A 12-year-old boy presented to the University of New Mexico Hospital Sleep Disorders Center accompanied by his mother, who stated that “he moans so loudly when sleeping that we are afraid bears will attack us when we’re camping, thinking he’s a hurt, dying animal.” Since 3 years of age, the patient has exhibited periods of groaning and moaning while asleep, lasting for up to about 1 hour and occurring several times virtually every night. There does not seem to be a predilection for any particular time of night. His usual bedtime is 8:30 PM, but he estimates an initial sleep latency of about 2 hours. Once asleep, he does not awaken until 6:00 AM on weekdays and 6:30 to 7:00 AM on weekends. His family describes him as a restless sleeper, but they have appreciated no snoring or wheezing. He has no symptoms consistent with restless legs syndrome and no witnessed leg movements during sleep. His past medical history is significant for exercise-induced asthma beginning at age 10 years, initially severe but currently well controlled on inhaled fluticasone propionate/salmeterol xinafoate twice daily and albuterol as needed, and congenital talipes

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Address correspondence to: Lee K. Brown M.D., Professor of Medicine and Pediatrics, Executive Director, Program in Sleep Medicine, University of New Mexico Health Sciences Center, 1101 Medical Arts Avenue NE, Building #2, Albuquerque, NM 87102; Tel: (505) 272-6110; Fax: (505) 272-6112; E-mail: lkbrown@alum.mit.edu

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equinovarus (club feet), requiring multiple surgeries up until the age of 3 years. Family history is positive for snoring and sleep apnea but not for parasomnias. Physical examination revealed a body mass index of 18.8 kg/m², moderate retrognathia, and a long tongue. He had no visible tonsillar tissue, and the remainder of his general and neurologic physical examination was unremarkable.

Video-Polysomnography Sleep Study Data

Overnight video-polysomnography (PSG) was performed, with an expanded electroencephalograhic (EEG) montage as well as bilateral wrist extensor electromyography. The tracings in the Figure are representative of the patient’s behavior during an episode of moaning, and the audio/video recording of this behavior is available on the Journal of Clinical Sleep Medicine website at http://www.aasmnet.org/JCSM/. Initial sleep latency was 24 minutes, sleep efficiency was 90%, and sleep maintenance was 95%. There were 4 rapid eye movement (REM) cycles, and the proportions of the various sleep stages as a percentage of total sleep time were stage 1, 20%; stage 2, 39%; slow-wave sleep, 16%; and REM sleep, 20%. Sleep was fragmented by a mean of 27 arousals and 3 awakenings per hour of sleep, with 17 per hour related to respiratory events. Two clusters of recurrent expiratory groaning were observed, 1 lasting 10 minutes and the other 25 minutes. The central apnea index was 12.9 per hour of sleep, with almost all events occurring just before the onset of a cluster of expiratory groans or immediately following an individual expiratory groan. Obstructive events were also seen; they were not associated with snoring, and the apnea-hypopnea index (including hypopneas followed by arousal or awakening without desaturation) was 6.9 per hour of sleep. Oxyhemoglobin saturation nadirs were 83% for central apneas, 85% for hypopneas, and 88% for obstructive apneas. The expanded EEG montage demonstrated no epileptiform activity.

What is Your Diagnosis?

Sleep Medicine Pearls
Sleep-Related Groaning (Catathrenia)

The PSG and video data demonstrate a repetitive pattern of non-REM (NREM) sleep-related groaning or moaning initiated by a sudden single augmented breath followed by a prolonged expiratory groan and then bradypnea (either a central apnea or a hypopnea without evidence of obstruction), sometimes terminated by an EEG arousal. These prolonged groaning expirations with bradypnea were occasionally followed by mild arterial oxyhemoglobin desaturations, rarely falling to as low as 90%. End-tidal PCO₂ was slightly higher (42-44 mm Hg) during periods of sleep-related groaning, as compared with during quiet sleep (38-40 mm Hg). The heart rate accelerated during the groan and then slowed during the central respiratory pause. These findings are characteristic of sleep-related groaning, or catathrenia, a parasomnia defined in the International Classification of Sleep Disorders, second edition.1

Sleep-related groaning was first reported in the medical literature in 1983, when De Roeck and coworkers described a young man who had expiratory groaning during REM sleep.2 In 2001, Vetrugno et al reported 4 additional patients with onset of the disorder between 5 and 16 years of age.3 Episodes would usually begin 2 to 6 hours after sleep onset, emerging more often from REM than from NREM (stage 2) sleep. The individual expiratory groans lasted from 2 to 20 seconds each and occurred in 2- to 60-minute clusters several times each night. These patients denied discomfort, respiratory distress, or even awareness of their groaning, and neither wheezing nor stridor was observed. These investigators coined the term catathrenia—from the Greek words kata, below or under, and threnia, to lament—to describe this sleep-related behavior. As of 2006, a total of 28 patients with catathrenia have been reported in the medical literature.2-7

Catathrenia is usually a chronic condition, occurring almost nightly over the course of many years. An EEG arousal with or without position change often signals the end of a groaning cluster, but overall sleep architecture is usually normal. A recent account of 12 new cases estimated a prevalence of 0.3% of patients referred to a sleep center over a 4-year period.8 An analysis of these 12, plus 9 additional cases from other centers, revealed a mean age of presentation in young adulthood (31.4 ± 8.1 years), a modest male predominance (13 men, 8 women), and a mean duration of symptoms before presentation of about 11 years.6 Seven patients had hypersomnia as assessed by the Epworth Sleepiness Scale. A family history of catathrenia was present in 3 patients, and pedigrees from 2 of these suggested an autosomal-dominant mode of inheritance. Other parasomnias (bruxism, somnambulism, somniloquy, sleep terrors) were reported in family members of 8 of the 21 patients.

Sixteen of the 21 patients underwent a single full-night laboratory 15-channel digital video-PSG. Catathrenia was observed in 12; the expiratory groan was characterized as a monotonous vocal noise, fairly constant in character in each subject, but varying in pitch and timbre between subjects. Some episodes were isolated, lasting about 10 seconds, whereas others occurred in short sequences lasting up to 4 minutes. No significant electromyographic activity was noted in the rectus abdominis or intercostal muscles during the expiratory phase. The majority (89%) of the 36 recorded episodes emerged from REM sleep, and there was no relationship with sleep posture. All of their patients had fewer than 5 apneas and hypopneas per hour of sleep, all had oxyhemoglobin saturations greater than 90% the entire night, and none had significant periodic limb movements of sleep. Pharmacologic treatment with clonazepam (2 patients), gabapentin, pramipexole, and trazodone (1 patient each) resulted in no sustained improvement in the parasomnia. Other authors have reported that trials of carbamazepine, paroxetine, and dosulepine (a tricyclic antidepressant) failed to suppress catathrenia in their patients.9 Continuous positive airway pressure (CPAP) has been reported to be ineffective in 2 patients5 but salutary in 1 patient with concurrent obstructive sleep apnea. The differential diagnosis for catathrenia includes somniloquy, sleep-related laryngospasm, stridor, nocturnal asthma, and moaning associated with epileptic seizures. Snoring is easily distinguished from catathrenia because snoring is a supraglottic sound occurring during inspiration.

Pathogenesis remains obscure. De Roek et al suggested a number of possibilities, including functional occlusion of the vocal cords during REM sleep with forced expiration to overcome this resistance or functional and/or anatomic lesions involving neurologic structures that control ventilation.2 Iriarte and colleagues speculated that the predilection for catathrenia during REM sleep might relate to asynchronous activation of the diaphragm and the oropharyngeal muscles during this sleep stage.7 Other authors have implicated the possible activity of a central pattern generator.8

In our view, studies of nonverbal sound production in the squirrel monkey may be instructive in understanding the pathogenesis of catathrenia.9 Calls and cries in this animal are stereotyped in acoustic structure and genetically determined and may be elicited by stimulation of neurons in a variety of locales. In 2 of these locations, the forebrain anterior cingulate cortex and the midbrain periaqueductual gray and laterally bordering tegmentum, stimulation results in stereotyped vocalization not accompanied by any associated motivational behavior. In particular, the midbrain periaqueductal gray seems to function as a collector of all descending vocalization-controlling limbic pathways; furthermore, vocalization is facilitated by injecting a number of neurotransmitters into this site, including GABA, agonists. Given the strong association between GABAergic neurons and the mechanisms producing both REM and NREM sleep, it is conceivable that excess sleep-associated facilitation of neurons in the periaqueductal gray could result in catathrenia in patients so disposed. The bradypnea that follows may simply be part of the prolonged expiration, as has been postulated by Sanders in his theory unifying the mechanisms of central, mixed, and obstructive sleep apneas,10 or may reflect the action of a variety of known neural circuits such as the Breuer-Hering reflex, the diving reflex, or other upper-airway reflexes.11-15

In our patient, a magnetic resonance imaging study of the brain and formal olotaryngologic examination were negative. Due to the presence (albeit mild) of obstructive sleep apnea on PSG, we repeated the nocturnal PSG with CPAP titration. A pressure of 8 cm H₂O somewhat unexpectedly resulted in the resolution of the catathrenia as well as the patient’s obstructive sleep apnea. On subsequent follow-up, the family reports that the patient wears his CPAP nightly, they do not hear any catathrenia, and the patient is experiencing less daytime sleepiness.

Pearls

1. Expiratory moaning or groaning during sleep must be dif-
ferred from inspiratory sounds such as snoring or stridor and should prompt consideration of a diagnosis of sleep-related groaning (catathrenia).

2. The diagnosis can be made solely on the basis of a reliable history or can be confirmed by PSG. The latter may be helpful to rule out a concurrent disorder such as obstructive sleep apnea, and to document the effect of CPAP treatment.

3. Sleep-related groaning (catathrenia) most frequently begins in adolescence and young adulthood, has modest male predominance, and is usually a chronic disorder. Daytime symptoms are uncommon and consist mainly of hypersomnia.

4. The pathogenesis is obscure but may involve the facilitation during sleep of acoustically stereotyped, genetically programmed vocalizations. Studies in the squirrel monkey suggest that such nonverbal utterances can originate in the midbrain periaqueductal gray.

REFERENCES


