arousal triggered by OSA, PLM, or environmental noise that was misinterpreted (given his pre-established mind set) as the sound of an intruder in his bedroom. Regardless of the proximate cause of arousal, he was primed to use his gun for self-defense. The complex clinical scenario of this case calls attention to Pressman's review on factors that pre-dispose, prime, and precipitate NREM parasomnias (Pressman 2007a).

Another geriatric case of sleep violence associated with combined severe OSA (O2 nadirs 80%) and severe PLMs (index, 112/h), and with preserved REM-atonia, was reported in a 79-year-old man with a 2-year history of injurious dream-enacting behaviors (Schenck et al. 1989a, 1989b).

Periodic limb movement disorder (PLMD) Pseudo RBD

A carefully documented case series established PLMD as another aggressive and injurious dream-enacting disorder mimicking RBD, and reinforcing how vPSG is mandatory not only for diagnosing RBD, but also for identifying or excluding other disorders mimicking RBD (Gaig et al. 2017). This series comprised 15 men and 2 women, with median age of 66 years (range, 48–77). Kicking during sleep was reported in all 17 patients, punching was reported in 16 patients, assaulting the bed partner in 2 patients, falling out of bed in 5 patients, and shouting in 10 patients. Injuries were sustained in 3 bed partners and in one patient.

vPSG documented frequent, vigorous PLMs of the lower limbs, upper limbs, and trunk. The median PLM index was 61/h, with the median PLM index during NREM sleep being 62/h, and the median PLM index during REM sleep being 39/h. Abnormal behaviors (e.g., punching, groaning) occurred immediately after some of the arousals in 71% (12/17) of patients. RBD/REM-without-atonia/OSA were excluded.

Bedtime dopaminergic therapy, prescribed in 14/17 patients, was fully effective in all 14 treated patients, with control of both the abnormal sleep behaviors and unpleasant dreams—as is also true in the successful treatment of RBD and OSA Pseudo-RBD. Follow-up vPSG in 7/14 treated patients documented a robust decrease in the median PLM index from baseline (109 vs. 19, p = .002) and absence of abnormal behaviors during the arousals.

The reports just discussed on OSA Pseudo-RBD and PLMD Pseudo-RBD as being predominantly older male conditions with VBS and dream-enacting behaviors, when considered in the context of RBD with VBS as being a predominantly older male condition with dream-enacting behaviors, suggests an underlying vulnerability of older men to VBS and dream-enacting behaviors across at least 3 different sleep-related diagnostic categories.

Sexsomnia

Sleep related abnormal sexual behaviors (sexsomnia, sleepsex) are officially classified primarily as a subtype of Confusional Arousals, and also SW, i.e. NREM parasomnias (American Academy of Sleep Medicine 2014). In the first classification of sleep related disorders and abnormal sexual behaviors and experiences, 31 published cases of sexsomnia (as a parasomnia) were identified, with a striking clinical profile consisting of male predominance (81%), chronicity, with a mean duration of over 9 years, and subsequent amnesia for the sexsomnia in all patients (Schenck et al. 2007). A full range of sexuality was expressed, including masturbation, sexual fondling, sexual intercourse, sexual vocalization/ shouting, and sexual assault. Agitated/assaultive behavior was reported in 45% of cases, and legal consequences in 35% of cases. Bed partners often experienced physical injuries (ecchymoses, lacerations) from the sexual assaults, and to a lesser extent the patients were also physically injured (bruised penis; fractured digits). vPSG helped identify the underlying diagnosis for the sexsomnia which was a NREM parasomnia in 90% of cases (confusional arousals, n = 26; SW, n = 2), and comorbidity with OSA as a sexsomnia promoter was found in 4 cases. A sexsomnia update on the cumulative world literature of 49 cases has been published (Schenck 2015), with similar findings, including 7 cases with OSA promoting the sexsomnia.

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In the largest published case series on sleep related violence evaluated as sleep forensics referrals to a single center over a period of 11 years, sexual assault was the most common criminal allegation among the 351 referrals, accounting for 41% (145/351) of the cases (Cramer Bornemann et al. 2018). Of the 351 referrals 110 were accepted following thorough case review. In general, cases not accepted were declined on the basis of little or no merit or contamination by alcohol intoxication. Sexual assault comprised 52 of the 110 accepted cases (47%). Of those cases accepted, the proposed initial claim that a sleep phenomenon was operant was supported in approximately 50% of cases, mostly NREM parasomnia.

Sleep related dissociative disorder

As described above, 7 of the 100 consecutive patients with sleep related injury were diagnosed with Sleep Related Dissociative Disorder, which is considered to be a psychiatric parasomnia (Schenck et al. 1989a, 1989b). With one exception, all patients were female and had childhood and subsequent histories of sexual, physical and emotional abuse, often occurring at night around bedtime. There was frequent self-mutilation with lit cigarettes and knives, including genital mutilation, cutting one's tongue and various other body parts, shaving one's head, and sustaining a variety of recurrent injuries from agitated behavior with running into furniture, and crashing through glass windows or doors, jumping from bed, and sustaining ecchymoses, fractures, and lacerations requiring stitches. vPSG can document the diagnostic finding of behaviors emerging during sustained EEG wakefulness during sleep-wake transitions, or from N1 or N2 sleep (Schenck et al. 1989b). Often there is a 30-60 s lag between the start of EEG wakefulness and the start of the dissociative behavior, which is in stark contrast to a NREM parasomnia episodes that emerges abruptly with an arousal, and usually from N3 sleep. However, positive findings may not be detected during all vPSG studies, and so Sleep Related Dissociative Disorder is often diagnosed by clinical history, and with the vPSG excluding other disorders causing sleep related injury, although some patients can have mixed disorders that pose a therapeutic challenge.

Trauma-associated sleep disorder (TSD)

A proposed novel parasomnia encompassing features of RBD (viz. modest REM-without-atonia) with nightmares and aggressive/violent sleep behaviors (Mysliwiec et al. 2014). Four male soldiers, 22–39 years old, were reported with TSD, who had no prior parasomnia history. Three soldiers developed TSD from their active duty traumatic experiences, and one soldier from a relationship breakup. It is most likely the TSD represents a form

of "REM sleep arousal disorder" triggered by nightmares and immediate post-arousal violent dream-enacting behaviors. This perspective is shared in a subsequent report by the authors of the original TSD paper: "Hyperarousal, as opposed to neurodegenerative changes in RBD, is a component of TSD that likely contributes to overriding atonia during REM sleep and the comorbid diagnosis of insomnia" (Mysliwiec et al. 2018). The authors propose that TSD incorporates an inciting traumatic experience and clinical features of trauma-related nightmares and disruptive nocturnal behaviors as a novel parasomnia.

A critical review of TSD, with discussion as to whether or not it represents a truly unique condition has recently been published (Rachakonda et al. 2018). The authors describe the overlapping features and differences between TSD, PTSD and RBD. A major identified difference between TSD and PTSD is the presence of symptoms during wakefulness in the latter. The authors call for additional research, with the use of TSD diagnostic criteria in diverse clinical populations.

PTSD with self-inflicted gunshot wound during a nightmare

A 45 year-old Vietnam War combat veteran presented to an Emergency Room on account of an unintentional self-inflicted gunshot wound (Coy 1996). He had awakened from a nightmare involving Viet Cong soldiers and when he got out of bed he thought he saw the enemy soldiers in trees outside his house and also in his home. He loaded his rifle and roamed his house looking for the enemy soldiers, but tripped over furniture, and when he fell the weapon was discharged and the bullet struck his foot.

Nocturnal scratching disorder

A case series was reported on two females and one male who presented to a sleep disorders center with complaints of injurious and exclusively nocturnal scratching that was the only parasomnia complaint in one patient, and was part of a high frequency parasomnia symptom complex in two patients (Schenck and Mahowald 2007). The patients underwent extensive evaluations, including overnight, hospital-based, vPSGs with seizure montage and fast paper speeds. Neither clinical nor EEG seizure-like activity, sleep disordered breathing or PLMs was found in any patient. The hourly spontaneous arousal frequency ranged from 25/h to 34/h.

Case vignettes from this series

A 28 year-old married Caucasian man reported a 1.5 year history of nightly, exclusively perianal scratching with excoriation and bleeding, and he demonstrated perianal scratching throughout NREM sleep during his vPSG study. Multiple evaluations by various specialists had not detected

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parasites, colorectal problems, dermatologic or psychiatric disorders. Hypnotherapy, corticosteroid creams, clomipramine and antihistamines were ineffective. However, 0.5 mg clonazepam and 20 mg paroxetine therapy resulted in about 50% improvement.

A 26 year-old African-American single female, with a male bedpartner, reported longstanding nocturnal scratching and other parasomnia behaviors (SW, sleep related eating) developed keloids from the vigorous scratching of her shoulders, back and buttocks. vPSG findings were unremarkable. Bedtime therapy with clonazepam, 0.5 mg, fully controlled the nocturnal scratching and other parasomnia behaviors.

A 50 year-old single Caucasian female reported a 15 year history of injurious nocturnal scratching (with excoriation and bleeding), bruxism and sleep terrors. vPSG study revealed 55% sleep efficiency, but no behaviors. Treatment outcome was not available.

The presumptive diagnosis for the abnormal nocturnal scratching in all three patients was confusional arousals manifesting as a high-frequency NREM parasomnia that was either an exclusive symptom or part of a NREM parasomnia symptoms complex. Perianal scratching can be the sole nocturnal (injurious) scratching behavior.

Miscellaneous disorders

Rhythmic movement disorders can at times result in recurrent injuries, especially head banging (jactation capitis nocturna) (Whyte et al. 1991). Sleep related eating disorder can be associated with injurious behaviors involving ecchymoses from running into walls or furniture on the way to the kitchen; lacerating digits while cutting food; impulsively consuming or spilling very hot beverages or oatmeal, with scalding injuries; and internal injury from consuming toxic substances, e.g. ammonia compounds (Schenck et al. 1991). Nocturnal seizures can result in sleep related injuries (Schenck and Mahowald 2002). Insulinoma, caused by a rare endocrine tumor, can manifest with agitated nocturnal behaviors (extending to the early morning hours) with injury, as reported in a series of 3 patients (Suzuki et al. 2015). These patients had been treated with anticonvulsants for presumptive epilepsy, without benefit. vPSG was negative for any REM-without-atonia or any other sleep pathology. The authors recommended obtaining overnight and early morning blood sugar levels in patients with enigmatic agitated nocturnal behaviors.

Conclusion

SRV is a common phenomenon with a complex set of determinants and precipitants (Siclari et al. 2010) that span across a wide spectrum of sleep disorders, particularly the parasomnias, with forensic consequences (Cramer Bornemann et al. 2019; Ingravallo et al. 2014;

Mahowald & Schenck 1995). The entire human life span, and all of sleep, carry a risk for SRV. The association of SRV with appetitive parasomnias (sexsomnia, sleep related eating disorder), along with confusional and exploratory behaviors in POD is intriguing, and merits further research. The crucial role played by experienced sleep clinicians in evaluating parasomnia cases at accredited sleep disorders centers with hospital-based extensive vPG should be recognized for both clinical purposes (including the initiation of proper and usually effective therapy), and for forensic cases. A biopsychosocial model for helping understand violence with SW (Stallman and Bari 2017) can also be applied and tested for its utility across the spectrum of disorders causing SRV.

Abbreviations

BIPAP: Bilevel positive airway pressure; CPAP: Continuous positive airway pressure; ICU: Intensive care unit; iRBD: Idiopathic RBD; NREM: Non-REM; OSA: Obstructive sleep apnea; PTSD: Post-traumatic sleep disorder; RBD: REM sleep behavior disorder; SRV: Sleep related violence; ST: Sleep terrors; SW: Sleepwalking; TSD: Trauma-associated sleep disorder; vPSG: Videopolysomnography

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