

# Update on Sexsomnia, Sleep Related Sexual Seizures, and Forensic Implications

Carlos H. Schenck

## ABSTRACT

The first classification of sleep-related disorders and abnormal sexual behaviors and experiences was published in 2007. Parasomnias (abnormal sleep-related behaviors and experiences) and sleep-related epileptic seizures were the most frequent disorders, after Kleine-Levin syndrome (periodic hypersomnia with abnormal wakeful sexual behaviors). The first two conditions were named sexsomnia (sleepsex) and epileptic (ictal) sexsomnia, respectively. Sexsomnia usually emerges during confusional arousals (CAs) from delta non-REM sleep (N3 sleep), either associated or unassociated with obstructive sleep apnea (OSA). We now report an additional 22 cases of sexsomnia and 3 cases of ictal sexsomnia (temporal lobe epilepsy; bupropion-induced seizures) published from 2007-2015, based on a literature search in PubMed and Embase, and also separately for Turkish language publications. Eighteen of the 22 additional cases of sexsomnia had sufficient data provided to allow for comparative analysis. (The 4 other additional cases involved sexsomnia emerging with Parkinson's disease). The demographics of the second group of 18 sexsomnia cases were comparable to those of the first group of 31 cases (published in 2007), in regards to male gender predominance (67% vs. 81%); age at presentation (40 yrs vs. 32 yrs); age of onset (33 yrs vs. 26 yrs); and mean duration of sexsomnia in males (5.6 yrs vs. 8.3 yrs). The female groups were too small to compare. The distribution of sexual behaviors across the groups was generally comparable in regards to sexual vocalizations, masturbation, fondling, and intercourse/attempted intercourse. Amnesia for the sexsomnia by the affected person was 89% vs. 100%. Video-polysomnographic studies were conducted in nearly all patients in both groups, and provided important diagnostic findings in almost all patients. The mean number of parasomnias per patient was  $1.8 \pm 1.4$  vs.  $2.2 \pm 1.0$ , respectively, with the range extending up to 5 parasomnias per patient. In both groups, a non-REM sleep parasomnia (disorder of arousal [DOA]) was the main cause of the sexsomnia (78% vs. 90%). There was a comparable percentage in each group having obstructive sleep apnea (OSA) as the presumed trigger for a DOA with sexsomnia (17% vs. 13%), and there was control of both sexsomnia and OSA with nasal CPAP in 100% (4/4) of treated cases. Overall treatment efficacy was 82% (n=18) in the 22 patients in the combined groups (n=53) for whom treatment was reported. Nine novel findings on sexsomnia were identified. An abstract on 41 consecutive cases of sexsomnia evaluated at a single sleep center in the U.K. was recently published, and the findings are highly congruent with the 53 cumulative cases in the world literature reported herein. Thus, there are now 94 total cases of sexsomnia reported in the world literature. The forensic implications of sexsomnia are discussed.

**Key Words:** sexsomnia, sleepsex, sexual behaviors of sleep, temporal lobe epilepsy, ictal orgasm, epileptic sexsomnia, polysomnography, non-REM sleep parasomnia, confusional arousals, obstructive sleep apnea, REM sleep behavior disorder, clonazepam, SSRI, bupropion, shift work, circadian sleep disorder, Parkinson's disease, forensic sleep medicine

**DOI Number:** 10.14704/nq.2015.13.4.873

**NeuroQuantology 2015; 4:518-541**

**Corresponding author:** Carlos H. Schenck

**Address:** Minnesota Regional Sleep Disorders Center, Department of Psychiatry (R7), Hennepin County Medical Center, 701 Park Ave., Minneapolis, MN 55415, USA.

**Phone:** + 612-873-6288

**e-mail** ✉ schen010@umn.edu

**Relevant conflicts of interest/financial disclosures:** The author declares that the research was conducted in the absence of any commercial or financial relationships that could be construed as a potential conflict of interest.

**Received:** September 1, 2015; **Accepted:** September 28, 2015

**eISSN** 1303-5150



www.neuroquantology.com

## Introduction

The first classification of sleep-related disorders and abnormal sexual behaviors and experiences was published in 2007, and encompassed a broad range of sleep-related disorders along with a broad range of associated sexual behaviors and experiences, involving the affected person and/or another person (usually the bed partner) (Schenck *et al.*, 2007). Kleine-Levin syndrome (periodic hypersomnia with abnormal wakeful sexual behaviors) and parasomnias (abnormal behaviors and experiences accompanying sleep) were the most frequently reported conditions, followed by sleep-related epileptic seizures. The latter two conditions were called sexsomnia (i.e. a sexual parasomnia) and epileptic (ictal) sexsomnia, respectively, which have overlapping and divergent clinical features. Sexsomnia most often emerges during confusional arousals (CAs) from slow-wave (delta) non-REM sleep (N3 sleep), which could be associated or unassociated with obstructive sleep apnea (OSA). Episodes of sexsomnia emerging with CAs take place in the bed of the sleeping person. Sexsomnia can also rarely occur during sleepwalking (SW). Both CAs and SW are classified as Disorders of Arousal from non-REM sleep, with sexsomnia recognized as a variant of CAs (American Academy of Sleep Medicine, 2014). Sleepsex, atypical sexual behavior during sleep, sexual behaviors during sleep, and sleep related abnormal sexual behaviors are other synonymous terms. Adverse physical and/or psychosocial effects from the sexsomnia are common, along with their forensic consequences (Schenck *et al.*, 2007). Videopolysomnography (vPSG), i.e., the objective, multichannel, physiological monitoring of sleep, with time-synchronized audio-visual recording, is critical for identifying the cause of the sleep-related sexual behaviors (and for ruling-out other causes), and for properly directing the initiation of specific and generally very effective therapy.

An update on this topic will now be presented, focusing primarily on additional published cases of sexsomnia and ictal sexsomnia, and their forensic consequences. The cumulative literature reflects a growing international attention to this still under-recognized and yet clinically and medical-legally important topic.

## Material and Methods

A PubMed and Embase literature search was conducted from 2007-2015, which identified peer-reviewed journal articles and published abstracts related to sleep and sex and sleep related sexual seizures. A separate literature search on this topic was conducted for Turkish language publications by S. Cankardeş, Istanbul. All identified publications were critically reviewed and the reported cases were classified.

## Case Samples

A total of 22 additional published cases of parasomnias with abnormal sleep-related sexual behaviors (i.e. sexsomnia) were identified, along with three additional cases of ictal sexsomnia, since the 2007 report by Schenck *et al.* Therefore, to date there have been 53 cases of sexsomnia published and 10 cases of ictal sexsomnia published in the peer-reviewed medical literature, for a total of 63 cases. The rate of publishing on sexsomnia in the peer-reviewed literature has increased by >50% over the past 7 years compared to the previous 21 years. Table 1 provides a summary of key findings from the additional cases and compares them with the previously published cases. Four of the 22 additional cases of sexsomnia did not have sufficient data provided to allow for comparative analysis; therefore, 18 of the additional 22 cases were used for comparative analysis). The demographics of the second group of 18 cases are similar to the demographics of the first group of 31 cases, in regards to male gender predominance, age at presentation, and age of onset and duration of sexsomnia in males. The female groups were too small to compare. The distribution of sexual behaviors across the groups was generally comparable. The frequency of sexsomnia episodes was reported for 9 of the 18 patients, with the following distribution: nightly, n=2; 2-4 nights weekly, n=2; one night weekly, n=1; 2-3 nights monthly, n=3; "frequently," n=1. The more recent group of 18 patients had substantially fewer sexual assaults, sexsomnia with minors, and legal consequences compared to the original reported group of 31 patients. Amnesia for the sexsomnia by the affected person was 88.9% vs. 100%, with two females in the former group presenting with the complaint of spontaneous sleep orgasms, which to date is the only form of sexsomnia with subsequent recall by the affected person. vPSG studies were conducted in nearly all

patients in both groups, which provided important positive and negative diagnostic findings in virtually all patients. There was a comparable number of mean parasomnias per patient, with the range extending up to 5 parasomnias in two patients. For both groups, a non-REM sleep parasomnia (disorder of arousal [DOA]) was the predominant diagnosis, including a comparable percentage in each group having obstructive sleep apnea (OSA) as the presumed trigger for a DOA with sexsomnia, which was strongly supported by the control of both sexsomnia and OSA in 100% of cases treated with nasal continuous positive airway pressure (nCPAP) therapy. Overall treatment efficacy was 82% (n=18) in the 22 patients in the combined groups (n=53) for whom treatment was reported.

The additional published cases of sexsomnia since 2007 will now be described in detail, with pertinent commentary, in order to highlight novel findings, and to further elucidate the phenomenology of this still nascent clinical area that is situated at the intersection of sleep medicine, sexual medicine, and forensic medicine.

## **I) Parasomnias with sleep-related sexual behaviors**

### **Sexsomnia cases reported from 9 countries (United States, Spain, Holland, Italy, France, Turkey, Australia, Brazil, United Kingdom)**

#### **A) Peer-reviewed journal articles (publications 1-8)**

##### **1) A Case of Sexsomnia with OSA-Associated Confusional Arousals (Schenck *et al.*, 2008)**

A 32 year-old married man experienced the concurrent emergence of sexsomnia and OSA, with subsequent control of both sleep disorders with nCPAP monotherapy. He had presented to the Minnesota Regional Sleep Disorders Center with his wife with the chief complaint of sexually groping and fondling her while asleep during the previous four years. This sleep related sexual activity began at the same time as snoring began, which became progressively louder over time, and his wife started complaining that “he tried to hump me while he was asleep.” His wife reported that he was somewhat insistent with his sleepsex initiatives with her, but was never aggressive or violent, and he always responded promptly to her limit setting. “On some occasions he would be awakened by his wife in the midst of a sexsomnia episode, and then he would recall having a sexual

dream involving the two of them.” His sleepsex repertoire was the same as the sexual repertoire during their waking lives. Sexsomnia frequency was four nights per week. He had total amnesia for these sexsomnia events. They reported a normal, regular, conventional wakeful sex life. No trigger for sexsomnia occurrence could be identified (e.g. stress, sleep-deprivation, sexual deprivation). There was no prior history of parasomnia, psychiatric disorder, paraphilia, alcohol or drug abuse, or family history of sleep or sexual disorder. He had been employed continuously since high school. They had been married for ten years, without any marital problems, apart from increasing stress related to the sexsomnia that eventually posed a risk to the marriage.

Overnight vPSG at an accredited, hospital-based sleep disorders center documented OSA, with an Apnea Index of 19/hr, and oxygen saturation nadir of 78%. nCPAP therapy completely normalized sleep continuity and hemoglobin oxygen saturation during the second half of the single-night vPSG study. No sexsomnia or other parasomnia behaviors were documented during the vPSG study.

The diagnoses were both OSA and Sexsomnia emerging during CAs induced by OSA. Treatment of these two temporally linked sleep disorders was initiated with nCPAP therapy, with the expectation that one therapy could control both disorders.

At one month and three-month follow-up visits, the wife reported complete control of her husband's snoring and sexsomnia with ongoing nightly nCPAP therapy. However, on nights when the nCPAP mask came off his face, there would be some mild sexual groping and fondling during his sleep. This indicated an ongoing propensity for sexsomnia emerging with CAs whenever the patient's OSA therapy was interrupted. Overall, the wife was very pleased and was optimistic about the future of their marriage.

A notable feature of this case, apart from the close association of the emergence of sexsomnia with snoring/OSA and their concurrent and enduring response to nCPAP therapy, is that the patient reported sexual dreams involving himself and his wife whenever his wife awakened him during a sexsomnia episode. Dreaming with sexsomnia has rarely been reported. This raises the question of whether for this patient the occasional episodes



of dreaming with presumed OSA-induced sexsomnia emerged during arousals from REM sleep, which is the sleep stage most frequently associated with dreaming.

This case calls attention to the need to question patients with snoring and diagnosed OSA about sexsomnia, and vice versa. Patients with OSA and other forms of sleep-disordered breathing may comprise a large and greatly under-recognized subgroup of sexsomnia patients. Patients should be told that sexsomnia is a medical (sleep-related) problem, and not a primary psychological or psychiatric problem, although there could be psychological consequences.

2) A sexsomnia case from Spain was reported [translated and summarized by the author, CHS] in which a 33-year-old male presented to a neurology sleep clinic with the chief complaint of sexsomnia for one year with repeated attempts to remove his pajamas and fondle his bed partner; there was no self-touching of genitals or masturbation (Penas-Martinez *et al.*, 2008). These episodes generally occurred in the first part of the night, and lasted around five minutes, without orgasm being achieved. The frequency of these episodes was two nights monthly, without any identified precipitant, and with full subsequent amnesia. There was no personal or family history of parasomnia. Medical history included hypertension, asthma, and nasal polyps. He was a former smoker. Neurological and medical evaluations were unremarkable.

vPSG during two non-consecutive nights documented various N3 arousals with diffuse generalized theta activity, consistent with a non-REM sleep parasomnia. No sexual or other parasomnia behaviors occurred during the vPSG study. Sleep architecture (i.e. distribution and cycling among the sleep stages) was normal. However, the presence of snoring, apneas, and hypopneas was mentioned in this report, but unfortunately no data were provided, including any presence of oxygen desaturations. The authors did state that there was a major postural component to the snoring and sleep disordered breathing, with attenuation achieved while the patient lay on his side.

The presumed diagnosis (not stated by the authors, but inferred from their comments) was sexsomnia during confusional arousals from N3 sleep. The findings of abrupt arousals from N3 sleep, which is an abnormal PSG finding, suggests

that perhaps the patient had a prior, unrecognized, and unobserved history of CAS without clinical consequence, which may have predisposed the subsequent emergence of sexsomnia.

Therapy with clonazepam, 1 mg at bedtime (a standard therapy for non-REM sleep parasomnias, including sexsomnia) was initiated. At two-month follow-up, the patient reported a decreased frequency of sexsomnia episodes, despite intermittent use of clonazepam. However, since the baseline sexsomnia frequency was two nights per month, and since there was only a two-month follow-up to assess treatment efficacy, with intermittent clonazepam use, no firm conclusion can be drawn about clonazepam treatment efficacy.

3) A sexsomnia case from Holland was reported in which a 30-year-old man, without any prior parasomnia history, developed *de novo* sexsomnia on a nightly basis for four weeks upon starting escitalopram [SSRI] therapy, 10 mg/day (standard dose), for control of a major depressive episode (Krol, 2008). His sexsomnia featured full intercourse with his bed partner, and complete subsequent amnesia. The sexsomnia ceased six days after discontinuation of escitalopram, consistent with the pharmacologic wash-out period for this drug. (Escitalopram was also not beneficial for his depression). There was no recurrence of sexsomnia at five-month follow-up during subsequent therapy with high-dose duloxetine, 90 mg/day. In contrast to escitalopram, duloxetine (a serotonin-norepinephrine reuptake inhibitor) was notably effective in controlling his major depressive episode.

This is the first reported case of medication-induced *de novo* sexsomnia, in which a standard dose of an SSRI (escitalopram) immediately induced nightly sexsomnia with full sexual intercourse and subsequent amnesia, and with prompt resolution of the sexsomnia upon discontinuation of the escitalopram. For this patient, it was a specific medication effect, since subsequent long-term therapy with a different class of antidepressant (duloxetine) at a high dose did not induce sexsomnia. The sexsomnia may have been an idiosyncratic medication reaction in this male patient, since in another report to be described below, the same SSRI escitalopram was therapeutic in controlling sexsomnia in two female patients.



A partial translation of this article from Dutch into English (courtesy of Michael A. Corner, PhD, Amsterdam) will now be provided:

"He called up three weeks later [after starting escitalopram therapy], distressed. He told that he apparently had had nightly sexual intercourse with his [female] partner, 'apparently' because she had completely surprised him with this information in the morning. She had found the timing - consistently about an hour after falling asleep - somewhat unusual, but further noticed nothing special about her friend's [sexual] behavior. He had caressed her (also genitally) and undressed her, and they had sex 'like always,' in which the patient and his girlfriend sometimes changed position. The patient reported having no recollection whatsoever of his nightly amorous approach. He found it frightening to have exhibited behavior at night of which he was not conscious and over which he had no control.

The following week he repeated this behavior on six of the seven nights. A few times he had a vague recollection in the morning, without being certain that it really had happened. His [girl] friend succeeded twice in wakening him, upon which, embarrassed, he deliberately got out of bed and went back to sleep a little later. According to the girl friend, he answered "no" on several occasions when she explicitly asked him if was sleeping, leading her to assume that the patient was awake and aware of their having intercourse. Here too, the patient was unable to remember any such thing the next morning.

Furthermore, because the depressive episode failed to go into remission, escitalopram treatment was discontinued at the end of the fourth week. Six days later the patient had the last of his nighttime 'episodes'. In the following five months this behavior didn't recur.

A week after stopping escitalopram, the patient started using duloxetine (60 mg), later raised to 90 mg. After another eight weeks his depression was completely in remission.

Upon questioning it became apparent that the patient had never before any trouble with parasomnias, also not as a child. A brother of his as a child had somnambulism, which disappeared well before puberty.

Standard laboratory investigation didn't turn up any deviations. A neurologist found no indications during a screening examination of any neurological insults.

Neither were there any indications of a dissociative disturbance. On the basis of the clinical picture, a diagnosis of *parasomnia* as made."

4) Three sexsomnia cases were reported from Italy, all involving males aged 32, 42, and 46 years, who had been referred to a sleep disorders center because of sleep-related sexual episodes (Della Marca *et al.*, 2009).

A 42-year-old man had a 12-year history of sexsomnia emerging with OSA, as documented by vPSG study, with an AHI (apnea-hypopnea index) of 38.5/hr, but without any parasomnia behavior occurring during the vPSG study. His sexsomnia involved initiating sex with his wife that occasionally led to full intercourse. Treatment with nCPAP resulted in both control of his OSA and a marked reduction of his sexsomnia, with the sexsomnia frequency going from 2-3 nights weekly to 3-4 nights yearly. Therefore, the diagnosis of OSA-induced CAs with sexsomnia was justified, with nCPAP monotherapy controlling the interlinked OSA and sexsomnia.

A 32-year-old man had a 4-year history of sexsomnia emerging as a non-REM sleep parasomnia. His wife reported that he would initiate sex during sleep that occasionally led to full intercourse. He had a childhood history of SW. During his vPSG study, there were three non-sexual minor motor events emerging from N3 (slow-wave) sleep, which supported the diagnosis of sexsomnia emerging as a variant of non-REM sleep parasomnia (CAs, SW). Therapy was not mentioned.

A 46-year-old man, without any prior parasomnia history, nor any family history of parasomnia, presented with a "recent onset" history of aggressive and violent sleep behaviors *and* sexual behaviors during sleep, with full intercourse, at times associated with sexual dreams. The wife provided all the details, since the man had no recall. These events occurred during the second half of the night and early morning hours. vPSG documented REM sleep without atonia, without any associated behaviors during REM or non-REM sleep. The diagnosis was REM sleep behavior disorder (RBD) with sexsomnia. Standard therapy of RBD with clonazepam, 2 mg at bedtime, was not effective in controlling sexsomnia. Carbamazepine and dopamine agonist therapies were also not effective.

This case represents the fourth reported case of sexsomnia presumably emerging with RBD, with the other three cases reported from a single tertiary sleep center (Guilleminault *et al.*, 2002). However, this case is the most compelling of the four cases for linking sexsomnia with RBD, as the wife described his aggressive and violent sleep behaviors and sexual behaviors emerging during the second half of the sleep period, which is when REM sleep predominates. The association with (sexual) dreaming also supports REM sleep as the sleep stage promoting the sexsomnia. Also, his male gender and middle age (46 years old) are typical for RBD, besides the aggressive and violent sleep behaviors. Finally, he had no prior history of non-REM sleep parasomnia. The other three sexsomnia cases diagnosed with RBD did not have dream-enacting behaviors (atypical for RBD), involved younger males (atypical for RBD), and also did not demonstrate any sexual or non-sexual behaviors in REM sleep during their vPSG studies (Guilleminault *et al.*, 2002). Their sexsomnia episodes by history did not emerge mainly in the second half of the night, as did the sexsomnia episodes in this Italian patient. The only positive objective finding in all four cases of presumed RBD with sexsomnia was the intermittent loss of REM-atonnia.

The possible association of sexsomnia and RBD remains an intriguing open question. Sexual behaviors during REM sleep have not yet been documented during vPSG studies, which would establish that sexsomnia can be a variant of RBD besides being a variant of CAs and SW. In the largest study to date on vPSG monitoring of Parkinson disease (PD) patients (n=457), despite 210 (46%) of these patients demonstrating RBD, none had any sexual behaviors during REM sleep during their vPSG studies (Sixel-Döring *et al.*, 2011). However, these elderly, medicated PD-RBD patients represent only one subgroup of all RBD patients, and so the lack of any sexual behaviors during REM sleep cannot be generalized for all RBD patients, thus underscoring the need for further research in this area.

Finally, from a Freudian psychoanalytic perspective, it is surprising that RBD is not a sexual parasomnia (or that sexsomnia is not RBD), since RBD is a dream-enactment disorder which for Freud would constitute the continuation of the "royal road" (beginning with dreaming) for wish fulfillment and sexual acting-out during sleep. However, a controlled study on

dreams in RBD (Fantini *et al.*, 2005) found a major shift in bias towards certain types of dreams (e.g. confrontational, aggressive), and away from other types of dreams (viz. sexual). Therefore, the dreams to be enacted in RBD are rarely sexual. This study supports the many anecdotal observations found in large and smaller case series on RBD in which sexual dream enactment is virtually absent.

#### 5) Two sexsomnia cases were reported from France, involving married women who were 36 years old and 40 years old (Bejot *et al.*, 2010).

Both patients reported traumatic sexual experiences during adolescence, involving "sexual abuse" at age 16 years in one patient, and witnessing a mother's rape at age 15 years in the other patient. Both patients reported current sexsomnia, with sexual moaning, "dirty talk", masturbation, sexual assaults of their bed partners, with sexual intercourse, and subsequent complete amnesia. The 36-year-old woman had a childhood-onset history of SW with current persistence, along with a 6-year history of sexsomnia. The 40-year-old woman had a 5-year history of sexsomnia in which she violently attacked her husband sexually--and also violently attacked herself sexually during sleep. Some examples include: "she was violently masturbating and inserting a deodorant stick into her vagina and anus. During another one, she tried to push marbles into her husband's anus. One time, he was awakened by intense pain caused by 3 padlocks placed around his penis and testicles. In another one, his penis was placed into the cover of a mincing machine."

During their vPSGs, both patients had multiple, abrupt, spontaneous arousals from N3 (slow-wave) sleep, without any associated behaviors. These findings led to the diagnosis of non-REM sleep parasomnia (CAs) as the basis for the sexsomnia in each patient. Therapy with the SSRI escitalopram, 10 mg/day for each patient (one with depression, one without depression) resulted in complete control of sexsomnia that was maintained at 9 months and 2-year follow-up, respectively. No rationale was given for the SSRI therapy of the sexsomnia. (However, in the second patient, escitalopram efficacy was not maintained after two years; substitution of lamotrigine, 1,000 mg at bedtime, was beneficial at 4-year follow-up [Isabelle Arnulf, personal communication]).



Both these females had sexsomnia associated with CAs, a non-REM sleep parasomnia, the most common cause of sexsomnia. Their childhood traumatic histories might have predicted sexsomnia emerging from a nocturnal, sleep related dissociative disorder (Schenck *et al.*, 1989a; 1989b), a psychogenic disorder emerging during the nocturnal sleep period with a wakeful EEG. This condition can feature the reenactment of past sexual traumatic experiences. A heightened suspicion for nocturnal sexual dissociation could be justified in these two cases, since sexual abuse often occurs at night and in a bedroom. This underscores the importance of conducting vPSG studies to objectively document, to the extent possible, any underlying sleep related disorder promoting the sexsomnia.

**6) Two cases of sexsomnia embedded in a longstanding, complex history of five parasomnias affecting each patient, were reported from Italy (Cicolin *et al.*, 2011).**

These cases represented the first two cases of a novel category of Parasomnia Overlap Disorder involving REM Sleep Behavior Disorder (RBD) and a non-REM sleep parasomnia in the same patient (Schenck *et al.*, 1997; American Academy of Sleep Medicine, 2014). Sexsomnia as a non-REM sleep parasomnia was found in patients who also had RBD. Furthermore, legal charges were involved on account of sexsomnia in one of these patients, with eventual acquittal.

The first case involved a 60 year-old married woman who presented to a sleep disorders center on account of RBD of four years' duration, with her husband complaining of her repeated, aggressive, dream-enacting behaviors. She also had a childhood-onset, lifelong history of SW, sleep talking, and some recurrent episodes of sleep related eating (Schenck *et al.*, 1991; American Academy of Sleep Medicine, 2014).

vPSG demonstrated RBD behaviors with loss of the customary REM-atoniality, and also non-REM sleep parasomnias emerging from N3 sleep. She also had an episode of sexsomnia with masturbation (placing a hand under her panty to masturbate) during N3 sleep, which lasted several minutes. The masturbation was preceded by a hypersynchronous delta EEG pattern, and during the episode, the EEG pattern showed the persistence of delta rhythms with increasing

alpha activity, thus confirming that the sexual behavior occurred during sustained sleep. Treatment was not mentioned.

The second case involved a 41-year-old man with childhood-onset, lifelong SW, sleep (night) terrors, and sleep talking, and a six-year history of violent RBD episodes with dream-enactment. His ex-wife and current girlfriend both confirmed repeated sexsomnia episodes, with sexual fondling and sexual intercourse. He faced a legal sexual assault charge from repeated sexual fondling of the 11-year-old daughter of his current girlfriend one night while he was reportedly asleep, and for which he was completely amnesic.

He underwent five vPSGs as part of the forensic sleep medicine evaluation related to his legal charge, which confirmed RBD, with behaviors emerging from REM sleep and loss of REM-sleep muscle atonia. He also had parasomnia behaviors that emerged from non-REM sleep, but solely non-sexual behaviors were documented by vPSG. He therefore had documented Parasomnia Overlap Disorder, with the sexual behaviors presumably emerging from non-REM sleep. He pleaded innocent to the charge of sexual assault by using the Parasomnia Defense, with sexsomnia/sleepsex being considered a non-insane automatism. The testimonies of his ex-wife and current girlfriend about his history of recurrent sexsomnia episodes, together with the vPSG findings, the absence of any prior history of sexual assault or deviance, and the lack of any motivation for intentionally engaging in sexual assault, resulted in his acquittal. The patient subsequently always slept with an infrared door alarm on his bedroom door, as a safeguard for aborting any future parasomnia episode, including sexsomnia associated with SW.

These two cases illustrate the common clinical scenario of sexsomnia being embedded within a longstanding, complex "Parasomnia Multiforme." In fact, apart from sexsomnia emerging with OSA or emerging as an ictal sexsomnia, sexsomnia typically emerges late in the course of a preexisting, longstanding non-REM sleep parasomnia, and is usually the last parasomnia to emerge in a series of previously established parasomnias. This documented observation has important legal implications: a first-time alleged sexsomnia episode that results in a legal sexual assault charge should be viewed

with great skepticism, and raises the strong suspicion of a "sexsomnia excuse" for intentional sexual assault behavior. Furthermore, the role of any alcohol use or intoxication (or illicit drug use) in alleged sexsomnia behavior should be explored, since alcohol intoxication and illicit drug use cannot absolve culpability for any subsequent sexual assault or other criminal behavior.

7) A report from Turkey on two cases of spontaneous orgasm during sleep (Özcan *et al.*, 2012) was translated from Turkish to English for this manuscript by S. Cankardeş, Istanbul:

"First case was a 37 year-old, married, employed woman. She presented to the psychiatry clinic in 2008 with the symptom of orgasms during sleep [of unspecified duration] without any arousal or sexual dream content. Sleep orgasms had appeared on most nights. With an ictal orgasm prediagnosis, a brain MRI was performed, but no pathology found. A sleep EEG [not with polysomnography] was performed, but no epileptic activity was observed even though the patient reported that she had experienced orgasm [in her sleep]. Patient was informed about having a parasomnia. Clonazepam therapy, 0.5 mg, was offered, but she refused treatment.

Second case was a 48 year-old, married, housewife. She presented to a menopause clinic with the complaint of sleep orgasm. She had sleep orgasm [of unspecified duration] at least 2-3 times in a month without any sexual dream content or arousal. She also had sleep orgasms after she had sexual intercourse. She reported that her 77-year-old mother had the same problem, which she discovered three years previously, when her mother wanted to take a shower before surgery because she had experienced orgasm during sleep on the preceding night [she was complying with the Muslim full ablution religious ritual, to become "pure" again after engaging in any form of sexual activity--even spontaneous orgasms--that had made her "impure"]. This was when she learned that her mother also had [spontaneous] orgasms at night. Clonazepam therapy (in liquid form) was initiated, 0.375 mg at bedtime, and the sleep orgasms ceased within one month. Although prior to this therapy an EEG and brain MRI were recommended, they were not performed.

Since both patients presented with the complaint of spontaneous sleep orgasms, they had awareness of their sexsomnia, and so sleep orgasm (in females) is the only form of sexsomnia reported to date in which there is recall of the sexsomnia by the affected person.

Both patients had normal mental health and medical histories and examinations. Both patients also did not have histories of any sexual abnormality." [No information was provided about a history of parasomnias or other sleep disorders].

8) Four cases of sexsomnia were reported from Spain, involving patients who presented to a multi-disciplinary hospital sleep clinic (Arino *et al.*, 2014). None of the patients had a psychiatric or sexual disorder history. vPSG was performed in all 4 cases.

The first case was a 38-year-old male who came with wife; they reported a stable 17-year marriage, with conventional, satisfactory sex occurring 2-3 times weekly. There was a negative personal and family medical history, except for nasal septoplasty five years previously. There was no history of traumatic sexual experiences. Parasomnia history was positive only for isolated sleeptalking as a child.

The wife described her husband's seven-year history of sexsomnia: he would sleep for 2-3 hours, then while still asleep he would abruptly attempt sexual intercourse for 10-30 minutes. The sexsomnia behavior differed from his waking sexual behavior, in that it was more vigorous, forceful sex with "lewd language" while trying to penetrate his wife. His wife observed an erect penis during each episode. The frequency of the sexsomnia was once weekly.

The wife reported one notable episode in which he immobilized her by placing his arm around her neck in a stranglehold. "Although he attempted penetration, she never permitted it because she believed that the episodes were not normal sexual behaviour and that her husband's actions were involuntary...The patient had awakened in a confused state during some of these episodes and was surprised by his wife's account of his behaviour."

There was complete amnesia for the sexsomnia, and no apparent associated dreaming. Also, there was no identified trigger, such as stress or prior sexual stimuli, for these weekly sexsomnia episodes.



There was a 20-year history of snoring, apneas observed by his wife, and excessive daytime sleepiness (EDS), which resulted in a nasal septoplasty five years earlier, without a prior PSG being performed, but there was no benefit for the snoring, EDS or sexsomnia from the septoplasty.

A waking EEG was unremarkable. vPSG detected sleep-disordered breathing, with an Apnea-Hypopnea Index of 13/hr, which increased to 40/hr while lying supine. No other abnormality was found, including the absence of any parasomnia behavior.

The presumptive diagnoses were sleep-disordered breathing (SDB), and sexsomnia as either an isolated parasomnia and/or a comorbidity of SDB with CAs. The patient refused treatment of the SDB and also refused clonazepam therapy of sexsomnia. However, nCPAP therapy should have been offered first as a potentially effective monotherapy of these two sleep-related disorders before considering clonazepam therapy.

The second case involved a 41-year-old female with a one-year history of sexsomnia. Her husband observed sleep masturbation three nights weekly, without involving him, even though they slept in the same bed. Sometimes she would achieve orgasm from her sleep masturbation. The episodes occurred around 5:00 a.m. There was complete subsequent amnesia for the masturbation, and "she felt ashamed" when her husband described these episodes to her. Her husband also observed repetitive and periodic non-sexual limb movements during sleep, especially in the lateral decubitus position. Upon questioning, she reported occasional symptoms of Restless Legs Syndrome (RLS) while resting, especially at night. There was no history of snoring, apneas, EDS or insomnia. Their waking sexual activity was described as being "regular, satisfactory, and pleasant sexual intercourse, with no sexual problems when awake."

Parasomnia history was positive for childhood SW until age 14 years.

There was a one-year history of metrorrhagia, of unknown origin, causing recurrent iron deficiency with periodic therapy with oral iron supplements.

vPSG was performed, which did not detect apneas, epileptiform activity, parasomnia

behaviors, or any other abnormalities. However, there was a PLM index of 24/hr throughout the night. The PLM index with microarousals, partially disrupting sleep, was 7/hr. Also, with foot movements there was sudden abduction of the lower limbs. On two occasions during the vPSG study, there were more prolonged movements, accompanied by repetitive arm movements. Of note is that she also placed her hand on her genitals for a few seconds but did not masturbate.

Although not stated explicitly, the presumed diagnosis appeared to be sexsomnia associated with RLS, PLMs and other periodic movements of sleep serving as precipitating factors, with a prior history of non-REM parasomnia (i.e. SW) serving as a predisposing factor. Treatment with pramipexole and an iron supplement improved RLS, but did not benefit the sexsomnia, as reported at 90-day follow-up. The patient was then lost to long-term follow-up.

The third case involved a 43-year-old male whose live-in partner of the past year observed four episodes of sexsomnia that occurred in the context of a more complex, non-sexual parasomnia disorder, which also dated back to childhood, when he experienced frequent sleeptalking and shouting, occasionally with distressing nightmares.

During the preceding year, when he started cohabitating with his current partner, he had started rotating shift work. His partner would observe that two hours after his falling asleep, he would occasionally sit up abruptly, with confusion and fear. He could engage in a partially coherent conversation with her for several minutes, always with his eyes open, and he would move and at times swat at something imaginary in the air. He appeared to be asleep and never had recall for these events.

The four similar episodes that involved the added component of sexual behavior occurred after he had slept 2-4 hours. He attempted to initiate sex with his partner by fondling her body and genitals. The partner rejected the advances, since to her they were inappropriate and involuntary behavior on his part. He never displayed aggressive or insistent sexsomnia behavior, "but merely moved to the other side of the bed, expressing his frustration and annoyance in such terms as 'no fun.'" There was total amnesia for these four sexsomnia episodes. These four episodes posed no problems for the couple's

relationship. The couple reported satisfactory conventional waking sex, and he had no history of any sexual disorder.

Of particular importance is that these four sexsomnia episodes occurred on a day when he had changed shifts at work.

The patient reported similar sexsomnia episodes with a former partner, who had on occasion consented to his advances, with both of them achieving orgasm. These episodes had been described to him by the previous partner, as he had only vague recall of the end of the sex acts. The history strongly suggests that he was not engaged in shift work while having sexsomnia with his former partner.

vPSG was performed for two nights, and no parasomnia activity was detected. Sleep-disordered breathing was found, with an AHI of 16/hr and 7/hr, respectively, during the two studies. OSA was especially common while supine, and particularly in REM sleep: 37/hr and 40/hr, respectively, during the two vPSG studies. Neither PLMs nor EEG epileptiform activity was detected, and normal REM-atonias was documented.

The patient refused therapy of both sleep-disordered breathing and sexsomnia.

From a diagnostic perspective, this case presents interesting possibilities, alone and in combination, to account for the emergence of sexsomnia. The patient had a childhood-onset, complex parasomnia history that persisted throughout adulthood up to the time of referral. Therefore, sexsomnia may have represented another non-REM sleep parasomnia in his evolving parasomnia history. He also had documented sleep-disordered breathing that could have promoted sexsomnia during CAs, as described above in other cases, and as previously published. And finally, rotating shift work was an apparent circadian rhythm disturbance trigger that promoted all four observed sexsomnia episodes. Therefore, this case reveals some of the complexity that can surround sexsomnia, both for individual patients and for the spectrum of affected patients. NREM parasomnia, sleep disordered breathing, and circadian rhythm disturbance may each have played a role in triggering sexsomnia in this patient.

The fourth case involved a 28-year-old male whose bed partner of nine months observed complex non-sexual and sexual parasomnia

activity. During 3-6 nights weekly, while they were asleep, he would abruptly sit up in bed or turn towards her and shake her before uttering incoherent phrases, such as "look at that house" or "the devil". They could engage in conversations, and occasionally he would cry out or laugh. He also displayed mimicking behaviors such as driving a car, having a conversation, or searching for something in the bed. His partner believed that he seemed to be acting out a dream. During these episodes, his eyes remained open and he appeared restless and worried. His partner would generally be able to calm him by telling him to go back to sleep. He never left the bed during these episodes.

Similar, but exclusively sexual episodes, occurred 2-3 times monthly, when he would masturbate or touch his partner with the intent to initiate sex. She reported that he was never aggressive or violent, but "it isn't him", describing the behavior as "more lewd and vulgar." His penis was always erect during these episodes. She would always refuse sex during these nocturnal episodes, and he would immediately cease the sexual advances without any complaint.

All the reported parasomnia episodes, both sexual and non-sexual, occurred no more than once nightly, 2-3 hours after sleep onset. He was always subsequently amnesic for these episodes, and would feel tired the next day. There was never any reported nightmares or erotic dreams. The couple's waking sex life was regular and mutually satisfactory.

Of note is that past partners had observed similar sexual and non-sexual sleep behaviors.

The patient reported a history of SW and sleeptalking, and a brother had SW.

vPSG study did not reveal any parasomnia behaviors, nor SDB, PLMs, EEG epileptiform activity, nor any other abnormality. The diagnosis was sexsomnia associated with CAs and other non-REM sleep parasomnias.

Treatment was directed both to improve sleep habits (establish regular sleep hours, and obtain 8 hrs of total sleep nightly), and to start taking clonazepam, 0.5 mg q HS. This dual therapy was beneficial, as the CAs decreased in frequency to 1-2 nights weekly, and the sexsomnia frequency was decreased to 1-2 episodes monthly.

## **B) Published abstracts on Sexsomnia (publications 9-11)**

**9) Familial sexsomnia for the first time has been reported involving a father and a son in an abstract from Australia (Kennedy *et al.*, 2010).**

The son, 28 years-old, along with his female partner, described a history of abnormal sleep-related sexual behavior consisting of "rough automaton-like intercourse that usually occurred about 60 to 90 minutes after they retired for sleep." The frequency of this sexsomnia was almost nightly and was not influenced by scheduled sexual intercourse before sleep onset.

"Sleep hygiene" interventions were initially attempted for two months, but were ineffective for controlling the sexsomnia. The second treatment option that was tried for 20 days was a 1.0 mg dose of clonazepam at bedtime, which proved to be efficacious in completely stopping the sexsomnia.

One month after the patient was successfully treated, his father, 58 years old, presented at the same clinic with a lifelong history of sexsomnia. His condition was also subsequently treated successfully with a 1.0 mg bedtime dose of clonazepam.

vPSG studies in both the son and the father reportedly were consistent with slow-wave sleep parasomnias, but without any details provided. Both patients had little or no memory of events, and were difficult to wake up.

**10) A published abstract from Brazil provided the first description of sexsomnia with Parkinson's disease (PD) in four patients, but without age or gender being stated (Neto *et al.*, 2012).**

All four patients had amnesia for the sexsomnia, and the bed partners reported that the sexsomnia involved sexual intercourse, sexual vocalization and/or masturbation during sleep.

Of note is that in all four cases, the sexsomnia began with the initiation, or with dose increase, of pramipexole therapy of PD. Also, two of the four patients had impulse dyscontrol disorders during wakefulness, involving hypersexuality, compulsive eating, and excessive spending--all of which have been previously reported side effects of dopamine receptor agonist therapy of PD and RLS. Therefore, two of the four patients treated with pramipexole developed impulse dyscontrol disorders during

sleep and wakefulness as behavioral side effects. Additionally, RLS, OSA, RBD, and insomnia (related to depression) were documented in these patients. vPSG data were not given in the abstract. None of the patients had a prior history of parasomnia or sexual disorder. This abstract did not contain sufficient information to be included in Table 1.

**11) A recently published abstract from a tertiary care sleep clinic in London, United Kingdom reported on the largest case series to date on sexsomnia (Muza *et al.*, 2015).**

This was an observational case series study of individuals presenting during a six-year period (2008-2014) with symptoms suggestive of sexsomnia. Of the 41 patients identified, the mean age was 32 years (range, 22-50 years); 90.2% (n=37) were male, and 9.8% (n=4) were female. Most were married and were Caucasian. The sexsomnia behaviors predominantly involved masturbation in 3 of the 4 females; and sexual intercourse, fondling and groping in 22 of the 37 males. In 11 cases there was aggression and violence, with forensic consequences in one case. Amnesia was reported by all patients. Self-reported triggers for sexsomnia episodes included sleep deprivation (n=8); stress (n=6); contact with a bed partner (n=2); and alcohol use (unspecified amount; n=3). 73.2% (30/41) of patients had a past history of another parasomnia, mainly non-REM sleep parasomnias such as SW and sleeptalking. Forty of the 41 patients underwent an overnight vPSG, and in 70% (28/40) there were sudden arousals from N3 (slow wave) sleep, strongly supporting the clinical history of a non-REM sleep parasomnia. Sexual behaviors did not occur during the vPSG studies. In the remaining 30% (12/40) of patients, positive findings during the vPSG included loss of REM sleep atonia, EEG epileptiform features, and mild OSA (no data were provided). However, the clinical correlates, if any, of the latter three positive findings were not provided in the abstract. Virtually all the findings reported on the 41 patients in this abstract from a single sleep center are highly congruent with the cumulative clinical profile of sexsomnia involving 53 patients reported as single cases and case series in the world-wide, peer-review literature on sexsomnia, as summarized and described in this manuscript and in table 1. Since most of the quantitative



findings contained in table 1 summarizing the world literature through 2015 were not contained in the just-described abstract by Muza *et al.* (2015), findings from the latter abstract were not included in table 1. However, the following updated data in the world literature on sexsomnia, incorporating the data from the Muza *et al.* (2015) abstract, are as follows: i) n=90 reported cases; ii) male predominance: 82% males (n=74), 18% females (n=16); iii) mean age at evaluation, 32-35 years; iv) amnesia for the sexsomnia in 97.8% (88/90); v) aggression and violence during sexsomnia episodes in 32% (29/90); vi) forensic consequences in 14% (13/90). The 4 reported cases of sexsomnia emerging with Parkinson's disease (Neto *et al.*, 2012) are excluded from this analysis on account of insufficient data being provided, except for there being a total of 94 reported cases of sexsomnia in the world literature, with 97.9% (92/94) amnesia for the sexsomnia.

It is important to emphasize that all these cases represent individuals who presented for medical evaluation and therapy, primarily at sleep clinics, on account of sexsomnia-related complaints. A separate issue is the range and prevalence of sexsomnia behaviors in society at large, and their negative and/or positive consequences.

A rare example of sexual behavior during sleep associated with REM Sleep Behavior Disorder (RBD, a parasomnia of dream-enactment) has been briefly mentioned: "In one patient, a behavior resembling sexual intercourse with an imaginary partner and accompanied by a disgusting comment occurred on a single night, as reported by his wife". (Fernandez-Arcos *et al.*, 2016). And in the RBD section of the International Classification of Sleep Disorders, 3rd Edition (American Academy of Sleep Medicine, 2014), sexual/sexualized behaviors were briefly mentioned: "Rarely there can be smoking a fictive cigarette, masturbation-like behavior, pelvic thrusting, and mimics of eating, drinking, urinating, and defecating. The eyes usually remain closed during an RBD episode, with the person attending to the dream action and not to the actual environment."

### **A case of sexual hypnagogic hallucinations with out-of-body experience**

Since parasomnias involve abnormal sleep related experiences besides abnormal sleep related behaviors, a case study from Brazil on abnormal sleep related sexual experiences at sleep onset will be described (Coelho *et al.*, 2011). A 46-year-old man presented with a six-year history of sexual hypnagogic hallucinations (HH)—and "out-of-body-experiences" triggered by the start of sex with his wife, mainly in the evenings. While floating up in air, he had an out-of-body experience (OBE) in which he was seeing the sex he was having with his wife in bed at that moment—but he had already stopped the actual sexual activity with his wife in bed. Therefore, he was hallucinating the sex with his wife, since the sex had stopped when he started hallucinating. The OBE & sexual HH had been present continually for six years.

His wife repeatedly complained, since the sexual activity with her husband would be interrupted when he started to have sexual HH and OBE. For the husband, the sexual activity with his wife in bed continued without interruption since he had a rapid, fluid transition from actual sex to hallucinatory sex (combined sexual HH/OBE), as an abnormal narcoleptic brain-mind state, during a wake-sleep transitional period that was initiated by actual sexual activity.

A diagnosis of narcolepsy-cataplexy (NC) was established after a formal sleep medicine consultation and sleep lab studies (vPSG and a daytime multiple sleep latency test that confirmed excessively short sleep latencies across five nap opportunities, with REM-onset sleep during all five naps), and the determination of a pathologically low CSF Hypocretin-1 level. Therapy of NC with daytime methylphenidate (and subsequently with modafinil) and bedtime amitriptyline was highly effective in controlling excessive daytime sleepiness and the sexual HH and OBE.

### **II) Ictal Sexsomnia**

An additional case of Epileptic Sexsomnia has been reported (Pelin *et al.*, 2012) since the 2007 report by Schenck *et al.* Also, during this subsequent time period a case of bupropion-induced ictal orgasmic sexsomnia has been reported (Şengül *et al.*, 2014). Furthermore, a third case of presumed ictal sexsomnia has been reported, in a postmenopausal woman (Demir, 2014). Therefore, three additional cases of ictal

(or presumed ictal) sexsomnia have been reported since 2007 (all from Turkey), for a total of 10 cases in the world literature.

The additional case of epileptic sexsomnia (Pelin *et al.*, 2012) involved a 31-year-old married man with an eight-year history of masturbation during sleep that was embedded in a complex set of wakeful and sleep related epileptic symptoms, with the sleep related symptoms, besides masturbation, also including laughing, unintelligible vocalizations, clapping his hands and banging on the wall 3-4 times nightly at any time during the sleep period, and with duration of approximately 15 minutes. It was virtually impossible to abort these episodes, which sometimes emerged from daytime naps. Video-EEG (not vPSG) documented an episode of masturbation during sleep associated with hypersynchronous delta waves (2-3 Hz) that were most prominent in the anterior temporal and temporal regions. There was no epileptiform activity detected during the sleep masturbation. However, right temporal EEG neuronal hyperexcitability was detected in periods during sleep apart from the masturbation episode. The initiation of carbamazepine therapy (in conjunction with prior topiramate therapy) completely stopped the sleep related masturbation and the other abnormal sleep related behaviors. He then developed what is commonly referred to as the "temporal lobe personality disorder" that is often found in "temporal lobe epilepsy" (complex partial seizure disorder), characterized by suspiciousness, jealousy, "emotional viscosity", irritability, and aggression that was finally controlled with the addition of aripiprazole therapy to his medication regimen. The patient had a completely negative premorbid psychiatric, neurologic and medical history.

Although in this case frank EEG epileptiform activity was not documented during the recorded sleep masturbation episode, the most compelling reasons that the authors provided, and also as contained in the case description, for why temporal lobe epilepsy (rather than a parasomnia) was the most likely etiology for the sleep related masturbation included: i) multiple nightly episodes; ii) right temporal neuronal hyperexcitability during sleep; iii) no prior parasomnia history, such as SW, sleep terrors, sleeptalking, etc.; iv) presence of the typical "temporal lobe personality disorder"; v) prompt and full response to the anticonvulsant

carbamazepine (a first-line drug for temporal lobe seizures). Also, in regards to the "hypersynchronous delta EEG activity" (HSD) documented during the sleep masturbation episode in this patient, although a number of authors over the years have touted this finding to be a common, specific, and virtually diagnostic feature of a non-REM parasomnia, in fact HSD should now be regarded as a non-specific finding that is also found in other (non-parasomnia) conditions, including respiratory arousals from sleep (Pressman, 2004). With the patient just described with epileptic sexsomnia, the HSD activity documented predominantly in the anterior temporal and temporal regions may have been a marker of epileptic activity related to the right temporal neuronal hyperexcitability as CNS manifestations of temporal lobe epilepsy.

The case of bupropion-induced orgasmic seizure (Şengül *et al.*, 2014) was translated from Turkish to English for this manuscript by S. Cankardeş, Istanbul:

"RA is a 67 year-old, high-school graduate, married woman. She is a housewife with two children and living with her husband. She came to our clinic with her husband one year ago. Her complaints were anxiety, worries, tediousness, insomnia, irritability, claustrophobia and being unable to do the housework. These complaints had been present for six months and they had worsened. She had presented to psychiatry clinics several times previously. She was taking sertraline 100 mg/day, and alprazolam 1mg/day.

Mental status examination revealed motor symptoms of anxiety (frequent wringing of the hands, swinging her feet, inability to sit still), and irritability. She complained of insomnia and there were depressive themes in her thought content. Generalized anxiety disorder was diagnosed. Routine blood chemistry and a complete blood count were normal. Sertraline dose was reduced to 50mg/day, and the alprazolam dose was reduced to 0.5 mg/day. Venlafaxine therapy, 37.5 mg/day, and quetiapine therapy, 25 mg/day, were started. During follow-ups, the venlafaxine dose was gradually increased to 150 mg/day, and the quetiapine dose was increased to 100mg/day. Sertraline and alprazolam were discontinued. The patient achieved remission for one year with this combined venlafaxine and quetiapine therapy. Because of weight gain (in one year her weight increased from 89 kg to 95 kg) and sedation, the quetiapine dose was reduced 25



mg/day. However, due to her complaints of anergy, lack of motivation, sedation and weight gain, the dose of venlafaxine was reduced to 75 mg/day, and bupropion HCl therapy, 150 mg/day, was added. Venlafaxine was gradually reduced to 37.5 mg/day, and then discontinued after a month. The dose of bupropion HCl was increased 300mg/day. The motivation and level of functioning of the patient increased, she began to lose weight, and the sedation was reduced. However, during the third month of bupropion therapy, 300 mg/day, the patient started to complain of spontaneous orgasms every night at any time during sleep, without any stimulation or sexual dream content. Brain MRI was normal. An EEG [presumably a waking EEG, without a sleep component being mentioned], performed with a preliminary diagnosis of ictal orgasm, revealed sharp slow wave activity in both frontal and left temporal lobes. These findings indicate a possible frontal attack. Bupropion was then discontinued. The patient's sleep orgasm frequency went down to once every two weeks, and disappeared in three months. A subsequent EEG, performed when she was no longer taking bupropion, did not show any epileptiform activity."

In this case there was apparently no history of prior seizures or head trauma, nor any prior history of parasomnia, although these negative findings were not explicitly stated. There was also no comment about any family history of epilepsy.

Bupropion has been reported to induce seizures (generally grand mal seizures) during wakefulness, but usually at doses of 400-450 daily (the highest allowed dose). In the case reported above, the (presumably ictal) sleep orgasms occurred while the patient was taking 300 mg/day of bupropion (and also 25 mg/day quetiapine). However, sleep is a state that is known to facilitate seizures on account of increased synchronized EEG activity, and so the combination of the sleep state and the 300 mg bupropion dose (presuming no prior history of seizures) may have conspired to lower the seizure threshold and produce seizures, with the only seizure manifestation being ictal orgasm during sleep, without any waking ictal manifestation. The specificity of sleep-related orgasm as the sole manifestation of recurrent, medication-induced, ictal events is difficult to explain.

The third case of (presumed) ictal sexsomnia, published as a letter-to-the-editor (Demir, 2014), was also translated from Turkish to English for this manuscript by S. Cankardeş, Istanbul:

"The patient is a 69-year-old, married housewife. She suffered from spontaneous orgasms during sleep for 12 years, but had never sought help because of feeling ashamed. She finally went to a gynecologist because her orgasms started to be severe enough to rupture the veins in her eyes, and after each orgasmic sexsomnia episode she had whole body pain. She reported that upon awakening there were contusions on her genitals and on her body. Immediately before these sleep-related orgasms she would usually have a dream in which she was climbing a tree and when climbing down the tree her vagina would be rubbed on the tree. Her orgasmic sexsomnia attacks occurred once a week, and at most twice weekly.

The gynecologist referred her to a psychiatrist. Mental status examination did not reveal any psychopathology. Complete blood count, blood chemistry, thyroid function tests, and serum lipid profile were all normal, along with serum levels of vitamin B12, folate, estradiol, follicle stimulating hormone, luteinising hormone, prolactin, and testosterone; urine test results were also normal. The presumptive diagnosis was ictal orgasm during sleep. MRI of the brain was normal. An EEG did not show any epileptiform activity. Polysomnography was not conducted on account of very limited availability in the province where she resided. Clonazepam therapy, 1 mg at bedtime, was started, and at 3-month follow-up there was no further complaint of orgasm during sleep.

During her clinical interview, she reported being sexually normal in her waking life, with the ability to experience both clitoral and vaginal orgasms. She denied masturbating. On account of her husband's neurologic disorder, they had not engaged in sex for the past 15 years. Orgasm episodes during sleep had emerged at any time of the night.

Discussion: When compared with other case reports in the literature, our case is the oldest reported patient with sexsomnia. Although the patient's EEG showed no abnormality, this negative finding does not completely eliminate epileptic seizures as the cause of orgasmic sexsomnia. Because the patient had benefit from clonazepam [a

benzodiazepine anticonvulsant], the diagnosis of epileptic seizures could not be excluded."

Comments: The recurrent, stereotypical dream occurring before (and/or during) most of the sleep orgasms strongly suggests a nocturnal seizure rather than a parasomnia, especially since dreaming with sexsomnia (as a parasomnia) has very rarely been reported, and when it has been reported, there was never a stereotypical quality to the dream. In this case, the woman was always climbing a tree in her recurrent dream, and would always rub her vagina on the tree on the way down, which was followed by an orgasm during sleep (or the dream occurred during the orgasm), with subsequent awakening. Therefore, the sequence of symptoms associated with the orgasm (followed by awakening with immediate realization of having had an orgasm during sleep) began with a stereotypical dream involving a genital area (*viz.* vagina) being rubbed against a tree.

The author's claim that this case involved the oldest reported patient with sexsomnia is correct for either sexsomnia or ictal sexsomnia. Furthermore, the author's comment that although the patient's EEG showed no abnormality, this negative finding did not eliminate epileptic seizures as the cause of orgasmic sexsomnia is also true, and calls attention to the important point that seizures (*i.e.* ictal events) are primarily a clinical diagnosis based on the patient report of symptoms, and a negative cortical EEG (especially during wakefulness, when for this patient the clinical events always occurred during sleep) does not diminish the probability of nocturnal seizures causing the sleep orgasm.

The full response to clonazepam does not clarify the diagnosis, since clonazepam is an anticonvulsant that also controls parasomnias, including sexsomnia. So based on the treatment response, her sleep orgasms could have been a parasomnia or a seizure event, with the same medication being able to control either abnormality.

It appears that this was the first parasomnia that this 69-year-old woman had experienced in her life, beginning at the age of 57 years, although there was no explicit mention in the publication about there being a negative past sleep history. If true, then this would be extremely unusual for sexsomnia (as a parasomnia), since in the updated review of the

world literature reported herein, sexsomnia predominantly emerges in patients with either (i) an established, longstanding history of multiple parasomnias (up to five in some patients), and then sexsomnia becomes the latest parasomnia to appear in these patients, or (ii) or with obstructive sleep apnea, and the successful therapy of sleep apnea (with nasal CPAP) also controls the sexsomnia. But in this case, there was no mention of snoring or daytime sleepiness or other signs suggestive of sleep apnea. Therefore, her history was very atypical for the two most common clinical scenarios reported with sexsomnia, which also lends support to the diagnosis of ictal orgasmic sexsomnia, together with the other reasons stated above.

Additionally, this patient eventually ruptured superficial ocular veins and had whole body pain immediately after her sleep orgasms, which have never been reported in sexsomnia. These symptoms are extreme for orgasms during wakefulness and for sleep orgasms (as a parasomnia), and are much more consistent with an ictal (epileptic) event, with generalized increased body pressure from tonic seizures being suspected to cause the whole body pain and the ruptured eye veins, particularly in the context of having a stereotypical dream involving her vagina immediately before (and/or during) most of the sleep orgasms. Therefore, the preponderance of the clinical information contained in this published letter strongly supports the diagnosis of ictal sleep orgasm more than sexsomnia as a parasomnia. This would also include the recurrent, stereotypical dream that immediately preceded the onset of sleep orgasms, since stereotypical dreams and nightmares are often associated with nocturnal temporal lobe seizures (Silvestri *et al.*, 2004), and can be considered as "seizure equivalent" experiences. Various other types of abnormal dreams and dream-enacting behaviors associated with nocturnal seizures have been reported, as reviewed (Schenck, *et al.*, 2002). Unfortunately, in this case, polysomnography with full EEG could not be performed, and so we don't have the complete data available, and a definitive diagnostic basis of the sleep orgasm remains indeterminate.

## Results and Discussion

### Novel findings from the additional Sexsomnia cases described herein



The 22 additional cases of sexsomnia described above both reinforce and further clarify the clinical profile of sexsomnia cases compiled and categorized in the first classification of sleep and sexual disorders (Schenck *et al.*, 2007). Moreover, nine novel findings related to sexsomnia were found, which expands knowledge on sexsomnia and helps point the way to future clinical research.

The nine novel findings on sexsomnia include:

- 1) Spontaneous sleep orgasms as parasomnia events, in two female cases (Özcan *et al.*, 2012). Previously reported cases of sleep orgasms involved ictal sexsomnia events, as reviewed (Schenck *et al.*, 2007), and also as described above (Demir, 2014).
- 2) Sexual dreams during sexsomnia episodes, in two cases (Schenck *et al.*, 2008; Della Marca *et al.*, 2009).
- 3) Medication (SSRI)-induced sexsomnia (Krol, 2008).
- 4) Successful SSRI therapy of sexsomnia (Bejot *et al.*, 2009).
- 5) Sexsomnia with a history of sexually traumatic events in adolescence, in two female cases, who both demonstrated non-REM sleep parasomnias during their vPSG studies, rather than a nocturnal sleep related psychogenic dissociative disorder (Bejot *et al.*, 2010).
- 6) Sexsomnia associated with Parasomnia Overlap Disorder, in two cases, with sexsomnia embedded in a longstanding, complex set of five parasomnias (REM sleep behavior disorder and non-REM parasomnias) (Cicolin *et al.*, 2011).
- 7) Sexsomnia emerging with a change in shift-work schedule, indicating that a circadian rhythm disturbance can trigger sexsomnia in a predisposed individual (Arino *et al.*, 2014).
- 8) Familial sexsomnia, affecting a son and father (both adults) (Kennedy *et al.*, 2010), and a daughter and mother (both adults) (Özcan *et al.*, 2012).
- 9) Sexsomnia emerging with Parkinson disease, in four patients, with the sexsomnia emerging with initiation or dose increase of pramipexole therapy of the PD, and in two patients the sexsomnia also emerged in tandem with wakeful disinhibited behaviors (Neto *et al.*, 2012).

### Additional clinical findings

In the clinical setting of a sleep disorders center, the two most common causes of sexsomnia are i) a non-REM sleep parasomnia (Disorder of Arousal), consisting of Confusional Arousals (CAs) in most cases and SW in a lesser number of cases; there is usually a longstanding history of parasomnias, often with a childhood-onset, preceding the emergence of sexsomnia, as discussed above; and ii) Obstructive Sleep Apnea (OSA) triggering CAs with associated sexsomnia. The typical history involves the onset of sexsomnia with the onset or increase of snoring and observed apneas, along with daytime sleepiness. The strong association of sexsomnia with CAs, in which the individual engages in sex with the bed partner (and/or oneself) and does not leave the bed to seek a sex partner, is in line with what is known about Disorders of Arousal with problematic (including legal) consequences, viz. the predominant role played by physical proximity and contact (Pressman *et al.*, 2007a). As reported herein, only 6.1% (3/49) of reported cases of sexsomnia involved SW episodes with sexsomnia. In 93.9% (46/49) of reported cases, the affected individuals remained in bed during CAs when they engaged in sexsomnia. Also, the role of OSA triggering CAs with sexsomnia is in line with what is known about sleep-disordered breathing arousal reactions predisposing to DOA parasomnia events during non-REM sleep (Espa *et al.*, 2002). The one case reported and discussed herein of changes in shift work schedule (as a circadian rhythm disturbance) triggering repeated sexsomnia episodes (Arino *et al.*, 2014) is similar to the release of another instinctual behavior during sleep, viz. eating, that can be triggered by a circadian rhythm disturbance (Schenck *et al.*, 1993). This supports the current concept of circadian rhythm "misalignments" between the "master clock" in the hypothalamic Suprachiasmatic Nucleus and the recently discovered peripheral clocks distributed throughout the major organs (e.g. alimentary organs) and tissues (e.g. adipose) (Summa *et al.*, 2015), thus promoting the inappropriate timing in the release of instinctual behaviors, such as eating and sex during sleep--which is another instinctual behavior.

Problems with sexsomnia include the following: i) disrupting the sleep of the bed partner; ii) physical injury to the bed partner or to oneself from aggressive sexual behaviors; iii) psychological disturbance to the bed partner





from offensive sleepsexalking; iv) psychological disturbance to the bed partner from the inappropriate time of sex, the inappropriate type of sex, and the non-consensual nature of the sexual behaviors (since the bed partner is asleep).

Some of the negative psychological consequences for the bed partner include shock, worry, alarm, anger, annoyance, and bewilderment. Also, there can be accusations of sexual assault/rape, even in a marriage. Some of the psychological disturbances to the sexsomnia include shame, guilt, confusion, and even despair experienced within the context of amnesia for the event: being told about one's objectionable, involuntary, and even aggressive sexual behaviors during sleep after awakening in the morning can be very disturbing. And there is often a major negative impact on a marriage or other intimate relationship.

Nevertheless, as previously reported (Schenck *et al.*, 2007), sexsomnia can have some positive consequences for the bed partner together with the negative consequences. Sex can be experienced as more pleasurable, even "kinky" in a positive manner. Sex can be less hurried than waking sex. The sexsomnia can be a gentler and more amorous lover, and more oriented toward satisfying his partner when he is asleep compared to when he is awake. Unfortunately, the sexsomnia is not aware of these positive sexual behaviors because he is asleep and has subsequent amnesia for these behaviors.

To date, there has not been any reported association of sexsomnia with increased sex drive, sexual deprivation, or sexual perversion (paraphilia).

Besides pharmacotherapy for sexsomnia related to a non-REM parasomnia, or nCPAP therapy of OSA, the clinician should consider referral of the patient and spouse/significant other to a psychologist or psychiatrist for one of two reasons (or both): i) explore the marital/interpersonal relationship as a possible contributing factor to the sexual parasomnia; ii) optimally address any adverse consequences (personal and interpersonal) from the sexsomnia.

The almost universal amnesia associated with sexsomnia, along with the almost universal bed partner-driven clinical referral, may help explain the occasional patient refusal to engage in the proposed therapy. There may also be various

degrees of denial, apart from the amnesia, of having this condition, which can be reflected by the refusal to initiate therapy.

The issue of penile erection (NPT--nocturnal penile tumescence) associated with sexsomnia needs to be addressed. In a number of reports described above, the female partner observed penile erections in the male sexsomnia during the sexual activity, which included at times intercourse with climax. However, NPT is a well-documented and reportedly exclusive REM sleep phenomenon, which poses an explanatory problem, since sexsomnia is predominantly a non-REM sleep related disorder. This quandary is carefully addressed by Andersen *et al.*, 2007 from a basic science-clinical medicine perspective. The authors propose that sexsomnia is an arousal phenomenon *from* non-REM sleep, and not during non-REM sleep, with NPT (and vaginal lubrication) emerging in the context of other autonomic nervous system (ANS) activations during multi-faceted arousals, such as enhanced cardiorespiratory responses, sweating, etc. This arousal-driven ANS activation from non-REM sleep could help explain the penile erections (and clitoral/vaginal engorgement) with sexsomnia as a Disorder of Arousal from non-REM sleep.

The curious and striking finding of male predominance in sexsomnia in the peer-reviewed medical literature needs to be better understood and explained, since one possible explanation is referral bias for clinical evaluation. Do males and females in the United States and Western Europe (the geographical regions represented in most of the reported cases) have different thresholds for identifying sexsomnia as a problem in their bed partners, and do they have different personal criteria for determining when clinical intervention should be sought? What are the boundaries for normal and abnormal sexual behavior surrounding sleep? These are mainly bed-partner related questions, since the sexsomnia are usually amnesic for their episodes. Psychological, interpersonal, sociological, cultural and religious factors need to be considered in this discussion.

The case of sexual hypnagogic hallucinations with OBE emerging as a longstanding symptom of narcolepsy-cataplexy (Coelho *et al.*, 2011) calls attention to a related sexual narcoleptic condition called "orgasmic cataplexy" in which the state of orgasm (an intense emotional state) triggers an immediate

cataplectic response, with generalized or focal muscle paralysis lasting seconds or minutes, and which can be very disturbing to the affected individual and the partner, until they are educated about the benign, albeit peculiar, basis for this phenomenon.

### Epidemiology of Sexsomnia

An epidemiologic study was published from Norway that estimated lifetime and current prevalence of various parasomnias in the general population, defined as the person having experienced the specific parasomnia at least once during the previous three months (Bjorvatn *et al.*, 2010). This was a population-based, cross-sectional study. One thousand randomly selected adults (51% female), 18 years and above, participated in a telephone interview. Lifetime and current prevalence of sexual acts during sleep were 7.1% and 2.7%, respectively. About 12% of all responders reported having five or more parasomnias (reminiscent of the study of Cicolin *et al.*, 2011, in which the two sexsomnia patients each had five parasomnias). The authors cautioned that the data needed to be interpreted with caution due to methodological issues, such as a low response rate to participate in the telephone interview, and the single questions used in the survey.

The frequency of sexsomnia in a general sleep clinic patient population was reported for the first time in an abstract from Canada (Chung *et al.*, 2010). In this study, sexsomnia was defined as a parasomnia-like behavior characterized by individuals performing sexual acts (e.g., masturbation, sexual intercourse with a bed partner) during sleep. A retrospective chart review of patients undergoing PSG evaluation was conducted. Patients had been asked about various sleep-related symptoms. Patients answering "yes" to having initiated sexual activity with a bed partner while asleep were considered to have symptoms of sexsomnia. Charts from 832 consecutive patients (428 males and 404 females) were reviewed. The frequency of reported sexsomnia was 7.6% (63/832; males, 11.0% vs. females, 4.0%). Only 6% of patients reporting sexsomnia also reported other symptoms of parasomnia, which is highly divergent from the cumulative published clinical reports on sexsomnia, and is difficult to explain. However, those patients endorsing sexsomnia did admit to greater use of illicit drugs (sexsomnia,

15.9% vs. no sexsomnia, 7.7%) and of alcohol use (sexsomnia, 41.3% vs. no sexsomnia, 27.4%), but statistics on these differences were not reported. Although this study had major limitations on account of its retrospective design, the results on illicit drug and alcohol use being more common in patients reporting sexsomnia merit further attention in future prospective studies.

Internet-based surveys of sexual behaviors during sleep provide another useful method for better understanding sexsomnia, its problems, its contributing and associated features, and its consequences (negative and positive). To date, three internet-based surveys have been published (Mangan, 2004; Trajanovic *et al.*, 2007; Mangan *et al.*, 2007), with the first two studies previously being cited and discussed (Schenck *et al.*, 2007), although the second study was cited as an abstract prior to its publication as a peer-reviewed journal article. In the third published study on internet-based research on sexsomnia (Mangan *et al.*, 2007), two sources of data were collected from the website <Sleepsex.org>. First, among visitors to this website, there were 157 responders to a link for completing a 28 item web-based survey on sexsomnia. Second, there were 69 responders to links for the same website sent by email to 409 visitors to the <Sleepsex.org> website, a 16.9% response rate. Among the web-based responders, 74% were male, and among the email responders, 59% were male. Combining these two data sets, 69% of all responders were male. While acknowledging the limitations of this research design, such as the lack of direct clinical evaluations, the authors pointed out the following strengths of their web-based survey: "it supported access to a population that is often too ashamed or scared to present in a clinical setting and second...it provided easy access to persons with a rare condition...Also, since sexsomnia sometimes involves adults coming into contact with minors...the legal implications of reporting this to a health care provider are serious...In short, given the increased sense of anonymity that the Web-based survey provided, people probably disclosed facts about their experiences that they might not have revealed in a face-to-face interaction." Furthermore, the authors stressed the ease of reaching participants across the internet. This should encourage web-based surveys across countries that could shed important new light on cultural, gender, and other differences related to sexsomnia.



A recent review article has been published in Turkish that touched on epidemiologic issues (Sarisoy *et al.*, 2014). Selected passages from that article were translated into English by I. Dedecan, Istanbul: "Page 81: It is difficult to detect the actual epidemiology of this disorder because (1) it is not well-known by health-workers, (2) diagnostic criteria are not clear enough, (3) practitioners other than sleep medical specialists are not looking for the characteristic symptoms of the disorder while they are getting the patient's medical history, (4) the patient or the partner are not likely to share the sexual symptoms of the disorder with the doctor because they are ashamed (Yilmaz, 2011)"..."Page 82: Approach to the patient with sexsomnia suspicion: It is assumed that there are a lot more cases of sexsomnia than the detected ones. When faced with a case of atypical sexual behaviors that are harmful to himself/herself or other people, the incidents should be examined thoroughly. The patient and the family should be evaluated about the present and past sleep disorders. The information should be gathered from the bed partner or the family: timing of the sexual behavior, frequency, any related traumas, level of amnesia, the attitude of the patient when awakened after the incident, the attitude of the patient in regards to past sleep disorders if there are any, and the relation of sexsomnia with daily activities (stress, alcohol, sleep deprivation, etc.). Because of the possibility of the existence of sexsomnia behaviors related to complex partial seizures, EEG should definitely be applied as well as polysomnography (Guilleminault *et al.*, 2002; Pelin *et al.*, 2012)." And lastly from this report, "Maybe the most important necessity about sexsomnia is that the doctor should ask questions to the patient about sexsomnia when he/she is aware of and suspecting sexsomnia. The reason for this is that the patient and/or their partners are not eager to talk about sexsomnia unless they are asked, or if there is a legal issue."

### Forensic aspects of sexsomnia

Recent publications that provide a current update on a new intersecting branch of (sleep) medicine and the law will be discussed. It is evident that there is a pressing need for establishing an international consensus regarding the optimal sleep medicine evaluation of forensic parasomnia cases, including those involving purported sexsomnia.

A book chapter written by a leading expert in the parasomnias forensic field utilized a medicolegal, case-based approach in analyzing putative sexsomnia cases presenting in legal settings (Cramer Bornemann, 2013). The author compiled and integrated his arguments from the following clinical-scientific sources: current clinical knowledge on sexsomnia; the basic and clinical sciences of state dissociation and sleep-related behaviors; the neuroscience of "central pattern generators" and "fixed action patterns" (first described by ethologists); clinical behavioral analysis of each case presented, including associated clinical conditions and influences; the role of *Mens Rea* in forensic analysis; and process fractionation as a tool to assess cognition. Furthermore, the author provided the following contemporary definition of Sleep Forensics: "The application of the principles and tools of neuroscience as applied to somnology and sleep medicine that have been widely accepted under international peer-review to the investigation in understanding unusual, irrational, and/or bizarre human behaviors associated with criminal allegations which is to undergo further examination in a conflict resolution legal atmosphere and/or courtroom." In addition, the author made the following comments on the utility of polysomnography, and of technical scientific data, presented in the courtroom in medicolegal cases, "Even frank sleepwalking during a formal sleep study would only indicate that the individual was a sleepwalker--not that sleepwalking was involved at the time of the crime. Thus, a diagnostic tool as polysomnography would not be temporally associated with any questions related to *mens rea* in a criminal allegation... In a court of law, the undisciplined use of scientific technical data is a real concern especially given the public misperception that science is a field that deals with absolute certainties when in actuality it is a field that reflects probabilities of occurrence."

A recently published abstract from the United States focused on the interface of sexsomnia and sleep forensics (Cramer Bornemann *et al.*, 2014). The reported study gathered data over seven years (2006-2013), in which a sleep forensics team comprised of three sleep medicine physicians known for their contributions to the field of parasomnias were consulted by the legal community to review criminal cases involving a potential sleep disorder (262 total cases). Parasomnias were the

most prevalent sleep disorder subtype implicated (131 cases), of which sleep-related abnormal sexual behavior, i.e. sexsomnia, was the most common condition implicated (103 cases). Virtually all the sexsomnia perpetrators were male (102/103), with an age range of 18-55 years, while gender of the victim was virtually always female (99/103), with an age range of 3-17 years in 70.9% (73/103). 86% of the victims knew the perpetrator as a family member, significant other, or friend. Sexsomnia behavior was divided into three subtypes: i) inappropriate touch, in isolation or combined with touching the breasts/genital regions (n=65); ii) sexual contact, in isolation or combined with oral/genital/anal regions(n=37); and iii) indecent exposure (n=1). Proximity between victim and perpetrator during the course of the behavior was: i) confined to the bed (n=47); ii) confined to the bedroom (n=19); or iii) began outside of the bedroom (n=37).

The authors concluded that this was the first published methodical analysis of parasomnias in a formal medico-legal arena and underscored the forensic implications of violent parasomnias that are common from the perspective of sexual assault. Analysis from such forensics data provides further insight into sleep-related abnormal sexual behaviors to enhance public safety, and provides an important avenue to improve the legal system's understanding, or lack thereof, of these sleep-related conditions.

A peer-reviewed journal article focused on a systematic review of medical-legal case reports on sleep-related violence and sexual behavior in sleep (SBS) (Ingravallo *et al.*, 2014). Nine case reports were found in which SBS (ranging from sexual touching to rape) was the defense used during a criminal trial, and in which information about the forensic evaluation of the defendant was provided. Victims were usually unrelated young girls or adolescents in SBS cases. In most cases the criminal events occurred 1-2 hours after the defendant's sleep onset, and both proximity and other potential triggering factors were reported. Eight of the nine cases resulted in acquittal, with the verdict not stated for the other case. The forensic evaluations widely differed from case to case, calling attention to the need to establish an international consensus on forensic evaluations of forensic parasomnia cases, including those with SBS. The same conclusions and recommendations were stated by different authors in three other recent publications, including an emphasis that sexsomnia should be

recognized as a *bona fide* sleep disorder (Morrison *et al.*, 2014; Banerjee, 2014; Organ *et al.*, 2015). In fact, sexsomnia is a recognized variant of Confusional Arousals (and Sleepwalking) from non-REM sleep in the International Classification of Sleep Disorders, 2<sup>nd</sup> (2005) and 3<sup>rd</sup> (2014) editions, published by the American Academy of Sleep Medicine.

The issue of the use and abuse of alcohol in the context of evaluating both clinical cases of sexsomnia (and other parasomnias), and forensic cases involving purported sexsomnia (and other parasomnias) needs to be addressed. Although the literature at first glance appears to be controversial, the preponderance of valid scientific evidence weighs heavily in favor of stating unequivocally that the abuse of alcohol disqualifies the valid use of the "parasomnia [sexsomnia] defense" in medical-legal cases (Pressman *et al.*, 2007b; Pressman *et al.*, 2013; Pressman *et al.*, 2015). Also, apart from the rare clinical case in which the patient, and importantly also a spouse or "significant other" serving as an observer, report that modest consumption of alcohol on occasion can trigger an episode of parasomnia (including sexsomnia) in a person already demonstrating a propensity for this parasomnia, alcohol use or abuse cannot be considered to be the cause of sexsomnia.

Finally, in the Turkish review article on sexsomnia described above, medicolegal aspects were touched upon (Sarisoy *et al.*, 2014), which will now be described, courtesy of the translation into English by I. Dedecan, Istanbul: "Medicolegal Aspects: When sexsomnia is considered as a type of parasomnia, items of Turkish Criminal Code related to the criminal liability of the sexsomnia patient are listed below:

Item 32, article 2: By 'the person who is less able to recognize one's behaviors is to be sentenced for 25 years instead of for life' and part of their sentence can be diminished.

Item 34: By 'the person who is significantly less able to recognize one's behavior or not be able to understand the legal meaning and consequences of his actions because of a temporary cause, or under the effect of alcohol/drug taken involuntarily is not to be sentenced', and they might not get sentenced at all.

There are some legal cases related to sexsomnia in Turkey too. A case had been sent to



the clinic to be evaluated about whether he had sexsomnia or not for a legal case. After the evaluation and literature review, it was decided that the person did not have sexsomnia. As it was thought that the topic is not well-known (or not known at all) in Turkey, we were encouraged to do this review study.

This study's limitations were that very few cases are published, sexsomnia is not clearly named in diagnostic systems, and there is not enough information about it. There might be legal aspects of sexsomnia as well as its psychosocial aspects. When sexsomnia is suspected, the patient should be evaluated for other possible comorbidities too. Experts should be very cautious when they are diagnosing sexsomnia in order to prevent legal exploitations."

### **Conclusion and Outlook**

Further multi-pronged research is needed across the basic and clinical sciences and other disciplines to better understand the full scope of

determinants (predisposing and precipitating factors) and consequences related to sexsomnia (including epileptic sexsomnia), along with the forensic consequences. For example, the basic research related to the neurobiology of sexual desire (Kim *et al.*, 2013) may apply to a substantial extent to the underlying mechanisms of sexsomnia. The world literature needs to be expanded so that national, cultural, religious, and gender differences related to all aspects of sexsomnia can be better understood, which will also benefit the clinical management of the patients. (The recent review article on sexsomnia published in Turkey, as cited above, is a good example [Sarisoy *et al.*, 2014]). The interpersonal psychology and the sociology of sexsomnia merit careful study in order better understand the dynamics resulting or not resulting in presentation for clinical evaluation and management. The epidemiology of sexsomnia is at present poorly known, and needs to be systematically and comprehensively studied.

*Please see next page for Table 1.*

**Table 1.** Data from 18 Published Cases of Sexsomnia from 2007-2015 Compared and Combined with Data from 31 Previously Published Cases from 1986-2007<sup>1,2</sup>

Category	N=18	N=31	N=49
Gender, % (n)			
Male	66.7% (12)	80.6% (25)	75.5% (37)
Female	33.3% (6)	19.4% (6)	24.5% (12)
Age, years, mean±SD (n)			
Total	39.6± 9.5 (18)	31.9±8.0 (30) <sup>3</sup>	34.8± 9.6 (48)
Male	37.6± 8.9 (12)	32.1±8.5 (24) <sup>3</sup>	33.8± 8.7 (36)
Female	44.2±10.1 (6)	30.8±6.4 (6)	37.5±10.9 (12)
Age, sexsomnia onset, years, mean±SD (n)			
Total	33.0± 5.8 (10) <sup>4</sup>	25.9± 8.7 (17)	28.5± 8.6 (27)
Male	32.1± 6.3 (7)	27.4± 7.9 (15)	28.9± 7.9 (22)
Female	35.5± 5.0 (3)	14.5± 3.5 (2)	27.1±11.9 (5)
Duration, sexsomnia, years, mean±SD (n)			
Total	5.0± 3.5 (8) <sup>5</sup>	9.5± 6.1 (8)	7.3± 5.8 (16)
Male	5.6± 4.1 (5)	8.3± 6.5 (6)	7.1± 5.7 (11)
Female	4.0± 2.6 (3)	13.0± 4.2 (2)	7.6± 5.6 (5)
Sexsomnia behaviors, % of patients (n)			
Masturbation	25.0% (4) (4 female)	22.7% (7) (4 male, 3 female)	23.4% (11) (7 female, 4 male)
Sleep orgasms (spontaneous)	11.1% (2) (2 female)		4.1% (2) (2 female)
Sexual vocalizations, talking, shouting	18.7% (3) (2 female, 1 male)	19.3% (6) (4 female, 2 male)	19.1% (9) (6 female, 3 male)
Fondling another person	31.2% (5) (5 male)	45.2% (14) (13 male, 1 female)	40.4% (19) (18 male, 1 female)
Sexual intercourse/ attempted intercourse	62.5% (10) (8 male, 2 female)	41.9% (13) (13 male)	48.9% (23) (21 male, 2 female)
Total # sexsomnia behaviors	24	40	64
Agitated/assaultive sexsomnia behaviors, % (n)	22.2% (4)	45.2% (14)	36.7% (18)
Sexsomnia with minors, % (n)	5.5% (1)	29.0% (9)	20.4% (10)
Legal consequences, % (n)	5.5% (1)	35.5% (11)	24.5% (12)
Amnesia for sexsomnia,%(n)	88.9% (16)	100.0% (31)	95.9% (47)
Video-Polysomnography,%(n)	83.3% (15)	83.9% (26)	83.7% (41)
Total # parasomnias	32	71	103
Mean # (±SD) per patient (range, 1-5)	1.8± 1.4	2.2± 1.0	2.1± 1.2
Final diagnosis, sexsomnia etiology, % (n)			
Disorder of Arousal (DOA) <sup>6</sup> , % (n)	77.8% (14)	90.3% (28)	85.7% (42)
Obstructive Sleep Apnea	16.7% (3)	12.9% (4)	14.3% (7)
REM sleep behavior disorder	5.5% (1)	9.7% (3)	8.2% (4)
Other conditions <sup>7</sup>	16.7% (3)	-	6.1% (3)
Treatment efficacy, % (n) (controlling sexsomnia)			
Clonazepam at bedtime	75.0% (3/4)	90.9%(9/10)	85.7%(12/14)
Nasal CPAP at bedtime	100.0% (2/2)	100.0% (2/2)	100.0% (4/4)
SSRI (escitalopram)	100.0% (2/2) <sup>8</sup>	-	100.0% (2/2) <sup>8</sup>
SSRI discontinuation	100.0% (1)	-	100.0% (1)
Other therapies <sup>9</sup>	0.0% (0/2)	-	0.0% (0/2)

<sup>1</sup> Schenck CH, Arnulf I, Mahowald MW. Sleep and Sex: What Can Go Wrong? A Review of the Literature On Sleep Related Disorders and Abnormal Sexual Behaviors And Experiences. *Sleep* 2007; 30: 683-702.

<sup>2</sup> Four other cases of sexsomnia (emerging with Parkinson's disease) published after 2007 were excluded from table 1 on account of insufficient data being provided (Neto, *et al.*, 2012). Therefore, a total of 22 new cases of sexsomnia has been published since 2007.

<sup>3</sup> N=1, age not reported (male patient).

<sup>4</sup> Age of onset not known for 8 patients (5 males, 3 females)

<sup>5</sup> Duration not known for 10 patients (7 males, 3 females)

<sup>6</sup> DOA (Non-REM sleep parasomnia): Confusional Arousals, n=13 and n=26 for each group, respectively; Sleepwalking, n=1 and n=2 for each group, respectively. The obstructive sleep apnea (OSA) data are contained within the DOA data, since the OSA events presumably induced confusional arousals that triggered sexsomnia episodes.

<sup>7</sup> Other conditions: SSRI (Serotonin Specific Reuptake Inhibitor)-induced (n=1); combined disorders: RLS (restless legs syndrome)/periodic leg and arm movements of sleep/other abnormal movements of sleep/prior history of NREM parasomnia (SW) (n=1); combined disorders: obstructive sleep apnea, NREM parasomnia, shift work related circadian rhythm disturbance (n=1).

<sup>8</sup> In one of the two patients, escitalopram efficacy was not maintained after two years; substitution of lamotrigine therapy at bedtime was beneficial and maintained at 4-year follow-up [Isabelle Arnulf, personal communication].

<sup>9</sup> Carbamazepine, dopaminergic agonists (n=1); dopaminergic agonist (n=1).

## References

- American Academy of Sleep Medicine. International classification of sleep disorders, 2nd edition. Westchester, Illinois, 2005.
- American Academy of Sleep Medicine. International classification of sleep disorders, 3rd edition. Darien, Illinois, 2014.
- Andersen ML, Poyares D, Alves RSC, Skomro R, Tufik. Sexsomnia: abnormal sexual behavior during sleep. *Brain Res Reviews* 2007; 56: 271-282.
- Arino H, Iranzo A, Gaig C, Santamaria J. Sexsomnia: parasomnia associated with sexual behaviour during sleep. *Neurologia* 2014; 29 (3): 146-152.
- Banerjee D. A case-based review of sleep-related criminal behaviours, treatment & outcomes. *Sleep and Biological Rhythms* 2014; 12: 7.
- Bejot Y, Juenet N, Garrouty R, Maltaverne D, Nicollet L, Giroud M, Didi-Roy R. Sexsomnia: an uncommon variety of parasomnia. *Clin Neurol Neurosurg* 2010; 112 (1): 72-75.
- Bjorvatn B, Gronli J, Pallesen S. Prevalence of different parasomnias in the general population. *Sleep Medicine* 2010; 11:1031-1034.
- Chung SA, Yegneswaran B, Natarajan A, Trajanovic N, Shapiro CM. Frequency of sexomnia in sleep clinic patients. *Sleep* 2010; 33: A226.
- Cicolin A, Tribolo A, Giordano A, et al. Sexual behaviors during sleep associated with polysomnographically confirmed parasomnia overlap disorder. *Sleep Med* 2011;12: 523-528.
- Coelho FMS, Moszczynski A, Narayansingh M, Parekh N, Pradella-Hallinan M. Sexual hypnagogic hallucinations and narcolepsy with cataplexy: a case report. *Sleep Science* 2011;4(3): 110-112.
- Cramer Bornemann MA. Sexsomnia: a medicolegal case-based approach in analyzing potential sleep-related abnormal sexual behaviors. In: Kothare SV, Ivanenko A, eds. *Parasomnias*. New York: Springer Science + Business Media 2013; pp. 431-461.
- Cramer Bornemann MA, Mahowald MW, Schenck CH. Sexsomnia and sleep forensics: the interface between sleep-related abnormal sexual behaviors and the law. *Sleep* 2014; 37: A210.
- Della Marca G, Dittoni S, Frusciante R, et al. Abnormal sexual behavior during sleep. *J Sex Med* 2009; 6 (12): 3490-3495.
- Demir EY. Uykuda Orgazm Olan Postmenopozal Kadın Hasta. [Post-menopausal female patient with orgasm during sleep]. *Düşünen Adam: The Journal of Psychiatry and Neurological Sciences* 2014; 27 (3): 270-271. [Turkish]
- Espa F, Dauvilliers Y, Ondze B, Billiard M, Besset A. Arousal reactions in sleepwalking and night terrors in adults: the role of respiratory events. *Sleep* 2002; 25: 32-36.
- Fantini ML, Corona A, Clerici S, Ferini-Strambi L. Aggressive dream content without daytime aggressiveness in REM sleep behavior disorder. *Neurology* 2005;65(7):1010-1015.
- Fernandez-Arcos A, Iranzo A, Serradell M, Gaig C, Santamaria J. The clinical phenotype of idiopathic REM sleep behavior disorder at presentation: a study in 203 consecutive patients. *Sleep* 2016 (in press).
- Guilleminault C, Moscovitch A, Yuen K, Poyares D. Atypical sexual behavior during sleep. *Psychosom Med* 2002;64(2):328-336.
- Ingravallo F, Poli F, Gilmore EV, Pizza F, Vignatelli L, Schenck CH, Plazzi G. Sleep related violence and sexual behavior in sleep: a systematic review of medical-legal case reports. *J Clin Sleep Med* 2014; 10: 927-935.
- Iranzo A, Santamaría J. Severe obstructive sleep apnea/hypopnea mimicking REM sleep behavior disorder. *Sleep* 2005;28(2):203-206.
- Kennedy GA, Elzo F. Case studies of familial sexsomnia. *Sleep and Biological Rhythms* 2010; 8: A73-A74.
- Kim SW, Schenck CH, Grant JE, Yoon G, Dosa P, Odlaug BL, Schreiber LRN, Hurwitz TD, Pfaus JG. The neurobiology of sexual desire. *NeuroQuantology* 2013; 2: 332-359.
- Krol DG. Sexsomnia during treatment with a selective serotonin reuptake inhibitor. *Tijdschr Psychiatr [J of Psychiatry]* 2008; 50 (11): 735-739. [Dutch]
- Mangan MA. A phenomenology of problematic sexual behavior occurring sleep. *Arch Sex Behav* 2004; 33: 287-293.
- Mangan MA, Reips U-D. Sleep, sex, and the Web: surveying the difficult-to-reach clinical population suffering from sexsomnia. *Behav Res Methods* 2007; 39 (2): 233-236.
- Morrison I; Rumbold JM; Riha RL. Medicolegal aspects of complex behaviours arising from the sleep period: a review and guide for the practising sleep physician. *Sleep Medicine Reviews* 2014; 18(3):249-260.
- Muza RT, Lawrence M, Williams A. The reality of sexsomnia: characteristics and manifestations of sexsomnia. *Sleep* 2015; 38: A250.
- Neto SMA, Pena-Pereira M, Sobreira ES, Chagas M, Fernandes RM, Eckeli AL, Tumas V. Sexsomnia in parkinson's disease patients. *Sleep* 2012; 35: A253-A254.
- Organ A, Fedoroff JP. Sexsomnia: sleep sex research and its legal implications. *Current Psychiatry Reports* 2015; 17(5): 34. doi: 10.1007/s11920-015-0568-y.
- Özcan H, Taşın C, Tandoğan M, ÖZakşit G. Uykü Esnasında Orgazm Olan İki Kadın. [Two women having orgasms during sleep]. *Düşünen Adam Psikiyatri ve Nörolojik Bilimler Dergisi* 2012; 25: 187-188. [Turkish]
- Pelin Z, Yazla E. Abnormal sexual behavior during sleep in temporal lobe epilepsy: a case report. *Balkan Med J* 2012; 29: 211-213.
- Penas-Martinez ML, Guerrero-Peral AL, Toledano-Barrero AM, et al. Sexsomnia: descripción de un nuevo caso. [Sexsomnia: description of a new case]. *Revista de Neurologia* 2008; 47 (6): 331-332. [Spanish]
- Pressman MR. Hypersynchronous delta sleep EEG activity and sudden arousals from slow-wave sleep in adults without a history of parasomnias: clinical and forensic implications. *Sleep* 2004;27(4):706-710.
- Pressman MR. Disorders of arousal from sleep and violent behavior: the role of physical contact and proximity. *Sleep* 2007a; 30: 1039-1047.
- Pressman MR, Mahowald MW, Schenck CH, Bornemann MC. Alcohol-induced sleepwalking or confusional arousal as a defense to criminal behavior: a review of scientific evidence, methods and forensic considerations. *J Sleep Res* 2007b;16(2):198-212.
- Pressman MR, Mahowald MW, Schenck CH, Cramer Bornemann MA, Banerjee D, Buchanan P, Zadra A. Alcohol, sleepwalking and violence: lack of reliable scientific evidence. *Brain* 2013; 136(Pt 2):e229. doi: 10.1093/brain/aws248.
- Pressman MR1, Grunstein RR, Mahowald MW, Schenck CH, Montplaisir JY, Bornemann MC, Zadra A, Buchanan PR. Alcohol and sleep review: flawed design, methods, and statistics cannot support conclusions. *Alcohol Clin Exp Res* 2015;39(5): 941-3. doi: 10.1111/acer.12712.



- Sarisoy G, Özdin S. Seksomnia: Bir Gözden Geçirme (Sexsomnia: A Review). J of Mood Disorders 2014; 4 (2): 80-87. [Turkish]
- Schenck CH, Milner DM, Hurwitz TD, Bundlie SR, Mahowald MW. A polysomnographic and clinical report on sleep-related injury in 100 adult patients. American J Psychiatry 1989a; 146: 1166-1173.
- Schenck CH, Milner DM, Hurwitz TD, Bundlie SR, Mahowald MW. Dissociative disorders presenting as somnambulism: polysomnographic, video and clinical documentation (8 cases). Dissociation 1989b; 2: 194-204.
- Schenck CH, Hurwitz TD, Bundlie SR, Mahowald MW. Sleep-related eating disorders: polysomnographic correlates of a heterogeneous syndrome distinct from daytime eating disorders. Sleep 1991;14(5):419-431.
- Schenck CH, Hurwitz TD, O'Connor KA, Mahowald MW. Additional categories of sleep-related eating disorders and the current status of treatment. Sleep 1993; 16: 457-466.
- Schenck CH, Boyd JL, Mahowald MW. A parasomnia overlap disorder involving sleepwalking, sleep terrors, and REM sleep behavior disorder in 33 polysomnographically confirmed cases. Sleep 1997; 20:972-981.
- Schenck CH, Mahowald MW. REM sleep behavior disorder: clinical, developmental, and neuroscience perspectives 16 years after its formal identification in *SLEEP*. Sleep 2002;25: 120-138.
- Schenck CH, Arnulf I, Mahowald MW. Sleep and sex: what can go wrong? A review of the literature on sleep related disorders and abnormal sexual behaviors and experiences. Sleep 2007; 30: 683-702.
- Schenck CH, Mahowald MW. Parasomnias associated with sleep-disordered breathing and its therapy, including sexsomnia as a recently recognized parasomnia. Somnology 2008; 12: 38-49.
- Şengül MCB, Hancı E. Uyku sırasında orgazm: Bupropiona bağlı bir nöbet olgusu. (Orgasm during sleep: bupropione induced seizure). Pam Med J 2014; 7(2):155-157. [Turkish]
- Silvestri R, Bromfield E. Recurrent nightmares and disorders of arousal in temporal lobe epilepsy. Brain Res Bull 2004;63(5):369-376.
- Sixel-Döring F, Trautmann E, Mollenhauer B, Trenkwalder C. Associated factors for REM sleep behavior disorder in Parkinson disease. Neurology 2011;77(11):1048-1054.
- Summa KC, Turek FW. The clocks within us. Scientific American 2015; 312 (2): 51-55.
- Trajanovic NN, Mangan M, Shapiro CM. Sexual behaviour in sleep: an internet survey. Soc Psychiatry Psychiatr Epidemiol 2007; 42: 1024-1031.
- Yılmaz H. Uykuda Seks Hastalığı. Uyku Fizyolojisi ve Hastalıkları içinde, Kaynak H, Ardış S (editörler). Türk Uyku Tıbbı Derneği Yayını, 2011:345. [Turkish]