Section

Introduction
If the notions of dream and nightmare are centuries old, going back to ancient Egyptian and Jewish civilizations, the distinction between nightmares and parasomnias is recent. It is interesting to note that already in 1932 Kouretas and Scouras [1] reported the complete absence of muscle tone during nightmares in one of their patients, but Kleitman in his seminal monograph places “nightmares, night terrors, somniloquy, bruxism and jactitation, enuresis, numbness and hypalgia, and personality dissociation” under the terminology “parasomnia” [2]. The term “parasomnia”, derived from the greek “para” meaning “around” and the latin “somnus” meaning “sleep”, had been coined in 1932 by the French researcher Henri Roger who published a monograph entitled “Les Troubles du Sommeil-Hypersomnies, Insomnies, and Parasomnies” [3]. In this monograph, Roger gave an excellent description of an episode of sleep terror in a child, and also described a somnambulistic episode. Kleitman’s review of previous literature shows that the difference between nightmares and sleep terrors was not well perceived before Roger; rather, many syndromes now understood to be within the rubric “parasomnia” had been investigated independently.

As parasomnias became distinguishable from nightmares, a possible link between such episodic nocturnal phenomena and seizure disorders was proposed. Henri Gastaut in Marseille, France, wanted to understand whether a relationship existed between the common parasomnias and seizures. To resolve this question, the Marseille team monitored individuals with abnormal nocturnal behavior and normal matched controls during nocturnal sleep. In 1965, Gastaut and Broughton reported some of their findings and hypothesized that most “episodic phenomena,” as they called events, were non-epileptic in nature and occurred out of NREM sleep, predominantly during slow wave sleep [4]. These authors confirmed the clinical description of sleep terrors reported by Kanner in 1935 [5]. However, they indicated that many of these phenomena occurred during NREM sleep stages 3 and 4, including enuresis. They even performed cystometric recordings during sleep in children and reported that the enuretic episodes occurred when the cystic pressure waves reached 120 cm H2O. Jacobson et al. [6] and Kales and Jacobson [7] focused more on sleepwalking episodes and had similar findings to Gastaut and colleagues, indicating the occurrence of events during NREM sleep, particularly stages 3 and 4. They emphasized subjects’ “indifference to environment” during episodes, and the “complete amnesia” of the events the following morning. The authors made the interesting observation that a sleepwalking event could be induced by having a known somnambulistic child stand up in slow wave sleep.

These studies were presented and discussed at the 15th European Meeting on Electroencephalography held in 1967 in Bologna, Italy, a meeting organized by Lugaresi and Gastaut and attended by European and North American researchers. Thereafter, in 1968 Broughton questioned the pathophysiological mechanisms underlying sleepwalking, sleep terrors, and head-banging [8]. He dissociated them formally from nocturnal epilepsy, emphasizing the well-demonstrated presence of sleep disturbance and occurrence in NREM sleep – and more particularly stage 4 sleep; he termed such events “disorders of arousal”. He also differentiated nocturnal enuresis from the other phenomena.

During the same period several authors emphasized the familial nature of the disorders. Kales and Jacobson [7] indicated that sleepwalking was found in families; in 1980, Kales et al. suggested a two-threshold multifactorial mode of inheritance [9]. Bakwin (1970)
found that monozygotic twins were concordant for sleepwalking considerably more than dizygotic twins \((p = 0.06)\) [10]. In 1972, Hallstrom suggested an autosomal dominant pattern of heredity for sleep terrors [11]. None of these studies fully explained observed patterns of inheritance, despite the fact that familial patterns were seen in parasomnias. In 1971, Klackenberg, in his classic longitudinal study which followed about 200 children from birth in 1955 until age 16 in Stockholm [12], made an interesting observation: children who would become somnambulistic later in life had much more restless sleep at the age of 4–5 years. Another important finding of this longitudinal study was that somnambulism and sleep terrors could be very rare or chronic. In the “chronic” or “habitual” sleepwalkers, enuresis occurred from 5 until 16 years of age and represented 2.5% of the sample. This number is of interest, as another epidemiologic study found similar results. Ohayon \textit{et al}., based on a representative sample of the adult population, found that sleepwalking affects 2.5% of the population [13]. Hublin \textit{et al}., on the other hand, indicated that 25% of children with a history of sleepwalking will sleepwalk as adults [14].

The following 20 years focused on pharmacological treatment of parasomnias with emphasis on the use of benzodiazepines, with variable results. A short report by an English orthodontist was completely ignored. Timms, who performed rapid maxillary expansion on children with a narrow hard palate, but who did not routinely perform sleep studies, described clinical amelioration of symptoms that today would suggest the presence of sleep-disordered breathing [15]. He also described the elimination of chronic sleepwalking events following rapid maxillary expansion.

In 1999, Ohayon \textit{et al}. in their epidemiological studies on sleepwalking and sleep terrors found that obstructive sleep apnea syndrome was the most common sleep disorder associated with parasomnias between the ages of 15 and 24 years [13]. In 2002, Espa \textit{et al}. reported a similar association in a small number of subjects focusing on sleepwalking, as did Guilleminault \textit{et al}., investigating sexual violence and confusional arousal [16,17]. Around the same time, Ohayon \textit{et al}. reported an important relationship between bruxism and the presence of sleep-disordered breathing (SDB) [18]. Goodwin \textit{et al}., in the pediatric Tuscon sleep cohort, also identified a frequent association between SDB and sleepwalking in 2004 [19].

Investigations of sleep EEG in parasomnias became more focused. In 1995, Zucconi \textit{et al}.., using a cyclic alternating pattern (CAP) scoring system, indicated that various parasomnias were associated with an abnormally elevated CAP rate [20]. This finding was confirmed by Guilleminault \textit{et al} both in children and in young adult sleepwalkers [21,22].

Another type of investigation of the sleep EEG was performed by Gaudreau \textit{et al} [23] and Guilleminault \textit{et al} [24] using quantified EEG. Both of these groups showed that subjects with chronic parasomnias had a significant decrease in delta power during the first NREM–REM sleep cycle, and lower than expected delta power in the second cycle. Guilleminault \textit{et al} compared quantified EEG and CAP analyses and showed that the results were concurrent when these two analyses were applied to the same subjects [21]. The conclusions drawn by these authors were that sleep EEG analyses indicated an “instability of NREM sleep.” The investigations of Guilleminault \textit{et al}. of children and adults [25] with chronic parasomnias showed that: (1) the individuals with chronic parasomnia presented such an instability of NREM sleep every night, with or without the presence of the parasomnia; (2) the subjects presented with another sleep disorder (most commonly SDB) which was responsible for the instability of NREM sleep; (3) the presence of an additional factor (e.g. stress, alcohol, fever, sleep deprivation) was needed to provoke the parasomnia episode; and (4) treatment of the underlying sleep disorder (including rapid maxillary distraction as noted by Timms) alleviated the instability of NREM sleep and eliminated or improved the parasomnia.

Recent work on bruxism suggests that teeth-grinding may be an attempt to protect against the occurrence of sleep apnea, which raises the question of whether this symptom should be placed in the same category as the others, despite the fact that it is also associated with NREM sleep instability. Understanding of parasomnias has evolved over time and will continue to evolve. As suggested by Guilleminault \textit{et al}. to the extent that parasomnias are triggered by unstable NREM sleep caused by SDB, the heritability of parasomnias may be related to the underlying familial facial traits involved in the development of SDB [26].

Epileptic disorders were shown to be rarely involved in abnormal behavior during NREM sleep, but when sleep-related seizure disorders are present, specific seizure entities are implicated. In sleep-related seizure disorder, polysomnographic studies have demonstrated that a frontal lobe locus is most common, and less frequently the mesiotemporal
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region is involved. Pedley and Guilleminault [27] tried to chronicle distinguishing characteristics between NREM parasomnias and sleep-related seizures, but such differences are based on statistical analyses and may not be true for every event. The stereotypy of the abnormal behavior of a seizure disorder is the most constant finding. Seizures are also more likely to occur out of stage 2 NREM and during the second half of the night. Studies from the 1970s and 1980s have demonstrated that nocturnal seizures represent a bit less than 1% of the abnormal behavior during sleep.

Finally, nocturnal polysomnography has allowed the dissociation of NREM from REM sleep abnormal behavior. The initial description of what is now known as “REM sleep behavior disorder” (RBD) came from Japanese researchers. It was related to the disappearance of the normal atonia of REM sleep reported during delirium secondary to alcohol intoxication [28]; it was again described by Japanese researchers in psychiatric patients [29]. In 1986, Quera-Salva and Guilleminault presented cases of olivo-ponto-cerebellar degeneration and REM without atonia associated with the acting out of dreams [30].

In the same year, Schenk et al. reported the presence of other neurological syndromes, particularly parkinsonianism, and called it a “new category of parasomnia” [31]. In 1987 these authors presented idiopathic cases and named the syndrome “REM sleep behavior disorder” [32]. Aspects of these cases resembled the 1965 report by Jouvet and Delorme of cats and investigations of the Mayo Clinic group [35] which indicate that this association is common. Are other neurodegenerative diseases involved in RBD? Alzheimer’s Disease has been reported in association with RBD, but this seems rare to exceptional; other neurodegenerative diseases are involved more frequently. Whether idiopathic RBD truly exists is still an unanswered question.

References


Section 1

Chapter 2

Epidemiology of parasomnias

Maurice M. Ohayon

Introduction

Parasomnias are sleep disorders characterized by abnormal behavioral or physiological events occurring at different sleep stages. These disorders have seldom been investigated in the adult general population although, as we will see, they are very frequent. Many of these parasomnias are accompanied with behaviors that can be potentially dangerous to the sleeper or to others, leading sometimes to dramatic consequences, such as self-mutilation, murder or suicide.

Parasomnias presented in this chapter are those included in the latest International Classification of Sleep Disorders (ICSD-2) [1]. They are divided into three main categories: disorders of arousal, parasomnias associated with REM sleep, and other parasomnias. There are 15 possible diagnoses of parasomnias. For many of them, information about their prevalence and incidence is limited or inexisten.

Disorders of arousal

Arousal parasomnias occur mainly during NREM sleep. This group consists of confusional arousals, sleepwalking and sleep terrors. These parasomnias occur primarily in childhood and normally cease by adolescence. A same individual may experience more than one type of arousal parasomnias. However, epidemiological figures for these co-occurrences are yet to be determined.

Confusional arousals

Confusional arousals, or sleep drunkenness, correspond to a mental confusion occurring upon waking, be it at night or in the morning. Physiologically speaking, cerebral reactivation to external stimuli is altered in the transitional period from NREM sleep to wakefulness. The sleeper appears awake but behavior may be very inappropriate, marked by memory deficits, disorientation in time and space and slow mentation and speech. An episode can be triggered by a forced awakening. Complex behaviors may occur, resulting in potentially dangerous situations for the sleeper and others.

Confusional arousals have received little attention in epidemiological studies. In children, Laberge et al. [2] have estimated that about 17% of children between 3 and 13 years are experiencing occasional or frequent episodes of confusional arousals. In our study, we have estimated their frequency in the general population to be 4.2%; decreasing from 6.1% in the 15–24 age group to 3.3% in the 25–34 and stabilizing around 2% after 35 years old [3]. No other figure exists for comparative purposes. Although confusional arousals are suspected to have a genetic component, genetic epidemiological prevalence is yet to be determined.

The phenomenon, however, has generated much more interest from a forensic standpoint. The first review of the literature devoted to assaults committed in a state of confusional arousal was completed in 1905 by Gudden [4]. It included 18 cases among which 10 resulted in homicides. In 1943, a similar review by Schmidt [5] covered 35 cases (15 homicides and 20 bodily injuries). Thirty later years, A. Bonkalo [6] reviewed cases of murder committed during confusional arousals that occurred from 1791 to 1974. He identified approximately 20 cases, mostly perpetrated by men. More recently, B. Raschka [7] has brought the case of a man of 54 years that, for no apparent reason, had assaulted two police officers shortly after they had awakened him in his car. Polysomnographic findings indicated that the man suffered from obstructive sleep apnea syndrome. It was concluded that the combination...
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of this disorder with alcohol intake and sudden arousal had prompted the violent behavior.

Several studies and case reports [7–10] have underscored the association between sleep apnea and violent behavior. One of these found that cerebral hypoxia provoked by recurrent breathing pauses during sleep could, over the long term, cause excessive daytime sleepiness, road accidents, impulsive behaviors at awakening, and even cerebral damage [10]. Being suddenly awakened when in a relative state of cerebral anoxia can produce a confusional state that can translate into violent behavior [7].

Precipitant factors of confusional arousals include sleep deprivation, consumption of alcohol, hypnotics or tranquilizers prior to bedtime, or a sudden awakening from sleep [3,11,12].

Sleepwalking

Sleepwalking is a parasomnia that occurs in NREM sleep stages 3 and 4, usually in the first third of the principal sleep episode. This disorder is characterized by automatic behavior during which the sleepwalker is usually unaware of his surroundings and almost entirely unresponsive to external stimuli. Sleepwalkers may leave their bed, walk around their bedrooms and perform semi-purposeful acts. They may also carry out complex motor automatisms, such as driving a car. There is always complete amnesia for the episode at awakening.

Sleepwalking is more common in childhood, with a prevalence ranging from 5 to 30% for occasional or frequent episodes of somnambulism [13,14]. The disorder, however, can also appear for the first time in adulthood [10,15]. The prevalence of sleepwalking oscillates between 2 and 5% in the adult general population according to some authors [14,16–18]. As for confusional arousals, prevalence significantly decreases with age from 4.9% in the 15–24-year age group to 0.5% in the ≥65-year age group [18]. A strong familial occurrence has often been reported in sleepwalking; for example, in a prospective study, Abe et al. reported a sleepwalking occurrence of 14% in children aged between 8 and 10 years who had one of their parents with a sleepwalking history and 2% of sleepwalking in children with non-sleepwalking parents [19].

It is not uncommon for sleepwalkers to inflict injury to themselves or others during an episode. Rauch and Stern described two cases of self-inflicted injury [20]. One involved a 34-year-old man who fell 20 feet into some bushes, lacerating his head and fracturing his tenth thoracic vertebra. The other involved a 20-year-old man who smashed through a glass door, resulting in a severe laceration of his right wrist. Several cases of murder while sleepwalking have also been documented. Studying 100 patients who claimed amnesia for their crime at the time of hospitalization, Hopwood and Shell identified one case of somnambulistic homicide: a fireman who battered his wife to death with a shovel during a sleepwalking episode [21]. More recently, Broughton et al. described the extraordinary case of a man who, during a sleepwalking episode, drove his car several kilometers to the home of his wife's parents before killing them [22].

Sleep terrors

Sleep terrors, too, are parasomnias that occur in NREM sleep stages 3 and 4. Typically, these episodes begin with an abrupt awakening from sleep accompanied by a panicky scream. The individual is generally inconsolable during an episode. At awakening, there is complete or partial amnesia of the frightening dream.

Like sleepwalking, sleep terrors occur more commonly in childhood, but less frequently than sleepwalking. Between 1 and 6.5% of children are afflicted by sleep terrors [23,24], and as many prepubertal children have recurrent episodes [25]. Occurrence of a few episodes of sleep terrors in childhood is estimated to occur in 20–30% [26]. In the adult general population, the prevalence of sleep terrors has been estimated to be about 1% for weekly episodes [27] and around 2% for less frequent episodes [18,27].

One of the earliest reports of murder during a sleep terror episode is the case of Esther Griggs, who in 1858 threw her baby out of a window. She was apparently dreaming her house was on fire at the time and was actually attempting to save her baby. We can find in the literature several cases of sleepwalking/sleep terror episodes that resulted in murder or serious injury [28–30]. The individual typically has no or only fragmentary memory of the dream being acted out. From the accounts of those who remember their dreams in part, certain recurring themes emerge. These include defending oneself against attacks from others or beasts, trying to escape a danger, or trying to protect a loved one against potential danger.

Sleepwalking and sleep terrors can be triggered by stress, sleep deprivation, alcohol ingestion, and
almost all sedative medications (hypnotics, tranquilizers, antihistamines, stimulants).

Parasomnias associated with REM sleep

This group of parasomnias is composed of three disorders occurring essentially during REM sleep. These disorders are nightmare disorder, recurrent isolated sleep paralysis and REM sleep behavior disorder (RBD).

Nightmare disorder

Nightmares are frightening dreams occurring generally during REM sleep and often resulting in awakening the dreamer. This disorder is frequent in children. It affects boys and girls equally. Nightmares occur always or often in 2–11% of children and now and then in 15–31% [31]. About a third of adults with recurrent nightmares have onset of the symptom during childhood [16]. Nightmares have been reported to occur at least once a week in 5% of the adult population [32] and always or often in 1–5% [26]. Women are 2–4 times more frequently affected than men. There are persistent genetic effects on the disposition to nightmares in about 45% of childhood and 37% of adult nightmares [33].

Recurrent isolated sleep paralysis

Sleep paralysis is one of the main symptoms associated with narcolepsy, but it can also occur individually (i.e. isolated sleep paralysis). Téllez-López et al. [34] found that 11.3% of their general population sample had sleep paralysis episodes at least sometimes. Where more narrowly defined populations are concerned, Goode [35] and Everett [36] observed rates of 4.7% and 15.4%, respectively, for self-reported sleep paralysis in medical students, and Bell et al. [37] noted a prevalence of 41% in Black Americans. In a study of adults living on the northeast coast of Newfoundland, Ness [38] reported a rate of 62% for “old hag” attacks, as sleep paralysis is popularly known in that part of Canada. An epidemiological study performed with 8085 subjects between 15 and 99 years of age found that 6.2% had at least one episode of sleep paralysis in their lifetime; 0.8% experienced severe sleep paralysis (at least one episode per week); and 1.4% moderate sleep paralysis (at least one episode per month) [39].

REM sleep behavior disorders

REM sleep behavior disorders (RBD) were first described in the late 1970s by Japanese researchers and labeled as such by Schenck et al. [43]. This sleep disorder is characterized by a loss of generalized skeletal muscle REM-related atonia and the presence of physical dream enactment. The individual has no consciousness of acting out a dream, but is generally able to recount the dream upon awakening. This syndrome often results in behavior dangerous to oneself or others [40–42]. Their initial sample [40] included four men and one woman, all aged 60 years or over. Four of them had neurological disorders. Most of these cases had excessive slow-wave sleep for their age. This is not, however, a sine qua non condition. Indeed, Tachibana et al. [43] reported seven cases of RBD without neurological or psychiatric disorder that had a normal quantity of slow-wave sleep. In all studies, this disorder is observed almost exclusively in men.

According to Schenck and Mahowald [44], prodromal symptoms of RBD appeared 10–40 years before the full manifestation of the disorder in 25% of the studied cases. This prodrome is characterized by sleep talking, yelling, or limb-jerking during sleep.

The mechanism underlying this disorder is not yet fully understood. Polysomnographic recordings of individuals with RBD showed a reduction of the tonic phenomena of REM sleep and the activation of the phasic phenomena. Destruction of the brainstem regions responsible for the REM sleep atonia has been hypothesized as responsible of such phenomena.

The prevalence of RBD in the general population is little-documented. Ohayon et al. [45] estimated it at 0.5% in the general population using minimal criteria proposed by the International Classification of Sleep Disorder (ICSD). However, as many as 2% of the general population reported experiencing violent behaviors during sleep, with a male predominance [45].

Other parasomnias

This group of parasomnias comprises sleep-related dissociative disorders, sleep enuresis, catathrenia, exploding head syndrome, sleep-related hallucinations, sleep-related eating disorder, unspecified parasomnia, parasomnia due to a drug or substance and parasomnia due to a medical condition.

Epidemiological data on this group of parasomnias are scarce. Prevalence in the general population is unknown for most of the parasomnias. Catathrenia,
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or sleep-related groaning, is a newly described parasomnia occurring in both REM and NREM sleep [46].

Sleep enureis

Sleep enuresis, also called nocturnal bedwetting, is characterized by recurrent involuntary voiding during the night. It is considered primary when the individual never experienced a dry period for at least six consecutive months.

Sleep enuresis is relatively common in childhood (≥5 years old) and progressively decreases with age. At 4–5 years of age, the prevalence of sleep enuresis is estimated to be between 20 and 30%. At 7 years, it decreases to about 10%, and further decreases to around 3% in 12-year-olds. The prevalence during adolescence and adulthood is around 2% [31,47,48]. Sleep enuresis in prepubertal children is twice as frequent among boys than girls. In adulthood, the prevalence is comparable in both sexes. However, in the elderly, this problem affects women more frequently than men.

Sleep enuresis has a strong hereditary component. When both parents have a history of sleep enuresis in childhood, 77% of the children suffer from enuresis. When only one of the parents has a positive history, nearly half the children have sleep enuresis [1].

Sleep-related hallucinations

Sleep-related hallucinations refer to hallucinations occurring at sleep onset (hypnagogic hallucinations) or upon awakening (hypnopompic hallucinations). Hypnagogic hallucinations are common in the general population with a prevalence of 25–37% [49,50]. The prevalence of hypnopompic hallucinations is around 7–13% [49,50]. Both hypnagogic and hypnopompic hallucinations significantly decrease with age, being twice less frequent in elderly subjects (≥65 years). These hallucinations are also more frequent among women than men. About 30% of those experiencing sleep-related hallucinations are frightened by them [50].

Conclusions

Our knowledge about the prevalence and incidence of parasomnias is still limited compared to dyssomnias. Most of the available data are based on the main symptoms of parasomnias, not the diagnoses.

Information about the associated factors and triggers of an episode is also limited. As shown, parasomnias are frequent in the general population; more than 30% of individuals experience at least one type of parasomnia. The consequences on daytime functioning and the emotional distress experienced following a parasomnia episode are undocumented at the epidemiological level. Therefore, it is difficult to ascertain what proportion of these individuals are in need of treatment.

At the genetic level, there is growing evidence that many parasomnias have a genetic component. Therefore, epidemiological genetic links need to be studied. Ethnicity also could play a role in some parasomnias. However, with the exception of sleep paralysis, the role of race has not been investigated.

References

Chapter 2: Epidemiology


