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Idiopathic bilateral abductor cord palsy-can it be the cause of obstructive sleep apnea?

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A B S T R A C T

We describe a case of 27 year old male who presented with history of breathing difficulty with episodes of apnea for 8 years. He was suspected to have obstructive sleep apnea and a nocturnal polysomnography showed Apnea-Hypopnea index of 34/hour and the lowest oxygen saturation of 78% and severe snoring in the night. Continuous positive airway pressure (CPAP) machine was advised from local hospital and he used for 2 month. Indirect laryngoscopy done showed bilateral abductor cord palsy. Contrast enhanced magnetic resonance imaging (CEMRI) of brain and contrast enhanced computed tomography (CECT) of chest and neck was normal study. Posterior cordotomy of left cord was done using coblation with resolution of symptoms of OSA.

Repeat nocturnal polysomnography showed apnea/hypopnea index 5 and lowest oxygen saturation of 95%.

1. Introduction

Bilateral vocal cord palsy (BVCP) could be associated with normal quality of voice and may have some impact on respiratory symptoms [1]. Bilateral vocal paralysis usually present with acute respiratory distress and stridor getting worse on exertion and require urgent surgical intervention but rarely they may present with chronic airway obstruction with history of nocturnal snoring and obstructive sleep apnea [2]. On searching the literature very few studies have described this association in the literature. In 1987, Ruff et al. [3] was the first to report a case of sleep apnea caused by bilateral vocal cord palsy with type I Chiari malformation but. Mc Brien et al. [4] reported case of Shy-Drager syndrome who presented with bilateral vocal cord palsy, nocturnal stridor and sleep apnea. Luaazy and Hasse ae al [5] described a patient with breathing difficulty, snoring and severe OSA caused by bilateral vocal cord palsy of unknown origin. Bonilla et al. [6] reported a case of OSA in a child with epiglottic aplasia. Sameer et al. [7] also reported a case of child with sleep apnea and growth retardation who had OSA caused by bilateral vocal cord palsy.

The cause of snoring and obstructive sleep apnea is believed to be caused by airway obstruction at the level of pharynx and anatomical obstruction caused by adenotonsillar enlargement, macroglossia, low position of the hyoid bone or excessive length of the soft palate are few of the causes [8]. However, in some situations, the noisy night time breathing may be produced by narrowing of the glottic structures [9] and Gillespie et al. in his study reported that laryngeal sleep apnea is similar to pharyngeal sleep apnea and may respond to nasal continuous positive airway pressure (CPAP) [10].

There are various causes for bilateral abductor cord paralysis, with iatrogenic injury post thyroidectomy surgery being the most common cause.[9]There are various treatment options for Bilateral abductor cord paralysis like arytenoidectomy, cordotomy, laterofixation of vocal fold, reinnervation procedures, botulinium toxin injection [11].

We hereby present a case of isolated bilateral abductor cord palsy as a cause for nocturnal stridor and obstructive sleep apnea was diagnosed to be caused by idiopathic bilateral abductor cord palsy that was surgically treated using coablation assisted posterior cordotomy (CAC).

2. Case report

A 27-year-old male presented with complaints of long-standing history of snoring associated with noisy breathing at night for the last 8 years. His friends noticed that he had apneic episodes at night along with mouth breathing. He had no symptoms of excessive daytime sleepiness or loss of concentration, early morning headache. In view of above complaints, he went to an ENT surgeon who advised him Polysomnography (PSG). A level 3 full-night polysomnography was done which showed Apnea-Hypopnea index of 34/hour and the lowest saturation of 78%. (Fig. 4). Hence, he was diagnosed to have Obstructive sleep apnea (OSA) and was advised to use Continuous positive airway pressure (CPAP) machine with pressure range at 8−11cm of water which he used for 2 months. His CPAP AHI was 3/hour bit in spite of using CPAP, patient had no relief and hence came to our Institute with main concern being noisy breathing at night.

His STOP BANG score was 4 and Epworth sleepiness score was 5 points. Nasal endoscopy showed no deviation of septum or spur with mild inferior turbinate hypertrophy. Clinical examination

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relieved grade 3 Friedman tongue position. Indirect laryngoscopy showed bilateral abductor cord palsy with cords in para median position with minimal flickering movement of left true vocal cord [Fig. 1]. Speech pathologist opinion was sought and perceptual voice analysis showed normal voice with maximum phonation duration of 26 seconds.

Contrast enhanced magnetic resonance imaging (CEMRI)of brain was normal, and contrast enhanced computed tomography (CECT) of chest and neck which showed glottic narrowing and no other significant finding [Fig. 2]. His thyroid hormone profile was eu-thyroid and his BMI was 22.3.

With no cause being found on imaging we diagnosed it as case of idiopathic bilateral abductor cord palsy.

Unilateral co ablation assisted right posterior cordotomy and arytenoidectomy was performed under general anaesthesia with preservation of inter arytenoids area. [Fig. 3]. Intra venous dexamethasone (10 mg) was given immediately after the completion of the operation and then 5 mg every 6 h for 2 days, along with systemic antibiotics and humidified oxygen to prevent postoperative airway edema.

Postoperatively patients snoring, sleep apnoea and breathing difficulties completely disappeared and his quality of sleep improved. His STOP BANG score post-operatively was 1 and Epsworth sleepiness score was 0 points. A nocturnal polysomnography was repeated after two weeks of surgery. His AHI score had decreased from 34 to 4.6 and oxygen desaturation index decreased from 44.3 to 5.1 and no of desaturations decreased from 244 to 45. (Fig. 4). He had no aspiration of swallowed foods but he complained of breathy voice and his maximum phonation time was 2–3 sec. His BMI was 22 postoperatively. He was referred for speech opinion. After 1 month of speech therapy his voice improved and his maximum phonation was improved to 9 sec.

3. Discussion

OSA is defined as a cessation of air flow for at least 10 seconds combined with a 4% or greater de saturation of arterial blood oxygenation during sleep as a result of an obstruction of the upper airway [10]. The prevalence of OSA is 24% among men and 9% among women [12]. The most frequent causes of OSA is the collapse of velo-pharynx and collapse at the level of base of tongue or hypopharynx occurring during sleep [8]. Very rarely congenital and acquired factors compromising the glottic airway can present with symptoms of obstructive sleep apnoea. Sleep-related laryngeal obstruction presenting as OSA has been reported in patients with laryngomalacia and vocal fold paralysis [6,7,9]. However role of larynx as contributory factor for OSA is usually overlooked by clinicians.

The mechanism of snoring in patients with bilateral vocal cord paralysis is similar to that caused by obstruction at the pharyngeal level and the only difference being the level of the flow limitation. In order to push the obstructed glottis, the patient creates a higher inspiratory pressure that leads to dynamic compression making inspiration increasingly difficult. The duration of inspiration becomes longer; air flow diminishes and with superimposed loss of tonicity of the tensor muscles causes OSA [13].

The most common causes of bilateral abductor vocal cord palsy include surgical trauma (44%), malignancies (17%), endotracheal intubation (15%), with neurologic causes and idiopathic cases accounting for 12% each [14]. The usual complaints of a patient with BVFP are respiratory distress which increases with exertion, normal voice and may sometimes cause sleep related laryngeal

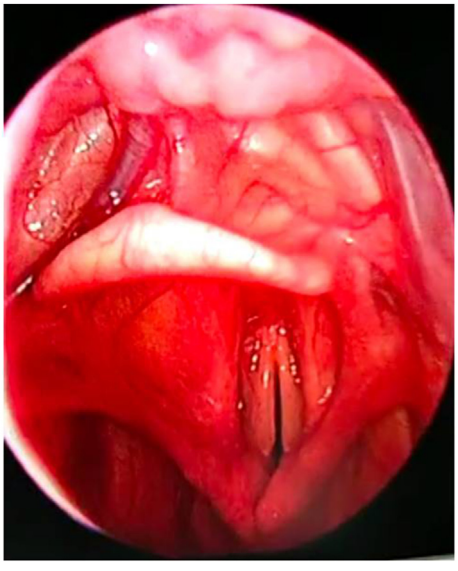

Fig. 1. Endoscopic picture showing bilateral vocal cords in paramedian position with minimal flickering of left vocal cord.

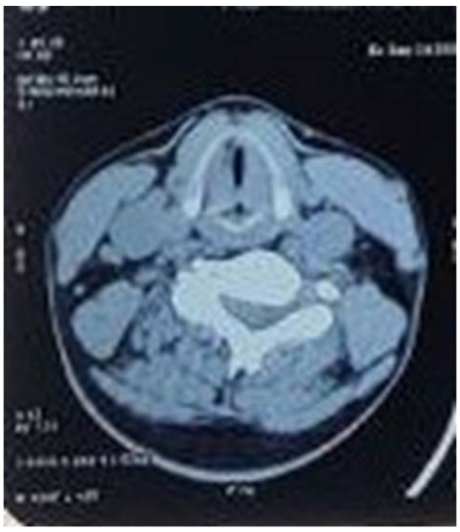

Fig. 2. CT neck showing bilateral cords in paramedian position with reduced glottis chink and no vocal cord lesion.

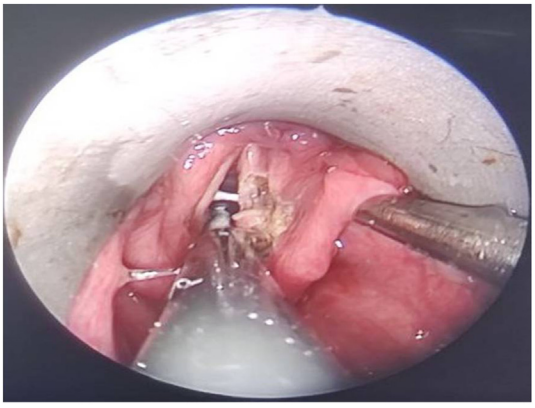

Fig. 3. Endoscopic picture showing posterior transverse cordotomy with artyenoidectomy of right vocal cord using coablation wand.

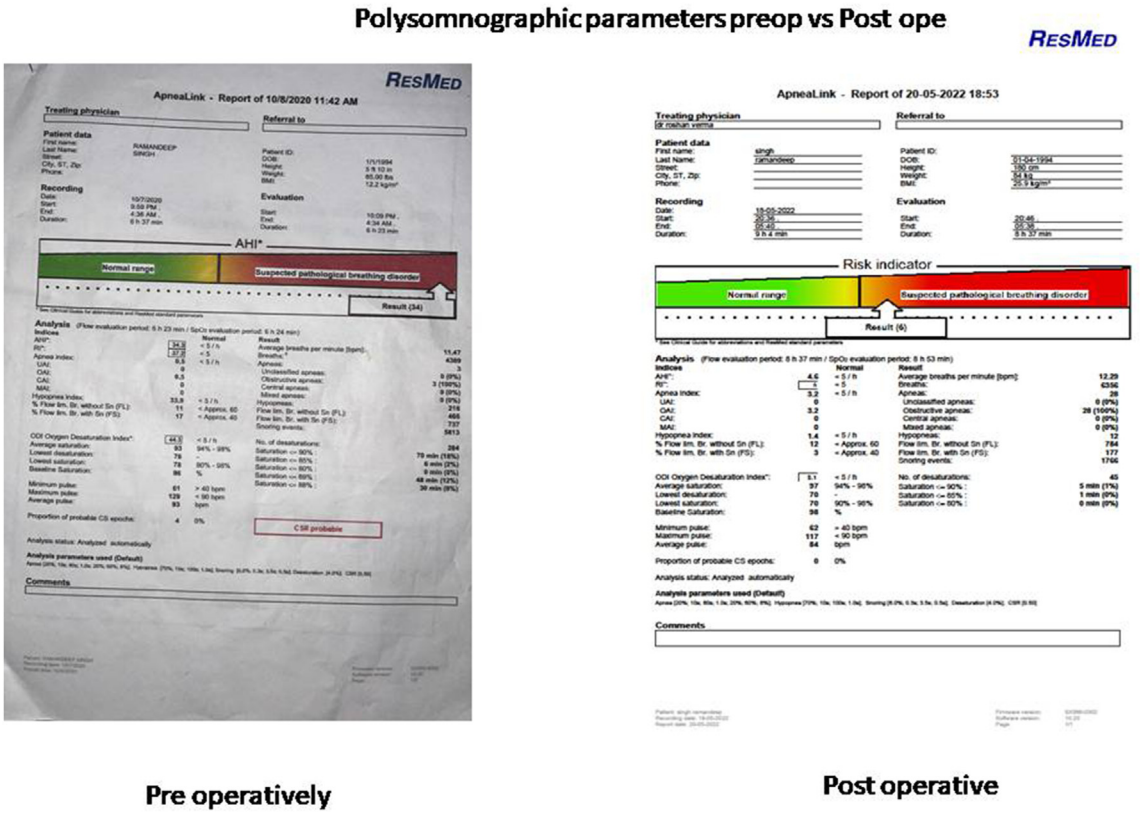

Fig. 4. Polysomnographic findings preoperative and post operative.

stridor and distress [14].

The management of vocal cord paralysis in association with OSA may include nasal continuous positive airway pressure (CPAP) or surgery. Surgical Options include tracheostomy, laterofixation of the vocal cord, Laser arytenoidectomy amd cordectomy and coablation assisted arytenoidectomy and posterior cordectomy. Newer approaches include re innervation techniques and botulinum toxin (Botox) injections into the vocal fold adductors, laryngeal pacing In our patient we performed coablation assisted arytenoidectomy and posterior cordectomy to widen the glottic aperture [15].

We performed coablation assisted arytenoidectomy and posterior cordectomy in our patient. A study by Hu et al. showed that 14 patients with bilateral vocal cord palsy were treated by coblation assisted arytenoidectomy [16]. Benninger et al. in his study concluded that coablation assisted cordectomy was a safe and efficient approach to treating bilateral vocal fold immobility [17].

As the laryngeal causes of OSA are rare, laryngeal examination is usually not done and all attention is focussed at the velopharyngeal and base of tongue level in sleep clinics. This report tends to highlight the importance of laryngeal examination in the sleep clinics and we emphasize that laryngeal examination should be performed routinely to evaluate the cause of sleep-related stridor.

4. Conclusion

This report tends to highlight the importance of laryngeal examination in the sleep clinics and we emphasize that laryngeal examination should be performed when CPAP pressure needs are high, or when the CPAP-AHI values still high in spite of good CPAP titration.

CRediT authorship contribution statement

Roshan Verma: Conceptualization, Conceptualized the paper, managed the patient, Writing – review & editing, edited the manuscript. **Reshma Raj:** Written initial paper, managed patient.

Declaration of competing interest

None to declare.

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