

Sexual Behavior in Sleep, Sleepwalking and Possible REM Behavior Disorder: A Case Report

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Seven cases of sexual behavior during sleep (SBS) have been recently reported. The subjects had histories of behavioral parasomnias as well as positive family histories of parasomnia. A 27 year-old man with a history of sexual behavior during sleep was reported. His sleep history disclosed sleepwalking (SW) since 9 years of age. He also developed episodes of highly disruptive and violent nocturnal behavior with dream enactment at age 20 years, which often resulted in physical injuries either to himself or his wife and infant. His wife also reported episodes of amnesic sexual behavior that began 4 years before referral. During the episodes, the patient typically procured his wife, achieving complete sexual intercourse with total amnesia. Physical and neurological diagnostic workups were unremarkable. Family history disclosed sleepwalking in his brother. He was put on 2mg/day of bedtime clonazepam with a remarkable clinical improvement. This case involves either the combination of violent and non-violent sleepwalking with SBS, or the superimposition of presumed REM sleep behavior disorder (RBD) on top of preexisting SW in a man who also developed SBS in adulthood. Thus, this is a case report of probable parasomnia overlap syndrome.

CURRENT CLAIM: The subject of this report was a young male with probable parasomnia overlap syndrome: a combination of violent and non-violent sleepwalking with sexual behavior during sleep (SBS), with the superimposition of presumed REM sleep behavior disorder with preexisting sleepwalking, who also developed SBS in adulthood.

Motor parasomnias are complex motor behaviors occurring during non-REM sleep, REM sleep, or transitional states. They are classified as either arousal disorders, sleep-wake transition disorders, parasomnias associated with REM sleep or other parasomnias (ICSD, 1997). Sleepwalking is one of the most frequent parasomnias occurring during NREM sleep, with variable degrees of complexity and duration. It is more prevalent in childhood, usually peaking at 11 to 12 years of age. REM sleep behavior disorder (RBD) is a recently described parasomnic disorder and is characterized by vigorous motor activity which occurs exclusively during REM sleep; it consists of often injurious dream-enactment motor activity associated with vivid dreaming. Chronic RBD is most common in older men with a mean age of onset at around 48 years; in some cases there is a familial predisposition. Sleep studies should disclose atypical REM sleep with intermittent loss of the typical REM atonia.

More recently, a subgroup of RBD patients with non-REM parasomnias (sleepwalking and sleep terrors) were described and named as Parasomnia Overlap Syndrome (POS) (Schenck and Mahowald, 1996; Schenck et al., 1997). In those studies, a group of patients with a history of RBD, sleepwalking and sleep terror with good responses to benzodiazepines were reported.

Seven consecutive cases of patients with amnesic engagement in sexual behavior while asleep, which was called "sexual behavior during sleep" (SBS), have recently been reported. Age of onset was early adulthood and some subjects also had histories of behavioral parasomnias and positive

family histories of parasomnia. Shapiro et al. (1996) proposed SBS to be considered a new parasomnia.

We now report a case of a 27-year-old man with a history of sexual behavior during sleep. His personal sleep history disclosed sleepwalking (SW) since 9 years of age, which used to occur once or twice a month. He also developed episodes of highly disruptive and violent nocturnal behaviors with dream enactment at age 20 years, which resulted in physical injuries to himself (broken leg), his wife and their infant child. These disruptive episodes typically occurred once a week. His wife also reported episodes of amnesic sexual behavior that began 4 years before referral. During the episodes, the patient typically procured his wife, achieving complete sexual intercourse with total amnesia. Episodes of sexual behavior during sleep occurred once a month. His wife remained in bed with him after the episodes. The patient did not consent to being videotaped. He denied use of any medication, but his wife observed that alcohol use could possibly precipitate some episodes (he had the habit of drinking only on weekends). A family history revealed that sleepwalking was present in his brother. There was no history of psychiatric disorders and no report of marital discord. There was no family history of mental disorders. He scored 4 points in the Epworth Sleepiness Scale.

METHODS

Physical and neurological diagnostic workups, including EEG, were unremarkable. A hospital-based overnight sleep study was carried out.

RESULTS

Physical and neurological diagnostic workups, including EEG, were unremarkable. A hospital-based overnight sleep study showed the following features—total sleep time: 432 min; sleep efficiency index: 0.92; sleep latency: 13.9 min; REM latency: 68 min; stage 1: 2.1%; stage 2: 53.7%; stage 3: 3.5%; stage 4: 3.1%; REM sleep: 37.6%. No tonic or phasic electromyographic abnormalities were recorded during REM sleep. There were no rapid stage shifting or frequent arousals and neither apnea nor periodic limb movement events were recorded. The percentage of REM sleep was increased and REM latency was reduced, as was delta sleep percentage. There were no other clinical parasomnia behaviors documented during NREM-REM sleep.

The patient was put on 2mg/day of bedtime clonazepam which produced a remarkable clinical improvement of the disruptive episodes and of the sexual behavior in sleep. A presumptive clinical diagnosis of overlap syndrome was assigned on the basis of his clinical and polysomnographic data.

DISCUSSION

This case involves either the combination of violent sleepwalking (beginning at age 20 years) and non-violent sleepwalking (beginning at age 9 years) with SBS, or the superimposition of presumed REM sleep behavior disorder (RBD) on top of preexisting SW, in a man who also developed SBS. It is possible that additional PSG studies would have documented motor abnormalities during REM, which is necessary to establish the final diagnosis of RBD. Therefore, given the available data, only the diagnosis of "possible RBD" is warranted.

A differential diagnosis with psychogenic dissociative disorder would be warranted. Dissociative disorders may present during the sleep period and display a wide variety of complex behaviors (Bokey, 1993); however, the events appear in a wakeful state by EEG criteria (Schenck et al., 1989). The patient, as already mentioned, had neither signs nor history of psychological or psychiatric disturbance.

A subgroup of RBD patients with Non-REM parasomnias (sleepwalking and sleep terrors) have been reported to have the Parasomnia Overlap Syndrome (POS) (Schenck et al., 1997). In this paper, 33 patients with a polysomnographically confirmed final diagnosis of RBD, sleepwalking and sleep terror, have been described. The forty POS cases reported in the literature to this date (as reviewed by Schenck et al., 1997) show a clear male preponderance and a positive therapeutical response to benzodiazepines. What these POS cases have in common with other parasomnias are positive responses to benzodiazepines.

Our case reported herein may qualify to have the POS, because of his history of SW, SBS, and the clinical characteristics of violent nocturnal behaviors suggestive of RBD. This is in keeping with a motor-behavioral dyscontrol extending across all states of NREM and REM sleep (Blanco and Garay, 1995).

In this case report, there is an overlap with another possible parasomnia, i.e., sexual behavior in sleep. Amnestic sexual behavior during sleep has been proposed as a parasomnia (Hurwitz et al., 1989; Shapiro et al., 1996). Shapiro describes seven patients (six males) with SBS; all had a previous history of parasomnias and three of them also had positive family histories of parasomnias. Additional clinical and demographic features of sexual behavior in sleep will be described as this newly proposed parasomnia becomes diagnosed more frequently.

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