Sleep Breath
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CASE REPORT

Effect of CPAP therapy on catathrenia and OSA: a case report and review of the literature

Murat Songu · Hikmet Yilmaz · Ali Vefa Yuceturk · Kivanc Gunhan · Aysun Ince · Ozgur Bayturan

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Abstract
Introduction Catathrenia is a rare, idiopathic, sleep-related respiratory condition characterized by irregular groans, which occur during prolonged expiration in sleep. The origins of catathrenia remain inexplicable, the long-term prognosis unexplained. Moreover, empirical treatment with neither pharmacological nor non-pharmacological approaches was satisfactory.
Case report We report a case of catathrenia with concurrent obstructive sleep apnea complicated with pulmonary hypertension and reviewed the literature.
Discussion Treatment with nasal continuous positive airway pressure resulted in marked improvement of catathrenia, obstructive sleep apnea, daytime dyspnea, and pulmonary hypertension for our patient. We think that nasal continuous positive airway pressure can be an option for the treatment of this infrequent but sometimes very disturbing sleep disorder.

Keywords Catathrenia · Nocturnal groaning · Moaning · Obstructive sleep apnea · Pulmonary hypertension · Continuous positive airway pressure

Introduction

Catathrenia (nocturnal groaning) is a rare, idiopathic, sleep-related respiratory condition characterized by high-pitched, monotonous, irregular groans, which occur during prolonged expiration in sleep. Catathrenia was first reported in the medical literature in Edegem, Belgium, in 1983 and was included in the recent International Classification of Sleep Disorders, 2nd edition (ICSD-2) [1, 2].

Obstructive sleep apnea (OSA) is a significant medical problem affecting up to 4% of middle-aged adults [3]. The most common complaints are loud snoring, disrupted sleep, and excessive daytime sleepiness. Pulmonary hypertension (PH) is a common complication in patients with moderate to severe OSA with a prevalence of approximately 20% [4]. The potential mechanisms of daytime PH in OSA are elevated daytime pulmonary vascular tone secondary to hypoxic pulmonary vasoconstriction, hypoxia-induced endothelial dysfunction, and pulmonary vascular remodeling [5, 6]. Transthoracic Doppler echocardiography (TTE) is an excellent non-invasive screening test for the patient with suspected PH. In this case, ($4v^2$+estimated right atrial pressure) to measure estimated systolic pulmonary arterial pressure (PAPs), which is also known as the simplified Bernoulli equation. According to the available data mild

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M. Songu (✉)
Department of Otorhinolaryngology/Head and Neck Surgery,
Dr. Behcet Uz Children's Hospital,
Izmir, Turkey
e-mail: songumurat@yahoo.com

H. Yilmaz · A. Ince
Department of Neurology, Celal Bayar University Medical Faculty,
Manisa, Turkey

A. V. Yuceturk · K. Gunhan
Department of Otorhinolaryngology/Head and Neck Surgery,
Celal Bayar University Medical Faculty,
Manisa, Turkey

O. Bayturan
Department of Cardiology,
Celal Bayar University Medical Faculty,
Manisa, Turkey

PH can be defined as a PAPs of approximately 36–50 mmHg or a resting tricuspid regurgitant velocity of 2.8–3.4 m/s (assuming a normal right atrial pressure). It should be noted that also with this definition, a number of false positive diagnoses can be anticipated especially in aged subjects and confirmation with right heart catheterization is required in symptomatic patients (NYHA—New York Heart Association Class II-III).

We herein review the literature while reporting a case of catathrenia with concurrent OSA complicated with pulmonary hypertension and point out that nasal continuous positive airway pressure (nCPAP) is an option for treatment.

Case report

A 40-year-old female was followed-up yearly in the outpatient clinic of cardiology for 4 years. She underwent operation for ventricular septal defect at age nine. She did not have any complaints until last year and she had been receiving no medication since 5 years. She was admitted with a progressive dyspnea on exertion, which she had for the past year. Transthoracic Doppler echocardiography was performed. There was no significant flow across the operated VSD patch and the estimated PAPs calculated 51 mmHg according to the simplified Bernoulli equation representing moderate PH [7]. Since her systolic pulmonary arterial pressure was increased and symptoms (NYHA class II-III) occurred, she was admitted to the hospital for further evaluation of PH. Arterial blood gases revealed mild daytime hypoxemia. Right heart catheterization was carried out in order to confirm the PH and to assess the severity of the hemodynamic impairment. Systolic pulmonary arterial pressure measured 52 mmHg, diastolic pulmonary arterial pressure was 20 mmHg, mean pulmonary arterial pressure measured 43 mmHg, pulmonary vascular resistance was 5.6 mmHg/L/min (Wood unit) and pulmonary capillary wedge pressure was 15 mmHg. High-resolution CT of the chest revealed minimal dilatation of pulmonary artery and the pulmonary function tests were normal. Otorhinolaryngologic examination upon admission revealed that she might have been suffering OSA complicated with PH and a polysomnography (PSG) was performed in the sleep research center.

The patient's history was detailed and was realized that, since age seven, she has exhibited periods of loud groaning while asleep, occurring several times every night and for the last 3 years her husband described episodes in which the patient stops breathing and then gives a loud gasp or snort when aroused by the apnea. The groaning recurred every night unassociated with insomnia, dreaming, or daytime sleepiness. She had no recall of vivid dreams and did not show abnormal motor behavior during sleep. There was no family history of parasomnia or violent behavior during sleep. She complained of restless sleep and tiredness during the daytime.

Physical examination revealed body mass index 36 kg/m^2. Her neurologic and routine laboratory examination was unremarkable. There were no obvious psychological problems or mood disturbances. The patient denied use of any medication. Otorhinolaryngologic examination revealed unrestricted mouth opening, normal dentition and Mallampati grade III airway. Fiberoptic examination of the upper airways with static and dynamic vocal cord evaluation were normal.

Polysomnography was carried out using a 16-channel digital polygraph (Embla A10©, Somnologica 3® software Flaga-Medcare, Reykjavik, Iceland). Sleep and respiratory events were recorded on videotape, and manually scored in 30-s epochs. Sleep stages were identified according to standard criteria (Rechtschaffen and Kales 1968) and the scoring of arousals was based on ad hoc guidelines of the American Sleep Disorders Association [8, 9].

Total sleep period was 377 min, sleep efficiency was 98%, and sleep onset latency was 44 min. There were six rapid eye movement (REM) cycles, and the proportions of the various sleep stages as percent of total sleep time were stage 1, 2.4%; stage 2, 48.3%; slow wave sleep, 32.2%; REM sleep, 17.1%. Sleep was fragmented by 84 arousals, 75 of which were associated with respiratory events, seven of which were associated with limb movements, and two were spontaneous. Apnea-hypopnea index (AHI) was 38 per hour of sleep. The expanded EEG montage demonstrated no epileptiform activity. Nocturnal PSG at presentation is shown in Fig. 1a. The groaning episodes started 3 min after falling asleep, with duration of about 2–10 s and total number of 2 to 33 in every cluster. The groaning occurred during expiration only and nine clusters of recurrent expiratory groaning were observed, three of which occurred in non-REM sleep stage 2, two in slow wave sleep and finally, two clusters occurred in REM sleep. Figure 2 demonstrates 120-s epoch of a polysomnographic segment showing two groaning episodes. Clusters were lasting with duration of about 2–8 min. Each event was characterized by a deep inspiration, followed by a short expiratory phase and then a long period where the breathing signals were significantly reduced. During these noise expiratory phases, decreased EMG activity in rectus abdominis and intercostalis muscles was detected. The body position seemed to have no influence.

Due to the presence of severe OSA on PSG, we repeated nocturnal PSG with manual nasal continuous positive airway pressure (nCPAP) titration. During the night, with use of nCPAP (11 mbar), AHI improved to 3.8 per hour of sleep and the groaning sound unexpectedly disappeared completely. Total sleep period was 329 min, sleep efficiency

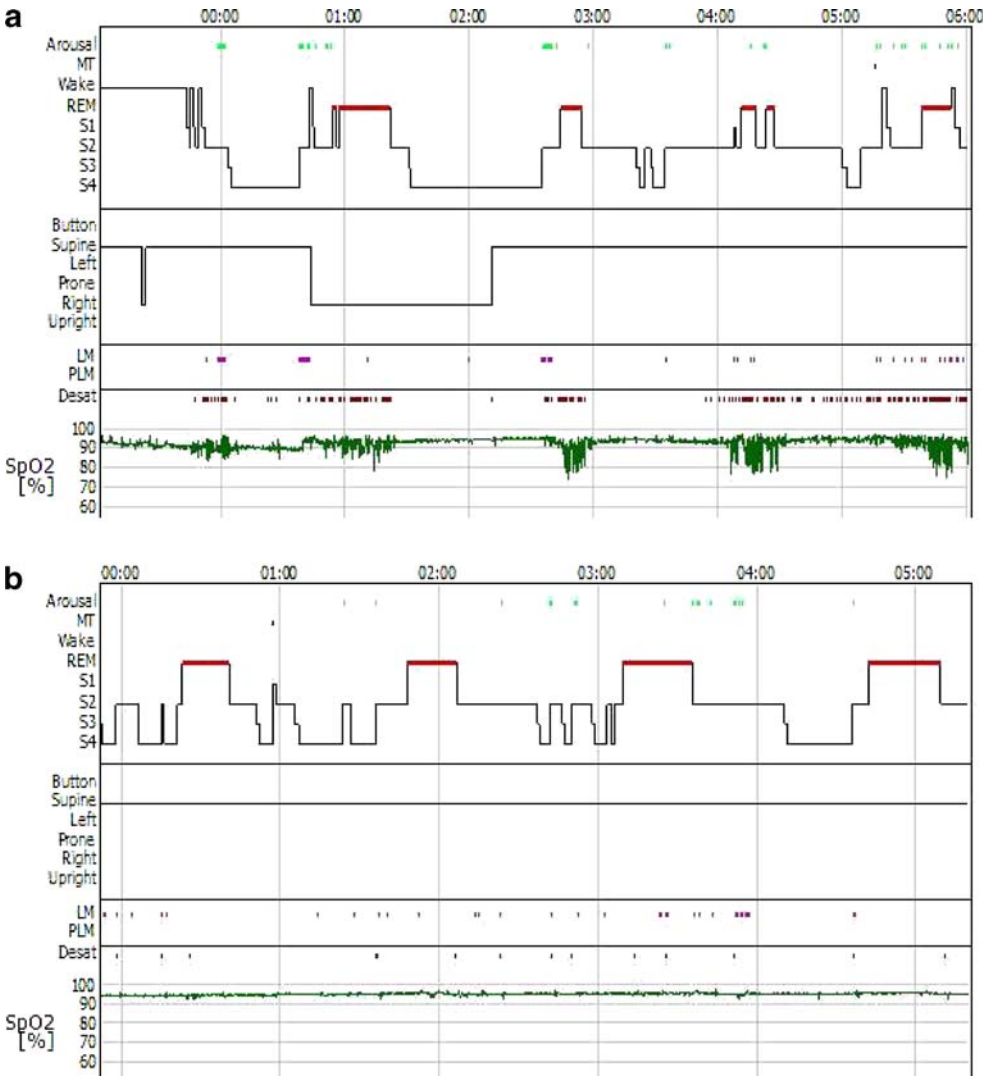

Fig. 1 **a** Nocturnal PSG at presentation. **b** Nocturnal PSG with CPAP titration

was 100%, and sleep onset latency was 2 min. There were four REM cycles, and the proportions of the various sleep stages as percent of total sleep time were stage 1, 0.5%; stage 2, 42.9%; slow wave sleep, 29.2%; REM sleep, 27.5%. Sleep was fragmented by 32 arousals, 11 of which were associated with respiratory events and 21 of which were associated with limb movements. Nocturnal PSG with CPAP titration is shown in Fig. 1b. She tolerated the nCPAP perfectly, and she was asymptomatic the next morning. Pharmacologic medication for pulmonary hypertension neither during her follow-up in the hospital nor after her discharge was administered. Seven days after starting nCPAP therapy, TTE was performed and the estimated PAPs using peak tricuspid regurgitant jet velocity was 31 mmHg, representing 20 mmHg of improvement. On follow-up, her family reported that she wore her nCPAP nightly, they did not hear any groaning, and the patient is experiencing less daytime sleepiness.

Discussion

Sleep-related groaning was first reported in the medical literature in 1983 when De Roeck and Van Hoof reported the case of a young male [1]. In 2001, Vetrugno et al. reported four additional patients with onset of the disorder but did not mention any treatment strategies [10]. In the same year, Pevernagie et al. reported ten patients of catathrenia, five with mild OSA and two with moderate OSA [11]. Empirical treatment with neither pharmacological (either dosulepine, trazodone, clonazepam or paroxetine were unsuccessful in eight patients) nor non-pharmacological approaches (nCPAP was inefficacious to two patients with mild OSA) was successful. Brunner and Gonzalez reported eight patients in 2004 without any mention of the applied therapy [12]. In 2005, Oldani et al. described 12 patients and reviewed the literature highlighting that no effective treatment is available [13]. Finally in 2006, Iriarte et al. demonstrated a patient of

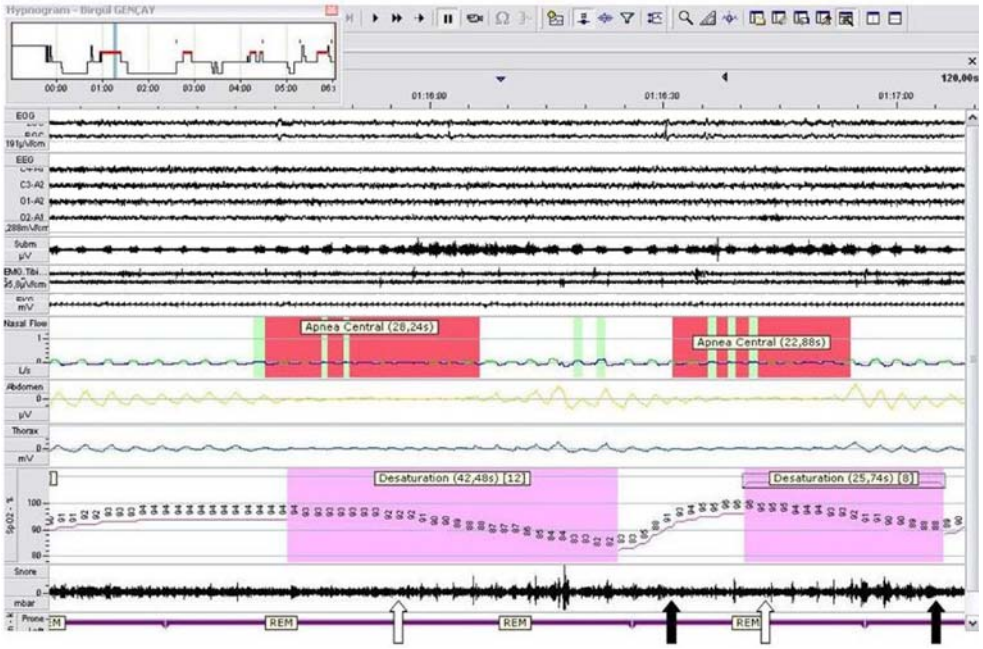

Fig. 2 Expiratory groans. A polysomnographic segment of 120-s epoch showing EEGs (C3-A2, O1-A2, C4-A1, O2-A1), left and right eye movements (LOC-A2; ROC-A1), chin, left and right tibialis EMGs, snoring, oronasal airflow, thoracic and abdominal effort channels, oxygen saturation. *White arrows* indicate where the groaning start and *black arrows* indicate where the groaning stop

catathrenia and concurrent moderate OSA. They applied nCPAP therapy, which effectively controlled groaning and snoring [14]. As of 2007, 36 patients (eight of them in abstract form) have been described in the medical literature with the term of catathrenia and eight of these patients were suffering mild or moderate OSA.

The origins of catathrenia remain inexplicable and the long-term prognosis unexplained. A possible mechanism of pathogenesis could be a functional REM sleep-related narrowing of upper airways during expiration but the wake laryngoscopic investigation cannot rule out a functional sleep-related obstruction of the upper airways during expiration. De Roeck and Van Hoof (1983) suggested three possibilities, including functional occlusion of the vocal chords during REM sleep with reactive forced expiration to overcome this resistance, critical narrowing of peripheral airways during expiration in REM sleep, or functional and/or anatomical lesions involving neurological structures that control ventilation [1]. Other authors (Vetrugno et al. 2001, oral presentation; Pevernagie et al. 2001; Brunner and Gonzalez, 2004) demonstrated the occurrence of groaning also during non-REM sleep, and excluded the association with identifiable neuroradiologic lesions [10–12]. Iriarte et al. mentioned an asynchronous activation of the diaphragm and the oropharyngeal muscles during REM sleep [14]. Ortega-Albas et al. suggest that post-inspiratory neurons, located between the dorsal and ventral respiratory groups, are active during the early expiratory phase and play a key role in breathing generation during sleep, are responsible for groaning [15].

Catathrenia often begins with a deep inhalation, followed by a short expiratory phase during which a typical monotonous sound is produced and then by a long expiratory period where the breathing signals are reduced, repetitively recurring during NREM sleep stages and REM sleep. Catathrenia reveals social and familial problems. Usually patients are all unaware, but typically, parents and bed-partners are troubled and alarmed by the nocturnal noise. The first symptoms usually appear during adolescence or early adulthood and in our case, the patient was exhibiting periods of loud groaning while asleep since age seven. The condition may be familial, but without any other neurological, psychiatric pulmonary, otolaryngologic disease or a history of alcohol or substance abuse.

In OSA, the apnea-associated triggers of multiple periodic alveolar hypoxia and intrathoracic pressure swings lead to repetitive rises of pulmonary artery pressure during sleep. The stimulus for PH is thought to be hypoxic pulmonary vasoconstriction and subsequent vascular remodeling [16]. It has been suggested that daytime hypoxemia, as seen in our patient, is important in the development of pulmonary hypertension and nocturnal hypoxemia (especially profound during REM sleep) in obstructive sleep apnea. The consequences of sleep-related hypoxemia include peaks of pulmonary hypertension due to hypoxic pulmonary vasoconstriction, generally observed in patients with marked daytime hypoxemia. Nasal CPAP leads to resolution of episodic nocturnal desaturation and rapid improvement in daytime hypoxemia. Also, effective nCPAP therapy has a beneficial influence on pulmonary

hemodynamics [17]. In the literature review, we have found neither a case report nor an article about the rapid resolution of the PH in 1 week. Therefore we need more experimental and non-clinical research studies on the resolution period of pulmonary hypertension.

In the previous reports, empirical pharmacological treatment of catathrenia with dosulepine, trazodone, clonazepam, paroxetine, carbamazepine gabapentin, and pramipexole has been unsuccessful or refused [11, 13]. nCPAP may be an option for the treatment of this disturbing condition. nCPAP has been reported to be ineffective in two patients with concurrent mild OSA (Pevernagie et al. 2001) but beneficial in one patient with concurrent moderate OSA (Iriarte et al. 2006) [11, 14].

Conclusion

When the literature is reviewed this is the first report of which catathrenia, OSA and pulmonary hypertension are observed as one. Furthermore, a successful treatment option is suggested for catathrenia for the second time in the literature: CPAP. Treatment with nocturnal CPAP, somewhat unexpectedly resulted in marked improvement of catathrenia, OSA, daytime dyspnea, and pulmonary hypertension for our patient. We think that CPAP can be an option for the treatment of this infrequent but sometimes very disturbing sleep disorder.

References

1. De Roek J, Van Hoof E, Cluydts R (1983) Sleep-related expiratory groaning. A case report. J Sleep Res 12:237
2. American Academy of Sleep Medicine (2005) International classification of sleep disorders, diagnostic and coding manual, 2nd edn. American Academy of Sleep Medicine, Westchester, IL, pp 165–167
3. Victor LD (1999) Obstructive sleep apnea. Am Fam Physician 60:2279–2286
4. Ogawa A, Emori T, Sumita W et al (2006) Continuous positive airway pressure ameliorated severe pulmonary hypertension associated with obstructive sleep apnea. Acta Med Okayama 60 (3):191–195
5. Sajkov D, Wang T, Saunders NA et al (1999) Daytime pulmonary hemodynamics in patients with obstructive sleep apnea without lung disease. Am J Respir Crit Care Med 159:1518–1526
6. Faller DV (1999) Endothelial cell responses to hypoxic stress. Clin Exp Pharmacol Physiol 26:74–84
7. Yock PG, Popp RL (1984) Noninvasive estimation of right ventricular systolic pressure by Doppler ultrasound in patients with tricuspid regurgitation. Circulation 70:657–662
8. Rechtschaffen A, Kales A (1968) A manual of standardized terminology, techniques, and scoring systems for sleep stages of human subjects. Brain Information/Brain Research Institute UCLA, Los Angeles
9. American Sleep Disorders Association—The Atlas Task Force (1992) EEG arousals: scoring rules and examples. Sleep 15:174–184
10. Vetrugno R et al (2001) Catathrenia (nocturnal groaning): a new type of parasomnia. Neurology 56(5):681–683
11. Pevernagie DA, Boon PA, Mariman ANN et al (2001) Vocalization during episodes of prolonged expiration: a parasomnia related to REM sleep. Sleep Med 2:19–30
12. Brunner DP, Gonzalez HL (2004) Catathrenia: a rare parasomnia with prolonged groaning during clusters of central or mixed apneas. J Sleep Res 13:107
13. Oldani A, Manconi M, Zucconi M et al (2005) 'Nocturnal groaning': just a sound or parasomnia? J Sleep Res 14:305–310
14. Iriarte J, Alegre M, Urrestarazu E et al (2006) Continuous positive pressure as treatment for catathrenia (nocturnal groaning). Neurology 66:609–610
15. Ortega-Albas JJ, Diaz JR, Serrano AL et al (2006) Continuous positive airway pressure as treatment for catathrenia (nocturnal groaning). Neurology 67(6):1103 author reply 1103
16. Presberg K, Dincer HE (2003) Pathophysiology of pulmonary hypertension due to lung disease. Curr Opin Pulm Med 9:131–138
17. Fletcher EC (1990) Chronic lung disease in the sleep apnea syndrome. Lung 168(Suppl):751–761