Atypical Catathrenia Mimicking Sleep Choking Sound of Obstructive Sleep Apnea

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Catathrenia is a rare type of sleep-related breathing disorder with benign course and generally does not require treatment. However, the psychological burden on patients and bed partners is not light. In most cases, due to the sound characteristic of identifiable harmonics, it can be easily diagnosed; however, in some cases, it has non-typical sounds, and polysomnography is essential to diagnose. Here, I report a middle-aged woman with choking sounds like an obstructive sleep apnea patient during sleep, with typical polysomnography features of catathrenia.

Keywords
Catathrenia; Obstructive sleep apnea; Polysomnography.

A 56-year-old woman was consulted at the sleep clinic for snoring and choking sounds during sleep. She was overweight (height 160 cm, weight 60 kg, and body mass index 23.4 kg/m²). Otherwise, she was healthy. Habitual sleep time was between 23:00 and 06:00. Her sleep partner reported mild snoring, apnea, and choking during sleep, but the patient denied these. She reported unrefreshing sleep, fatigue, and excessive daytime sleepiness (Epworth sleepiness scale, 15). STOP–BANG score was 3, and Mallampati score was 3. As clinical symptoms suggested obstructive sleep apnea (OSA), polysomnography (PSG) was performed. Total sleep time was 417 minutes, and sleep efficiency was 84.8%. PSG revealed a normal range of apnea or hypopnea (apnea–hypopnea index, 4/h). Arousal index was 11.4/h. Periodic limb movement was absent. Rapid eye movement (REM) without atonia was not observed. A few dozen repeated short choking sounds occurred twice, both during REM sleep, lasting for 13 and 34 seconds at AM 01:12 and 01:13, respectively (Fig. 1A). Series of choking sounds were recorded while she exhaled a long breath after taking a deep breath (Supplementary Audio 1 in the online-only Data Supplement). The patient did not seem in pain, and there was no change in her facial expression. During events, there was no respiratory flow or respiratory effort (Fig. 1B). Diagnosis of catathrenia was made, and although therapeutic continuous positive airway pressure (CPAP) therapy and multiple sleep latency test were discussed, the patient refused CPAP and further evaluation.

Catathrenia, so-called expiratory groaning, is a rare condition that is categorized as a sleep-related breathing disorder in the International Classification of Sleep Disorders—Third Edition [1]. Catathrenia occurs predominantly during REM sleep, but has also been identified in non-REM sleep [1]. Catathrenia may co-occur with OSA, but has not been reported with significant morbidity [1,2]. Although the pathophysiology of catathrenia is largely unknown, the origin of catathrenia is different from snoring: catathrenia arises from laryngeal, and snoring from guttural sources [3,4]. Monotonous vocalization resembling groaning is characteristic [1,4,5]; however, sound characteristics may be diverse. Sound analysis of five catathrenia reported two sound characteristics: monotonous sinusoidal with normal pitch.
Fig. 1. The hypnogram (A) and 60-second excerpt from overnight polysomnography (B). The hypnogram showed decreased N3 sleep but relatively normal sleep architectures. Some bouts of hypopnea were recorded but within the normal range of apnea–hypopnea index, 4/h. The arrowhead indicates time points of the onset of catathrenia (A). Thermistor and nasal pressure transducer signal are nearly absent, and respiratory efforts of thorax and abdomen are also absent. The episode can be scored as central sleep apnea if expiratory sound as catathrenia is absent. Repeated short choking sound during the event is against central apnea. Note the decreased heart rate and slightly decreased oxygen saturation during the event (B). Typical polysomnographic findings of catathrenia resemble those of central apnea, but the sound characteristics may be diverse.
or sawtooth shaped with high pitch sound [4]. Iriarte et al. [5] reviewed previous cases and grouped them into two different types of catathrenia. One is long-lasting soft-intensity sounds like sirens occurring during REM sleep, which are associated with central apnea and non-responsive to CPAP. Another is short-lasting loud-intensity sounds occurring during both non-REM and REM sleep, which are associated with normal or apnea/hypopnea and responsive to CPAP therapy. This case may be grouped as the first one according to Iriarte’s classification, but the sound character is quite different.

Although catathrenia is generally considered a benign disorder that does not require treatment, it may be a frustrating concern and source of anxiety in some patients and their bed partners [2], especially young adults. As the sound characteristics of catathrenia may be atypical, PSG is the only golden tool to diagnose catathrenia and evaluate its possible comorbid sleep disturbances. PSG features of catathrenia mimicking central apnea should be considered as diagnostic reference when the sound character is atypical, especially when occurring during REM sleep.

**Supplementary Audio Legend**
Audio 1. First episode depicted in Fig. 1.

**Supplementary Materials**
The online-only Data Supplement is available with this article at https://doi.org/10.17241/smr.2023.01634.

**Ethics Statement**
This study followed the principles of the Declaration of Helsinki and was approved by the Inje University Busan Paik Hospital Institutional Review Board (BFIRB#: 2022-06-020) with a waiver of informed consent.

**Availability of Data and Material**
All data generated or analyzed during the study are included in this published article.

**Conflicts of Interest**
The author has no potential conflicts of interest to disclose.

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**REFERENCES**