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CASE REPORTS

Severe Chronic Sleep-Related Neurogenic Tachypnea

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We report a chronic and severe case of sleep-related neurogenic tachypnea that occurred after head injury and preceding the use of medications known to suppress the respiratory system. Failure to acknowledge or explain the patient's sleep behavior had previously contributed to her depression and anxiety. She accepted that her respiratory abnormality in sleep is of little prognostic significance. Her medication was not changed, and no treatment was initiated. The patient took comfort from the polysomnography evidence shown to her and was greatly reassured.

Key Words: Sleep Disorder, Neurogenic Tachypnea, Respiratory rate, pontine lesion

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Sleep-related neurogenic tachypnea is a rare condition and, at present, is classified as a proposed sleep disorder (*International Classification of Sleep Disorders* 780.59-4). It is characterized by a sustained increase in respiratory rate during sleep, which occurs at sleep onset; is maintained throughout sleep, and reverses immediately upon wakefulness. In 2 published abstracts,[1,2] 8 patients are described, and sleep-related neurogenic tachypnea is associated with multiple sclerosis, lateral medullary syndrome, and benign intracranial hypertension. In a more recent series of 3 patients,[3] all had complaints of daytime sleepiness. On polysomnography, 2 had features of obstructive sleep apnea, but tachypnea persisted despite therapy with continuous positive airway pressure. There were some features of autonomic dysfunction on electrocardiogram criteria. There are no published data on the persistence of this disorder over time.

HISTORY AND EXAMINATION

Patient History

A 70-year-old woman was referred to our sleep centre in 2001 for investigation of rapid respiration at night and a complaint of unrefreshing sleep. She had a fall in 1980 when she hit the base of her skull and lost consciousness. A radiograph of the skull revealed no fracture. Over subsequent days, she had further falls and developed progressive spasticity in her legs. Magnetic resonance imaging of her head was normal, and old fractures of her T2 and T12 vertebrae were reported as being unrelated to her neurologic symptoms. No final diagnosis was made. She subsequently complained of unrefreshing sleep, and her husband was concerned by her rapid respiratory rate at night. A limited respiratory study conducted at her local hospital 4 years previously demonstrated long periods of the night when her respiratory rate was 38 to 42 breaths a minute and other periods of the night when it was 13 to 16 breaths a minute. Her respiratory disturbance index was 4 per hour, and her oxygen desaturation index was 3 per hour (mean Sp_{O_2} 92%). The pulmonologist responsible for her care ascribed her sleep disturbance to carryover of a daytime anxiety state into sleep.

She was referred to a specialist in geriatric psychiatry 3 years after this study, who agreed that she was very anxious. At that time, she had had a low mood with suicidal thoughts. She was prescribed trazodone for its potential anxiolytic properties. Her complaint of unrefreshing sleep persisted, and her husband's concern over her sleep and breathing problem initiated her referral to several other medical specialists. Numerous further investigations failed to provide a diagnosis, and this contributed to her low mood and anxiety. Over the following years, she was prescribed medications for spasticity, anxiety, depression, and difficulty falling asleep. She found clomethiazole to be useful to help her to sleep at night but obtained little symptomatic relief from other medications. These included diazepam, dihydrocodeine, and propranolol.

Clinical Presentation

She appeared well, and, on examination, her cardiovascular and respiratory systems were unremarkable. She had increased tone in her quadriceps with upgoing plantar reflexes and normal upper limbs. There were no other neurologic signs.

Sleep Investigations

Polysomnography confirmed a normal respiratory rate of 14 to 16 breaths per minute before sleep onset and in wake after sleep onset. The mean respiratory rate during sleep (whether non-rapid eye movement or rapid eye movement) was 40 ± 2 breaths per minute (mean increase from baseline, 167%) (see Figure 1) with normal pulse oximetry (mean Sp_{O_2}, 93%) and an apnea-hypopnea index of 0. Audio recordings confirmed vocalization during sleep with no stridor. Sleep macrostructure was normal, with an arousal index of 20 per hour.

Disclosure Statement
This was not an industry supported study. Drs. King, Shneerson, and Smith have indicated no financial conflicts of interest.

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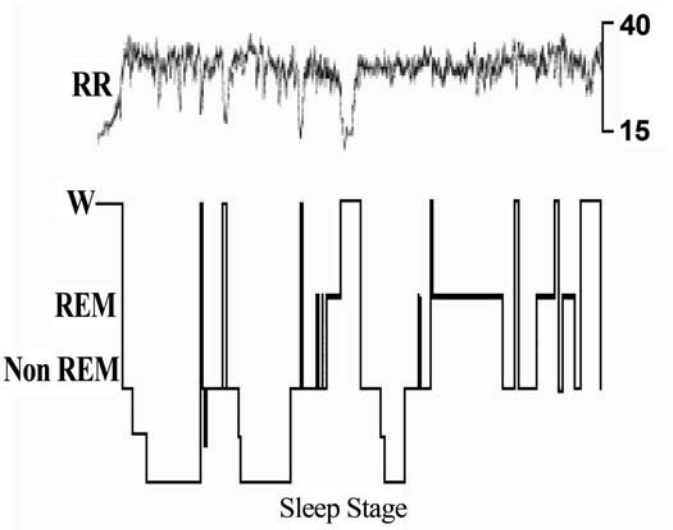

Figure 1—An overnight hypnogram and trend curve of respiratory rate (RR) demonstrating a dramatic difference between wake RR and sleep RR. Significant changes in RR are evident at the transition from wake to sleep and from sleep to wake. Similar changes in RR are seen between sleep and prolonged arousals from sleep (3-15 seconds). W refers to wakefulness; REM, rapid eye movement sleep.

She reported mild daytime somnolence (Epworth Sleepiness Scale score, 13). The patient and her husband were relieved by these findings. They found it easier to accept the abnormal respiratory pattern as part of her underlying neurologic disorder rather than as a manifestation of anxiety, which they had always maintained, was a secondary phenomenon.

Summary

We report a chronic (documented over 4 years) and severe case of sleep-related neurogenic tachypnea that occurred after head injury and before the use of medications known to suppress the respiratory system. Failure to acknowledge or explain the patient's sleep behavior contributed to her developing depression and anxiety. Her mental state improved once a diagnosis was achieved.

DISCUSSION

Sleep-related neurogenic tachypnea is rare, and its pathophysiology is unclear. The rapid respiratory rate may be due to loss of the inhibitory tone from higher centers, particularly the cerebral cortex, or the medullary respiratory center. A similar pattern of rapid breathing is seen during wakefulness in central neurogenic hyperventilation, in which central neural structures regulating respiration retain their sensitivity to reflex and chemical stimuli. This condition is related to pontine lesions and, particularly, necrosis of the medial pons.[4] An alternative theory suggests that a lesion in the reticular activating system may affect respiratory control and also lead to daytime sleepiness.[3]

Our patient with chronic sleep-related neurogenic tachypnea has had no cardiopulmonary complications and has seemingly retained respiratory chemosensitivity and cortical control of breathing, suggesting that the expression of a local medial pontine lesion is possibly unmasked when she is asleep. Our patient's disorder has persisted for at least 7 years, with no evidence of deterioration, and does not seem to be an important prognostic concern. Documenting the abnormal pattern of respiration at night and explaining to the patient that this is a recognized disorder was valuable in the management of this individual, and we would support the retention of this disorder in the *International Classification of Sleep Disorders*.

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