

Sleep and Sex: What Can Go Wrong? A Review of the Literature on Sleep Related Disorders and Abnormal Sexual Behaviors and Experiences

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Study Objectives: To formulate the first classification of sleep related disorders and abnormal sexual behaviors and experiences.

Design: A computerized literature search was conducted, and other sources, such as textbooks, were searched.

Results: Many categories of sleep related disorders were represented in the classification: parasomnias (confusional arousals/sleepwalking, with or without obstructive sleep apnea; REM sleep behavior disorder); sleep related seizures; Kleine-Levin syndrome (KLS); severe chronic insomnia; restless legs syndrome; narcolepsy; sleep exacerbation of persistent sexual arousal syndrome; sleep related painful erections; sleep related dissociative disorders; nocturnal psychotic disorders; miscellaneous states. Kleine-Levin syndrome (78 cases) and parasomnias (31 cases) were most frequently reported. Parasomnias and sleep related seizures had overlapping and divergent clinical features. Thirty-one cases of parasomnias (25 males; mean age, 32 years) and 7 cases of sleep related seizures (4 males; mean age, 38 years) were identified. A full range of sleep related sexual behaviors with self and/or bed partners or others were reported, including masturbation, sexual vocalizations, fondling, sexual intercourse with climax, sexual assault/rape, ictal sexual hyperarousal, ictal orgasm, and ictal automatism. Adverse physical and/or psychosocial effects from the sleepsex were present in all parasomnia and sleep related seizure cases, but pleasurable effects were reported by 5 bed partners and by 3 patients with sleep related seizures. Forensic consequences were common, occurring in 35.5% (11/31) of parasomnia cases, with most (9/11) involving minors. All parasomnias cases reported amnesia for the sleepsex, in contrast to 28.6% (2/7) of sleep related seizure cases. Polysom-

nography (without penile tumescence monitoring), performed in 26 of 31 parasomnia cases, documented sexual moaning from slow wave sleep in 3 cases and sexual intercourse during stage 1 sleep/wakefulness in one case (with sex provoked by the bed partner). Confusional arousals (CAs) were diagnosed as the cause of "sleepsex" ("sexsomnia") in 26 cases (with obstructive sleep apnea [OSA] comorbidity in 4 cases), and sleepwalking in 2 cases, totaling 90.3% (28/31) of cases being NREM sleep parasomnias. REM behavior disorder was the presumed cause in the other 3 cases. Bedtime clonazepam therapy was effective in 90% (9/10) of treated parasomnia cases; nasal continuous positive airway pressure therapy was effective in controlling comorbid OSA and CAs in both treated cases. All five treated patients with sleep related sexual seizures responded to anticonvulsant therapy. The hypersexuality in KLS, which was twice as common in males compared to females, had no reported effective therapy.

Conclusions: A broad range of sleep related disorders associated with abnormal sexual behaviors and experiences exists, with major clinical and forensic consequences.

Keywords: Sleep, sleep disorders, sexual behavior/experience, parasomnias, sleep related/nocturnal seizures, NREM sleep parasomnias, confusional arousals, Kleine-Levin Syndrome, polysomnography/sleep EEG, forensic medicine

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INTRODUCTION

THE RECENTLY REVISED *INTERNATIONAL CLASSIFICATION OF SLEEP DISORDERS-2ND EDITION (ICSD-2)* CONTAINS A SUBSTANTIALLY UPDATED SECTION ON THE parasomnias, which encompass sleep related behaviors, emotions, perceptions, dreaming, and autonomic nervous system events during entry into sleep, within sleep, or during arousals from sleep."¹

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Instinctual behaviors are often released inappropriately, as seen with sleepwalking (locomotion), REM sleep behavior disorder (aggression), sleep related eating disorder (feeding), and confusional arousals (sexual behaviors).

A classification of sleep related disorders associated with abnormal sexual behaviors and experiences, to our knowledge, has not been published. We now present such a classification for several reasons. First, there is growing awareness that abnormal sexual behaviors can emerge during sleep, described as "sleepsex," "atypical sexual behavior during sleep" and "sexsomnia."²⁻⁴ The *ICSD-2* recognized this phenomenon as a parasomnia classified as a variant of confusional arousals (and sleepwalking).¹ Second, there is an expanding set of sleep disorders and other nocturnal disorders known to be associated with abnormal sexual behaviors, or the misperception of sexual behaviors. Third, the cause of sleepsex can often be identified after clinical and polysomnographic (PSG) evaluations, and can then be effectively treated. Fourth, the forensic aspects of abnormal sleep related sexual behavior have commanded increas-

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ing attention. Fifth, new data on periodic hypersomnia (Kleine-Levin syndrome), including its range and frequency of abnormal sexual behaviors have recently been published. The question of how to distinguish normal from abnormal sleep related sexual activity also needs to be considered.

METHODS

Computerized literature searches were conducted for English language publications, including foreign language publications with English abstracts. Old Medline (1950-1965), and Medline, Embase, and PsycInfo searches (1966-2006) used the following key words, restricted to humans: Sleep and sex/sexual behavior; parasomnia and sex/sexual behavior; epilepsy/seizures and sexual behavior/hypersexuality; nocturnal frontal lobe epilepsy and sexual behavior; Kleine-Levin syndrome (KLS) and sexuality/hypersexuality. In addition, information on sleep and sexual, or pseudo-sexual (“sexualized”) behavior from peer-reviewed journal articles that were not detected by the computerized literature searches was gathered by the authors. Data were analyzed to categorize and describe the known associations between sleep, sleep related disorders, and abnormal sexual behaviors and experiences. Data on patients with sleepsex parasomnias and with sexual seizures were also tabulated.

Current textbooks on sleep medicine were reviewed for descriptions and citations related to sexual activity during sleep or sexual behaviors associated with sleep disorders. Additionally, the large-scale Kinsey and Hite reports were reviewed.⁵⁻⁹ The Kinsey reports, conducted more than 50 years ago, remain the most comprehensive, systematic studies on sexual behavior in males (n=5,000) and females (n=6,000) that utilized direct person-to-person interviews with a semi-structured format.^{5,6} A subsequent book, with a question-and-answer format, was published by the Kinsey Institute in 1991.⁷ The Hite reports, from 30 and 25 years ago, gathered extensive data on the sexual behavior and experiences of thousands of women and men in the general population by means of mailed anonymous essay questionnaires.^{8,9}

The table of contents and the index in each of the 3 Kinsey reports and the 2 Hite reports (containing only a table of contents) were reviewed to identify all sleep and dream entries related to sexual behavior and experience, including orgasm.

Finally, a comprehensive review of the world literature on KLS, including its association with hypersexuality and deviant sexuality during recurrent hypersomnia phases, was gathered by one of the authors (IA), as described in a separate section on KLS.

RESULTS

Kinsey and Hite Reports

The topic of sleep and sexual behavior was not specifically covered in either of the 2 Kinsey books on males and females^{5,6} or in the subsequent Kinsey Institute book,⁷ apart from considerable attention being paid to “nocturnal emissions” or “wet dreams” in males (Chapter 15: Nocturnal Emissions).⁵ These events were described as being “generally accepted as a usual part of the sexual picture.”⁵ It was also acknowledged that nocturnal emissions, i.e., spontaneous ejaculations during sleep, could be strongly associated with sexual dreams in some people, but rarely or never accompanied by dream recall in other people. Sleep ejaculations

were sometimes reported to occur from daytime naps.

There was no mention of any elicited or spontaneous reports of either sleep masturbation, sleepsex with a bed partner, or sexual sleeptalking in the Nocturnal Emissions chapter. In a discussion of the physiologic origins of the nocturnal emissions, sleep related mechanisms were not mentioned.⁵

Chapter 14 on male masturbation did not cover sleep related (i.e., involuntary) masturbation.⁵ The observation was made that “human sexual behavior represents one of the least explored segments of biology, psychology, and sociology.” Kinsey described his study as a “fact-finding survey” to discover what people do sexually (given the lack of adequate scientific information). First-hand interviews were used to obtain the data. However, sleep related sexual behavior was not addressed (other than nocturnal emissions), despite the claim that “all kinds of persons and all aspects of human sexual behavior are being included in this survey... [without] preconception of what is rare or what is common... the story of the sexual behavior of the American male, as we find him.” The following sexual outlets were systematically addressed in Part III (“Sources of sexual outlet”): masturbation, heterosexual petting, pre-marital intercourse, marital intercourse, extra-marital intercourse, intercourse with prostitutes, homosexual outlet, and animal contacts. Although Part II addressed the topic of “Factors affecting sexual outlet,” in which nine factors were discussed in separate chapters, sleep was not discussed, apart from nocturnal emissions. In the section on “Individual Variation,” Kinsey commented on the broad range of variation in human sexual behavior, which from our current-day perspective most likely also applies to human sexual behavior during sleep.

In Kinsey’s book on female sexual behavior,⁶ the entry for sleep in the index pertained to nudity and also to sexual responses, with the latter referring only to nocturnal sexual dreams and to orgasms; 99% of sleep orgasms in females were associated with sexual dreams.

In the *Kinsey Institute New Report on Sex*,⁷ a question-and-answer format was used, based on letters containing questions sent to the Kinsey Institute. The topic of sleep was not addressed in the title of any of the 19 chapters. In the “involuntary sexual behaviors” section of Chapter 6 (“The Sexual Adult”), the topic of spontaneous nocturnal orgasms in males and females, described to occur during REM sleep, was restricted to five questions and the corresponding answers. The index did not have an entry for “sleep,” but did have 4 entries for “Rapid Eye Movement (REM) sleep, sexual arousal during”; 5 entries for “nocturnal emissions (wet dreams)”; and 3 entries for “nocturnal penile tumescence (NPT).” No question or answer addressed the topic of any other sexual behavior during sleep (involving either an individual alone or together with a bed partner), or sleep vocalizations with sexual content.

The Hite Reports, like the Kinsey Reports, had no information on sleep and sexual behavior in women or men, and did not cover the topic of nocturnal emissions.^{8,9} Sleep was not included among the six questions that discussed the context for achieving orgasms in women. No write-in question discussed orgasm during sleep or dreams. At the end of the questionnaire for women, respondents were asked “is there anything on your mind you would like to speak about which was left untouched by this questionnaire?” Similarly, male respondents were asked at the end of their questioning: “please add anything you would like to say that was not mentioned.”⁹ There was no response in which sleepsex was reported.

Other Reports

Data from a 28-item internet survey of sexual behavior in sleep were gathered over a 3-month period, as recently reported.¹⁰ There were 219 respondents (69% male; mean age, 30.4 yrs, range 15-67 yrs), 92% of whom reported multiple episodes of **sexsomnia**. Sexual intercourse during sleep was reported by 48%. Precipitating factors for sexsomnia included physical contact with another person in bed (64%), stress (52%), fatigue (41%), alcohol use (14.6%), and drug use (4.3%). Sexsomnia with minors during sleep was reported by 5.9% (10 males, 3 females), with legal repercussions occurring in most cases. Almost half (47%) reported having another sleep disorder. No information on degree of recall or amnesia for the sexsomnia was provided.

The relationship between specific sexual activity (i.e., masturbation) and its subsequent effect on sleep has been explored with PSG monitoring in one study.¹¹ This study involved 10 volunteers (5 men, 5 women; mean age, 25.1 years) without sleep complaints, psychopathology, or sexual dysfunction. Three conditions were examined: sleep following masturbation with or without orgasm, and sleep after reading neutral material. The PSG sleep parameters did not reveal any effect of masturbation on sleep. The authors pointed out that a significant sleep effect might have occurred after masturbation or coitus in a more natural (i.e., nonlaboratory) setting, or among older subjects, insomniacs, or people experiencing tension/anxiety before sexual activity, with sexual activity then serving an anxiolytic, and perhaps sleep-enhancing, role. (On the other hand, coitus can also promote difficulty in falling asleep.¹²) The authors also called attention to an anecdotal literature without PSG monitoring that revealed wide individual differences after orgasm, particularly among women, ranging from enhanced sleepiness to hyperarousal. Another study examined the effects of viewing an erotic film during the daytime on the subsequent night's sleep and dreams in 10 young adult men.¹³ No sleep disturbance was documented, but reduced dream recall was found, along with other dream changes. A third study examined the effects of a brief period of sexual arousal, without climax, on sleep related erections (SREs) in 12 men who viewed a sexually explicit video (with associated erections) for 5 minutes before sleep.¹⁴ No adverse effects on SREs were found.

The effect on nocturnal penile tumescence (NPT) and sleep architecture from 10 consecutive days of sexual abstinence followed by a night of sexual intercourse was evaluated in 10 married men, 22-32 years old.¹⁵ No increase in NPT or sleep disturbance was found.

A questionnaire study of nocturnal orgasm in college women focused only on spontaneous sleep orgasms and "sexual excitement in dreams" without any mention of self-induced sleep orgasm (i.e., sleep masturbation) observed by a bed partner or roommate.¹⁶

Among the current sleep medicine textbooks, one has the following listings in the index¹⁷: 1) sexual activity, in arousal disorders and in hypnagogic hallucinations; 2) sexual content of dreams; sex hormones; penile erections; penile tumescence; penis. Another book¹⁸ has one listing for nocturnal penile tumescence (NPT), but no listing for sex/sexual behavior or orgasm. A third book¹⁹ has a listing for sexual activity, sleep related; two listings for sexual abuse, dissociated memories; one listing for NPT; and no listing for orgasm.

Table 1 contains a classification linking sleep related disorders

Table 1—Sleep Related Disorders and Abnormal Sexual Behaviors and Experiences

- I) Parasomnias with abnormal sleep related sexual behaviors (sexual vocalizations/talking/shouting, masturbation, fondling another person, sexual intercourse with or without orgasm, agitated/assaultive sexual behaviors):
 - A) Confusional arousals (with or without obstructive sleep apnea)
 - B) Sleepwalking
 - C) REM sleep behavior disorder (RBD) (?)¹
- II) Sleep related sexual seizures (sexual vocalizations/ moaning/ shouting, masturbation, libidinal hyperarousal, genital arousal, ictal orgasm, sexual automatisms, agitated/assaultive sexual behaviors)
- III) Sleep disorders with abnormal sexual behaviors during wakefulness and wake-sleep transitions:
 - A) Kleine-Levin syndrome (broad range of hypersexual and deviant sexual arousal and behaviors)
 - B) Severe chronic insomnia (increased libido, genital arousal, compulsive sexual behaviors)
 - C) Restless legs syndrome (masturbation, rhythmic pelvic/coital-like movements)
- IV) Special clinical considerations:
 - A) Narcolepsy (compelling sexual hypnagogic/hypnopompic hallucinations and REM-onset dream attacks, cataplectic orgasm)
 - B) Sleep exacerbation of persistent sexual arousal syndrome (genital-sensory sexual arousal without increased libidinal arousal; sexual behaviors)
 - C) Sleep related painful erections and increased sexual activity (increased sexual behaviors—masturbation and intercourse)
 - D) Sleep related dissociative disorders (pelvic movements and other sexualized behaviors, attempted reenactments of past sexual/physical abuse scenarios)
 - E) Nocturnal psychotic disorders (sexual delusions/hallucinations after awakenings)
 - F) Hypersexuality after nocturnal awakenings
 - G) Miscellaneous (naps; [REM] sleep erections and sexual vulnerability; medication-induced states) (masturbation, sexual intercourse, sexual hypnagogic/ hypnopompic hallucinations with or without sleep paralysis)

¹Three patients with histories of abnormal sleepsex were diagnosed with RBD, but polysomnographic (PSG) monitoring did not document any sexual or nonsexual behaviors during REM sleep; only REM sleep without atonia was documented by PSG monitoring.

with a broad range of **abnormal** sexual behaviors and experiences. Each category will be presented, with a focus on objective data, experiential aspects, clinical and legal consequences, and response to treatment.

Parasomnias With Abnormal Sleep Related Sexual Behaviors And Sleep Related Sexual Seizures

Table 2 contains data on 31 published cases of parasomnias and 7 published cases of epilepsy with abnormal sleep related sexual behaviors and experiences. Eight peer-reviewed journal articles and one peer-reviewed abstract were the sources of the 31 parasomnia cases,^{2-4,20-25} and seven peer-reviewed articles were the

Table 2—Data from 31 Published Cases of Parasomnias and 7 Published Cases of Epilepsy with Abnormal Sleep Related Sexual Behaviors & Experiences

Category	Parasomnias (n=31)	Sleep Related Epilepsy (n=7)	Category	Parasomnias (n=31)	Sleep Related Epilepsy (n=7)
Gender, % (n)			Agitated/assaultive sleep related sexual behaviors,%(n)	45.2% (14)	14.3% (1)
Male	80.6% (25)	57.1% (4)	Sleepsex with minors, %(n)	29.0% (9)	0%
Female	19.4% (6)	42.9% (3)	Legal consequences from sleepsex, % (n)	35.5% (11)	0%
Age, years, mean ±SD (n)			(2 with adults; 9 with minors ⁴)		
Total	31.9 ± 8.0 (30) ¹	37.7 ± 8.5	Other consequences, % (n)		
Male	32.1 ± 8.5 (24)	34.0 ± 3.5	I) Adverse: Physical		
Female	30.8 ± 6.4 (6)	42.7 ± 11.6	Self	6.4% (2)	71.4% (5)
Age, sleepsex onset, years, mean ±SD (n)			Other	61.3% (19)	0%
Total	25.9 ± 8.7 (17) ²	32.0 ± 9.6	Adverse: Psychosocial		
Male	27.4 ± 7.9 (15)	27.0 ± 9.1	Self	67.7% (21)	57.1% (4)
Female	14.5 ± 3.5 (2)	38.7 ± 5.6	Other	80.6% (25)	14.3% (1)
Duration, sleepsex, years, mean ±SD (n)			Total	100.0% (31)	100.0% (7)
Total	9.5 ± 6.1 (8) ³	12-16 yrs (n=3), brief (n=4)	II) Positive, self/other (with or without adverse consequences)	12.9% (4)	57.1% (4)
Male	8.3 ± 6.5 (6)	12,16 yrs (n=2)	Polysomnography (PSG), % (n)	83.9% (26)	28.6% (2)
Female	13.0 ± 4.2 (2)	12 yrs (n=1)	Sleep EEG (without PSG)	0%	14.3% (1)
Sleepsex behaviors, % of patients (n)			No PSG or sleep EEG	16.1% (5)	57.1% (4)
Masturbation	22.6 % (7) (4 male, 3 female)	14.3% (n=1) (1 male)	Total # parasomnias ⁵	71	N/A (not reported)
Sexual vocalizations, talking, shouting	19.3% (6) (n=2 moaning; n=4 talking) (2 male, 4 female)	28.6% (2) (n=1 moaning; n=1 shouting) (2 male)	Mean # (±SD) per patient (range, 1-4)	2.2 ± 1.0	
Fondling another person (13 male, 1 female)	45.2% (14)	0%	Final diagnosis, sleepsex etiology ⁶ , % (n)		
Sexual intercourse (13 male, 0 female)	41.9% (13)	0%	Disorder of arousal (DOA) (CAs, n=26; SW, n=2) (OSA comorbidity, n=4)	90.3% (28)	N/A
Sexual hyperarousal (experiential)	0%	28.6% (n=2) (male, female)	RBD	9.7% (3)	N/A
Ictal orgasm	N/A	42.9% (n=3) (1 male, 2 females)	Sleep related seizures	N/A	100% (7)
Ictal sexual automatisms	N/A	14.3% (n=1) (male)	Treatment efficacy ⁷ : (controlling sleepsex), % (n)		
Total # sleepsex behaviors	n=40	n=9	Clonazepam at bedtime	90.0% (9/10): (6/7-DOA; 3/3-RBD)	
Amnesia for sleepsex, % (n)	100% (31)	28.6% (2)	Nasal CPAP at bedtime	100.0% (2/2-OSA)	N/A
Recall of sleepsex, % (n)	0% (0)	71.4% (5)	Control of nocturnal seizures ⁸	N/A	100% (5)

¹N=1, age not reported.

²Age of sleepsex onset was known in 54.8% (17/31) of patients, and was unknown or not reported in 45.2% (14/31) of patients.

³Duration of sleepsex reported on n=8 patients; n=8 had only one reported episode of sleepsex and n=1 had two reported episodes within 1 month, so duration is not applicable; n=14, duration not known.

⁴Adult males assaulted 9 girls (8-15 years old [n=8], and a “teenage girl” [n=1]).

⁵Confusional arousals [CAs] (n=24); Sleepwalking [SW] (n=21); Sleptalking and vocalizations, sexual & nonsexual (n=15); Sleep terrors (n=7); REM sleep behavior disorder [RBD] (n=3); sleep related eating disorder (n=1). [A history of enuresis was not included in these data].

⁶In all 3 patients with RBD, no behaviors (sexual or nonsexual) were documented in REM sleep. OSA=obstructive sleep apnea.

⁷An additional patient with a DOA responded to clonazepam, but with remission maintained after clonazepam was discontinued. Another patient with a DOA did not respond to limited therapy consisting of low-dose (25 mg) clomipramine at bedtime. Therefore, these two cases were kept separate from the treatment outcome data. For the remaining n=17, treatment was not mentioned. [CPAP: continuous positive airway pressure].

⁸Control of nocturnal seizures was achieved with anticonvulsant medications. N=2, treatment outcome was not mentioned.

sources of the 7 sleep related seizure cases.^{3,26-31} Two parasomnia articles were published in a foreign language (German,²² Dutch²³), each with an English abstract. These two articles were translated into English (courtesy of R. Ghedini [German to English] & M. Corner [Dutch to English]).

Both the parasomnia and seizure groups had notable findings; caution, however, should be taken when comparing the data between the two groups, since the latter had a small size. The parasomnia group has a strong male predominance. Age of onset of “sleepsex” in both groups was during early adulthood. Sleepsex

Table 3—Terminology of Abnormal Sleep and Sex¹

Terms	Definitions and Comments
Sleep related abnormal sexual behaviors (SRASB)	From <i>ICSD-2</i> , ² a variant of confusional arousals and sleepwalking, with or without associated obstructive sleep apnea.
Sleepsex, sexsomnia, sexual behavior in sleep (SBS)	Synonymous, interchangeable terms that encompass all abnormal sexual behavior and experiences surrounding sleep: within NREM or REM sleep, and during sleep-wake transitional states. The etiologies include parasomnias and other sleep disorders. At present, SRASB should be considered a subset of these terms.
Epileptic sleepsex/sexsomnia/SBS, sleep related sexual seizures	Synonymous terms, analogous to those listed above, but with an epileptic etiology; encompass ictal and postictal sexual automatisms, genital and sexual hyperarousal, rhythmic pelvic thrusting, and ictal orgasms.
Sleepsex moaning, sleepsex talking, sleepsex shouting	Sexual vocalizations and verbalizations during sleep (including profanities), as isolated phenomena or emerging with other SBS.
Epileptic sleepsex moaning, sleepsex talking, sleepsex shouting	Analogous to the terms listed above, but with an epileptic etiology.
Sleepsex snoring	Snoring during sleepsex, associated with obstructive sleep apnea.
Hypersexuality with Kleine-Levin syndrome; chronic, severe insomnia; or restless legs syndrome	Excessive, inappropriate, or deviant sexuality during wakefulness, or wake-sleep transitional states, that is closely associated with these sleep disorders.
Sexual hypnagogic or hypnopompic hallucinations (including rape); sexual REM-onset dream attack	Usually associated with narcolepsy; prone to enduring convictions that the sexual experiences had actually occurred.
Cataplectic orgasm, peri-orgasmic cataplexy	Synonymous terms that refer to a cataplectic attack being triggered by orgasm.
Sleep exacerbation of persistent sexual arousal syndrome	Sleep exacerbating a wakeful sexual disorder.
Hypersexuality with sleep related painful erections (SRPE)	Excessive or inappropriate sexual activity associated with SRPE.
Hypersexuality with sleep related dissociative disorders	Rhythmic pelvic movements, other sexualized behaviors, and attempted reenactment of past sexual abuse scenarios.
Nocturnal sexual delusions and hallucinations	Emerge before sleep onset or after awakenings. Associated with primary psychotic disorders, Parkinson's disease (esp. with dopaminergic therapy), and other neurologic conditions.
Hypersexuality during nocturnal awakenings	De novo onset of nocturnal hypersexuality emerging with pallidotomy and Deep Brain Stimulation (DBS) therapies of Parkinson disease.

¹Covers the range of abnormal sleep related and sleep disorder-related sexual behaviors.²ICSD-2: International classification of sleep disorders, 2nd edition, 2005.

had been a longstanding problem in 8 parasomnia patients and 4 epilepsy patients prior to clinical intervention.

Four categories of sleepsex behaviors were found in the parasomnia group, and 5 categories of sleepsex behaviors and/or experiences were found in the sleep epilepsy group. In the parasomnia group, females almost exclusively engaged in masturbation and sexual vocalizations, whereas males commonly engaged in sexual fondling and sexual intercourse with females. There were 2 cases of sleep related homosexual behavior: i) A 16-year-old male (without reported sexual orientation) went sleepwalking into the bedroom of his aunt and uncle (who slept in the same bed) and started fondling his uncle's genitals.⁴ ii) A heterosexual Dutch man had a history of multiple forms of interpersonal sleepsex, involving sexual initiatives towards his brother, current wife, and ex-wife; he was also accused of fondling his 15-year-old daughter on one occasion.²³ Therefore, in at least one of these two cases, homosexual sleep behavior diverged from customary (wakeful) sexual orientation.

Memory recall after sleepsex diverged between the two groups:

all 31 parasomnia patients had full amnesia for their sleepsex episodes, whereas there was recall of sleepsex episodes in 5 of 7 patients with sleep related seizures.

Agitated or assaultive sleepsex behaviors, sleepsex with minors, and legal consequences affected a substantial number of parasomnia patients. Sleepsex was far more injurious to the bed partner than to the affected person in the parasomnia group. However, adverse psychosocial consequences were quite common in both patients and bed partners. In contrast, pleasurable experiences from the sleepsex were reported by 3 patients with sleep related seizures, and by the bed partners of 4 parasomnia patients and 1 sleep related seizure patient.

In the parasomnia group, histories of multiple NREM sleep parasomnias were common, with confusional arousals being predominant. Sleepsex was rarely the only parasomnia behavior in the longitudinal histories of these patients. Two cases also involved sleepwalking with "sleep driving"⁴ and one case also involved sleep related eating disorder.² Besides a clinical evaluation, PSG monitoring (without penile tumescence monitoring)

took place in the preponderance of cases. OSA comorbidity that promoted sleepsex was present in 3 cases, with snoring during sleepsex being a prominent feature. (The authors of one report⁴ commented on an additional case of sexsomnia with OSA that resolved with CPAP therapy).

Although 26 parasomnia patients with sexsomnia had PSG studies, sometimes on multiple nights, only 4 (15.4%) demonstrated sexual behaviors during sleep: 3 patients had sexual moaning during slow wave sleep (with one patient also attempting to remove his pajamas³); one patient, who slept with his wife during the PSG study, had the following event occur: “the video surveillance showed Mrs. D. initiating sexual foreplay, which led to intercourse, performed by Mr. D. while he was ‘drifting’ between stage 1 and wakefulness. The patient was not aware of this in the morning.”²⁴ Therefore, the only parasomnia sleepsex behaviors documented by PSG in the published literature involves sexual moaning, and one case of sexual intercourse initiated by the bed partner.

The apparent association of sexsomnia with alcohol use/abuse and drug abuse was as follows: limited use of alcohol on the night of sleepsex (therefore, no presumed link), n=1; sufficient alcohol use or abuse on the night of sleepsex (therefore, possible or probable contributing factor), n=8 (with n=1 also smoking marijuana, and n=1 also taking “speed”). In addition 3 had past histories of drug abuse, with established remission at the time of sleepsex. A total of 8 patients had identified psychiatric disorders, but without any presumed link with the sexsomnia. Another patient was experiencing “stress” and sleep deprivation on the night of committing sexual assault during sexsomnia.

Parasomnias, OSA, and seizure-induced sleepsex appeared readily amenable to therapy. The benzodiazepine clonazepam was effective in almost all treated parasomnia cases, CPAP was effective in all 3 treated OSA cases, and anticonvulsant medication was effective in all 5 treated seizure cases.

Abnormal Sleepsex Behaviors Found with Parasomnias and Sleep Related Seizures

Masturbation:

In the parasomnia group, 4 patients masturbated to climax on a recurrent basis during sleep, whereas three patients did not climax during recurrent episodes of sleep masturbation. In the nocturnal seizure group, one man had longstanding, recurrent episodes of “violent masturbation.”

Parasomnia masturbation had various clinical manifestations:

- A 34-year-old married man would spontaneously masturbate to ejaculation every night after he had been asleep 2-3 hours, and was not arousable. Nightly sleep masturbation occurred while he continued to engage in sexual intercourse with his wife every night before falling asleep.²⁰
- A 27-year-old man “usually ‘awoke’ with an ejaculation between 02:00 and 06:00 during the preceding 5 years. He broke 2 fingers when he tore off the restraints he used to avoid moving in bed. He also slept in a different bed or on the floor in futile attempts to avoid the undesired masturbation.³
- A 26-year-old married woman had a history of abruptly tearing off her clothing and masturbating violently during the first half of the night. Her masturbation was associated with soft to loud vocalization and occasional vaginal discharge. If her husband interrupted the episode of masturbation, it might recur a second or a third time

during the night. Any attempt to initiate intercourse after she was awakened was rejected, and she denied the behavior.³

- A 38-year-old woman “asked for professional help after her husband, to whom she was married for 2 months, had awoken repeatedly at night only to find his wife apparently masturbating in her sleep.”²⁴

The one patient with sleepsex masturbation associated with nocturnal seizures was a 31-year-old man with a 12-year history of masturbating while asleep.³ Episodes lasted from 3 to 15 minutes, and movements became increasingly violent the longer the episodes lasted. Bed partners would hear inarticulate moaning sounds together with movements of the feet or legs. There was confusion and disorientation whenever he awakened during an episode. Timing of these events was variable. No ejaculation was reported by the patient or the bed partner. Because of his sleep masturbation, the patient avoided sexual relationships for more than 8 years. During that time, he was aware of repeated bruising of the penis, along with a sore groin.

Sexual Vocalizations and Talking:

In the parasomnia group, sleeptalking and sleep vocalizations (brief or prolonged) affected 48.4% (15/31) of patients, with 19.4% (n=6) having sexual sleeptalking and vocalizations, and 29.0% (n=9) having nonsexual sleeptalking and vocalizations. Some examples include:

- A 26-year-old woman had soft to loud vocalizations during sleep masturbation.³
- A 27-year-old woman had sexual moaning during sleep at least three nights weekly for 15 years that would emerge within 15-20 minutes of sleep onset.³
- A 28-year-old woman had nightly sexual moaning and sexual fondling during sleep for 16 years that would appear within 20 minutes of falling asleep and disturb the sleep of her husband and children.³
- A 26-year-old woman would initiate foreplay with her bed partner while they were both asleep between 02:00 and 05:00 and would utter sexually provocative phrases while fondling him. Whenever he responded positively to her involuntary sexual overtures during sleep, she would then awaken and accuse him of forcing sex on her while she slept.³

Uttering profanities and sexually provocative phrases, or engaging in “dirty talk” accompanied sleepsex fondling or intercourse in three patients. In the sleep related epilepsy group, previous bed partners of one patient had noted inarticulate sexual moaning during sleep together with movement of the feet and legs. Another patient shouted while engaging in sleepsex.

Sexual Fondling:

Some examples include the following:

- A 23-year-old man would attempt to remove his girlfriend’s clothing and fondle her, without ejaculating, during the first part of the night. When he was awakened, he would be confused and disoriented.³
- An 18-year-old male was accused of putting his finger into the vagina of a nearby teenager at 6 a.m. while he was asleep with recent sleep deprivation.³
- A man would grab his wife’s buttocks during sleep while “grinding her from behind” with rhythmic pelvic movements.²²
- A 22-year-old man on two occasions within a month, while presumably asleep, fondled the breasts of one woman (before initiating intercourse), and inserted his fingers into the vagina of another

woman (and when she asked him to stop, “he did so and left the room without incident”).²⁵

- A 27-year-old man would frequently engage in cunnilingus on his wife while they both were asleep.⁴

Sexual Intercourse:

A diverse set of clinical presentations was reported:

- “During the episodes, the [27-year-old] patient typically procured his wife, achieving complete sexual intercourse with total amnesia...His wife remained in bed with him after the episodes.”²⁴
- A 43-year-old man engaged in nightly episodes of amnesic sleep-sex along with amnesic eating in his sleep. His current and previous girlfriends had sexual intercourse with him every night while he would be snoring and sound asleep for up to 30 minutes before ejaculation. Furthermore, “the repertoire of unconscious sexual activity by the patient was varied and included intercourse in different body positions, as well as oral sex, both given and received.”²²
- A 38-year-old man for 12 years had sexually assaulted his spouse during sleep between 03:00 and 05:00 at irregular intervals, but at least once every 15 days. He would tear off his wife’s clothes, fondle her, and initiate sexual intercourse. His wife observed that he was “not present” and “unresponsive” while acting violently, and one time attempted to choke her.³
- A 33-year-old man one night grabbed his wife in his sleep, tore off her bed clothes, and forced intercourse. Despite her active resistance, she could not “reach” him. He seemed “far away” and appeared “glassy-eyed.”³
- A woman whose husband would initiate and consummate sexual intercourse with her while asleep also described his total disregard for her menstrual status while engaging in sexsomnia; they never had engaged in sex during wakefulness while she was menstruating.⁴
- A woman described her husband as being more amorous and more aggressive during episodes of sexsomnia than during his wakeful sexual activity. On some nights “there is no stopping him.” Once when he grabbed her around the neck, she slapped him hard, which awakened him, and he immediately let go.⁴
- A man had his two current girlfriends independently confirm that he frequently engaged in sexsomnia. One girlfriend commented that he was a “different person” during sexsomnia, being a more amorous and gentle lover, and more oriented in sexually satisfying her.²
- A 29-year-old man for at least six years had sexually assaulted his bed partner and uttered profanities between 03:00 and 06:00 while he was asleep. He also moved his arms and legs excessively in sleep when he dreamt of fighting against “intruders.”⁴
- A 22-year-old man “after a night out drinking” fell asleep on the sofa in a separate room from a female (platonic) friend who an hour later “was woken suddenly feeling a hand fondling her breasts and then, in rapid succession, she felt his erect penis penetrating her anally, vaginally, and then being forced orally.”²⁵

Sleepsex Behaviors and Experiences Found Exclusively with Sleep Related Seizures

Sexual Hyperarousal.

i) A 38 year-old man had a right temporal lobectomy one year earlier after a 19 year history of intractable complex partial seizures that were devoid of any sexual manifestations.²⁶ One year after surgery, complex partial seizures reemerged at a rate of 2-

3 times monthly, with 2 de novo forms of sexual behaviors and experiences coming from sleep and wakefulness. He would “occasionally wake up during the night with a strong sexual desire, and he believed that this was brought about by a seizure.” An immediate post-awakening, post-ictal sexual hyperarousal was considered to be the most likely explanation. The second form of abnormal sexual behavior occurred while he was awake, about 20 minutes after the onset of a complex partial seizure. He would approach his wife to have sex, but he would not be insistent nor would he proceed if their children were present. If a seizure occurred after sex with his wife, 20 minutes later he would again desire sex with his wife. ii) A 32-year-old woman developed sleep related hypersexuality 3 weeks after undergoing removal of a right inferior temporal lobe lesion, for an 18-year history of intractable bitemporal complex partial seizures.²⁷ “She would wake in the middle of the night with intense sexual desire and rouse her husband. She stated a wish for her husband to have the same operation...The hypersexuality subsided 18 months after surgery.” The histopathology of the resected tissue identified a dysembryonic neuroepithelial tumor.

Ictal Orgasm.

- A 41-year-old woman developed frequent, brief episodes of sleep related somatosensory seizures that responded to phenobarbital therapy for 2 years. One night 5 months after discontinuing phenobarbital she “was awakened by the electrical [seizure] discharge, now with paresthesia in the right lateral abdominal and pubic regions and about her genitalia.”²⁸ A sensation of vaginal dilatation ensued immediately afterward, which inevitably brought about an orgasm, either pleasant or painful. During these episodes there was no confusion or memory loss...Normal marital sexual behavior was reported.” PSG with extensive sleep EEG documented 8 clinical seizures, with the first 4 emerging from slow wave sleep, and the remainder emerging from stages 1 and 2 sleep. The central-parietal regions exhibited sharp wave and spike paroxysmal activity followed by the patient awakening, with electrical sensations. Orgasm immediately ensued, with persistence of generalized paroxysmal EEG activity. Resumption of phenobarbital therapy immediately controlled her seizures.
- A 55-year-old woman had a 12-year history of “paroxysmal nymphomania” arising from sleep, when she awakened with “a feeling of being hot all over as if she were having coitus.”²⁹ She also had left somatosensory seizures and developed left hemiplegia.
- A 31-year-old man with temporal lobe epilepsy had nocturnal attacks in which he would awaken with a stereotyped sequence of motor, sensory, and experiential symptoms that began with twitching of the forehead and face which progressed down to the neck and left arm, followed by the sensation of “being just like an orgasm” and “sexual and intensely pleasurable,” but never associated with ejaculation.³⁰ There was a dramatic response to diphenylhydantoin therapy, with total cessation of nocturnal attacks and orgasmic sensations.

Ictal Sexual Automatism:

A 36-year-old man with a 16-year history of complex partial seizures with sexual automatisms underwent sleep EEG monitoring with sphenoidal leads.³¹ Sleep related seizures “consisted of the patient suddenly seizing the groin area with both hands, turning and thrashing about, and uttering obscenities. He continued to

manipulate his penis and then had lip smacking, snorting noises, and intermittent grimaces for 2 minutes, with postictal confusion for 5 minutes. EEG during the attack showed rhythmic slowing bilaterally.”

Other cases of nocturnal seizures associated with various nocturnal sexual automatisms and experiences have been published, but without mention of whether they were sleep related, and neither sleep EEG or PSG were performed.³²⁻³⁴ In another case, a 51-year-old man with a 21-year seizure history was documented to have genital automatisms arising from a wakeful complex partial seizure, but genital automatisms were not present during a documented sleep related seizure.³⁵ However, given his long and intractable seizure history, it is possible that some of his sleep related seizures involved genital automatisms.

Sleep Related Hyperkinetic Seizures with Sexualized Pelvic Thrusting

Repetitive pelvic thrusting that often resembles sexual (coital) behavior is a prominent feature of sleep related hyperkinetic seizures that are found most commonly in nocturnal frontal lobe epilepsy (NFLE), but also in nocturnal temporal lobe epilepsy (NTLE).³⁶⁻⁴⁰ In a study of 442 surgically treated epileptic patients, 5.6% (n=25) had exclusively sleep related hyperkinetic seizures.⁴⁰ These 25 patients reported sensory-experiential (nonsexual) auras emerging from sleep that immediately preceded the hyperkinetic automatisms. These 25 treatment-resistant patients underwent surgical resections of their epileptogenic zones; 18 were diagnosed with NFLE and 7 with NTLE. The 16 patients with Taylor’s dysplasia, as demonstrated by histopathology, were all free of their sleep related seizures after one year; the status of the other 9 patients was not reported. In contrast to the sleep related, sexualized, hyperkinetic seizures that are predominantly due to NFLE, discrete epileptic genital automatisms without a hypermotoric component is generally due to NTLE.³⁵

Legal Consequences, Including Sleepsex with Minors, in the Parasomnia Group

The legal outcome from the accusations and legal charges of sleepsex with minors were acquittal, N=2 (on the basis of sexsomnia); long-term probation, N=2 (including outpatient psychiatric monitoring); and unknown, N=6 (outcome from legal charges and completion of child protection agency investigations were still pending). Sexsomnia was the medical diagnosis that was also used as a legal defense in the cases involving minors. The following are pertinent examples:

- A 45-year-old man with lifelong sleepwalking was awakened one night around 02:00 by the screaming of his 14-year-old daughter’s girlfriend who was sleeping over at their home. She accused him of sexually fondling her while she was asleep. He was arrested that night, and claimed to be entirely amnesic for the episode.²
- An 18-year-old male with longstanding sleep terrors and sleepwalking was accused of placing his finger into the vagina of a female teenager who was sleeping in the vicinity.⁴
- A 39-year-old married man was charged with sexually touching his 9-year-old daughter while she was sleeping one night in bed with her parents, after leaving her own bed because of a nightmare. He claimed to have been sleeping at the time. He and his wife would

often initiate sex with each other during sleep⁴

- A 32-year-old man was accused of inserting a finger into the vagina of a 10-year-old girl, after falling asleep in bed with her (and another child) subsequent to drinking alcohol excessively and smoking marijuana. He had a history of sleeptalking, had one known episode of sleepwalking, and had a family history of parasomnia.⁴
- A man was accused by his 15-year-old daughter of kissing her breasts and penetrating her vagina with his finger while he was asleep, which his wife and ex-wife also reported his doing to them while he was asleep.²³
- A 35-year-old man was accused by his 11- and 8-year-old daughters of touching their genitalia (without penetration) while they were all apparently asleep.²¹

Another category of child molestation involving sexual fondling during sleep was reported in a 27-year-old man with severe obstructive sleep apnea.²¹ While napping on two separate days, his young daughter jumped into bed with him and fondled his genitalia until ejaculation, when he then awakened and became alarmed.

In one of the two adult cases with forensic consequences, a 27-year-old man faced criminal charges initiated by his wife because she claimed that he frequently assaulted her sexually while she was asleep by attempting cunnilingus and sexual intercourse. He was amnesic for these episodes, had a history of sleepwalking and sleep terrors, and had a family history of sleepwalking.⁴ In the other adult case, a 22-year-old man was acquitted “on the basis of automatism” that presumably occurred during sleep.²⁵

Physical and Psychosocial Consequences

In the parasomnia group, the bed partners often experienced physical injuries (ecchymoses, lacerations) from the sexual assaults, and to a lesser extent the patients were also physically injured (bruised penis; fractured digits). Moreover, both the patients and bed partners often experienced an array of adverse psychosocial consequences involving bewilderment, embarrassment, shame, guilt, despair, “shock,” alarm, anger, worry, annoyance, denial, feelings of inadequacy/low self-esteem, and reactive emotional distancing that led to some marital estrangement with marriage counseling sometimes being sought. In some cases, there was “low-grade guilt” over disturbing the sleep of family members from sexual moaning.³ One man was not convinced that he engaged in any sleep masturbation (that occurred nightly), but then became worried “about the effect of excessive drainage of seminal fluid on his health.”²⁰ His wife, on the other hand, was quite concerned about his sleep masturbation and “felt sexually redundant, inadequate, ‘cheated’ of her husband’s confidence.” Interestingly, they regularly engaged in sexual intercourse before falling asleep.

On the other hand, in 4 parasomnia cases, pleasurable aspects of the sleepsex were reported by the bed partners, with negative aspects also acknowledged:

- One woman commented that her boyfriend’s sleepsex was more “aggressive and dominant” than his waking sexual behavior, and she “found some aspects of the sleepsex pleasurable...and a little kinky,” such as “forceful albeit playful biting and ‘talking dirty,’ but “nonetheless she requested that the patient incorporate some of the nighttime sexual practices...into their conscious daytime lovemaking.”²
- Another woman “reported infrequent and hurried sex with her husband, whom she described as distant and reluctant during wakeful-

ness. Nocturnal sex was more satisfactory to her, even if associated with bruises at times.²³

- A woman who slept with a sexsomnia boyfriend commented on how he was a “different person during these activities—apparently, he is a more amorous and gentle lover and more oriented toward satisfying his partner when he is asleep.”²⁴
- A wife reported that her husband would initiate sex in his sleep about once monthly, during which times he would be “more aggressive and more amorous... than when he is awake.”²⁴

Likewise, in 4 epilepsy cases, pleasurable aspects of sleep related sexual seizures were reported by the three patients and one bed partner:

- A 41-year-old woman with nocturnal somatosensory seizures would experience pleasurable (or painful) orgasms together with vaginal dilatation during her seizures.²⁸
- A 31-year-old man with temporal lobe epilepsy would experience a sleep related “sensory phenomenon along with the twitching [that] was described as ‘being just like an orgasm...sexual and intensely pleasurable,’ but without ejaculation being achieved.”³⁰
- A 32-year-old woman with an 18-year history of complex partial seizures developed sleep related hypersexuality arising from sleep that began 3 weeks after surgery. “She would wake in the middle of the night with intense sexual desire and rouse her husband. She stated a wish for her husband to have the same operation.”²⁷
- A “wife had started to look forward to this happening,” referring to her husband’s post-ictal (psychomotor attacks) hypersexuality that occasionally emerged from sleep.³¹

Finally, it should be emphasized that in the parasomnia and epilepsy cases just described, psychopathology was uncommonly present and was not an identified contributing factor to the abnormal sleep related sexual behaviors and experiences. Furthermore, neither sexual deprivation/frustration nor a previous history of paraphilia or criminal sexual misconduct were reported.

Experiences with sleepsex, including psychosocial problems in both initiators and recipients of sleepsex, have been categorized in a web-based survey involving 121 respondents (mean age, 37 yrs; 65% female).⁴¹ Six distinct themes stemming from the problematic sleepsex, affecting the “initiators” and the “recipients”, were identified: (1) fear and a lack of emotional intimacy; (2) guilt and confusion; (3) a sense of repulsion and feelings of sexual abandonment; (4) shame, disappointment, and frustration; (5) annoyance and suspicion; (6) embarrassment and a sense of “self-incrimination.” Various methodologic limitations of this study were acknowledged by the author, such as lack of longitudinal information on sleepsex, absence of clinical evaluation and PSG, and the anonymous and non-verified nature of the reporting. Also, amnesia or recall for the sleepsex was not systematically addressed; there were comments of complete amnesia in some people, others “often had no recollection,” and others had “a high level of amnesia for the events.”

Sleep Disorders with Abnormal Sexual Behaviors During Wakefulness and Wake-Sleep Transitions

Sexual Disturbances in Kleine-Levin Syndrome (KLS) (Isabelle Arnulf)

KLS is a rare sleep disorder characterized by recurrent and unusually long episodes of hypersomnia.¹ Sleep episodes last 16-24

hours per day for days or weeks, and during periods of wakefulness are associated with behavioral and cognitive disturbances, often including megaphagia and to a lesser extent sexual disinhibition. These symptomatic episodes alternate with periods in which patients have normal sleep and behavior that usually last for months to years.

Herein are presented the results of an extensive meta-analysis of all 195 articles published about KLS in the Medline database (1962-2004). After having removed duplicate and doubtful cases, reviews and points of view, information about 186 patients from 139 articles were compiled.⁴² KLS affected mainly males (68% of the group), with a median age of onset of 15 years. The disease was self-limiting, but usually lasted between 4 and 8 years.

Data on KLS was also gathered using a standardized questionnaire and interview of 108 new patients and their parents taking part in a prospective research program on KLS at Stanford University (Arnulf I, et al. submitted manuscript).

Frequency of Hypersexuality

In the meta-analysis of the literature, we found that 41.9% (n=78) of patients had symptoms of hypersexuality, while 57.5% (n=106) did not report these symptoms and a single patient had decreased libido during his episodes.⁴² In contrast with other symptoms, that were equally frequent in both sexes (such as megaphagia, 75% in women vs. 82% in men; or cognitive disturbances, 93% in women vs. 95% in men), hypersexuality was twice as frequent in men (51%) as in women (24%, odds ratio: 3.4, P=0.002). The age at KLS onset was not different with (16±7 y) and without (17±10 y) sexual disinhibition. The presence of hypersexuality did not predict a worse prognosis (such as a longer disease course, or longer episodes) than its absence. This symptom was also present in 28% of 12 patients with KLS symptoms secondary to various brain lesions. Plasma levels of testosterone, measured in 16 of 186 KLS patients, were normal in 14 patients and mildly decreased in 2 patients, but never increased.

In the prospective KLS series involving 108 cases in which interviews used a structure questionnaire, 53% reported increased sexual drive, while 6% had decreased sexuality at least during an episode. Hypersexuality was more frequent in boys (58%) than in girls (35%).

Symptoms

Hypersexuality was briefly described in the second KLS patient of Kleine⁴³ in 1925. Later, Robinson and McQuillan, in 1951, reported on a 19-year-old officer-cadet who had disinhibited behavior during a KLS episode: “He indecently exposed himself in the ward, and while lying unclothed on the bed he masturbated, grinning broadly. Lewd remarks were made to the nursing staff. Once he urinated in the garden in the presence of a sister.”⁴⁴ In our meta-analysis, we observed that the changes in sexual behavior shared some similarities with the disturbed eating behavior, such as increased quantities (increased frequency of masturbation or of sexual intercourse, demanding intercourse several times daily⁴⁵), compulsions (with active and uncontrolled research of sex), lack of judgment in the choice of sexual partner (sexual advances were made to religious sisters, to the patient’s daughter⁴⁶ or sister,⁴⁷⁻⁴⁹ to a nurse “who is said to have been old enough to be his grandmother,”⁴⁴ and in three cases to other males by otherwise non-

homosexual male patients^{46,50,51}), inattention to the environment (such as masturbating in public), and absence of self-awareness of the inappropriateness of the behavior.

Garland, in 1965, reported that a 16-year-old teenager made “frequent and vigorous attempts at masturbation, whether or not the curtains round his bed were drawn, and this would be continued during questioning. He never achieved either orgasm or ejaculation.”⁵⁰ In our own prospective program on KLS, a young adolescent went to the beach with his family, and started to masturbate ostentatiously on the beach, to the great embarrassment of the grandparents, parents, and sister. Another reported masturbating 3 to 5 times a day, while one reported: “I masturbated excessively, to the point of bleeding.” A typical example of the compulsive component of KLS was observed a 39 year-old man, who, during his third episode of KLS, attacked 2 women sexually, and the same day attacked his daughter sexually. He showed exhibitionistic behavior and ran naked in the ward, threatening to attack the nurses sexually and fondled the female patients. During the sixth episode, he undressed himself, exposing himself to other patients, made advances to the nurses and patients, and threatened to sleep with them. During the seventh episode, he was confined to a male-only, locked psychiatric ward for 5 months, where he approached a male patient to have relations with him.⁴⁶

Intensity of Symptoms

The intensity of the abnormal sexual behavior may vary. Some behaviors were considered as mildly inappropriate in a given context of education or culture: patients used obscene words in front of the parents and doctors.⁵² One mother told us his son was “cursing like a trooper”; a 14-year-old teenager answered to the female doctor that his three wishes were: “1- to go to sleep; 2-money; 3: to have a ‘dick’ as long as the width of the bed.” A usually extremely polite, 13-year-old teenager answered the doctor asking him what he would be doing during the course of summer: “fuck, fuck girls.” Patients can demonstrate inappropriate sexual behavior in the clinical setting while transgressing interpersonal boundaries: the aforementioned 14-year-old teenager also put his hand on the female doctor’s thigh, while another patient fondled the female nurse during his EEG recording. Another had increased interest in pornographic magazines,⁵³ and one patient stared at a nurse’s body in tactless way.⁵⁴ In our prospective series, several teenagers reported that they frequently accessed pornographic material via phone services or the internet, leading one of them to spend more than \$500 in phone bills during an episode. A mother reported to us that her 15-year-old son, during the first episode, started masturbating when the urinary catheter was inserted in the emergency room, so everybody believed the catheter was responsible for the odd behavior. Even after the catheter had been removed, he continued to masturbate several times a day in front of people and used sexually explicit language with a curiously desperate voice (e.g., “I want to fuck you, I want to screw your ass, I want a blow-job”) without directing his comments at anyone in particular. A young father in India had a severe fever for 13 days, and after the fever subsided, he had an increased sexual urge and began using profanity.⁵⁵ The sexual disinhibition was obvious as “he would drag away his wife, and would start undressing her in the presence of everybody, unconcerned and completely oblivious of their great horror and embarrassment. Not only that, he made amorous advances to other females too.”⁵⁵ In another case, be-

haviors caused a disruptive and potentially dangerous, nuisance, involving a school boy who went to a neighbor’s house, masturbated into his neighbor’s underwear, stole football match tickets, and set fire to the drapes.⁴⁶

Associated Symptoms

Sexual disinhibition sometimes occurred simultaneous with other repetitive, stereotypical behaviors compulsions such as setting fire,⁴⁶ or with the compulsion to repeatedly turn the light switch on and off.⁴⁷ Patients with KLS also frequently experience a disturbed state of consciousness that they struggle to describe as “being between dream and reality.” One of them “admitted he had vivid dreams, or rather fantasies, of a violent sexuo-sadistic character.”⁴⁴ In almost all cases, however, the abnormal sexual behavior did not stem from sleep, as with sleepsex, but occurred during wakefulness in the daytime.

Hypersexuality in Women With KLS

The symptoms of sexual disinhibition were reported in women⁵⁸⁻⁶⁰ and in 3 prepubescent children.⁶¹⁻⁶³ One young lady was hospitalized because she was somnolent and for the two previous days had been “scratching her genitals in public, lifting her clothes over her head to warm her buttocks at the fire.”⁵⁸ During that KLS episode, she also made advances to male patients and staff. In our case series, the parents of a senior high school teenager noted a change in her daughter’s clothing choices. The teenager purchased tube tops, playboy tank tops, and thong swimwear; these choices were completely out of character for her. One woman reported that during her KLS episodes: “remember, this experience was in the early 1950s, a more innocent time than today. I usually wanted to masturbate. In junior high school, riding in car with several kids, I sat on a boy’s lap and kissed him and have no idea what things I was saying to him. At the same time I asked myself, ‘What am I doing here? This isn’t me.’ Feelings were flat, no embarrassment or shame, though I was aware of the disapproval of others.”

Hypersexuality and the Longitudinal Course of KLS

In a recent prospective study of 108 KLS patients (76% male), the presence of hypersexuality in 53% (n=57; 48 [84%] males) of cases conferred a two-fold lengthening of the median KLS clinical course (21.2 ± 1.7 yr vs. 10.2 ± 1.9 yr) ($P = 0.01$) (Arunulf I, et al. submitted manuscript). Age of KLS onset did not differ between the hypersexual and non-hypersexual KLS groups: 16.2 ± 4.1 vs. 15.5 ± 7.9 yr. This finding suggests that hypersexuality is associated with KLS disease severity.

Pathophysiology of KLS

The cause of KLS is unknown. Scarce neuropathological evidence and a possible association with HLA DQB1-201⁶⁴ suggests it could be an autoimmune encephalitis restricted to the hypothalamus and adjacent areas. While brain computed tomography and magnetic resonance imaging were normal in all reported patients with primary KLS, diffuse brain hypoperfusion, mostly focused on the thalamic and frontotemporal areas, has been reported.⁶⁵ It is therefore difficult to correlate the symptoms of hypersexuality

in KLS with a precise anatomical location. KLS is strongly associated with male sex and puberty. Hypersexuality affects more than half of the patients and is associated with a much longer disease course. This suggests that boys are more vulnerable to the disease and may suffer from more extensive or severe brain lesions than girls. Eventually, the association of instinctive behaviors (sleep, sex, aggression and eating) being intensively released (hypersomnia, hypersexuality, overeating) in the same patients points at a common origin from brain structures driving archaic behaviors. Similar symptoms of hypersexuality are observed in various neurological diseases, such as in the Kluver-Bucy syndrome, caused by lesions of the temporal lobes (and associated with increased oral exploration) in humans⁶⁶ and nonhuman primates,⁶⁷ of the paramedian thalamus or subthalamus nucleus,⁶⁸ of the orbitofrontal and medial frontal area.⁶⁹ Hypersexuality may also complicate the course of the disease in patients with Alzheimer disease or mania, and as a side effect of dopaminergic agents in Parkinson disease.⁷⁰ Some KLS behaviors, such as singing loudly while playing and banging on a piano continuously, are also observed in manic states. Finally, both deviant sexuality (including paraphiliac behaviors, e.g., exhibitionism) and hypersexuality are prominent manifestations of altered sexuality in KLS during its hypersomnia phases.

Sleep Disorders With Abnormal Sexual Behaviors During Wakefulness and Wake-Sleep Transitions

Severe Chronic Insomnia:

A published case described a 39-year-old woman who presented to a sleep disorders center with a lifelong history of severe insomnia associated with motor restlessness.^{71,72} She would sleeplessly walk around her house on most nights until 01:00-03:00 and rarely slept for more than 5 hours. In early adulthood, the insomnia became progressively worse. She felt that every day in her life there was “a motor running inside me all the time, revved up, with surges of electricity and high energy going all over me.” During the daytime, her mind and body felt speeded up, but she felt physically exhausted.

Beginning with puberty, she experienced problems with an excessive libido and genital hyperarousal that bothered her nearly every day. There were strong sexual sensations in the vulva, and general sexual heightening without erotic thoughts or altered consciousness that would only partly and briefly be relieved by having sex with her husband. Sometimes they had sex multiple times daily on many consecutive days. She could readily distinguish between this type of “hyper sex” (mechanical physical discharge, “pure lust,” devoid of emotion) from “emotional sex” which was intimate and pleasurable with her husband.

Successful control of her severe insomnia was only achieved with bedtime opiate therapy that also controlled her motor restlessness, along with her excessive libido and genital hyperarousal, without interfering with normal sexual relations with her husband. Remission of her insomnia, excessive libido, and genital hyperarousal has been maintained during more than 20 years of nightly opiate therapy, with relapses of these problems occurring whenever she did not take the medication.

Three additional cases of hypersexuality emerging during exacerbations of chronic insomnia in young adults (2 males; 1 disabled from insomnia) involved persistently heightened libido and

increased sexual behavior that lasted for days to weeks until the insomnia had subsided sufficiently (CHS & MWM, unpublished data).

Restless Legs Syndrome (RLS)

A case has been reported involving a 72-year-old man with a 4-year history of RLS (with abnormal sensations originating in the lower abdomen), who was documented by PSG to engage in rhythmic, pelvic, coital-like movements during relaxed wakefulness, at the wake-sleep transition, during infrasleep awakenings, and after the final morning awakening.⁷³ There were “intermittent periods (1-6 minutes) of stereotyped, repetitive, rhythmic body movements with side-to-side trunk motions or, when in a prone position, rhythmic pelvis forward and backward rocking, resembling coital movements, at a frequency between 2 and 3 per second.” The concurrent EEG showed alpha or alpha-theta activity, and the patient complained of typical restlessness in the lower abdomen. He could voluntarily suppress the pelvic movements for only brief periods of time. Pramipexole therapy at bedtime promptly controlled all sleep related sensory and motor symptoms, including the coital-like pelvic movements, which was sustained at one month follow-up. This case illustrates the convergence of RLS with a rhythmic movement disorder, both in the linked symptomatology and in the shared response to monotherapy. It also exemplifies how sexualized behavior can be linked with negative somatic sensations.

Another male patient with severe RLS, aged 64 yr, reported that he usually used masturbation at night to alleviate the painful sensation of RLS. He found long-lasting relief that could extend for the entire night (IA, personal communication). It is possible that orgasm-associated dopamine and opioid release mediated the therapeutic effect on RLS.

A study of 930 patients with moderate-to-severe RLS found that ropinerole-treated patients had significantly less disturbance in sexual activity than placebo-treated patients.⁷⁴ The mechanisms of efficacy were postulated to involve a direct dopaminergic effect and/or an effect derived from the control of RLS symptoms.

Special Clinical Considerations

Narcolepsy:

Hypnagogic and hypnopomic hallucinations (HH), often occurring with sleep paralysis (SP), can incorporate sexual content, with serious consequences. A series of four cases with PSG/MSLT confirmed narcolepsy, with the narcoleptic tetrad, was reported in three females aged 31-42 years and a man 39 years old.⁷⁵ In all four cases, compelling recurrent experiences of sexual molestation and rape began early in the course of their narcolepsy. One woman with the narcoleptic tetrad complained that after getting married, her father-in-law had continually molested her sexually in a bizarre manner. She also reported a history of HH without sexual content, recurrent sleep paralysis (including hypnopompic SP), and cataplexy. A second woman had SP and multi-sensory HH, including “astral travel experience” and paranoia about poisonings and plots. She described being raped by a policeman after going to bed, with detailed accounting of her state of undress and the sexual positions used. Following the rape, the policeman made her take drugs and then left. The patient saw a psychiatrist

for rape counseling. She described another rape that had taken place five years after the first rape. Her manager at work gave a party, and after she drank alcohol and went to a bedroom, her chairman came in and raped her—while she reported being paralyzed and mute. Other men were then let in her room and they silently raped her. When she “recovered her senses” she found that the rapists had put her clothes back on. She “permanently” lost respect for her supervisors and retired early from her position. She reported being raped on other occasions. She was eventually correctly diagnosed to have narcolepsy, and responded well to amphetamine therapy.

A third woman with severe HH had her narcolepsy substantially controlled with amphetamine therapy, but had a persistent belief that she had previously been repeatedly raped. The fourth patient was a 39-year-old man with lifelong HH who recalled being repeatedly fondled sexually by a friend when he was a young man. During naps he would partially awaken and feel he was elsewhere, “paralyzed by drugs,” while his friend was fondling him. When he was awake, he found that his friend always acted appropriately. Amphetamine therapy completely controlled his narcolepsy. However, he persisted in regarding his previous sexual molestations were real and wondered what drug was used to paralyze him.

Two other cases of recurrent sleep related sexual hallucinations in undiagnosed narcolepsy have been reported.⁷⁶ A 23 year old female vividly recalled being sexually assaulted in a car and went to the police to file charges, but then “she slowly realized that her experience of violation was a hallucination.” She then retracted her charges, and a sleep evaluation with PSG and MSLT confirmed narcolepsy-cataplexy. Clomipramine therapy, together with modafinil, fully suppressed the hallucinations. A 45 year old man had vivid experiences of sexual relations with the wife of his chief, and he told his colleagues about this, but they were skeptical. This led him to consider the possibility of hallucinations. After PSG-confirmed narcolepsy, clomipramine therapy, together with modafinil, fully suppressed the sexual hallucinations and their delusional interpretations.

In a study on SP associated with hypnagogic or hypnopompic hallucinations, in which the Waterloo Unusual Sensory Experiences Survey was administered to 1,273 undergraduate students, 13 respondents (all female) reported their SP experiences “as feeling very much like being sexually assaulted or raped.”⁷⁷

In the original description and naming of narcolepsy by Gélinau in 1881, the patient was a 38-year-old man with a two year history of very frequent sleep attacks, totaling up to 200 attacks daily.⁷⁸ This patient could not speak with Dr. Gélinau for even 30 minutes without falling asleep, and constantly needed his 13-year-old son at his side to keep awakening him, so he could attend to his successful business. A wide array of intense emotional states played a prominent role in triggering his sleep attacks. Gélinau’s male patient also spontaneously reported that his infant child “was conceived in a moment when the illness came over him.” He was convinced that sexual intercourse took place during one of his sleep attacks, which resulted in the conception of his child. This is an unlikely possibility, since even if he did have a REM-onset penile erection, the atonia of REM sleep would not have allowed him to initiate or participate in sexual intercourse. Gélinau’s patient most likely experienced either peri-orgasmic cataplexy, or he had a HH or vivid dream involving sexual intercourse with his wife during REM-onset sleep, which later became linked with his

firm belief that the conception of their baby had also taken place during that sleep attack or HH.

Sleep Exacerbation of Persistent Sexual Arousal Syndrome (PSAS)

PSAS, also called recurrent sexual arousal syndrome, is a rare condition with widely diverse causes. Recently, a unique case involving exacerbation of PSAS with genital arousal during drowsiness and sleep onset was reported.⁷⁹ A 66-year-old sexually active woman presented with a 5-year history of persistent, unpleasant, “overwhelming” internal sensations of the vagina and pelvis. These genital sensations became worse whenever she felt vibrations while riding in a car, or while reclining or falling asleep; these sensations would often disrupt her sleep several times nightly. Erotic thoughts did not accompany the genital arousal either while falling asleep, upon awakening, or during the daytime.

When she awakened after being asleep for less than an hour, lying in bed would become intolerable because of the insatiable urge for sexual release, always being on the verge of achieving orgasm. The genital sensations and urges were not accompanied by any subjective sexual desire (libido), and so a form of “mechanical sex” was recurring. The genital arousal “felt intrusive and unwanted.”

Sex with her male partner would reduce her symptoms for only 2-3 minutes after orgasm. Physical activity, cold compresses, and distraction techniques helped reduce her symptoms, as did overuse of analgesics. The longitudinal course of her PSAS was marked by progressive severity; after five years, more than 80% of her waking time was disturbed by her genital symptoms. The intensity of symptoms, however, had not changed in 3 years.

She had gone through menopause 14 years previously (9 years before developing PSAS), and hormone replacement therapy was discontinued after 4 years. There was no abnormal menstrual history and no psychiatric history. During drowsiness and sleep onset, vaginal pulse amplitudes increased by 95% of basal values, which together with other findings documented increased vaginal blood flow and congestion and corroborated the patient’s complaint of sleep exacerbation of her PSAS. Genito-sensory testing of the vaginal and clitoral areas documented reduced sensory function, pointing to an organic basis for her genital arousal disorder. The brain, spinal cord, and pelvic regions were intact upon testing. PSG was not performed.

Olanzapine, carbamazepine, clomipramine, paroxetine, and various other remedies had been ineffective; risperidone was effective upon initiation of therapy.

This case may represent a dissociated sleep state in which REM sleep genital arousal with increased vaginal blood flow inappropriately intruded into this woman’s hypnagogic and hypnopompic states. Alternatively, this case may represent a “restless vagina syndrome” akin to RLS, consisting of abnormal vaginal sensations while resting before sleep onset associated with the “urge” to have sex, with sexual activity transiently reducing the unpleasant vaginal sensations.

Sleep Related Painful Erections (SRPE) and Increased Sexual Activity

SRPE is an obscure parasomnia in which painful erections occur during sleep and disappear upon awakening. Increased sexual

activity can emerge during the course of SRPE as a (futile) form of self-therapy, as reported in a 65-year-old man who presented with a 10-year history of SRPE.⁸⁰ He would experience 3-5 painful erections nightly, and over time the pain spread over surrounding areas. As soon as he awakened, the erection and pain completely ceased.

Two years after SRPE onset, he sought urologic consultation and underwent a transurethral prostatic resection, despite a negative history for urinary retention. SRPE did not improve after the surgery. He eventually resorted to “increased sexual activity” (details were not provided) in an effort to avert or diminish painful nocturnal erections. When relief from SRPE was not obtained with increased sexual activity, the patient presented for sleep and medical evaluation. PSG monitoring with SRE monitoring demonstrated nocturnal sleep disruptions associated with 3 REM sleep erections lasting 3-8 minutes before culminating in full awakenings with prompt cessation of erections. Brain MRI revealed a left posterior cerebral artery compression on the anterolateral surface of the left hypothalamus, an area which when stimulated (in rats and presumably in humans) induces erections and disinhibits penile reflexes. The location of the vascular compression was thus implicated in the pathophysiology of this case of SRPE.

Another recently reported case involved a 45-year-old man who developed SRPE 2 years after surgical excision of a thoracic ependymoma.⁸¹ Various measures were successfully used to relieve the pain, such as urination and cold water application, along with “physical maneuvers,” but it is unclear whether the latter referred to masturbation or other sexual activity. He was reported to be satisfied with his sex life with normal erections, ejaculations, and orgasm during sexual intercourse 1-4 times weekly. Amitriptyline therapy was not effective, and the patient refused other therapies.

SRPE has been described in over 35 publications, including a controlled study of 10 patients,⁸¹ raising the possibility of sexual overactivity in some of these other cases of SRPE.

Sleep Related Dissociative Disorders

Sexualized (repetitive behavior without affect) and frankly sexual behaviors (with affect) can emerge with sleep related dissociative disorders.⁸³ A particularly striking episode was demonstrated by video-PSG in a 22-year-old woman, with a duration of seven minutes during well-defined EEG wakefulness.⁸³ She quietly lay awake in bed with eyes closed just prior to falling asleep. Then she gradually began jerking her head side-to-side, which proceeded to squirming in bed followed by a crescendo progression of pelvic thrusting, side-to-side movements, and other thrashing behaviors, along with moaning and groaning. Defensive, pained behaviors and moaning often accompanied the sexualized behaviors, and comprised a reenactment of a past abuse scenario that she later believed she had been dreaming about, when in fact she had been awake and remembering in a dissociated state. In her “dream” that she was acting out in bed, her older sister was repeatedly shoving a ruler into her vagina, stomach, and legs, which hurt her but sometimes also sexually aroused her.

In a case of animalistic nocturnal personality involving a 19-year-old male who would prowl around the house on all 4 limbs while growling for approximately 30-60 minutes per episode twice weekly while occasionally chewing on a piece of uncooked bacon,⁸² a thinly disguised recurrent sexualized “dream” during

his nocturnal dissociated states of wakefulness consisted of his being a large jungle cat approaching a female zookeeper who held a piece of raw meat in her hand which he wanted to pounce on and “snatch” it from her hand and eat it. However, “an invisible force field” prevented him from getting near to her, and he felt “frustrated” by being kept away and not having the raw meat. He would then “wake up” (i.e., snap out of his dissociated wakeful state) and be confused and groggy, and would only gradually re-establish contact with reality.

Nocturnal Psychotic Disorders

At least 2 categories associated with sexual delusions and hallucinations that may or may not be sleep related have been documented in the literature, one associated with a primary psychiatric disorder, such as schizophrenia, and the other with Parkinson disease (PD). A vivid example from the first category, involving schizophrenia, is found in the book, *The Professor and the Madman: A Tale of Murder, Insanity, and the Making of the Oxford English Dictionary*.⁸⁴ Dr. William Chester Minor, an American Civil War veteran who was committed to the Broadmoor Criminal Lunatic Asylum in Great Britain for more than 2 decades for committing murder while under the influence of (subsequently diagnosed) schizophrenia, provided more than 10,000 entries for the Oxford English Dictionary. Examples of nocturnal sexual delusions are contained in excerpts taken from the book:

“Already at the time he was admitted he had a detailed awareness of the curious happenings that plagued him at night—always at night. Small boys, he believed, were put up in the rafters above his bed; they came down when he was fast asleep, chloroformed him, and then forced him to perform indecent acts...” *It is a very dirty business,*” he screamed one morning, standing now only in his drawers... *“He made a pimp of me!”*

“...fears that he would be transported from his room at night and made to perform “deeds of the wildest excess” in “dens of infamy” before being returned to his cell by dawn. Once airplanes were invented...he incorporated them into his delusions. Men would then break into his rooms, place him in a flying machine, and take him to brothels in Constantinople, where he would be forced to perform acts of terrible lewdness with cheap women and small girls.”

PSG-documented, immediate post-REM sleep nocturnal delusions, including sexual delusions, were first documented by Arnulf et al. in 2000.⁸⁵ Ten patients with PD and visual hallucinations, 7 of whom also had delusions, were studied. Two of the patients had sexual delusions about the spouse being unfaithful. All had received long-term treatment with dopaminergic drugs, and 5 also with benzodiazepines, but none with anticholinergics or neuroleptics. (Of further interest is one patient with possible sexual RBD whose spouse observed aggressive sleep behaviors associated with moaning and penile erection). A subsequent report by Pacchetti et al. in 2005 on a consecutive series of treated PD patients found that 6 patients suffered from “erotomaniac-type delusions” that often emerged in the evening or after nocturnal awakenings in association with aggressive behaviors and clouded sensorium.⁸⁶ Nocturnal hallucinations were observed in 62 patients, but the content was not described, apart from distinguishing “minor” from “formed” hallucinations. Presence of RBD increased the risk of delusions and hallucinations, with an odds ratio of 2.73.

Hypersexuality After Nocturnal Awakenings

A case has been reported of a 57-year-old man with PD who underwent a pallidotomy, followed 2 years later by deep brain stimulation (DBS) therapy; within 5 months his wife noticed inappropriate hypersexuality, including when he “awoke in the middle of the night demanding sexual intercourse...”⁸⁷ No further sleep related details were given and PSG was not performed. Hypersexuality can be triggered by a variety of brain insults,⁸⁸ but apart from the case just cited, and the epileptic cases previously cited, we are not aware of any other descriptions of nocturnal or sleep related hypersexuality in neurologic patients with wakeful hypersexuality.

Miscellaneous Sleep and Sex Associations

Naps:

One of the authors (IA, unpublished case) evaluated a patient with PD being treated with dopaminergic therapy who reported various types of pleasant kinesthetic experiences and visual hallucinations upon awakening from both nocturnal sleep and daytime naps, associated with sleep paralysis. This 54-year-old woman had a mild, de novo form of PD. She reported out of body experiences, with her body floating above her bed, then moving downstairs up to the entrance door, and then being suddenly projected back in the bed. She also saw colorful sceneries, including blue skies and green landscapes, having the feeling of standing on the top of a church, and wonderful golden horses surrounding a chimney. During one early morning while sleeping in bed with her husband, and having her back turned to him, she suddenly woke with the internal sensation that he had just penetrated her from behind with his penis. She couldn't move because of her SP, but after a while she could turn and was surprised to see him sleeping. Also, after awakening from afternoon naps, she regularly experienced a man making love to her. Although she could not see his face, she had the feeling that it was someone familiar, but not her husband. She sometimes reached orgasm. An overnight PSG and next-day MSLT indicated SOREMPs and short sleep latency, indicative of “narcolepsy-like” phenotype, as described in several other PD patients. During wakefulness, the woman, who had menopause 4 years before, had regular, satisfying sex with her husband, and had no lover.

In another recent case (JE Tatman, unpublished data), a 32-year-old divorced male presented to sleep disorders center with the complaint of excessive sleepiness, nocturnal awakenings, and intermittent RLS. He underwent a PSG followed by a MSLT the next day. The PSG was abnormal for poor sleep efficiency and a prolonged >2 hour awakening. Sleep during his third MSLT nap was immediately preceded by masturbation that was recorded on videotape. He did not sleep during any of the 3 other MSLT nap opportunities. During 2 subsequent interviews he confirmed the masturbation incident during the MSLT and related it to being “very frustrated” about his poor nocturnal sleep the preceding night and the lack of sleep during the preceding 2 nap opportunities. He also reported frequent difficulty in initiating sleep or returning to sleep at home without prior sexual activity, either by masturbating or by having sex with a female partner. His ex-wife complained about his frequent insistence on having sex as soon as they got into bed at night in preparation for sleep.

Sleep Erections and Sexual Vulnerability:

An example is contained in a report of a study on men who reported being pressured or forced to have sex.⁸⁹ One man wrote that he came home “drunk from the bars” and passed out in a friend's bunk bed. He awoke to find an acquaintance—who wanted to date him—“riding” him in order to “get off.” Presumably these were REM sleep erections, and given the customary atonia of REM sleep, a male in REM sleep is in a demonstrated state of sexual vulnerability, with an involuntary physiologic penile erection accompanied by generalized muscle paralysis and an increased sensory threshold.

The above scenario has been played out with forensic consequences, as reported in 2001.⁹⁰ The report, translated from French by one of the authors (IA), reads thusly: “An unusual case of ‘rape by surprise’ during sleep without the victim being aroused was recently arbitrated in France. A 24-year-old man woke up a Saturday morning with painful anal lesions that made him suspect he had been raped during his sleep the night before. The legal medicine examination indeed reported on visibly recent tears of the anal margin. The young man described himself as heterosexual, a deep sleeper and not an alcohol drinker. On the preceding Friday afternoon, he was invited by his boss to have a sauna, then go swimming and finally to drink 7 beers (11.4 grams pure alcohol) between 9 p.m. and 1 a.m. As his boss had no access to public transportation at that time, the young man proposed for him to sleep in his sofa, while he slept in his mezzanine. The boss (who stayed in jail for 2 years before the judgment) reported that he indeed had anal sex with the young man soon after going to bed on that ‘alcoholic night,’ but felt he was a willing participant in having sex because he had a penile erection and did not react to his anal penetration....The court finally arbitrated that both the victim and the defendant were right, and the defendant was released.” Therefore the visible sexual arousal (penile erection, presumably during REM sleep) and lack of resistance to anal penetration convinced the boss that this was consensual sex on the part of the 24-year-old man.

Medication-Induced States:

In a report on amnesic sleep related eating associated with zolpidem therapy of insomnia,⁹¹ 4 additional patients were mentioned who had engaged in non-eating amnesic parasomnias induced by zolpidem, 2 of whom had engaged in sleepsex with their usual partners. The unusual feature of these two cases was that sleepsex had not occurred in their lives prior to the initiation of zolpidem therapy. In a third case involving zolpidem, a 43-year-old woman developed de novo sleepsex together with morbid sleep related eating (requiring bariatric surgery).⁹² During sleep medicine consultation prompted by progressive weight gain after the bariatric surgery, this was attributed to the initiation of nightly zolpidem therapy 4 years previously. Her husband observed not only the new-onset sleepsex but also her unprecedented sexual behaviors, such as being the initiator of sex and being aggressive, with groping and biting, for which she had no recall.⁹² In addition, in a series of 54 patients with narcolepsy-cataplexy treated with sodium oxybate, two male patients had to be withdrawn from the drug because of hypersexuality. In one case the patient was craving for the drug as its consumption was associated with enhanced erotic fantasy thinking. The other patient reported he was running

around in his house, naked, sexually aroused, with an erection after he took the drug. He was concerned it could embarrass his young daughter to see him in this state. Both patients had this sexual arousal while fully awake (Geert Mayer, cases presented during the symposium, "Narcolepsy: multi-symptoms therapeutic approach," at the European Sleep Research Society meeting, Innsbruck, Austria, September 13, 2006).

Forensic Considerations

Although an in-depth analysis of this topic is beyond the scope of this review, some clinical and medicolegal points pertinent to sexsomnia can be addressed. Sleepwalking with lewd behavior in public that prompted several arrests (from exposing, fondling, masturbating himself, but never in front of another person) was reported in a 21-year-old man with childhood-onset sleepwalking, and without any history of unusual, problematic or criminal sexual behavior during the daytime.⁹² Charges were dropped because of the apparent sleepwalking. Clinical and PSG evaluations later supported the diagnosis of sleepwalking with pseudo-paraphilia (i.e. pseudo-psychosexual disorder).

In regard to sleepwalking and the "sleepwalking defense," 2 additional medico-legal categories have been proposed, including their use in cases with sexual charges involving adults or minors: "Parasomnia with continuing danger as a non-insane automatism," and "Parasomnia with (intermittent) state-dependent continuing danger as a non-insane automatism."⁹³ The first category was proposed in response to a report entitled "Sleepwalking and Indecent Exposure."⁹³ A sleepwalker (or any other type of "parasomniac") who intentionally engages in any behavior that for him is known to trigger sleepwalking should be recognized as "carrying de facto full legal culpability for the consequences of any subsequent actions promoted by such behavior." The behavior in question was excessive alcohol consumption, and the case involved a 27-year-old man convicted of indecent exposure despite evidence indicating that he had been sleepwalking. Both this man and his mother reported that excessive use of alcohol was known to trigger his sleepwalking. The scientific evidence and forensic considerations of alcohol-induced sleepwalking or confusional arousals as a defense to criminal behavior have recently been critically reviewed.⁹⁴ In addition, factors that can predispose, prime and precipitate NREM parasomnias in adults have been recently reviewed, with forensic (including sexsomnia) implications.⁹⁵

Another proposed medicolegal category was contained in the report "An analysis of a recent criminal trial involving sexual misconduct with a child, alcohol abuse and a successful sleepwalking defence: arguments supporting two proposed new forensic categories."⁹⁶ This report was prompted by a Vancouver, British Columbia case in 1996 involving a 26-year-old man who drank 36 beers with a friend over a 10-hour period, and then—after his friend was injured and taken to a hospital, where the friend's wife joined him—he ended up in the same bed as his friend's 4-year-old daughter, who later was found "upset and crying...and saying, 'he took my panties off and he hurt me.'" Medical examination revealed an abrasion in the little girl's vagina. Therefore, there was no medical-legal doubt that the girl was "interfered with" by the defendant, who then invoked the sleepwalking defense. He had an observed history of childhood onset, recurrent sleepwalking, as did his mother and sister. In one prior dramatic episode, "the night before his marriage, when after he had been out drinking...he...got up and urinated in a basket next

to his sister's bed, splashing her...he had no recall of this..." In the subsequent forensic case, both the prosecution and defense agreed that there was no prior history of pedophilia, no prior criminal history, and the defendant was not faking sleep or amnesia. However, there was no mention of whether he had been evaluated or diagnosed with alcoholism, or whether he had received any DWI (driving while intoxicated) citations.

The court identified 5 additional factors, besides the personal and family history of sleepwalking, that supported the defendant's sleepwalking defense:

- disorientation upon awakening from sleep, as determined by the arresting police officers. (However, this could have been lingering intoxication after sharing 36 beers with his friend)
- amnesia for the event (this also could have been due to intoxication)
- presence of factors known to trigger sleepwalking—alcohol; fatigue; stress
- no apparent attempt to conceal the crime
- the crime was out of character for this person.

The court acquitted the defendant because he "was not conscious of what he was doing" at the time of the sexual misconduct. By legal criteria, he was not found to have any internal disease of the brain or mind. Rather, "fleeting disturbances of consciousness which are external to the person's individual, emotional and psychological make-up do not fall within the concept of a disease of the mind. The evidence is that this episode was triggered by external cause." The identified "external causes" consisted of (excessive) alcohol consumption, stress, and sleep deprivation.

This trial is of medical-legal importance because it raised the issue of whether a sleepwalker with a prior sleepwalking episode(s) provoked by alcohol excess should be held legally culpable for any subsequent alcohol-provoked sleepwalking with criminal misconduct. In our opinion, the Vancouver court did not adequately address the issue of alcohol use in this case, since we believe that it should have played a crucial role in determining whether the defendant's sleepwalking posed a "continuing danger." Instead, the final judgment considered the set of circumstances resulting in sexual misconduct during presumed sleepwalking to be so extraordinary and unique that the risk of recurrence was found to be negligible, and he was thus granted an outright acquittal. There was no mention of future culpability were he to once again drink alcohol excessively and engage in sleepwalking with criminal misconduct, either sexual or nonsexual. Furthermore, there was no recommendation for the defendant to obtain clinical consultations for possible alcohol abuse disorder and/or for his sleepwalking, and how to minimize its recurrence.

As a result of this Vancouver case, in 1998 an expanded category of "(Intermittent) State-Dependent Continuing Danger" was introduced with a focus on the legal consequences of behaviors willfully engaged in by a person who already knows that such behavior carries a definite risk for triggering a parasomnia recurrence (including criminal sleepsex).⁹⁶ The acquitted person, as part of the final judgment, would be informed that the "SW defense" would be unacceptable for any future SW episode resulting from such high-risk behavior. Guidelines for the proper methods for evaluating forensic parasomnia cases, including those involving presumed sexsomnia, have been developed and refined.⁹⁷ Finally, malingering should always be considered in clinical and medical-legal cases.⁹⁸

DISCUSSION

A broad set of problematic and clinically consequential sexual behaviors can be associated with parasomnias and other sleep disorders, and emerge during vulnerable sleep states, sleep-wake transitional states, or wakeful states. Furthermore, seizure disorders and other neurologic and medical disorders can manifest with abnormal sleep related sexual behaviors and experiences. A classification of these associations, as shown in Table 1, provides a framework for expansion and refinements as more cases (preferably from large case series) are published and further knowledge is gained in this area. Clinicians across various specialties should be informed about the clinical and medical-legal aspects of “sleep and sex” and consider questioning their patients about any sexual problems associated with their sleep disorders, or about any sleep and sexual problems associated with their neurologic or medical disorders. The latter comment has already been well-recognized in regards to the adverse (i.e., suppressing) effects on libido and sexual activity resulting from a range of chronic sleep disorders, such as OSA, RLS, insomnia, etc. This is a related but separate issue from the focus of this report. Questioning on altered sexuality (libido and behavior) related to a sleep complaint or disorder is strongly encouraged.

The striking male predominance in the published cases of sleepsex, as shown in Table 2, underscores the major sex discordances often found with adult parasomnias.¹ RBD is also male-predominant,¹ along with OSA “pseudo-RBD,”⁹⁹ and injurious sleepwalking and sleep terrors.¹ In contrast, sleep related eating disorder and sleep related dissociative disorders are female-predominant.¹ The basis for the male predominance in the reported cases of sleepsex is not known. The data in Table 2 indicate that sleepsex is often a longstanding disorder that carries major adverse physical, psychosocial, and legal consequences. However, 5 of the bed partners and 3 of the patients reported pleasurable aspects to the sleepsex. Fortunately, the reported sexual parasomnias have been very responsive to standard therapy, consisting of clonazepam for NREM parasomnias and presumed RBD, or CPAP therapy of OSA precipitating confusional arousals with sexual behaviors.

The complete amnesia for the sleepsex reported in all 31 parasomnia cases is contrasted by only 2 of the 7 sleep related epilepsy patients having amnesia for their nocturnal sexual episodes, suggesting that their memory systems were either activated and/or not suppressed by the seizure-related episodes. However, the very small sample size precludes any generalizations. Also, selection bias for complete amnesia in the 31 published parasomnia cases may have been present, for several reasons: only the most severe cases may have presented clinically with profound confusional arousals or sleepwalking linked with prominent sexual disinhibition; embarrassment in recalling sleepsex would lead to no reported recall; and in medicolegal cases, claiming amnesia for sleepsex would carry secondary gain. Therefore, the issue of amnesia or recall of sleepsex related to parasomnias or epilepsy remains an open question and more data need to be gathered.

Terminology

Based on current knowledge reflected in the published literature to date, as already cited in this report, we would advocate the use of the terms pertaining to abnormal sleep and sex listed in Table 3 in future reports in the sleep medicine and other clinical-

scientific literature. These terms, as indicated in the table, cover both etiologic and purely behavior-experiential descriptors. These terms should be considered sufficiently clear to be used in patient-related medicolegal reports. Further subtyping is facilitated by Table 3, such as identifying whether problematic events (presumably) occurred within NREM or REM sleep, during arousals from these sleep stages, during wake-sleep transitions, or during established wakefulness. Additionally, specific sexual behaviors and experiences can be listed under each term, as shown in Table 2, with further elucidations possible, such as describing seizure-related sexual automatisms (e.g., paroxysmal nymphomania). On the other hand, terms such as “restless vagina syndrome” as a variant of RLS should be avoided unless future reported cases justify the use of this term with sufficient documentation.

The topic of “sexualized” movements or behaviors, particularly repetitive pelvic thrusting, was discussed in relation to nocturnal frontal lobe or temporal lobe seizures, RLS, and sleep related dissociative disorders. These types of “sexualized” activity reflect restricted components of the sexual repertoire expressed in a repetitive manner and often without an affective component. The use of the term “sexualized” should be reserved for these behavioral phenomena that appear to be expressions of “central pattern generator” activity originating in the brainstem, as elucidated by Tassinari and colleagues.¹⁰⁰

The Evaluation and Management of Sleep Related Sexual Complaints

Patients with parasomnias, OSA, and RLS should be questioned (with their bed partners) about any associated sleepsex. We believe that a sleep-wake questionnaire for patients presenting to a sleep center should now include 2 questions on the topic of sleep and sex: “Has your libido or sexual activity changed, either while you are awake, or falling asleep, or during your sleep? Has your bed partner observed any sexual vocalizations or sexual behaviors on your part while you are asleep?” For those patients having sleepsex, the frequency, severity, longitudinal course, and predisposing and precipitating factors should be identified, along with any psychosocial and physical consequences. Covering the problematic sleepsex experiences identified by Mangan⁴¹ can be useful in distinguishing between sleepsex “initiator” and “recipient” problems: the initiators often feel “guilt, confusion, shame, disappointment, frustration, embarrassment and self-incrimination,” whereas the recipients often feel “fear, lack of emotional intimacy, repulsion, sexual abandonment, annoyance and suspicion.”

Screening psychological testing, e.g. Minnesota Multiphasic Personality Inventory, Beck Depression Inventory, should be administered, and formal psychiatric consultation should be considered on a case-by-case basis.

Time-synchronized video-PSG with seizure-montage and fast EEG speeds, and upper/lower limb EMG monitoring are strongly recommended in evaluating the complaint of sleepsex. At present SRE monitoring is not recommended, for the reasons given in the next section. Although the yield in identifying parasomnia behaviors and confirming the diagnosis by PSG is substantial, sexual behaviors during sleep have rarely been documented, as already described. Similarly, with epileptic sexual seizures, only 2 cases have documented sleepsex during PSG or sleep EEG monitoring, as described.

Management of this problem can occur along 2 dimensions: the medical management of any identified underlying sleep, medical, neurologic, or psychiatric disorder (or their therapies) promoting the problematic sexuality; individual and/or couple's psychotherapy to address any predisposing issues and/or to address the consequences of the problematic sexuality.

SRE Monitoring

The lack of SRE monitoring during PSG in any of the published sexsomnia cases allows only for speculation at present. Given the preponderance of published cases of sleepsex involving males diagnosed with a disorder of arousal from NREM sleep, the question of penile erections emerging during NREM sleep or immediately after an arousal from NREM sleep must be considered. A problematic issue is that penile erections during sleep in humans are predominantly, but not exclusively, a REM sleep phenomenon,¹⁰¹⁻¹⁰³ as erections can emerge for minutes during transitional NREM-REM sleep periods and be sustained during REM-NREM transitional sleep periods.¹⁰¹ Furthermore, medications such as trazodone can accelerate the onset of tumescence in NREM sleep prior to REM sleep, and also prolong erections into NREM sleep after cessation of REM sleep.¹⁰⁴ Various conditions, such as spinal cord injury, other neurogenic problems, and other disorders or medications can be associated with NREM erections representing dissociations of SRE from REM sleep.^{101,105} Therefore, the interpretation of NREM sleep erections may be the indeterminate. Penile erections may also emerge during partial arousals from NREM sleep as a manifestation of emerging wakefulness with sexual arousal. However, it is unlikely that full wakefulness is established, since complete amnesia occurred in all reported cases of sleepsex. There is precedent in the animal literature for penile erections during NREM sleep, as documented in the armadillo, which also had the absence of penile erections during REM sleep.¹⁰⁶ Rats, in contrast, demonstrate a pattern identical to humans with REM sleep penile erections.¹⁰⁷

Adults Co-sleeping with Teens and Children

A recommendation based on the published data is that adults or teens with NREM sleep parasomnias should be informed about the risks of co-sleeping, including co-sleeping with minors, especially after drinking alcohol or after sleep deprivation. There may be inadvertent touching during sleep that could precipitate sexual behaviors, resulting in legal and psychosocial consequences. A notable example is contained in the report on "sexsomnia,"³³ in which a 32-year-old male shared a bed with 2 children during a nap at a friend's home, "and was subsequently awakened by a friend who said that one of the children, a 10-year-old girl, claimed that he had inserted a finger into her vagina. Mr. F. could not recall the event, saying he had been asleep at the time."

Does "Sexual RBD" Exist?

The issue of "sexual RBD" is problematic, since sexual behaviors during REM sleep have not yet been documented by PSG, and the 3 reported cases of sexual RBD did not involve any dream-enacting behaviors.³ This is atypical for RBD, for which "dream-enacting behaviors" is a virtual hallmark.¹ Hundreds of RBD cases have been reported in the peer-reviewed literature, but

apart from these 3 cases, none has involved sexual behaviors.

An experimental animal model of RBD involving cats does not include sexual behavior.¹⁰⁸ In fact among a large series of cats with experimental RBD, no sexual behavior was ever observed during REM sleep, apart from one case of a female cat demonstrating the characteristic meowing while in heat (J-P Sastre, personal communication).

However, 2 additional cases support the possibility of sexual RBD. An elderly man with a typical history of RBD sodomized his wife in bed one night while he was clearly asleep, but it was not known whether he was dreaming at the time, and he later denied recall of the event. The presence of an erection suggests a REM sleep event (sexual RBD), which was a singular event for this man and his wife (CHS, MWM, unpublished case). The other case involved a previously described man with PD treated with dopaminergic therapy whose wife observed an erection while he moaned and moved side-to-side in bed.⁸⁴ It is known that patients with PD receiving dopaminergic therapy are prone to sexual and other forms of disinhibited behaviors during wakefulness.¹⁰⁹ Furthermore, male PD patients carry an elevated risk for subclinical and clinical RBD.¹¹⁰ The available clinical evidence would thus suggest that the most likely clinical scenario for identifying "sexual RBD" would be middle-aged and older male patients with PD and subclinical or clinical RBD, who are receiving dopaminergic therapy, and who also have waking hallucinations and delusions (a recognized complication of PD⁸⁶), particularly if erotic content is present.

Concluding Comments

The first synthesis of the literature assessing the effects of neurologic insults on human sexual behavior has recently been published, with six key brain regions (including the hypothalamus, also a key sleep center) being identified as mediating specific aspects of sexual behavior.⁸⁸ Although sleep was not specifically addressed in this review, the various conditions identified with hypersexuality in wakefulness or after awakenings (e.g. after pallidotomy and deep brain stimulation therapies of PD)⁸⁷ may be relevant to problematic sleep and sex.

A broad set of research studies should be encouraged by this review, such as obtaining normative data on the range of non-problematic and problematic sexual behaviors during sleep in both the general population and various clinical populations, particularly those patients who appear to be at highest risk, i.e., those with NREM parasomnias, OSA, other sleep disorders, nocturnal seizures, and PD. Nocturnal epilepsy monitoring should include not only a comprehensive EEG montage but also PSG with time-synchronized audio-videotaping of sleep behaviors. Finally, a review of the clinical correlates of erectile-sexual dysfunction, as detected during sleep with SRE monitoring, is beyond the scope of this review, but has recently been addressed comprehensively, with a proposed set of clinical indications for formal SRE monitoring in a sleep laboratory.^{107,111}

Note Added in Proof

Two cases of sexual behaviors in sleep (SBS) associated with PSG-confirmed parasomnia overlap disorder have been identified in a 41 y.o. man and 61 y.o. woman (Cicolin A, et al., personal communication & submitted manuscript). During the first SWS period in the PSG study, the woman attempted to remove her py-

jamás and began to masturbate while moaning, and was amnesic for the episode. In both cases, SBS were diagnosed as NREM parasomnias, and RBD was considered to be a separate parasomnia in both cases of parasomnia overlap disorder.

A case of hypersexuality induced by dopaminergic therapy of RLS has been identified in a 54 y.o. married man who developed new-onset, longstanding, compulsive (multiple times daily) masturbation after starting pramipexole therapy that effectively controlled his RLS, in contrast to prior therapy with levodopa that had been ineffective for RLS and had not induced any change in sexual arousal or behavior. Discontinuation of pramipexole immediately eliminated the compulsive masturbation. (Ramseyer J, et al., manuscript in preparation).

APPENDIX

I) Confusional Arousal With Sleepsex In Literature

In *Tess of the d'Urbervilles* (1891), a confusional arousal with sleepsex and rape comprises the pivotal point in Thomas Hardy's novel. The protagonist Tess had always rebuffed the persistent advances of a man named Alec. He finally was able to take advantage of his base desires when an exhausted Tess fell asleep rapidly and deeply inside a forest late at night, and forced himself on her, raped her and made her pregnant, with tragic consequences: "She was sleeping soundly, and upon her eyelashes there lingered tears. Darkness and silence ruled everywhere around... But, might some say, where was Tess's guardian angel? Where was the providence of her simple faith? Perhaps, like that other god of whom the ironical Tishbite spoke, he was talking, or he was pursuing, or he was in a journey, or he was sleeping and not to be awaked. Why it was that upon this beautiful feminine tissue, sensitive as gossamer and practically blank as snow as yet, there should have been traced such a coarse pattern as it was doomed to receive... many thousand years of analytical philosophy have failed to explain to our sense of order." Later Hardy referred to that sleep scene in which Tess "had been stirred to confused surrender..." and had acted with "inadvertence."

Tess had six risk factors that night promoting confusional arousals: her youth (the younger the age, the greater the risk for confusional arousals); acute sleep deprivation (waking up early and falling asleep at a "late hour" that night); physical exhaustion from walking many miles across changing terrain that day; forced awakening by her assailant; acute stress from being confronted by a jealous female who wanted to fight with her earlier in the evening; and major, longstanding emotional stress—her family and Alec (her assailant) were pressuring her to marry a man (i.e., Alec) she didn't love.

Strong homeostatic and circadian factors were thus promoting precipitous and deep sleep, and Tess collapsed on the ground in the forest and quite probably rapidly fell into delta sleep. Shortly afterwards a confusional arousal was elicited during a forced awakening when Alec rapes her. She does not resist him in her state of profound sleepiness, a stark contrast to her unwavering rejection of his advances while being awake.

II) Sleepsex in Movies

Two prominent examples, one comedic and the other dramatic, are contained in the following Hollywood feature films: i) *Planes*,

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Trains and Automobiles (1987, starring John Candy and Steve Martin). After an exhausting and aggravating day of not getting anywhere with any mode of transport, two strangers then have to share a hotel room with only one bed. John Candy and Steve Martin did manage to fall asleep. The next morning, John Candy has a confusional arousal and initiates sexual foreplay with a surprised and resistant Steve Martin. When John Candy finally awakens, he is shocked to realize what he had inadvertently done, and repeatedly denies any intentional wrongdoing. ii) *Brokeback Mountain* (2005, Oscar-nominated film directed by Oscar-winning best director Ang Lee, starring Heath Ledger and Jake Gyllenhaal). The first moment of physical and sexual intimacy between the two cowboy leads came suddenly out of sleep. While asleep in their sleeping bags inside a tent, Jake's character suddenly roused from sleep, turned over and lurched over his partner who was sleeping with his back towards him, who then also roused and they clasped their hands and then sat up and were kneeling in front of each other when they began passionate kissing.

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