CASE REPORT

Expiratory-Synchronized Sleep in a Quadriplegic Patient Using Inspiratory Neck Muscles To Breathe

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ABSTRACT

In a patient with C3 quadriplegia causing complete diaphragm paralysis who developed inspiratory neck muscles (INM) hypertrophy to sustain ventilation, spontaneous breathing deeply altered sleep architecture, relegating sleep to the expiratory phase of the ventilatory cycle. A polysomnographic recording performed during mechanical ventilation (without INM activity), showed that sleep was abnormal but unaffected by the respiratory cycle. During spontaneous breathing, the polygraphic recordings showed expiratory microsleep episodes, with inspiratory arousals synchronous to bursts of INM activity. This case report illustrates the powerful adaptability of the respiratory and sleep control systems to maintain each vital function.

KEYWORDS: Diaphragm, respiratory muscles, sleep, sleep-related respiratory disorders, tetraplegia, control of breathing, brain plasticity

Diaphragmatic dysfunction can lead to hypoventilation during sleep and particularly during rapid eye movement (REM) sleep, where the diaphragm alone has to cope with the burden of ventilation.[1] In a young adult quadriplegic who had learned to breathe with his inspiratory neck muscles, the use of these muscles during sleep caused cyclical arousals relegating sleep to the expiratory phase of respiration. This illustrates how adaptations to the complete loss of diaphragm function can occur to preserve vital functions, respiration, and sleep.

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CASE DESCRIPTION

A 28-year-old patient suffered traumatic C3 tetraplegia with central respiratory paralysis. Because of associated stretch lesions of the cervical roots, diaphragm pacing was contraindicated.[2] There was no evidence of head or medulla injury, particularly no pharyngeal sensorimotor abnormalities. The patient underwent a 2-year rehabilitation program including inspiratory neck muscles (INM) training. He achieved major INM hypertrophy, and became capable of sustaining spontaneous breathing for several hours by voluntarily activating INM (20 breaths/min^{-1} on average; minute ventilation 7 L/min^{-1}; baseline $PaCO_2$, 43 mm Hg). INM inspiratory activity persisted during feeding tasks, mental operations, or computer tasks requiring close attention, although it was weaker and at a slower pace. In view of these results, the question of autonomous ventilation during sleep arose.

Methods for Sleep Studies To evaluate the possibility of nocturnal weaning from the ventilator, polysomnographic recordings were obtained from 11:00 PM to 7:30 AM in the intensive care unit, with (night 1) and without (night 2) mechanical ventilation. The recordings included electroencephalogram (Fp1/Cz, Fp2/Cz, O1/Cz), electro-oculogram (EOG), and chin electromyogram (chin EMG). Respiration was monitored using a thoracic inductance plethysmography band and surface EMGs of INM from electrodes taped over the muscle masses of the right and left sternomastoid muscles. Sleep stages and microarousals were scored using standard criteria.[3,4] At the time of the study, the patient was given fluoxetine 20 mg/d for 2 years, as a treatment for depression.

Sleep During Mechanical Ventilation (Night 1) Almost instantaneously after the start of mechanical ventilation, INM activity disappeared while the patient was still awake. Alpha rhythm in O1/Cz had a regular 10 Hz frequency. NREM and REM sleep latency were 28 and 85 minutes, respectively. Total sleep time was 421 minutes (sleep efficiency, 87%). Slow wave sleep was dramatically reduced (4 minutes). INM EMGs were silent both in NREM and REM sleep (Fig. 1, left). Blood gases were normal under mechanical ventilation.

Sleep During Spontaneous Respiration (Night 2) Sleep latency was 152 minutes. Respiration slowed down slightly, with a lengthened expiratory time (awake, 0.8 ± 0.3 second; asleep, 1.2 ± 0.4 seconds). Sleep architecture was extremely unusual, with regularly recurring "sleep" epochs of 1 to 2 seconds' duration containing EEG theta activity, strictly confined to expiration (Fig. 1, right, top trace, downward arrow). Each inspiration (INM activity) was accompanied by signs of EEG arousal (see Fig. 1, right). Despite a likely contamination of the EEG signal by INM and perhaps masseter activity hampering the evaluation of EEG frequency, the Fp1/ Cz signal during inspiration—after the upward arrow on Fig. 1—exhibits a major decrease in amplitude as compared with the previous expiration—after the downward arrow. This suggests that inspiration was accompanied by the suppression of the theta rhythm visible during expiration. Furthermore, regular 10-Hz α waves are visible on the occipitocentral trace during the next expiration. The microsleep episodes also did not contain sleep spindles and K complexes. The patient remained in a quiet awake state containing 47 episodes of "expiratory microsleep" during 95 minutes. At the beginning of the night (first 30 minutes), these episodes recurred every 237 ± 202 seconds and lasted 0.89 ± 0.16 second. Then they tended to become more frequent and prolonged (76 ± 48-second interval, 1.55 ± 0.33-second duration during the last 30 minutes of the recording), suggesting an increased sleep pressure. The patient then asked to be put back on the ventilator. Stable sleep stage 2 appeared 18 minutes later. The characteristics of the subsequent sleep were

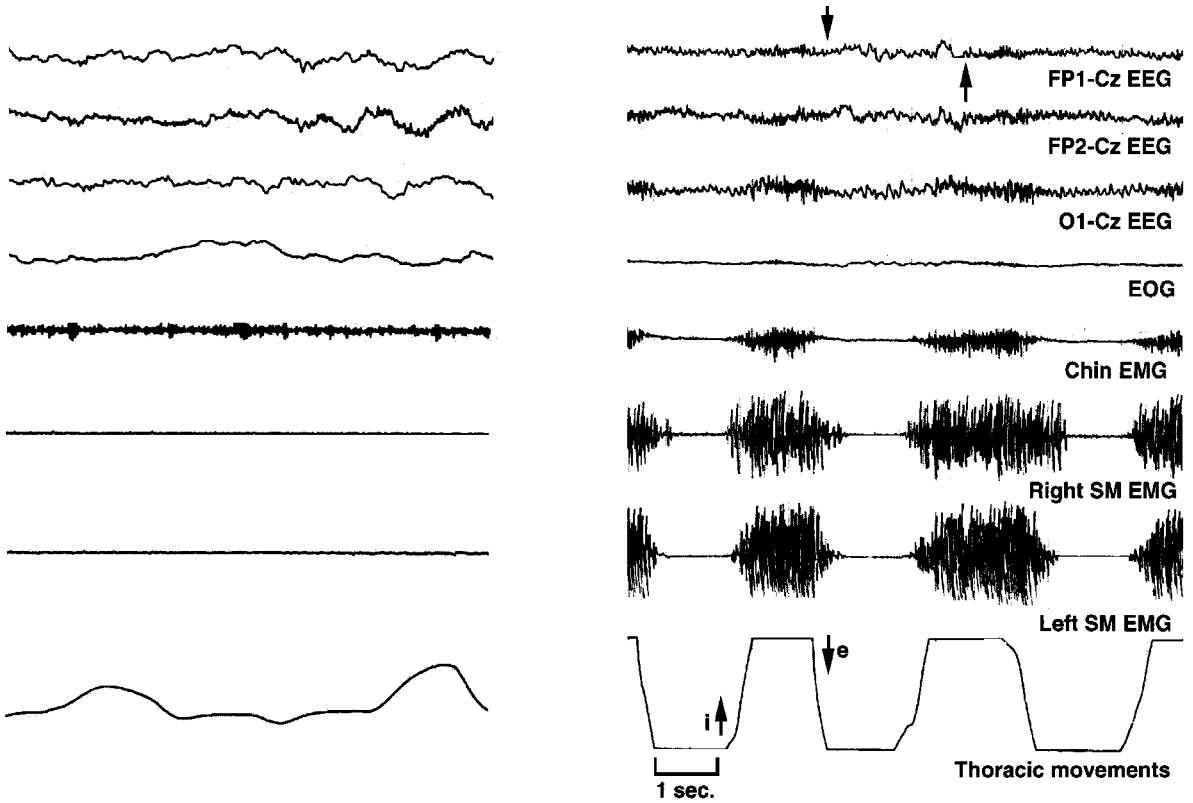

Figure 1 Sleep polygraphic recordings (from top to bottom, three electroencephalogram channels (Fp1/Cz, Fp2/Cz, and O1/Cz), one electro-oculogram channel (EOG), one chin electromyogram channel (chin EMG), and three respiratory channels, namely right (R) and left (L) inspiratory neck muscles surface electromyograms (SM EMG) and thoracic movements through a strain gauge mounted on an elastic belt). (Left) Recordings obtained during mechanical ventilation (night 1), showing that INM are inactive. (Right) An example of a microsleep episode during spontaneous breathing (night 2). On the top EEG trace, the downward arrow indicates the appearance of theta EEG waves (low frequency, high amplitude waves), as observed in light sleep stages, simultaneous with expiration. The upward arrow indicates an arousal (low amplitude EEG waves, when compared with previous theta waves, mixed with EMG activity) simultaneous with inspiration. Sleep is here confined to expiratory time. During the following cycle, the patient remains awake. On the bottom trace (respiratory movements), the upward arrow denotes thoracic expansion during inspiration (i), and the downward arrow denotes expiratory (e) thoracic deflation.

similar to those noted during night 1, including three cycles of NREM-REM sleep.

DISCUSSION

To begin with, it must be acknowledged that the contamination of the EEG signal by the inspiratory activity of neck muscles, visible in Figure 1, sets some limitation to the detection of arousals in our patient. In particular, it makes difficult the evaluation of EEG frequency. Nevertheless, we feel that, as described above, there were EEG elements sufficiently suggestive of inspiratory arousals and expiratory sleep.

Although far from a stable sleep state, the patient's vigilance was markedly decreased during night 2. The persistence of INM breathing suggests that it did not depend solely on the patient's learned,

voluntary control of breathing, but that some degree of automatism had taken place. Such a plasticity could be obtained by deriving the output of the central respiratory pattern generator (CPG), normally destined for the diaphragm, to the INM. It could also be the result of the neck muscles training program. The decrease in the force and frequency of INM contractions during mental tasks and reduced vigilance, resembling observations in congenital central alveolar hypoventilation,[5] suggests an INM control system different from the normal CPG, possibly suprabulbar.

During spontaneous breathing, "sleep" consisted of high amplitude theta waves as seen in sleep stage 1B (but this term cannot be used strictly speaking, each expiratory bout of "sleep" lasting less than 2.5 seconds[4]). The extreme brevity of the microsleep episodes, which has been described only in deeply sleep-deprived or narcoleptic sleepy adults,[6] is a striking feature of our observation. It implies that the source of inspiratory arousals was different from the long latency chemical or mechanical process involved, for example, in the termination of obstructive sleep apneas. The 400 milliseconds average difference between wake and sleep expiratory times sets the maximal delay of detection of the arousing phenomenon and suggests a reflex mechanism. The patient had sensory denervation of the rib cage and diaphragm, but an intact vagus nerve and sensory feedback from neck muscles. A unifying hypothesis to explain why INM contractions and expiratory microsleep episodes alternate can thus be constructed. First, the expiratory decrease of lung volume below functional respiratory capacity, unsurprising in a patient with complete diaphragm paralysis, could have triggered inspiration through the vagally mediated lung deflation reflex.[7] The corresponding afferent messages would in turn have stimulated the bulbar respiratory central pattern generator, presumably still functional and intact in this patient although made clinically "silent" by the spinal lesion. The resulting increase in respiratory drive would, through a corollary discharge[8] type of mechanism, be responsible for the arousal, the latter in turn permitting the expression of the wake-dependent INM contraction program.

Whatever the mechanisms at play, and although they were insufficient to achieve a life-compatible equilibrium in this patient, the present case provides a strong demonstration of the plasticity and intertwining of the neural control systems that regulate sleep and respiration.

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