Chapter 54

Isolated motor phenomena and symptoms of sleep

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INTRODUCTION

Sleep identifies a natural and healthy, temporary, and periodical state of rest, with suspension of the sensorial functions of the organs of sense, as well as those of the voluntary and rational soul. Several motor phenomena, however, occur during sleep, both physiological and pathological.

The International Classification of Sleep Disorders, second edition (ICSD-2) (American Academy of Sleep Medicine, 2005), lists sleep disorders within eight categories:

   I.   Insomnias
  II.   Sleep-related breathing disorders
 III.   Hypersomnias of central origin not due to a circadian rhythm sleep disorder, sleep-related breathing disorder, or other cause of disturbed nocturnal sleep
  IV.   Circadian rhythm sleep disorders
   V.   Parasomnias
  VI.   Sleep-related movement disorders
 VII.   Isolated symptoms, apparently normal variants, and unresolved issues
VIII.   Other sleep disorders.

Some periodic and aperiodic phenomena of sleep are included within different categories of the ICSD-2, without a unifying classification scheme. Here, these will be described with their salient characteristics and distinguishing features, ranging from simple movements or symptoms of sleep up to more complex behaviors classified within the parasomnias.

PHYSIOLOGICAL AND EXCESSIVE FRAGMENTARY HYPNIC MYOCLONUS

Myoclonus is part of normal sleep physiology representing a state-related paradoxical motor excitation.

Physiological fragmentary (or partial) hypnic myoclonus (PFHM)

PFHMs was first described by De Lisi in 1932 as sudden, arrhythmic, asynchronous, and asymmetrical brief twitches involving various body areas, in particular distal limb and facial muscles, occurring during sleep. Such electromyographic (EMG) activity in humans shows an inverse relationship with the degree of sleep EEG synchronization (Dagnino et al., 1969), prevailing equally during relaxed wakefulness (nonrapid-eye-movement (NREM) sleep stage 1) and during rapid-eye-movement (REM) sleep (Montagna et al., 1988). The muscle discharges of PFHM appear as isolated or bursts of motor unit action potentials with or without visible movement (Figure 54.1). They must originate in the muscle endplate (Buchthal and Rosenfalck, 1966) but are modulated by brainstem regions implicated in motor control during sleep. The motor inhibition typical of REM sleep is related to state-dependent activity of the pontine nucleus pontis oralis which, by exciting cells of the medullary nucleus reticularis gigantocellularis, inhibits the alpha motoneurons (Moruzzi, 1972). A physiological escape from these state-related motor inhibition hypothetically accounts for the PFHM, which is probably caused by descending volleys within the reticulospinal system impinging upon the spinal alpha motoneurons during REM sleep. Indeed, destruction of the reticulospinal tract in animals abolished the myoclonic activity that remained unchanged by lesioning the dorsal roots, red nucleus, or pyramidal tracts (Gassel et al., 1964).

In humans, PFHM represents a "simple" motor phenomenon during sleep, present mostly during stage I of NREM sleep and during REM sleep, in apparent contrast with experimental animals which show the highest peak of hypnic myoclonic activity during REM sleep

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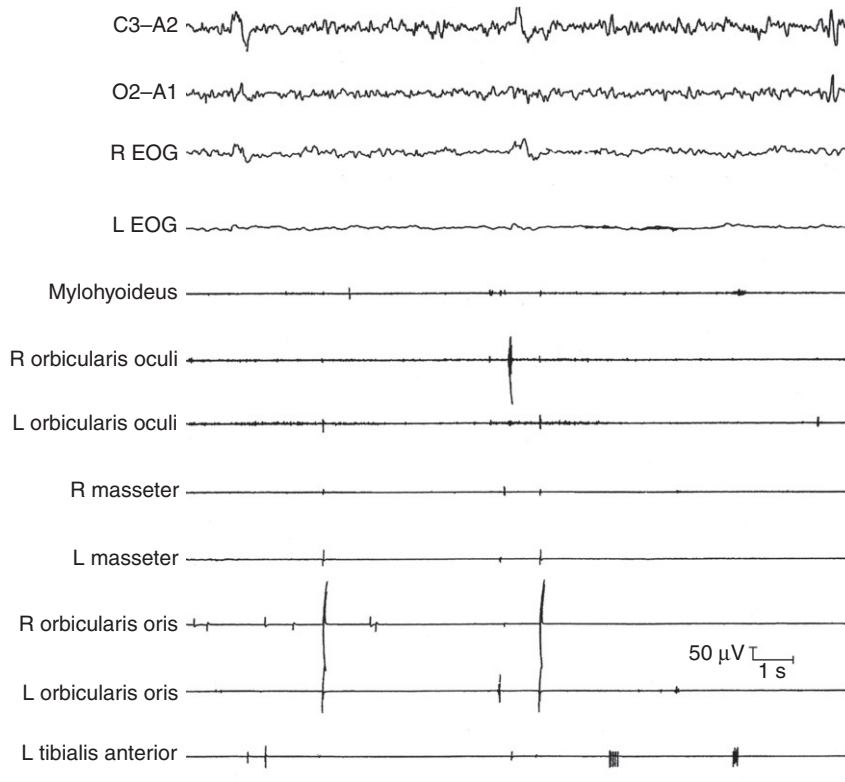
Fig. 54.1. Physiological fragmentary hypnic myoclonus during stage 1 NREM sleep involving face and limb muscles. EOG, electro-oculogram; R, right; L, left.

(Gassel et al., 1964). No PFHM was recorded from patients with complete peripheral nerve lesions or with spinal paraplegia. PFHM, absent on the affected side of patients with chronic ischemic hemiparesis and increased in patients with Parkinson's disease and Wilson's disease, was normally present in patients with tabe dorsalis in whom muscle proprioception input was probably abolished (Dagnino et al., 1969). A supraspinal origin of PFHM is therefore implicated by its absence in muscles that are completely paralyzed because of a peripheral nerve, spinal, or pyramidal lesion, its increase in extrapyramidal diseases, and by the fact that it remains unaffected by lesions of the dorsal roots that abolish muscle spindle afferent activity.

Excessive fragmentary hypnic myoclonus (EFHM)

EFHM has been reported as a pathological enhancement of PFHM in which small myoclonic twitches and fasciculations are present throughout sleep and associated with sleep apnea, excessive daytime drowsiness, and insomnia (Broughton and Tolentino, 1984; Broughton et al., 1985; Lins et al., 1993). EFHM may cause small movements of the fingers, toes, and/or corners of the mouth, without gross displacements

across a joint space. Similar motor activity during sleep has been reported in patients with restless legs syndrome (Coccagna et al., 1966) (Figure 54.2), in extrapyramidal syndromes (Tassinari et al., 1965), and in patients with REM sleep behavior disorder (Mahowald and Schenck, 2000), obstructive and central sleep apnea, narcolepsy, periodic limb movements during sleep and fatigue (Broughton et al., 1985). EFHM may also present as an isolated motor phenomenon during relaxed wakefulness, NREM, including stages III and IV, and REM sleep in which "quiver" movements recur throughout the body, affecting primarily the hands and face with some degree of sleep fragmentation. The twitches may occasionally awake the patient. They are absent during wakefulness and EEG–EMG back-averaging does not show any cortical potentials related to the twitches (Vetrugno et al., 2002).

These data suggest a selective impairment of motor control during sleep. However, the exact origin and significance of EFHM remain unclear and despite the myoclonus being a common finding in polysomnography, it is often asymptomatic. EFHM is now classified in the VII section of the ICSD-2 (Isolated symptoms, apparently normal variants, and unresolved issues) that lists sleep-related symptoms that are in the borderline between normal and abnormal sleep.
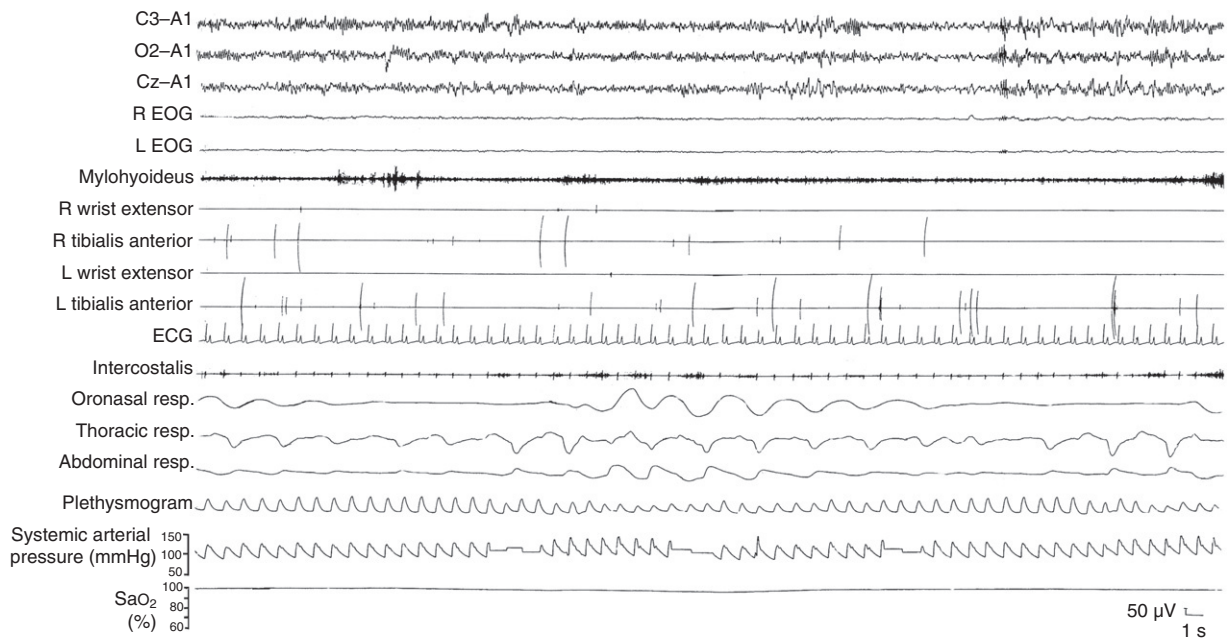
Fig. 54.2. Excessive fragmentary hypnic myoclonus during stage 2 NREM sleep in a patient with restless legs syndrome. EOG, electro-oculogram; ECG, electrocardiogram; resp., respirogram; Sao_2, oxygen saturation; R, right; L, left.

HYPNIC JERKS (SLEEP STARTS)

Hypnic jerks, or sleep starts, are normal physiological events occurring at the transition from wakefulness to sleep, often associated with sensory phenomena, such as a feeling of falling, unexplained alarm or fear, inner electric shock or light flash (Oswald, 1959; Gastaut and Broughton, 1965). An outcry can occasionally accompany the jerks, which are usually associated with autonomic activation (tachycardia, irregular breathing, and sudomotor activation). Jerks consist of nonstereotyped, abrupt, and brief flexion or extension movements, generalized, or, more frequently, segmental and asymmetrical with neck and/or limb muscles involvement. Usually EMG complexes last less than 250 milliseconds and are associated with K-complexes and vertex sharp waves on the EEG (Figure 54.3).

Hypnic jerks occasionally occur during light sleep, causing a brief arousal. Fatigue, stress, and sleep deprivation may facilitate the occurrence of the hypnic jerks, which may be misdiagnosed as myoclonic seizures. Sleep starts may occur without any motor activity with only visual, auditory, or somesthetic sensory phenomena. *Purely sensory sleep starts* without a body jerk have been described occurring exclusively at onset of sleep (Sander et al., 1998). The *exploding head syndrome* may also represent a variety of purely sensory sleep starts, even though it is classified amongst the "Other parasomnias" in ICSD-2. It was first described by Armstrong-Jones (1920) as a "snapping of the brain"

and is characterized by the sensation lasting some seconds that an explosive noise has occurred in the head, which wakens the individual from sleep. Patients describe this sensation variously as a terrifying "loud bang" or a "shotgun- or bomb-like explosion" (Pearce, 1989), the most common age of onset being over 50 years. The attacks may have variable frequency, up to more than one per night, for a few weeks or months, with possible prolonged or total remissions. Polysomnographic studies have documented the occurrence of these attacks during all sleep stages, including REM sleep (Sachs and Svanborg, 1991), and during the passage from wakefulness to sleep (Pearce, 1989).

Another related phenomenon at sleep onset, probably representing a variant of sensory sleep starts, is the *"blip" syndrome* in which patients experience momentary sensations of impending loss of consciousness particularly when relaxed, without any obvious cardiac, cerebral vascular, or epileptic basis (Lance, 1996).

It has been suggested that sleep starts with combined sensory, motor, and autonomic components represent dissociated elements of sleep during which "twilight" phenomena arise. A possible basis for these motor–autonomic–sensory phenomena is thought to be a delay in the reduction of activity in selected areas of the brainstem reticular formation as the patient passes from wakefulness to sleep (Mahowald et al., 1998). Hypnic jerks represent a normal accompaniment of sleep.
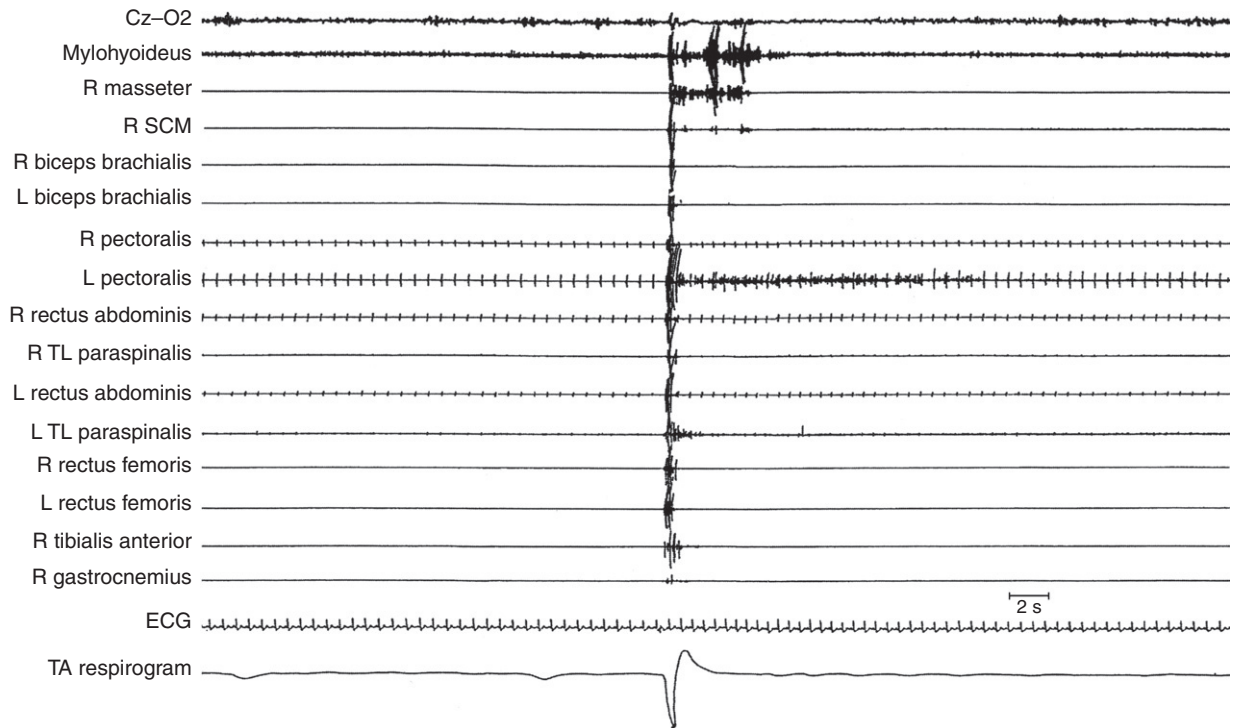
Fig. 54.3. Hypnic jerk in a normal subject at the transition from wakefulness to sleep, with abrupt neck, trunk and limb muscles involvement. SCM, sternocleidomastoideus; TL, thoracolumbar; ECG, electrocardiogram; TA, thoracoabdominal; R, right; L, left.

Intensified hypnic jerks, however, may cause sleep fragmentation and insomnia, a condition recognized since 1890 by Weir Mitchell (Mitchell, 1890; Broughton, 1988).

PROPRIOSPINAL MYOCLONUS AT THE WAKE–SLEEP TRANSITION

Propriospinal myoclonus (PSM) describes jerks that involve muscles innervated by many different segments of the spinal cord, myoclonic activity spreading up and down the cord via supposed propriospinal pathways from a more restricted source (Brown et al., 1991). PSM has been reported associated with infective myelitis (de la Sayette et al., 1996), cervical trauma (Fouillet et al., 1995), pharmacological treatments (ciprofloxacin, cannabis, interferon-α) (Benatru et al., 2003; Lozsadi et al., 2004; Post et al., 2004), syringomyelia (Nogues, 2002), and multiple sclerosis (Kapoor et al., 1992), but in many cases it remains idiopathic. PSM may progress into a severe and potentially fatal "myoclonic status" (Manconi et al., 2005).

In some patients PSM displays a striking relationship with vigilance level, as, independently of posture, it recurs in a semirhythmic fashion only during relaxation and drowsiness preceding sleep onset. This has been termed PSM at the wake–sleep transition (Montagna et al., 1997; Vetrugno et al., 2001b). Patients complain of sudden involuntary axial jerks occurring every night and impeding falling asleep. The spontaneous jerks remain restricted to the prehypnic wake period when the EEG alpha activity spreads to involve the anterior brain regions, but disappear as soon as spindles and K-complexes begin on the EEG. Sometimes PSM reappears during intrasleep wakefulness and upon awakening in the morning. Mental and sensory stimulations (simple arithmetic exercises or making a fist) during relaxed wakefulness stop the jerks concomitantly with the disappearance of the EEG alpha activity and independently of any postural changes. Findings on neurological examination, neurophysiological investigation (EEG back-averaging, somatosensory evoked potentials, transcranial magnetic stimulation motor-evoked potentials), and spinal and cranial magnetic resonance imaging (MRI) are usually normal. The myoclonic jerks occur as flexion or extension movements with prominent axial muscles involvement, may be single or repetitive, with or without agonist–antagonist relationship, and with EMG discharges lasting 100–300 milliseconds (Figure 54.4). In some of the jerks, myoclonic activity may be confined to the abdominal muscles, as in spinal segmental myoclonus, but usually the jerks spread at about 7–8 m/s up and down the cord to involve muscles in the legs and
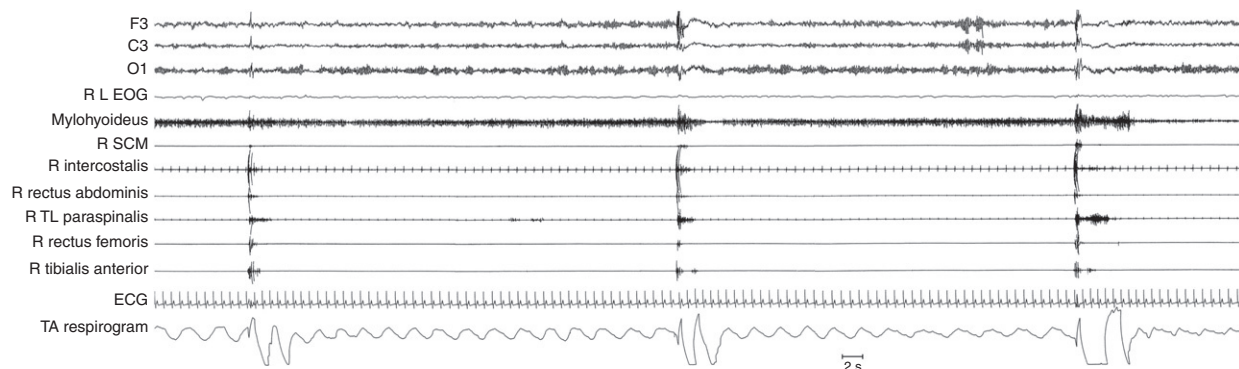
Fig. 54.4. Propriospinal myoclonus at the wake–sleep transition: quasi-periodic recurrence of axial jerks during wake–sleep transition occurring at a time when alpha activity is evident on anterior EEG leads. EOG, electro-oculogram; SCM, sternocleidomastoideus; TL, thoracolumbar; ECG, electrocardiogram; TA, thoracoabdominal; R, right; L, left.
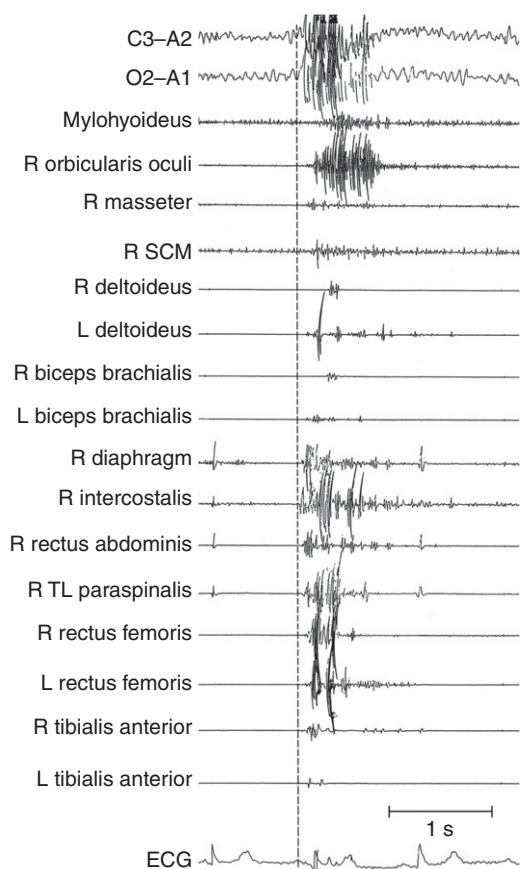

Fig. 54.5. EEG–EMG recordings of propriospinal myoclonus at sleep onset showing a rostrocaudal propagation of muscular activity starting in the right intercostalis muscle (dotted line). SCM, sternocleidomastoideus; TL, thoracolumbar; ECG, electrocardiogram; R, right; L, left.

neck (Vetrugno et al., 2000) (Figure 54.5). PSM may be spontaneous or triggered by stimuli. As a consequence of the slow conduction velocity of spinospinal pathways along which EMG activity supposedly spreads,

significant jitter occurs from jerk to jerk and among single muscles involved in the same jerk. There is no associated EEG abnormality routinely searched or by back-averaging.

PSM at sleep onset has also been found in patients with a long history of restless legs syndrome (Vetrugno et al., 2005). In these cases, PSM jerks arise during relaxed wakefulness but give way with the appearance of spindles and K-complexes on the EEG to typical periodic limb movements during sleep with characteristic EMG activity limited to leg muscles.

The occurrence of PSM only during relaxed wakefulness indicates that the predormitum and postdormitum periods represent peculiar states with intrinsic mental and neurophysiological characteristics (Critchley, 1955; Braun et al., 1997; Montagna and Lugaresi, 1998). Reduced sleep-related spinal inhibition during the wake to sleep transition may result in activation of several motor generators, including those associated with PSM. These state-dependent modulatory influences on motor control set PSM into motion, probably by releasing a spinal generator responsible for PSM.

Benzodiazepines – clonazepam (0.5–2 mg/day at bedtime) in particular – may reduce their intensity and frequency, but usually do not abolish the jerks of PSM at sleep onset.

BENIGN SLEEP MYOCLONUS OF INFANCY

Described for the first time in 1982 by Coulter and Allen, benign sleep myoclonus of infancy is a movement disorder with a self-limited course characterized by myoclonic jerks occurring only during sleep and presenting in the first month of life (Coulter and Allen, 1982; Di Capua et al., 1993; Vaccario et al., 2003). Movements are absent during wake and stop when the child is woken. A spontaneous resolution

occurs around the third or fourth month of life, always before the first year of age (Di Capua et al., 1993; Noone et al., 1995; Caraballo et al., 1998). Affected children show no increased risk of developing subsequent epileptic seizures (Caraballo et al., 1998; Vaccario et al., 2003) and are otherwise healthy newborns with normal psychomotor development and no neurological deficits.

Some 30% of the jerks of benign myoclonus of infancy involve the whole body, 20% the abdominal or proximal muscles, and 50% the arms or the legs. They often involved the whole limb, but on occasion prevail in the distal segments (Resnick et al., 1986). Ankle dorsiflexion myoclonus was described in the first two infants reported. The jerks frequently occur in clusters repeating at one per second (Resnick et al., 1986), last 40–300 ms, being briefer when occurring in clusters, and persist from several seconds to 90 minutes. Only one reported case exhibited prolonged jerks lasting for 12 hours, mimicking status epilepticus (Turanli et al., 2004).

The movements are present during all sleep states, although with higher frequency during NREM sleep (Resnick et al., 1986), but they do not awake the infant. No EEG correlates are noted and the EEG is normal during and after the episodes (Resnick et al., 1986). Benign neonatal myoclonus may be triggered by noise (Daoust-Roy and Seshia, 1992) and especially by rocking (Alfonso et al., 1995). The major differential diagnosis of benign sleep myoclonus of infancy is, of course, with epilepsy and particularly with two myoclonus epilepsy syndromes that can occur in the neonatal period: infantile spasms and benign infantile myoclonus (Lombroso and Fejerman, 1977; Kellaway et al., 1983; Donat and Wright, 1992). Benign sleep myoclonus of infancy can be distinguished from these conditions by history or by the observation of repetitive myoclonic limb jerks that occur only during sleep and stop upon awakening, by a normal EEG and neurological examination, and by a sustained normality on follow-up.

The mechanism of benign sleep myoclonus of infancy is unknown. It has been hypothesized to result from a benign disturbance of the brainstem control of sleep (Coulter and Allen, 1982) or from transient immaturity or imbalance of the serotonergic system (Resnick et al., 1986). Genetic factors might play a role, because this disorder has been described in siblings (Dooley, 1984; Tardieu et al., 1986; Vaccario et al., 2003).

NOCTURNAL LEG CRAMPS

Nocturnal leg cramps are sudden, involuntary contractions of the calf muscles that occur during the night or while at rest. Occasionally, muscles in the soles of the feet also become cramped. The cramps can last for a few seconds or up to 10 minutes, but the soreness may linger. The cramps can affect persons in any age group, but they tend to occur in middle-aged and older individuals, and cause recurrent awakenings from sleep associated with painful leg sensations; the discomfort is relieved by local massage, movement, or heat. Studies show that about 25–50% of adults, especially subjects aged 80 years or more, experience nocturnal leg cramps (Naylor and Young, 1994; Abdulla et al., 1999).

Polysomnographic recordings show increased EMG activity in the affected leg and associated awakening (Saskin et al., 1988). EMG studies suggest that cramps result from spontaneous firing of groups of anterior horn cells followed by contraction of several motor units at rates of up to 300 Hz (Miller and Layzer, 2005), but a distal origin in the intramuscular motor nerve terminals is also possible (Jansen et al., 1990).

Muscle cramps are a common consequence of vigorous exercise and are more frequent in patients with peripheral vascular disease (Abdulla et al., 1999). Nevertheless, in many cases nocturnal leg cramps occur independently of arterial circulation abnormalities as an idiopathic condition (Jansen et al., 1990). Medications (diuretics, nifedipine, β-agonists, steroids, lithium), some medical conditions (uremia, diabetes, thyroid disease, hypomagnesemia, hypocalcemia, hypokalemia, continuous motor unit activity syndromes such as neuromyotonia), and pregnancy (Hertz et al., 1992) are associated with nocturnal leg cramps. Nocturnal cramps in some cases may be a familial condition with an autosomal dominant pattern of inheritance (Lazzaro et al., 1981; Jacobsen et al., 1986; Ricker and Moxley, 1990).

Nocturnal leg cramps should not be confused with restless legs syndrome, which is characterized by an urge to move during repose and in the evening with or without a crawling sensation that is relieved by walking or moving around. Although uncomfortable, restless legs syndrome usually does not involve cramping. Conditions that mimic cramps include simple muscle strain, dystonias, ischemic or neuropathic claudication, nerve root disease, and PLMS. Muscle cramps are a feature of many myopathic and neuropathic conditions in which they are not usually restricted to the nighttime or necessarily to the legs. Although various medications, including vitamins of the B group and vitamin E, calcium blockers, potassium, magnesium citrate, gabapentin, and botulinum toxin may be effective, the most successful treatment is quinine sulfate (Man-Son-Hing et al., 1998; Butler et al., 2002; Miller and Layzer, 2005). Quinine, however, carries a high risk of hepatic and blood toxicity (Jung, 2004). Some patients also respond to local massage and leg movement.

RHYTHMIC MOVEMENT DISORDERS

Rhythmic movement disorder (RMD) is a group of stereotyped, repetitive movements involving large axial muscles, usually of the head, neck, and trunk, and sometimes also the legs, which typically occur immediately before sleep onset and are sustained into light sleep. Also known as jactatio capitis or corporis nocturna, headbanging, headrolling, bodyrocking, bodyrolling, *rythmie du sommeil*, the term RMD is preferred as different body areas may be involved in the movement activity. Rhythmic body movements may occur in any stage of sleep, including REM sleep, but most often during drowsiness persisting into light sleep (American Academy of Sleep Medicine, 2005). In the more common form of RMD, at least one of the following types of movement is present: the head is forcibly moved in a back and forward direction (headbanging type); the head is moved laterally while in a supine (lying on the back, face up) position (headrolling type); the whole body is rocked while on the hands and knees (bodyrocking type); the whole body is moved laterally while in a supine position (bodyrolling type). The stereotypic, repetitive movements occur with a frequency of 0.5–2.0 Hz and persist for a few minutes to many hours during sleep.

RMD is common in children, sometimes as a semivoluntary behavior utilized as a sleep-inducing aid, usually resolving by about 4 years of age (Klackenberg, 1971), but it may rarely persist into adulthood (Happe et al., 2000). RMD isolated to REM sleep has occasionally been reported in children (Walsh et al., 1981; Gagnon and De Koninck, 1985; Stepanova et al., 2005) and unusually persists into adult life (Anderson et al., 2006). It is also possible that RMD may appear *de novo* during adulthood. RMD may be associated with other disorders such as anxiety, attention deficit disorders, and autism, and occasionally skull injuries can occur in patients with associated intellectual impairment or autistic syndromes.

The treatment of RMD usually involves reassurance, but protective headgear may sometimes be necessary, particularly in children with learning disabilities. Benzodiazepines, levodopa or dopamine agonists, and tricyclic antidepressants have been tried with variable success.

The association of RMD with longlasting restless legs syndrome, although reported by Morgan in 1967 and emphasized by Walters in 1988, is still underrecognized. RMD may also occur in restless legs syndrome of recent onset (Lombardi et al., 2003).

Rhythmic feet movement, also called *hypnagogic foot tremor*, occurring during presleep wakefulness and light sleep may be considered a new kind of RMD arising in adults, in some cases associated with insomnia (Broughton, 1988), sleep apnea, PLMS, and restless legs syndrome (Wichniak et al., 2001). Hypnagogic foot tremor presents as rhythmic, oscillating movements of the whole foot or toes, occurring usually bilaterally but asynchronously in both legs at varying frequencies, between 0.5 and 3 Hz, with a mean duration of the EMG bursts of 500 ms and without a substantial sleep-disturbing effect (Figure 54.6).

Brief activations of the tibialis anterior in one leg alternating with similar activation in the other leg, so-called *alternating leg muscle activation* (ALMA), have also been described. Such activations, similar to rhythmic feet movements while falling asleep, occur at a frequency of 1–1.5 Hz, each lasting up to 0.5 s, with sequences of several to 20 seconds and recurring in all sleep stages but particularly during arousals (Figure 54.7). ALMA has been described in patients with sleep apnea, PLMS, those taking antidepressant medication (Chervin et al., 2003), and with restless legs syndrome (Vetrugno et al., 2005).

SLEEPTALKING

Sleeptalking (somniloquy) is one of the most common parasomnias and consists of verbal vocalization during sleep ranging from mumbled nonsense to coherent sentences, but without awareness of the event. The utterances may be brief, infrequent, and devoid of any emotional stress, or they may include long speeches. Somniloquy can sometimes be evoked by conversation with a predisposed sleeping individual. Multilingual or dominant bilinguals patients always use the dominant language during somniloquy episodes (Gastaut and Broughton, 1965; Pareja et al., 1999), and balanced bilinguals (those who have equal proficiency in both languages) may sleeptalk in either of the two languages (Pareja et al., 1999). Somniloquy occurs during NREM as well as REM periods (Rechtschaffen et al., 1962); in general, 20–25% of speeches are associated with REM sleep and 75–80% with NREM sleep (Arkin et al., 1970), but some individuals are more productive in REM sleep. Somniloquy occurs spontaneously, but in some instances it may be precipitated by factors such as febrile illness or emotional stress.

Somniloquy is common in both children and adults (Horne, 1992). About half of the children aged 3–13 years present somniloquy at least once a year, but less than 10% every night (Reimão and Lefévre, 1980; Simonds and Parraga, 1982; Laberge et al., 2000). Adair and Bauchner (1993) reported that somniloquy was more prevalent in children aged 4–5 years, but other studies have found no effect of age on the prevalence of somniloquy (Reimão and Lefévre, 1980; Simonds and Parraga, 1982; Laberge et al., 2000). Somniloquy is
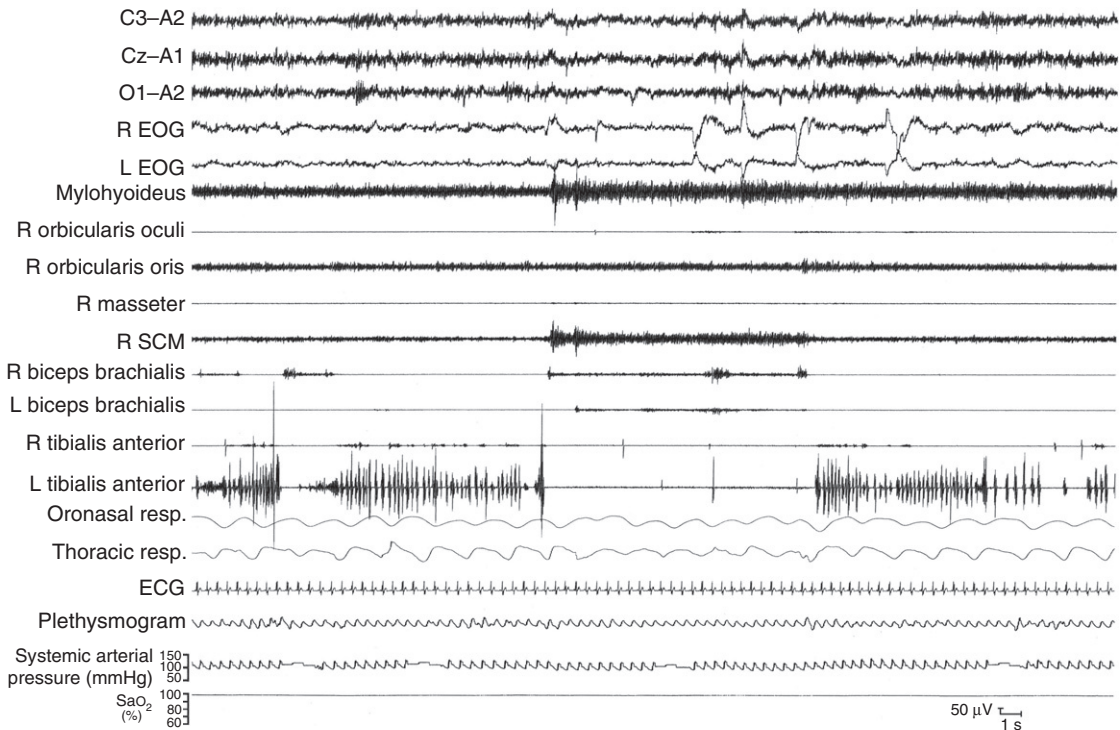
Fig. 54.6. Hypnagogic foot tremor occurring during presleep wakefulness on the left tibialis anterior muscle. EOG, electro-oculogram; SCM, sternocleidomastoideus; resp., respirogram; ECG, electrocardiogram; Sao_2: oxygen saturation; R, right; L, left.
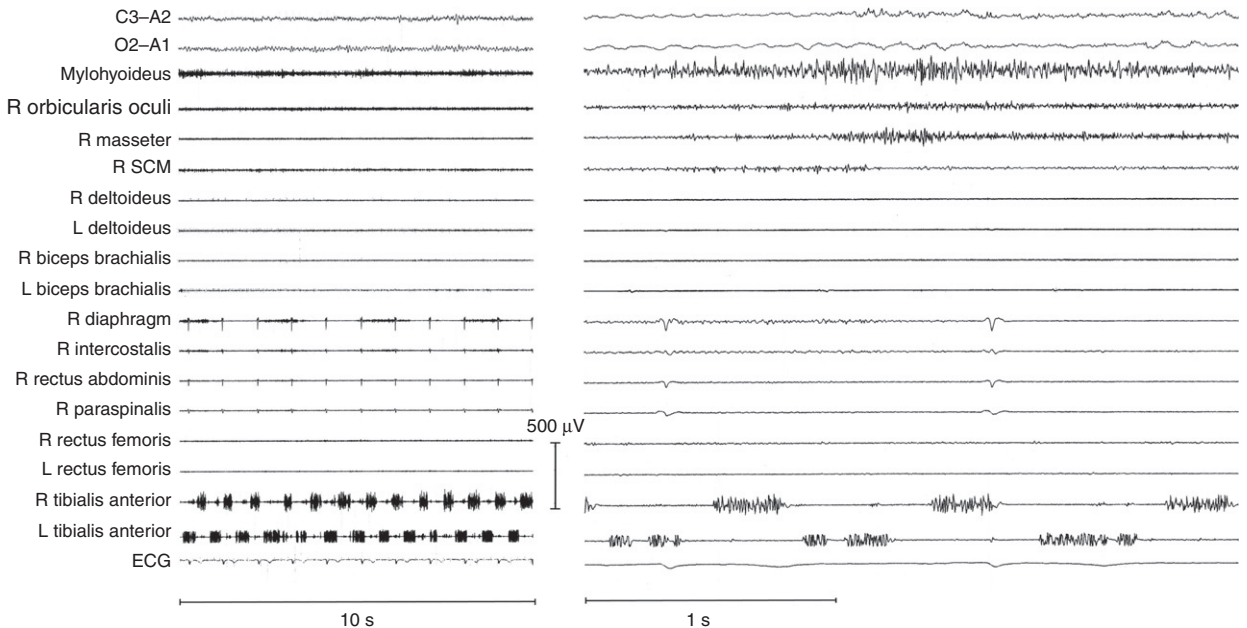

Fig. 54.7. Sequence of alternating leg muscle activation occurring on the right and left tibialis anterior muscles at a frequency of 1.5 Hz, each lasting about 0.5 s. SCM, sternocleidomastoideus; TL, thoracolumbar; ECG, electrocardiogram; R, right; L, left.

indeed reported by 24% of normal adults (Ohayon et al., 1997). There is no apparent sex difference, even though in one study somniloquy was more common in boys (Laberge et al., 2000). Preadolescents and adolescents sleeping badly present a higher incidence of somniloquy than the subjects who sleep well (Kahn et al., 1989; Manni et al., 1997). Children with sleep-disordered breathing and children with chronic headache are more likely to sleeptalk (Smeyers, 1999; Goodwin et al., 2004).

Somniloquy is usually an isolated phenomenon occurring in otherwise healthy subjects (Arkin, 1966), but it may be one of the clinical features accompanying obstructive sleep apnea syndrome (OSAS), sleepwalking (somnambulism), and sleep terrors (*pavor nocturnus*) (Gastaut and Broughton, 1965; Broughton, 1968), and REM sleep behavior disorder (RBD) (Schenck et al., 1987). There is a strong association between sleepwalking, night terrors, and somniloquy (Kales et al., 1980; Simonds and Parraga, 1982; Abe et al., 1984; Laberge et al, 2000), which suggests a common genetic background. Children are indeed likely to sleeptalk if one or both parents have a parasomnia such as sleepwalking (Abe et al., 1984). Somniloquy may be a prodrome of RBD (Pareja et al., 1996), but it usually represents a benign condition that resolves spontaneously.

CATATHRENIA (NOCTURNAL GROANING)

Catathrenia, from the Greek κατά θρήνος (like a groan), is a rare idiopathic condition characterized by high-pitched, monotonous, irregular groans that occur during prolonged expiration in sleep (Vetrugno et al., 2001a). The hallmark of catathrenia is that deep inspiration is followed by protracted expiration during which a sound is produced, repetitively recurring during NREM and especially REM sleep. Patients are unaware, but typically parents and bed partners are troubled and alarmed by the nocturnal noise that occurs very often, if not every night. The patients do not have respiratory distress or anguished expression during the groaning, or any abnormal motor behavior or recall of vivid dream. Coaxed to change posture during sleep, they may transiently stop to groan, only to start again later on the same night.

Physical and neurological examinations, laboratory and otolaryngological investigation including static and dynamic vocal cord endoscopic evaluation and neck and cranial computed tomography and MRI are generally normal, and no mood disturbance has been identified. On polysomnographic examination, the groaning usually starts 2–6 hours after falling asleep, and occurs during NREM and REM sleep, lasting 2–20 seconds and repeating in clusters for 2 minutes to 1 hour, many times per night, with the patient lying in any position in bed. A slight decrease in heart rate and arterial blood pressure with moderately positive intraesophageal pressure but no EMG activation of the diaphragm and intercostal muscles occurs during each expiratory groaning, which ends usually with a snort followed by a rebound in heart rate and arterial blood pressure, and without significant oxygen saturation variability, SaO_2 remaining normal at 95–98% (Vetrugno et al., 2001a) (Figure 54.8). During the groaning, the patients remain still and an EEG arousal irrespective of a change of posture often marks the end of a groaning episode. Treatment with benzodiazepines, antidepressants, or dopamine agonists has been unsuccessful or refused (Pevenargie et al., 2001; Vetrugno et al., 2001a; Oldani et al., 2005), and nasal continuous positive airway pressure has been proposed as an option to treat this condition (Iriarte et al., 2006; Guilleminault et al., 2008). The origin of this nocturnal groaning remains unexplained and the long-term
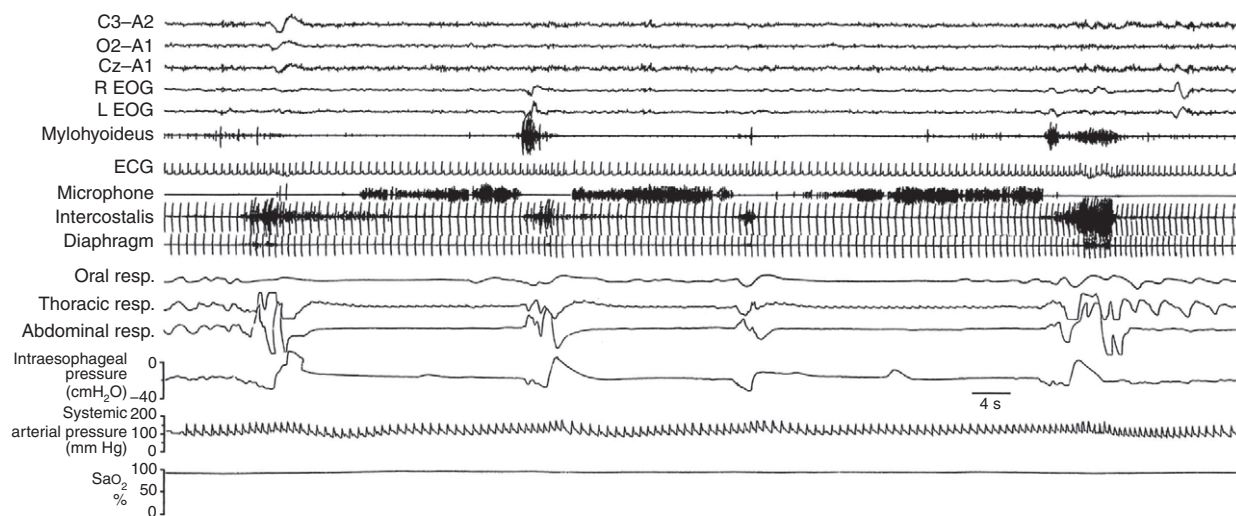

Fig. 54.8. Nocturnal groaning (catathrenia) arising during REM sleep. Groaning sounds (microphone) lasting 20–28 seconds follow one another in cluster during prolonged expiration, associated with slightly decreased heart rate and arterial pressure, and without appreciable SaO_2 modification. EOG, electro-oculogram; resp., respirogram; SaO_2, oxygen saturation; R, right; L, left.

prognosis unknown, as the normal wake laryngoscopic investigation cannot rule out a functional sleep-related obstruction of the upper airway during expiration.

SLEEP-RELATED LARYNGOSPASM

A laryngospasm is a spasm of the throat. Sleep-related laryngospasm refers to episodes of abrupt awakenings from sleep with an intense sensation of inability to breathe and stridor. If a laryngospasm occurs during sleep, the patient will typically wake up choking and jump out of bed, clutching the throat and rushing in panic to the bathroom or a window with acute fear of suffocation. Episodes last anywhere from a few seconds to five minutes. Patients typically experience laryngospasm only two or three times per year; the result is similar to a single episode of apnea, but these patients do not have apnea. Drinking water usually speeds the relaxation of throat muscles. Often patients with sleep-related laryngospasm have evidence of gastroesophageal reflux and respond to antireflux therapy (Thurnheer et al., 1997). Patients who suffer frequent attacks are afraid to go to sleep at night.

The infrequent, erratic and almost exclusively nocturnal occurrence and short duration of the attacks makes them hard to observe and to capture on polysomnography (Aloe and Thorpy, 1995). The differential diagnosis includes sleep-related neurogenic hyperventilation, nocturnal bronchial asthma, epilepsy manifesting as laryngospasm (Mahowald and Schenck, 1993), the sleep-related choking syndrome, the sleep-related abnormal swallowing syndrome, and nocturnal panic attacks (American Academy of Sleep Medicine, 2005).

SLEEP-RELATED CHOKING SYNDROME

This is a rare disorder. Unlike sleep-related laryngospasm, which usually only occurs two or three times a year, this condition causes the individual to have choking episodes almost every night, and sometimes several times in one night. This causes the individual to wake up with feelings of anxiety, fear, and even impending death. It is different from nightmares or night terrors, because the fear is always associated with the choking. These individuals do not suffer from sleep apnea (Thorpy and Aloe, 1989; American Academy of Sleep Medicine, 2005).

SLEEP-RELATED ABNORMAL SWALLOWING SYNDROME

People with this rare disorder have inadequate swallowing of their saliva with aspiration while sleeping. Saliva builds up in the mouth, then flows down the throat and is breathed into the lungs, the sleeper coughing, choking, and briefly arousing or completely waking up from sleep. Polysomnographic studies show brief episodes of coughing and gagging (Guilleminault et al., 1976; American Academy of Sleep Medicine, 2005).

SLEEP-RELATED NEUROGENIC TACHYPNEA

Sleep-related neurogenic tachypnea is a rare condition that was considered only as a proposed sleep disorder in the ICSD-1 but was excluded from the ICSD-2. It is characterized by a sustained respiratory rate occurring at sleep onset, maintained throughout sleep, and reversing immediately upon awakening. This disorder has been described in a few patients, associated with multiple sclerosis, lateral medullary syndrome, and benign intracranial hypertension (Broughton et al., 1988; Wilmer and Broughton, 1989). A patient with chronic sleep-related neurogenic tachypnea after head injury has been reported recently with polysomnographic documentation of the condition (King et al., 2005). Notably, tachypnea during sleep has been reported as a polysomnographic finding in patients with multiple system atrophy (Vetrugno et al., 2004).

SLEEP HYPERHIDROSIS (NIGHT SWEATS)

In sleep hyperhidrosis, more commonly known as "night sweats", profuse sweating occurs during sleep and requires the patient to change the bedclothes (Geschickte et al., 1966; Lea and Aber, 1985; Smetana, 1993). The sweating can cause an awakening because of the discomfort due to sweat sleepwear, and the patient may have to arise. Excessive daytime sweating may or may not be present. Excessive sweating during sleep may occur at any age, but most commonly in early adulthood. Some patients may have a lifelong tendency to sweat excessively during sleep, whereas in other patients it is a self-limited condition. Although uncomfortable, isolated nighttime sweating is not usually a sign of a serious underlying medical condition.

Polysomnography with quinizarin powder, which turns purple on contact with sweat, can be performed to demonstrate the affected body areas. Examinations are, however, aimed at revealing associated symptoms that enter into the differential diagnosis such as febrile illness, infections, menopause, diabetes insipidus, obstructive sleep apnea, gastroesophageal reflux disease, pregnancy, anxiety, medications, drug or alcohol abuse, hyperthyroidism, pheochromocytoma, cancer (especially lymphoma), hypothalamic lesions, epilepsy, cerebral and brainstem strokes, cerebral palsy, chronic paroxysmal hemicrania, spinal cord infarction, head injury, familial dysautonomia (Viera et al., 2003), primary hyperhidrosis,

and unilateral hemihidrotic hyperhidrosis in patients long confined to bed (Vetrugno et al., 2003). Idiopathic night sweats are at present classified in the group of miscellaneous secondary parasomnias in ICSD-2.

NOCTURNAL PANIC ATTACKS

Panic episodes arising during sleep are common, and may result in a sense of anxiety about falling asleep with a complaint of insomnia. Nocturnal panic attacks occur in 30–50% of patients with diurnal panic, but episodes arising exclusively from sleep appear to be rare (Hauri et al., 1989; Rosenfield and Furman, 1994; Schredl et al., 2001). During sleep studies, patients with nocturnal panic attacks do not usually report a dream. The episodes occur during NREM sleep, particularly at the transition from stage 2 to stage 3, and, in contrast to patients with sleep terrors, patients with nocturnal panic attacks are awake and hyperalert during the episode (Stein et al., 1993; Landry et al., 2002). Nocturnal panic attacks should be differentiated from the sleep terrors, nocturnal seizures, sleep apnea, and sleep-related abnormal swallowing, choking, and laryngospasm.

SLEEP-RELATED HALLUCINATIONS

Sleep-related hallucinations are included in this chapter because they may represent an isolated symptom of sleep (witness the hypnagogic hallucinations of drowsiness), and also because they are often associated in the motor manifestations, as, for example, in sleep paralysis (see below). Sleep-related hallucinations are hallucinatory experiences, principally visual, that occur at sleep onset (hypnagogic hallucinations) or on awakening from sleep (hypnopompic hallucinations). The term hypnagogic hallucinations was coined by Maury in 1848 to describe his own vivid hallucinations in the state of drowsiness, just before sleep. Sleep-related hallucinations are often vivid and terrifying, and are also recalled clearly; they are not perceived as dreams. Up to one-third of normal individuals may experience these hallucinations (Ohayon, 2000), anywhere from once in a lifetime to most nights, but they are particularly common in the narcolepsy–cataplexy syndrome. Vivid hallucinatory experiences are often associated with sleep paralysis, a temporary period of paralysis experienced prior to falling asleep or waking from REM sleep (Cheyne, 2005). Both hypnagogic and hypnopompic hallucinations are more common in younger persons and occur slightly more frequently in women than in men (Ohayon, 2000).

Complex nocturnal visual hallucinations represent a well-defined syndrome characterized by nocturnal visual hallucinations that occur upon waking during the night. Hallucinations consist of well-delineated, realistic, vivid, detailed, relatively immobile, silent, usually multicolor, and often oddly distorted images of people or animals, and occur several times a week. Described in 12 patients, mostly women (Silber et al., 2002, 2005), the age of onset of symptoms varied from 5 to 80 years (mean 40.2 years). The etiologies were different: idiopathic hypersomnia, β-adrenergic antagonists, dementia with Lewy bodies, visual loss from macular degeneration, anxiety disorder, and depression. No patient had symptoms suggesting psychosis. In only four patients, complex nocturnal visual hallucinations were the only parasomnia, whereas in the others they occurred together with sleepwalking, sleeptalking, RBD, sleep paralysis, and lucid dreams. The hallucinations occurred immediately after an arousal from NREM sleep (Kavey and Whyte, 1993; Silber et al., 2005) and the EEG during the episodes showed alpha rhythm without epileptic activity.

Complex visual hallucinations are also seen in pathological states such as Charles Bonnet syndrome (exclusively visual, well-formed hallucinations of people, places, and things, but devoid of emotional or threatening contents; they do not interfere with normal mental functions and occur in otherwise intact, elderly individuals with usually binocular visual loss), peduncular hallucinosis (striking visual images occasionally accompanied by tactile and auditory hallucinations, often of prolonged duration with disturbed sleep and related to brainstem or thalamus lesions; patients usually have insight into the hallucinations and cope with them well without features of paranoia or psychiatric disturbance), delirium tremens (variable, brief or usually almost continuous visual, occasionally polymodal, hallucinations with autonomic disturbances, motor agitation, and confusion), Parkinson's disease and Lewy body dementia (often in the evening and rarely polymodal, lasting some minutes usually with preserved insight, and related to widespread cortex and brainstem lesions, patients being not unconscious but normal or drowsy), migraine (simple visual, much rarer complex, hallucinations, the initial region of visual abnormality being usually near the fixation point, and the brilliant and flickering scintillations surrounding the negative scotoma making "fortifications" figures or spectra), schizophrenia (complex visual hallucinations usually in color and with auditory components, with animals and figures, but often with a delusional or hyper-religious character), epilepsy (usually brief, stereotyped, and fragmentary, with or without other seizure manifestations such as experiential phenomena, altered awareness, motor activity, and automatisms, often due to posterior temporoparietal lesions, patients being normal between episodes) (Teunisse

et al., 1996; Manford and Andermann, 1998; Barnes and David, 2001; Williams and Andrew, 2005).

Alice in Wonderland syndrome is a peculiar disorder in which the symptoms are remarkably similar to the distortion in body image and shape as experienced by the main character in Lewis Carroll's, 1865 novel, and first described in 1955 by the English psychiatrist John Todd. The patients have the feeling that their entire body or parts of it have been altered in shape and size, and usually experience visual hallucinations with impaired perception of time and place (also known as Lilliputian hallucinations). The majority of patients with the syndrome have a family history of migraine headache or have overt migraine themselves and, perhaps not coincidentally, Lewis Carroll himself suffered from severe migraine.

The visual imagery in the nightmares and RBDs occurs during sleep and not after waking, and is clearly recognized by the patients as dreaming. Sleep terrors arise out of NREM sleep, but patients recall little imagery and appear terrified and confused during the events.

A number of factors have been invoked to explain why complex hallucinations occur during sleep. Low ambient illumination may play a role, as the images vanish upon switching on the light. In most form of hallucinosis, however, including the Charles Bonnet syndrome, hallucinations are most prominent at the end of the day, which would support the importance of arousal and brainstem activity (Manford and Andermann, 1998).

SLEEP PARALYSIS

Sleep paralysis is one of the cardinal symptoms of the "narcoleptic tetrad" (Yoss and Daly, 1957): about 60% of people with narcolepsy experience sleep paralysis either rarely or frequently (even daily). However, recurrent isolated sleep paralysis (ISP), i.e., not associated with narcolepsy, is one of the lesser known and often benign forms of parasomnia.

ISP consists of brief episodes of transient inability to move when falling asleep (hypnagogic or predormital form) or, more usually, when wakening up, either during the night or in the morning (hypnopompic or postdormital form). Characteristically all of the skeletal muscles are virtually "paralyzed", resulting in inability to move the limbs, trunk, and head, or to speak; respiratory and ocular movements remain intact and consciousness is clear. Eye fluttering, moaning, numbness or tingling of the limbs, palpitation or sweating, and a sensation of struggling to move may be experienced during an episode, and also a feeling of pressure on the chest and difficulty breathing despite normal respiration function. ISP usually lasts from few seconds to several minutes. The muscle power returns to normal either spontaneously or if the individual tries hard to move or is touched by another person. The attacks leave no sequelae. Hallucinatory experience (visual hallucinations, unusual bodily sensations including floating and feeling of pressure on the body, sensing the presence of others, feeling external pressure on the chest, and hearing footsteps or odd sounds) can commonly accompany sleep paralysis, causing fear and making such episodes particularly alarming (Cheyne et al., 1999).

Sleep paralysis is such a vivid experience that it has been incorporated into popular folklore in many parts of the world, and may be interpreted as a supernatural experience, a nocturnal visitation of demons and devils, in a culturally distinct manner (Hinton et al., 2005). These cultural interpretations include "Kanashibari" in Japan (Fukuda et al., 1987), "ghost oppression phenomenon" in Hong Kong, China (Wing et al., 1999), "Old Hag" in Newfoundland (Ness, 1978), "*uqumangirniq* or *aqtuqsinniq*" among Inuit (Laws and Kirmayer, 2005) "bedridden by the witch" among African Americans in the USA, and visitation by *tokoloshis* (spirit of ancestors) among black South African people (Gangdev, 2004). In general, stress, anxiety, panic disorder, sleep deprivation or irregular or disturbed sleep–wake rhythm appear to precipitate attacks in some patients; ISP can be associated with the use of anxiolytic drugs, even after consideration for the concurrence of mental disorders.

Estimates of the prevalence of isolated ISP vary, depending on the sample under study and the ethnic, genetic, and cultural experience (reported prevalences of sleep paralysis can be changed by the terminology used) (Fukuda, 1993). Prevalence has been estimated as 6–62% in the general population (Dahlitz and Parkes, 1993; Ohayon et al., 1999), 18% in the elderly (Wing et al., 1999), and 29% among college student samples (Cheyne et al., 1999). Increased rates of ISP occur in adults reporting history of childhood sexual abuse and in individuals meeting criteria for post-traumatic stress disorder (Ohayon and Shapiro, 2000). The frequency of ISP may vary from a single attack in a lifetime to attacks every night. The age of onset of ISP is usually around the teens, and gender does not seem to be a factor. The occurrence of sleep paralysis in some families raises the possibility of a genetic mechanism (Dahlitz and Parkes, 1993; Wing et al., 1999).

It has been suggested that sleep paralysis consists of dissociated REM sleep activities that occur in full awareness: sleep paralysis occurs if awareness alone returns to the wake mode but muscle tone and perception continue in REM mode (Hishikawa, 1976). Video-polysomnographic recordings during sleep paralysis

document the characteristics of the dissociative state characterized by mixed EEG patterns of REM and wake, abundant alpha activity and atonia of antigravitational muscles.

Sleep position and timing appear to influence rates of sleep paralysis, with higher rates in individuals sleeping in the supine position and in patients reporting paralysis at the beginning and in the middle of sleep compared with those reporting episodes when waking up at the end of sleep (Buzzi and Cirignotta, 2000; Dahmen and Kasten, 2001; Cheyne, 2002; Girard and Cheyne, 2006).

The differential diagnosis of sleep paralysis includes: atonic generalized epileptic seizures (they occur during daytime and EEG shows epileptic abnormalities); drop attacks (they occur during wakefulness and in patients with vertebrobasilar vascular insufficiency); cataplexy (which does not occur at sleep onset and is a reaction to, rather than a cause of, emotional experience); familial hypokalemic periodic paralysis (which often occurs on awakening but lasts hours or days, is associated with low serum potassium levels during attacks, and is easily reversed by correcting the hypokalemia); dissociative or psychotic states (in which observation of the patient's movements during sleep can be conclusively diagnostic of psychogenic paralysis). In particular, difficulty waking up, for example because of intense fatigue, should not be mistaken for sleep paralysis. Occasionally, the symptoms associated with sleep paralysis may be mistaken for a psychotic state. It is possible for sleep paralysis to be the first presenting feature of the narcolepsy syndrome if the excessive daytime sleepiness has not been recognized or is concealed.

If sleep paralysis is frequent, serotoninergic agents seem to represent the most effective agents for treatment; in particular, amitriptyline has been used in conjunction with L-tryptophan as a precursor of brain 5-hydroxytryptamine (5HT, serotonin) (Snyder and Hams, 1982).

STATUS DISSOCIATUS

The concept of status dissociatus (SD) defines a clinical condition in which elements of one state of being (wakefulness, NREM sleep, and REM sleep) persist or are pathologically recruited into another state (Mahowald and Schenck, 1991, 2001). This condition is thus characterized by a breakdown of state-determining boundaries, in which every manifestation represents a "switching error" (Mahowald and Schenck, 2001). Polygraphically, SD is often characterized by the simultaneous admixture of elements of wakefulness, REM and NREM sleep, so as to make recognition of the proper wake or sleep stage difficult or even impossible.

The definition of SD is a broad one, but it is useful in the clinic, as it brings together under one concept several sleep disorders variously classified under different headings. The elemental forms of SD are cataplexy (intrusion of REM sleep muscle atonia into wakefulness), hypnagogic and hypnopompic hallucinations (dream mentation occurring during sleep), NREM parasomnias such as confusional arousals (wakefulness motor behavior occurring during deep sleep), REM parasomnias such as RBDs (persistence of muscle tone during REM sleep), and sleep paralysis (the persistence of REM sleep muscle atonia into wakefulness). Symptomatic SD has been reported after brain lesions, such as surgery for tegmental pontomesencephalic cavernoma (Provini et al., 2004) and paramedian thalamic syndromes (Montagna et al., 2002). The most complex forms of SD are characterized from a behavioral point of view by frequent muscle twitching (resembling excessive fragmentary myoclonus), diffuse and sometimes massive myoclonic jerks, vocalization, and complex motor behaviors associated with reports of dream-like mentation (Cochen et al., 2005). Agrypnia excitata, a syndrome characterized by prominent loss of slow-wave sleep and abnormal REM sleep without atonia, associated with autonomic and motor hyperactivity (Lugaresi and Provini, 2001; Montagna and Lugaresi, 2002), has been also viewed as a particular instance of SD.

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