ORIGINAL ARTICLE

Paradoxical vocal cord motion with obstructive sleep apnea: An incompletely understood association

Ketaki Utpat, Vinaya S. Karkhanis, Jyotsna M Joshi

Department of Pulmonary Medicine, T.N. Medical College and B.Y.L. Nair Hospital, Mumbai Maharashtra, India.

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Abstract

Paradoxical vocal cord motion (PVCM) is a condition characterized by abnormal adduction of the vocal cords during inspiration. Diagnosis is often challenging owing to its heterogeneous presentation. Nocturnal symptoms are relatively uncommon. Associated comorbidities include gastroesophageal reflux disease or laryngopharyngeal reflux, allergic sinus disease,and possibly obstructive sleep apnea (OSA).We report two cases of PVCM presenting with predominant nocturnal symptoms owing to coexisting OSA.

Keywords: Paradoxical vocal cord motion, Obstructive sleep apnea

Introduction

Paradoxical vocal cord motion (PVCM) is an umbrella term used for a distinct entity that encompasses multiple conditions, which share the common phenomenon of abnormal inspiratory adduction of the vocal cords[1]. This results in a wide spectrum of symptoms occurring from the resultant upper airway occlusion including breathlessness, stridor, and cough. In view of its heterogeneous presentation, numerous terminologies have been used to describe this syndrome[2]. The current literature states that PVCM can occur in all age groups including the pediatric population[3]. The gold standard for diagnosis is direct visualization of the glottis and demonstration of the paradoxical movement of the cords.[4]Both psychological and nonpsychological factors have been implicated in the etiopathogenesis of this disorder. The association between obstructive sleep apnea (OSA) and PVCM has been postulated earlier; however, the exact relationship between the two has never been precisely described. We report two cases of PVCM sharing several common features including their predominant nocturnal symptoms and the striking association with OSA.

Address for correspondence
Dr. J. M. Joshi
Professor and Head
Department of Pulmonary Medicine
T.N. Medical College and B.Y.L. Nair Hospital
Mumbai 400008, India
Email:drjoshijm@gmail.com

Case Reports

Case 1

A 14-year-old boy, a student, presented with a 4-year history of noisy breathing predominantly at night and exertional dyspnea. He had received antiasthma medications for his symptoms in the past without any relief. He also reported history of heartburns and regurgitation suggestive of gastroesophageal reflux disease (GERD). His physical examination was unremarkable. His hemogram and serum biochemistry including serum IgE showed normal findings. Chest radiograph (CXR) was within normal limits. Spirometry showed obstructive abnormality with forced vital capacity (FVC) of 1.62 L (49%), forced expiratory volume in first second (FEV1) of 1.06 L (38%), and FEV1/FVC ratio of 66% with

bronchodilator reversibility of 70mL and 7%. Flow volume loops showed flattening of the inspiratory loop (Figure 1) with Empey's index of 6.9 and the ratio of maximum expiratory to inspiratory flow at 50% of forced vital capacity (FEF50/FIF50) of 1. Flexible fiberoptic bronchoscopy (FOB) revealed paradoxical motion of the vocal cords with inspiratory adduction and visible posterior chink. A detailed psychiatry evaluation revealed that the patient was experiencing significant level of stress related to his academic activities. His child assessment test(CAT) report stated that "the Father figure is seen as weak and insecure and there are feelings of rejection and conflict with the Mother Figure."In view of his predominant nocturnal symptoms, he underwent a detailed neurological examination and computed tomography (CT) of brain, which were within normal limits. Nocturnal polysomnography (PSG) showed significant apnea–hypopnea index (AHI) of 19.2/h, which was corrected to 1.2/h on titration with continuous positive airway pressure (CPAP) therapy. For his GERD symptoms, he was evaluated with esophageal manometry that demonstrated reflux esophageal. Hence, the patient was treated with tablet pantoprazole (40mg once daily), CPAP therapy, and speech therapy. Psychological counseling was offered to the patient and his parents.

Case 2

A 11-year-old boy, a student, presented with 2-year history of noisy breathing predominantly at night, exertional dyspnea, and symptoms of allergic rhinosinusitis.His physical examination was unremarkable. Hemogram and serum biochemistry showed normal findings. Serum IgE was 2111 IU/mL (normal range, 200 IU/mL). CXR was within normal limits. His psychiatry evaluation was normal. Spirometry showed obstructive abnormality with

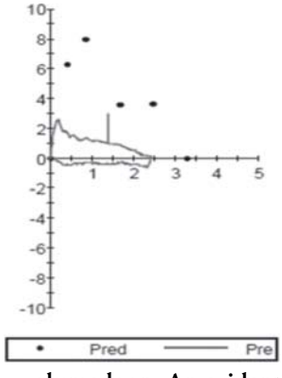

Figure 1: Flow volume loop. An evidentflattening of the inspiratory curve and expiratory curve is seen with a box-shaped pattern in case 1

FVC of 1.69 L (75%), FEV1 of 1.09 L (58%),and FEV1/FVC ratio of 64% with bronchodilator reversibility of 0%. Flow volume loop showed flattening of the inspiratory loopwith Empey's index of 10.44 and the FEF50/FIF50 ratio of 5, which confirmed PVCM occurring predominantly in the inspiratory phase of respiration. Flexible FOB revealed paradoxical motion of the vocal cords with inspiratory adduction and visible posterior chink (Figure 2). Neurological examination and CT of the brain were within normal limits. Nocturnal PSG showed AHI of 51.7/h, which was corrected to 2.4/h on titration with CPAP therapy. He was evaluated with esophageal manometry keeping in mind the possibility of silent GERD/laryngopharyngeal reflux (LPR), which, however, showed normal findings. Hence, the patient was treated with CPAP therapy and speech therapy along with combination of tablet montelukast (10mg) and tablet levocetirizine (5mg once daily)for treatment of allergic rhinosinusitis.

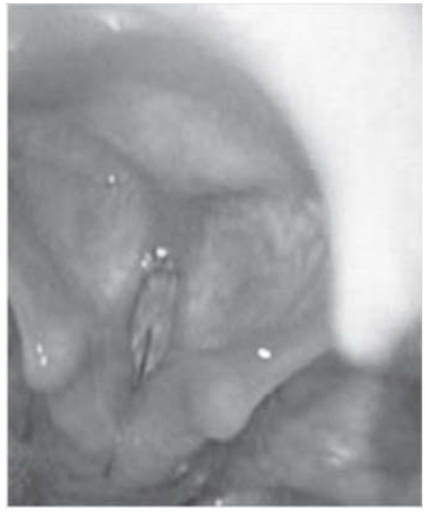

Figure 2: Inspiratory adduction of the anterior portion of the vocal folds with formation of a posterior cleft in case 2

Discussion

PVCM is a syndrome characterized by paradoxical adduction of the vocal cords during inspiration leading to paroxysms of glottis obstruction clinically manifesting as dyspnoea and noisy breathing[1]. Numerous labels have been used to describe this entity including PVCM, vocal cord dysfunction, irritable larynx syndrome, Munchausen's stridor, pseudoasthma, functional upper airway obstruction (UAO), and spasmodic croup. In

understanding the causative factors of this condition, focus has broadened beyond purely psychological factors to include a spectrum of disorders causing laryngeal irritation and, thereby, ultimately leading to paradoxical cord motion. Hence, the terminology "PVCM" can be said to be more appropriate than rest.[2] There is paucity of literature as far as the exact epidemiology of PVCM is concerned. Although, traditionally, PVCM has been reported more commonly in young women in the age group of 20 to 40 years, the current literature clarifies that it can occur in all age groups including children and adolescents[3]. The salient criteria for diagnosis include (1) clinical symptoms of dyspnea or noisy breathing, (2) direct visualization of inspiratory adduction of vocal cords, and (3) abnormal flow volume loop most commonly involving the inspiratory limb[4]. Our patients satisfied all the aforementioned diagnostic criteria.

The basic underlying pathophysiology is an accentuation of the glottic closure reflex. Various theories have been postulated for the same including a hyperresponsive larynx and an altered autonomic nervous system balance triggered by either an extrinsic or intrinsic stimulant[5]. Etiology of PVCM includes organic causes such as Arnold–Chiari malformation, cerebral aqueductal stenosis, motor neuron diseases, myotonic disorders, and laryngeal breathing disorders and nonorganic (functional) causes such as GERD, LPR, rhinitis, psychological causes, and environmental irritants.[6] Each factor may act separately or in association with other factors. PVCM commonly manifests in children and adolescents in the background of psychosocial stress[7]. These patients may exhibit peculiar traits such as high-performance standards and low-tolerance threshold for failures and may be victims of excessive parental pressures to succeed. Chronic GERD is also a common trigger for laryngeal irritation. It leads to retrograde movement of acidic gastric contents into the larynx and pharynx, resulting in irritation of cords. Similarly, allergic rhinitis and sinusitis lead to chronic postnasal drip, thereby leading to laryngeal hypersensitivity and triggering of glottic closure reflex. In pediatric population, the cause of PVCM has been more closely linked to GERD and allergic rhinitis. In our first patient, psychological stress and GERD were the contributing factors, which were suspected in the preliminary history and confirmed on a detail psychiatric evaluation and esophageal manometry, respectively. In the second patient, allergic rhinosinusitis was the precipitant as identified by the patient's symptoms and confirmed by a very significant serum IgE levels. Thus, both our patients are examples of PVCM in the pediatric age group triggered by commonly encountered precipitants in this age group.

Our patients, however, revealed a very unique presentation of their disorder in the form of predominant nocturnal symptoms, which is a very uncommon phenomenon in PVCM. Nocturnal stridor in PVCM may occur with organic neurological causes such as Chiari malformations, cerebrovascular accidents, meningomyelocele, multiple system atrophy, and autonomic dysfunction disorders[1,8]. However, these were systematically excluded in our patients by a detailed clinical and radiological assessment. On the other hand, our patients were diagnosed to reveal OSA by nocturnal PSG showing very significant AHI. This provided an explanation for the predominant nocturnal symptoms in the absence of any organic causes. It has been speculated that OSA and PVCM may be associated,[9] but the exact relationship between the two has not been described in literature till date. However, the definite link between the two entities can be deduced from various indirect facts. In majority of patients with OSA, the UAO during sleep can be attributed to the increased pharyngeal collapsibility. However, studies have shown that paradoxical glottic narrowing as seen in PVCM may contribute to UAO and OSA irrespective of the pharyngeal collapsibility.[10] This hints toward association of PVCM and OSA. Moreover, CPAP therapy, which is the prime treatment in OSA, has been described to be effective in PVCM as well.[2]This can be said to be an indirect evidence of the link between the two entities. So, our case reports can be summarized as examples of PVCM in pediatric population triggered by common intrinsic precipitants, namely, psychological stress, GERD, and allergic rhinosinusitis with OSA as the rare associated comorbidity, thereby leading to the rare nocturnal symptomatology. Management of PVCM involves a multidisciplinary approach with speech therapy and psychological counseling forming the cornerstones[11]. The treatment of coexisting comorbid conditions such as GERD, allergic rhinitis, asthma, and OSA is equally important. Hence, our patients were managed with speech therapy and psychological counseling, and the comorbidities in the form of GERD, allergic rhinitis, and OSA were addressed with antireflux, antiallergy, and CPAP therapy, respectively, to take care of all the components of the disorder.

PVCM should be considered in the differential diagnosis of dyspnea and noisy breathing in all age groups including the pediatric population. Diagnosis of this condition requires a high index of suspicion as the presentation is variable. Workup should involve an individualized but holistic approach, and all the potential contributing factors should be identified and addressed appropriately. We also hereby propose that the link between OSA and PVCM is an incompletely explored area of fertile research. Hence, OSA as a comorbidity of PVCM should be scheduled as a topic of interest in the future research trials.

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