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Effect of CPAP therapy on Catathrenia and OSA: A Case Report

Authors
Dr Tabassum Saher[1], Dr Mohammad Anamul Haque[2], Dr Nilofar Rasheed[3]
[1]Assistant Professor, Faculty of Physiotherapy, SGT University, Gurugram
[2]Physiotherapist, Prince Sultan Military Medical City, Riyadh, Kingdom of Saudia Arabia
[3]Assistant Professor, School of Nursing and Health Sciences, NIU University, Noida, India

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 and the long-term prognosis unexplained.*
Case Report: *We report a case of Catathrenia with concurrent Obstructive Sleep Apnea complicated with pulmonary hypertension and treat the case with Continuous Positive Airway Pressure (CPAP).*
Discussion: *Treatment with Nasal CPAP resulted in marked improvement of Catathrenia obstructive sleep apnea, daytime dyspnea and respiratory failure for our patient. We think that nasal continuous positive airway pressure can be an option for the treatment of this case and more better outcomes on Dyspnea and respiratory failure are expected if done on large population with periodic follow up.*
Keywords: *Catathrenia, Nocturnal Groaning, Obstructive Sleep Apnea, Pulmonary Hypertension.*

Introduction

Catathrenia is a very rare, idiopathic, sleep related respiratory condition characterized by high-pitched, irregular groans, which occur during prolonged expiration in sleep. In 1983 Catathrenia was first reported in the medical literature in Edegem, Belgium 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, (4v2 +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 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 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 48-year-old male was followed-up yearly in the outpatient clinic of cardiology for 6 years. He underwent operation for ventricular septal defect at age ten. He did not have any complaints until last year and she had been receiving no medication since 5 years. He was admitted with a progressive dyspnea on exertion, which he had for the past year. Transthoracic Doppler echocardio-graphy 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 his systolic pulmonary arterial pressure was increased and symptoms (NYHA class II-III) occurred, he 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. 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 lab. The patient's history was detailed and was realized that, since age ten, he has exhibited periods of loud groaning while asleep, occurring several times every night and for the last 3 years his wife 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. He 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. He complained of restless sleep and tiredness during the daytime. Physical examination revealed body mass index 36 kg/m2 . His 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 groaning episodes started 5 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. 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 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. He tolerated the nCPAP perfectly, and he was asymptomatic the next morning. Pharmacologic medication for pulmonary hypertension neither during his follow-up in the hospital nor after his 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, his family reported that he wore his 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[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]. Catathrenia reveals social and familial problems. 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, otolaryngo-logic 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[13]. 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 [14]. In the previous reports, empirical pharmacological treatment of catathrenia with dosulepine, trazodone, clonazepam, paroxetine, carbamazepine gabapentin, and pramipexole has been unsuccessful or refused[11]. 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].

Conclusion

This is a first case report of which catathrenia, OSA and pulmonary hypertension are observed as one. Furthermore, a successful treatment option is suggested for catathrenia i.e. 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.

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