CASE REPORT

Sleep Science

A case of positional central sleep apnea due to compression of the left vertebral artery on brainstem

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ABSTRACT

Studies evaluating the association between Central Sleep Apnea (CSA) and positional sleep apnea are not commonly described and are barely understood. We report a case of a 51-year-old-male with moderate Obstructive Sleep Apnea (OSA) and severe CSA probably secondary to brainstem compression, which responded to the adoption of strict lateral body posture. The addition of Continuous Positive Airway Pressure (CPAP) optimally resolved the remaining obstructive respiratory events. We suggest including Magnetic Resonance Imaging (MRI) in the work-up plan of patients with positional CSA.

Keywords: Central Sleep Apnea; Continuous Positive Airway Pressure; Posture; Sleep-Disordered Breathing; Magnetic Resonance Imaging.

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INTRODUCTION

According to International Classification of Sleep Disorders Third Edition, CSA is defined as cessation in airflow of 10 or more seconds in the absence of any inspiratory effort, while positional sleep apnea is said to be present when there is a 50% reduction in the Apnea-Hypopnea Index (AHI) during non-supine sleep. The association between these conditions is poorly understood and not commonly reported[1]. We reported a case of a 51-year-old male with moderate OSA and severe CSA probably due to respiratory center compression.

REPORT OF CASE

A 51-year-old male presented to our sleep facility with history of snoring, sleep-breathing pauses and 4 kg weight gain over the past months. He denied excessive daytime sleepiness (Epworth Sleepiness Score 3/24), nightmares, dream-enacting behavior or nighttime awakenings. He reported occasional alcohol consumption and denied tobacco use or drugs. There was also occasional consumption of caffeinated beverages. The past medical history included stage-2 chronic renal disease secondary to focal segmental glomerulosclerosis and high blood pressure. Current medications included folic acid, allopurinol, amlodipine, atorvastatin, and losartan. Physical examination revealed a body mass index of 27.78 kg/m^2, neck perimeter of 42 cm, Mallampati class III, large tongue, high palate and non-congestive nasal mucosa. The cardiovascular and neurological examination was unremarkable. Blood pressure was 140/90mmHg and pulse was 98 bpm.

In order to rule out a case of Obstructive Sleep Apnea, a Polysomnography was conducted. The parameters of sleep architecture, as shown in Table 1, were found to be within normal ranges, but the diagnostic study concluded moderate Obstructive Sleep Apnea with an Obstructive Apnea Index (OAI) of 17.4, and Severe Central Sleep Apnea, with a Central Apnea Index (CAI) of 31.6. It was also noted that Respiratory Disturbance Index improved by adopting a non-supine posture from 103 versus 16.5 events per hour, and this improvement involved both obstructive and central events.

Due to the patient's Apnea-Hypopnea Index (AHI) and our laboratory protocol, we decided to perform a split-night polysomnography. Titration with CPAP was performed in both supine and non-supine positions, revealing resolution of obstructive respiratory events but minimal changes in CAI (24.9), as well as an exacerbation of central hypopneas (Table 2). Additionally, a short-term trial of Bilevel Positive Airway Pressure (BiPAP) did not add any improvement.

As part of the study for CSA, we solicited an echocardiography and pro-BNP, the latter with unremarkable results. Echocardiography concluded mild hypertrophy of basal septum and abnormal left ventricle relaxation, but normal size and motility of both ventricles and preserved ejection fraction of 74%. No pulmonary hypertension was disclosed. Contrast-enhanced brain MRI was also requested, revealing a discrete bulbar compression on the left side by the left vertebral artery, suggesting the impairment of respiratory center on brainstem as the cause of CSA (Figure 1). No other cerebral, spinal or cerebellar alterations were reported, cerebellum was located above foramen magnum and no visible Chiari malformations were found.

A second polysomnography was performed two months later to study posture as a therapeutic option due to the previous study results. We observed a substantial reduction in CAI (1.86) in the left lateral position and no central events in the right lateral position; however, obstructive events persisted in those postures in the form of hypopneas. Four weeks later, an auto-CPAP titration with a pressure ranging from 4 to 10 cm H_2O was completed over five days, simultaneous to Home Sleep Apnea Testing (HSAT) with a cardio-respiratory device (Polygraphy), with strict lateral body position. The results were satisfactory as seen in Table 2. The patient is now in continuous monitoring.

DISCUSSION

Brainstem damage by vascular compression as a cause of CSA has been described previously in a few cases, as a giant vertebrobasilar aneurysm or a calcified vertebral artery[2-4]. In our patient, an anatomic disturbance in the ventrolateral medulla,

Table 1. Parameters of Sleep Architecture of Split-Night Polysomnography Study.

	Diagnostic		CPAP Therapy	
Parameter	Duration (minutes)	% of TST	Duration (minutes)	% of TST
Total bedtime (TBT)	156.5	-	342	-
Total Sleep Time (TST)	133	-	272	-
Sleep Latency	2	-	0	-
Period of awake after sleep onset (WASO)	12.5	-	68.5	-
NREM	126.5	95%	230.5	85%
N1	7.5	6%	43.5	16%
N2	72.5	55%	180	66%
N3	46.5	35%	7	3%
REM	6.5	5%	41.5	15%
Sleep Eficiency	90%	-	80%	-

NREM: Non-rapid eye movement sleep, REM: Rapid eye movement sleep.
N1, N2, N3: Stages 1, 2 and 3 of sleep, respectively.

Table 2. Results of diagnostic and therapeutic polysomnography and follow-up Polygraphy.

	Split Night Study				Second Polysomnography: Positional Therapy **				HSAT (Auto-CPAP + Polygraphy) ***	
	Diagnostic		CPAP Therapy*		Left Side		Right Side			
	N°Events	Index	N°Events	Index	N°Events	Index	N°Events	Index	N°Events	Index
AHI	--	56.4	--	50.5	--	23	--	15	15	2.3
RDI	--	58.2	--	50.5	--	28.8	--	23.9	--	--
Obstructive Apnea	4	1.8	0	0	0	0	0	0	0	0
Central Apnea	70	31.6	112	24.7	8	1.8	0	0	7	1.1
Mixed Apnea	35	15.8	1	0.2	2	0.4	0	0	3	0.15
Hypopnea	15	6.8	106	23.4	81	18.8	46	13.8	5	0.8
RERAS	4	1.8	0	0	21	4.8	29	8.7	--	--
Total Apnea	109	49.2	113	24.9	8	1.8	0	0	10	1.5

AHI: Apnea-Hypopnea Index, RDI: Respiratory Disturbance Index, RERAs: Respiratory Effort Related Arousal, HSAT: Home-Sleep Apnea Testing.
* CPAP was titrated during first polysomnography study and reached a pressure of 16 cm H_2O
** Positional Therapy consisted on adopting strict left and right lateral positions during a second polysomnography study.
*** HSAT was performed during a five-day ambulatory titration of CPAP in strict lateral posture during sleep. The Polygraphy included the following sensors: nasal pressure, respiratory inductance plethysmography, body position sensor and pulse oxymetry.

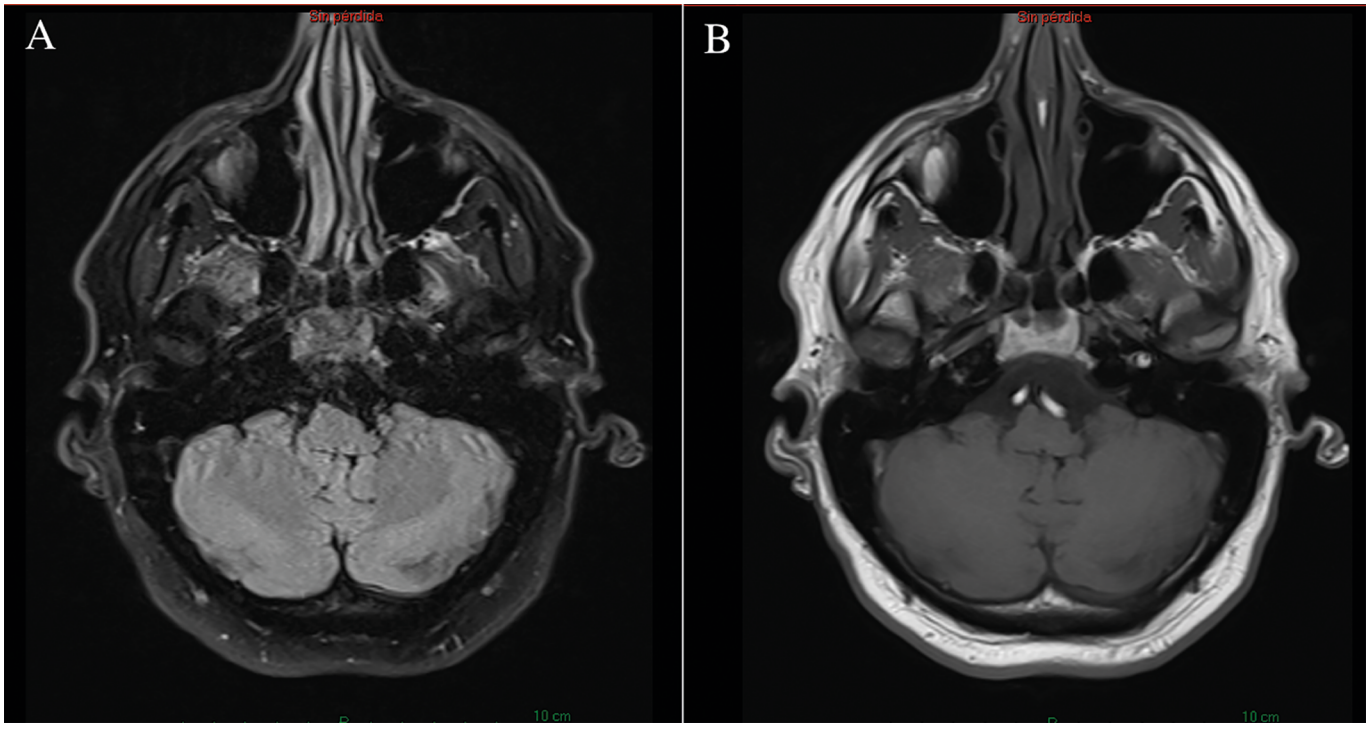

Figure 1. Brain MRI.

containing the Pre-Bötzinger complex, suggests that impairment of respiratory rhythm generation might be the cause of severe CSA[5].

This particular case of brainstem compression syndrome singularly presents as an isolated breathing disorder; unlike others more commonly described neurological disturbances: pyramidal tract signs, vertigo, dysphagia, sialorrhea, velar paresis, ataxia or tinnitus[6]. Additionally, the compression of the nucleus ambiguous may contribute to our patient's obstructive events due to an impact in the central nervous system regulation of pharyngeal tone during sleep[5].

Continuous Positive Airway Pressure is considered an initial option of therapy in patients with Congestive Heart Failure CSA and an option therapy for idiopathic CSA and end-stage renal disease CSA[7]. The rationale for this therapy in central apneas has not been entirely elucidated, but it may be related to the prevention of inhibitory reflex during airway narrowing and an increase in lung volume and O_2 stores[7].

In our patient, positive airway pressure was effective in reducing obstructive events in both the supine and non-supine positions, but paradoxically exacerbated central apneas and hypopneas; as opposed to a few reports that have shown improvement of central apneas by using CPAP and BiPAP in patients suffering from bulbar compression[2].

Sleep studies in this patient revealed a remarkable improvement in central respiratory events by adopting a lateral

posture, with slight predominance on the right side. Changes in CSA indexes by adopting a non-supine position have been described in Cheynes-Stokes Respiration (CSR) or Congestive Heart Failure. The probable mechanism involves changes in cardiac chamber dimensions, cardiac filling, cardiac output and other hemodynamic variables[7].

Although the patient's echocardiography described an abnormal left ventricle relaxation, which would suggest the presence of diastolic dysfunction, he does not met the criteria for Heart Failure. It is worth mentioning that diastolic dysfunction alone has not been associated with CSA[8]. However, this finding would suggest that the adequate response to postural control could also be influenced by these hemodynamic changes, as well as other mechanisms involving lung volume and chemoreceptor sensitivity[7]. The adoption of a lateral posture and an optimal response in sleep parameters have been described in other few cases of CSA[1,2].

Even though it could be speculated that the pressure generated by the left vertebral artery on the ventrolateral surface of the bulb might be mitigated in lateral postures due to gravity, studies evaluating changes in blood flow in vertebral arteries, regarding neck and head positions, have shown no remarkable differences[9]. Nonetheless, a mechanism by compression-induced ischemia cannot be ruled out. On the other hand, as reported by Watters et al., hypertension raises the risk for developing vertebrobasilar ectasia and brainstem compression[10].

Even though the brain MRI did not show any vessel dilatation, brain imaging could have missed a pulsatile intermittent compression caused by the vertebral artery. It is also worth mentioning that none of the drugs in the patient's treatment have reported adverse effects regarding sleep apneas, or any impairment in the respiratory drive.

In conclusion, combination therapy of positive airway pressure and strict body position resulted in an optimal response in the context of a patient with CSA and OSA, so we consider that surgical correction may be delayed unless these previous therapies fail or an additional neurological finding appears. Finally, we propose a MRI to be part of the diagnostic work-up to rule out any anatomic alteration of the brainstem in patients with positional CSA without cardiac involvement or CSR.

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