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Case Report

Catathrenia Presenting as Excessive Daytime Sleepiness

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ABSTRACT

Catathrenia is a rare condition and corresponds to the identification of a deep inspiration followed by a prolonged and monotone groaning, usually occurring on rapid eye movement (REM) sleep. Although it's clinical relevance is still uncertain.

In this report, the authors describe a case of catathrenia presenting with excessive daytime sleepiness and non-restoring sleep that was interfering with work performance as well as martial life. This condition was successfully treated with Positive Airway Pressure (PAP) despite the lack of diagnosis of Obstructive Sleep Apnea (OSA) by correction of sleep fragmentation.

Treatment with PAP is well established when catathrenia is associated with OSA but is unclear if no other sleep disorder is present. In the last International Classification of Sleep Disorders, catathrenia has been classified as an isolated symptom, however in this patient, the magnitude of repercussion on her daily life might suggest the need for a recategorization. We hypothesize that the pathophysiologic mechanism is related to upper airway ventilatory instability due to a high arousal index and that secondary sleep fragmentation explains excessive daytime somnolence and fatigue that are reversed with PAP treatment.

Keywords: Catathrenia; Excessive daytime sleepiness; Positive airway pressure

INTRODUCTION

Catathrenia is a rare condition, with a reported incidence between 0.17% and 0.40% [1]. It is diagnosed by polysomnography and corresponds to the identification of a deep inspiration followed by a prolonged and monotone groaning associated to a respiratory pattern of bradypnea, that mimics central apnea and usually occurs on rapid eye movement (REM) sleep [1,2]. These respiratory patterns may last between 2 and 49 seconds [3]. There has also been reported an association with electroencephalographic (EEG) arousals in 80% of the cases, either preceding or during the events [2,3].

Its clinical relevance is still unclear, but in this disorder there is a potential for high arousal index associated with ventilatory instability and consequent excessive daytime sleepiness that has been reported to occur in 44.7% of patients, as well as fatigue [2,4]. However, there seems to be different phenotypes to this condition, including patients with and without Obstructive Sleep Apnea (OSA) and variations on their REM sleep specificity [5].

OSA has been observed in 34% of the cases of catathrenia and positive airway pressure (PAP) seems to be an efficient treatment, but there has been poor patient compliance associated with its use because the patients remain unaware of their condition and for the lack of unequivocal evidence of physical harm associated with catathrenia [1,2,6]. Anxiety and social distress lead patients to seek medical counsel, because the groaning causes unrest and discomfort on their bed-partners, as well as being a social problem when it is associated with sexual connotation [1].

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In this report, the authors describe a case of catathrenia presenting with excessive daytime sleepiness without the diagnosis of OSA and treated successfully with PAP.

CASE REPORT

A 29-years-old female, non-smoker with no medical or drug abuse history sought a neurologist due to fatigue and sleepiness during the day, associated with non-restoring sleep that was interfering with her work performance. The family reported a loud groaning during sleep since she was 17-years-old and the relationship with her husband was becoming problematic because he could not rest at night.

On the first evaluation, she had an Epworth Sleepiness Scale (ESS) score of 13, her weight was 74 Kg and height was 165 cm (body mass index: 27.1 Kg/m^2). She had no history of stridor, laryngospasm, snoring, or previous diagnosis of OSA, restless leg syndrome, parasomnia, narcolepsy or seizures. Physical examination and family history were unremarkable.

A sleep study (level-2 polysomnography using SomnoScreen with DOMINO software 2.8.0) was performed and scored by a trained somnologist according to the American Academy of Sleep Medicine manual of 2018. During the study, the sleep efficiency was 91.4%, REM sleep was low (10.2%), the arousal index was elevated (25.1 events/hour), the respiratory disturbance index was normal (3.4 events/hour), the periodic limb movement index was normal (5.5 events/hour) and the oxyhemoglobin desaturation index was 0 events/hour. Catathrenia was evaluated in clusters forming catathrenia periods (CP). Nine CP were identified with a total duration of 8.3 minutes and the first period visualized was during non-rapid eye movement (NREM) sleep (N1), but there was a predominance of events on REM sleep (5.7 minutes; 69%). The catathrenia index was 7.5 events/hour in total sleep and 47.5 events/hour in REM sleep. An example is shown in Figure 1.

There were no abnormalities identifiable on a brain CT scan leading to a diagnosis of catathrenia. Treatment with PAP was proposed and accepted by the patient due to her excessive daytime sleepiness and family disturbance, despite the lack of criteria for an OSA diagnosis.

For two years the patient has been using PAP with a nasal mask, with a mean pressure of 8.5 cmH_2O (between 4 and 9 cmH_2O). The patient no longer reports fatigue or daytime sleepiness (EES score of 8), but still refers higher sleepiness as a passenger in a car for an hour without break and laying down to rest in the afternoon when circumstances permit. There was a significant improvement in her work performance and her husband confirms the disappearance of groaning during sleep.

The onset of catathrenia usually occurs during adolescence or early adulthood, as observed in this patient. Nevertheless, she only sought medical intervention after her husband complained about the severe disturbance caused by her sleeping noise as previously reported in similar cases [1-3,5,6].

Treatment with PAP is well established when catathrenia is associated with OSA [6]. However, the need for treatment is unclear if no other sleep disorder is present, due to inconsistent data [2]. Despite the frequency of poor patient compliance, in this case there was a good adherence to PAP treatment. This was because of multiple factors such as there being resolution of the disturbance to her husband sleep quality and by the improvement of her symptoms, including excessive daytime sleepiness.

In the third edition of the International Classification of Sleep Disorders (ICSD), catathrenia has been classified as an isolated symptom [7], however this patient highlights the need for a recategorization. We, as Guilleminault [5], believe that catathrenia is an uncommon symptom of Sleep Disordered Breathing, with consequent excessive daytime somnolence and fatigue. We hypothesize that the pathophysiologic mechanism is related to upper airway ventilatory instability due to a high arousal index. Secondary sleep fragmentation explains excessive daytime somnolence and fatigue that are reversed with PAP treatment, as well as the abolition of noise. Further studies on the evolution of this disorder over time and the long-term response to PAP are needed.

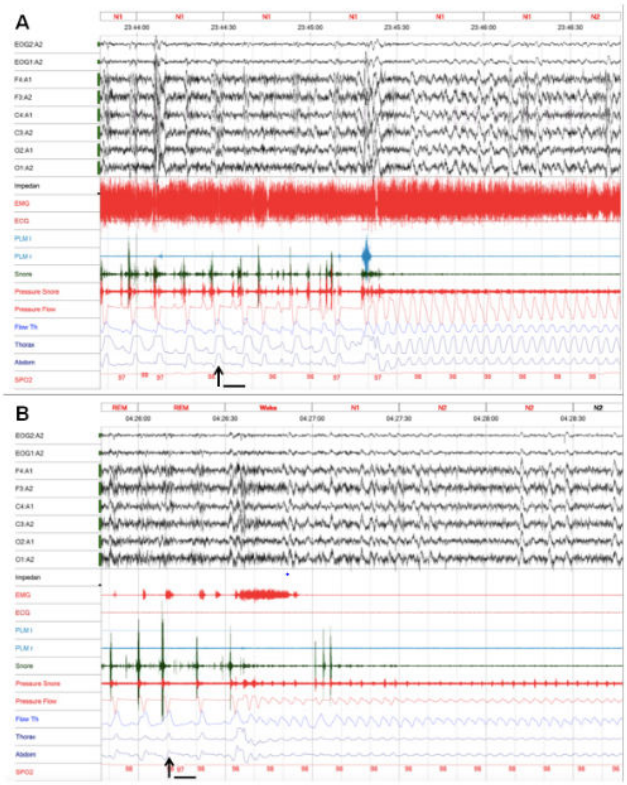

Figure 1: Two clusters of catathrenia events shown in N1 (A) and in REM sleep (B) characterized by a deep inspiration (arrow) followed by a prolonged expiration (line) and groaning.

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