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Research Article

Bilateral Congenital Choanal Stenosis and Changes in Sleep: A Case Report

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Abstract

Congenital choanal stenosis as a risk factor for the syndrome of Upper Airway Resistance (UARS) is not well described. The report case aim is to analyze disorders of sleep microstructure in a patient with choanal stenosis and UARS.

Man, 22 years, mouth breathing, with diagnosis of allergic rhinitis, Attention Deficit Hyperactivity Disorder and insomnia. Adenoid face, tonsils 2 + / 4 by Brodsky Classification, Modified Mallampati 1, high-arched palate, no craniofacial deformities. Obese Grade 1, 40cm of neck circumference, 98cm of waist circumference. Nasal endoscopy with bilateral choanal stenosis without other malformations. Epworth Sleepiness Scale = 2. In Polysomnography (PSG), there were null AHI, 9/h of respiratory disturbance index, by elevated RERA index. No oxyhemoglobin desaturation, reduced sleep efficiency, reduced percentage of REM sleep. Increased arousal rate (16/h), nasal cannula with permanent flattening of the curve and Cyclic Alternating Pattern (CAP) in stage 2 non-REM sleep.

The congenital bilateral nasal stenosis undiagnosed in the neonatal period is relevant. The consequence of this adaptation to airflow limitation in the upper airway is noticed by the adenoid face, neuromuscular and cognitive changes. Nasal flow is not well defined as a risk factor for Obstrutive Sleep Apnea Disorders. Also it is not related to significant oxyhemoglobin desaturation, or apnea / hypopnea. However, it can improve CPAP adaptation. The microstructure of sleep shows arousals and CAP. The later is an event of cerebral electrical activity with periods of activation and inhibition during the second phase of non-REM sleep. It's a partial activation of the brain and indicates instability of sleep, being related to reduce quality of sleep and also insomnia. It's possible that the increased number of awakenings compromises REM quality and quantity, causing a possibly non-restorative sleep and sleep fragmentation.

CAP inclusion in AASM manual may increase PSG sensitivity and diagnosis neglected disorders.

Introduction

The congenital bilateral nasal stenosis is a rare disease that occurs in 1:5000 newborns, mainly in women. Congenital choanal stenosis as a risk factor for Upper Airway Resistance Syndrome (UARS) is not well described. The AASM scoring atlas since 2007 defines RERAs as events associated with evidence of increased respiratory effort (and/or flattening of inspiratory flow) and an arousal at event termination may not meet diagnostic criteria for apnea or hypopnea. The Respiratory Disturbance Index (RDI) was defined as all breathing events (apneas+hypopneas+RERAs) divided by total sleep time. Although RERAs and RDI are both scientifically validated, scoring them is optional. However, when you are titrating CPAP, it is recommended that those events (that you may or may not score) should be eliminated for the "ideal" pressure. This is contradictory.

The UARS physical examination shows nasal obstruction and nocturnal polysomnography does not show apneas or hyponeas as Obstructive Sleep Apnea Syndrome (OSAS), it shows periods of increase in respiratory effort, sleep fragmentation, RERAs and flattening respiratory curve, which indicates airflow limitation. It is possible to prevent long-term consequences, if we diagnose and treat upper airway resistance syndrome.

The aim of this case report is to illustrate a case of a mouth breathing patient with UARS, showing the microstructure of sleep disorders.

Materials and Methods

Man, 22 years old, mouth breathing since childhood, with allergic rhinitis in treatment with fluticasone for 2 years, had the diagnosis of Attention Deficit Hyperactivity Disorder, in use of methylphenidate 10mg twice a day and carbamazepine 200mg two tablets before sleep. Reports having difficulty initiating and maintaining sleep without the use of carbamazepine. He complains of chronic insomnia, fatigue and snoring. Epworth Sleepiness Scale = 2. Adenoid face, tonsils 2 + / 4 by Brodsky Classification, Modified Mallampati 1, high-arched palate, no craniofacial deformities.

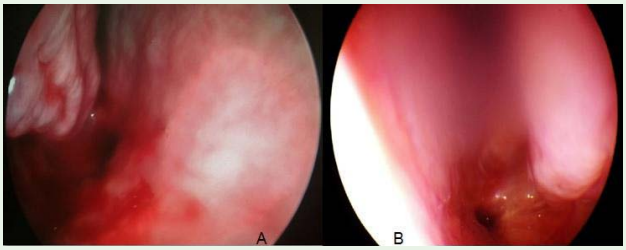

Figure 1: Endoscope view of choanal stenosis on the right (A) and on the left (B).

Obese Grade 1, 40cm of neck circumference, 98cm of waist circumference. Nasal endoscopy shows bilateral choanal stenosis (Figure 1 and 2) without other malformations.

Results

In Polysomnography (PSG), there were null AHI, 9/h of respiratory disturbance index, by elevated RERA index (Figure 3), no significant oxyhemoglobin desaturation, reduced sleep efficiency (Table 1) and reduced percentage of REM sleep (Table 2). Increased arousal rate (16/h), nasal cannula with permanent flattening of the curve and Cyclic Alternating Pattern (CAP) in stage 2 non-REM sleep.

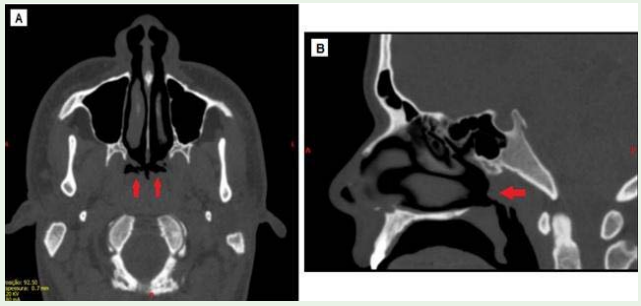

Figure 2: Axial (A) and Sagital (B) section of sinus computed tomography scan demonstrating bilateral membranous choanal stenosis (red arrow).

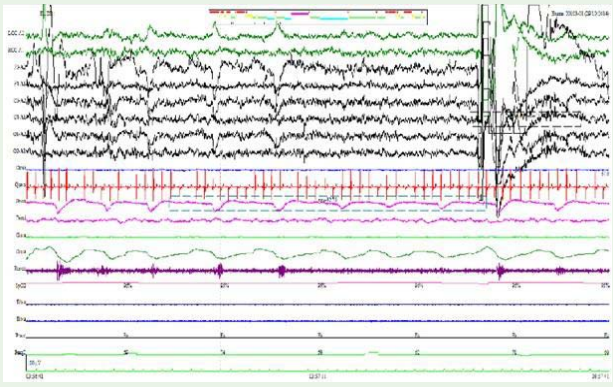

Figure 3: Respiratory effort- related arousal (RERA) in polysomnography.

Conclusion

The congenital bilateral nasal stenosis undiagnosed in the neonatal period is relevant. The consequence of this adaptation to airflow limitation in the upper airway is noticed by adenoid facies, neuromuscular and cognitive changes. Nasal flow is not defined as a risk factor for Obstructive Sleep Apnea Syndrome (OSAS) and it is not related to significant oxyhemoglobin desaturation, apnea or hypopnea. However, nasal flow can improve CPAP adaptation.

Table 1: Total sleep time percent in each stage. Total Sleep Time (TST) in minutes.

Sleep Stage	Percent of Tst in Each Stage	Simple Rate by Insomnia Brazilian Consensus
N1	15%	≤5%
N2	48%	50-55%
N3	26%	>15%
REM	11%	20-25%

Table 2: Sleep Parameters.

Sleep Parameters	Polissonography Result	Simple Rate by Insomnia Brazilian Consensus
Apnea + Hypopnea Index	1,93	≤5/hour
Hypopnea Index	1,93	
Respiratory disturbance index	9,4	
RERA Index	9	
Arousal Index	16	≤10/hour
PLMS Index	1,69	≤5/hour
Percent sleep efficiency	75,8%	>85%
Arterial oxygen saturation, mean value	96% (92 to 99%)	

Respiratory Effort-Related Arousal Index (RERA index; # of RERAs × 60 / TST); Respiratory Disturbance Index (RDI; (# apneas + # hypopneas + # RERAs) × 60 / TST); Arousal Index (ArI; number of arousals × 60 / TST); Percent sleep efficiency (TST / TRT × 100); PLMS Arousal Index [PLMSArI; Number of Periodic Limb Movements of Sleep (PLMS) with arousals × 60 / TST]; Total recording time (TRT; "lights out" to "lights on" in min); Total sleep time (TST; in min).

The AASM scoring manual since 2007 defines RERA and Respiratory Disturbance Index (RDI) (apneas+hypopneas+RERAs divided by total sleep time). Although RERA and RDI are both scientifically validated, scoring them is optional. However, for titrating CPAP, it is recommended that those events (that you may or may not score) should be eliminated for the "ideal" pressure.

We should be careful not to confuse the event (RERA) with

the syndrome (UARS). UARS was first described by Christian Guilleminault in children in 1982 and subsequently in adults. This syndrome is characterized not only by increased respiratory effort and airflow limitation during sleep associated with an increase in the upper airway resistance, but also by patients complains. They usually have daytime sleepiness, fatigue, snoring, difficulty to maintain sleep, cognitive impairment, anxiety and irritability. The physical examination shows nasal obstruction, increase in soft tissue and craniofacial abnormalities associated with decrease in the upper airway space. Nocturnal polysomnography shows periods of increase in respiratory effort, sleep fragmentation, RERAs and flattening respiratory curve, which indicates airflow limitation. It is important to prevent long-term consequences by UARS diagnosis and treatment.

Beside the microstructure of sleep shows arousals and CAP. The later is an event of cerebral electrical activity with periods of activation and inhibition during the second phase of non-REM sleep. It's a partial activation of the brain and indicates instability of sleep, being related to reduce quality of sleep and also insomnia. It's possible that the increased number of awakenings, compromises the quality and quantity of REM sleep, causing a possibly non-restorative sleep and sleep fragmentation. CAP inclusion in AASM manual may increase PSG sensitivity and diagnosis neglected disorders. More studies are necessary to define polysomnography scoring and patient management.

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