

# A case of vocal cord dysfunction in the emergency department

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## Abstract

We describe the case of a 78-year-old woman admitted to our emergency department for an acute onset of severe dyspnoea with inspiratory wheezing-like sounds. She denied fever, cough, voice change and pain. She referred a similar but less severe episode occurred spontaneously one year before, with complete resolution in few minutes without sequelae. On examination upper airway

obstruction was firstly excluded. She was initially treated as having asthma, without response. Parenteral high dose corticosteroids and antihistamines provided no benefit. Point-of-care-ultrasound resulted normal. Flexible laryngoscopy during the episode showed paradoxical vocal cord movement with adduction during both inspiration and expiration. This demonstrated that her dyspnoea was from Vocal Cord Dysfunction (VCD). VCD completely solved after administration of intravenous benzodiazepines.

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## Case Report

A 78-year-old woman presented to our emergency department for an acute onset at rest of severe dyspnoea with inspiratory wheezing-like sounds. She was affected by anxiety and depression, blood hypertension and dyslipidaemia. She denied smoking, allergies, asthma, cough, fever, and voice changes. She reported a similar episode few years ago but less severe with complete resolution at home in few minutes without sequelae. Further investigations were not planned with her general practitioner in absence of recurrent episodes. On admission she was afebrile, displaying no skin rashes. Blood pressure, heart rate, peripheral oxygen saturation and respiratory rate were 155/88 mmHg, 100 bpm, 98% while breathing in room ambient air, and 18 breaths/min, respectively. Upper airway obstruction was immediately excluded: both the tongue and the uvula were completely normal. Heart sounds and chest exam were normal. Electrocardiogram showed sinus tachycardia. Point-of-care ultrasonography and chest X-ray resulted normal. She was initially treated as having asthma, without response. Parenteral high dose corticosteroids and antihistamines provided no benefit. Flexible laryngoscopy during the episode showed paradoxical vocal cord movement with adduction during both inspiration and expiration, excluding oedema and signs of inflammation or infections (see Video 1). This demonstrated that her dyspnoea was from Vocal Cord Dysfunction (VCD). A brain CT scan was performed, and it excluded cerebral lesions or masses. A neurological evaluation performed during the episode excluded impairments of the cranial nerves and demonstrated a progressive improvement of the phonation with exercise. Laboratory findings, including full blood counts, renal and hepatic function, serum electrolytes, coagulation time and C-reactive protein were all within the normal range. A RT-PCR nasopharyngeal swab for SARS-CoV-2 resulted negative. VCD completely solved after administration of intravenous benzodiazepines (delorazepam 2 mg). The patient was discharged after 48 hours of observation in good clinical condition, completely asymptomatic, with a diagnosis of “spontaneously occurring VCD”.

## Discussion

VCD is a functional cause of respiratory distress. Firstly described by Dunglison in 1842 as “hysterical croup”,<sup>1</sup> and later termed as “Munchausen’s stridor”,<sup>2</sup> it is now recognized as a clinical disorder.<sup>3</sup> VCD is a respiratory condition characterized by paradoxical adduction of the vocal cords during the inspiratory phase of the respiratory cycle with resultant airflow limitation at the level of the larynx, that can cause sudden onset of inspiratory stridor and dyspnoea in the absence of identifiable neurological abnormality.<sup>4</sup> This is most likely caused by a laryngeal hyperreactivity due to and increased sensitivity of the laryngeal sensory receptors and exaggerated glottic closure and cough reflexes following a number of triggers, including upper respiratory infections, occupational exposures, talking, laughing, singing, acid reflux, cough, foods, physical exertion, exercise, post-nasal drip, weather changes, emotional stressor, odours, strong scents and irritants,<sup>5,6</sup> that must be investigated in all the patients.

The prevalence is unknown because of under- and overdiagnosis, but the exertional VCD seems to be the most common phenotype, triggered by maximal athletic activity or routine exercise, and presenting as dyspnoea on exertion associated with inspiratory stridor.<sup>7</sup>

The diagnosis should be based on a careful clinical history, a physical examination, and a direct laryngoscopy. Performing a laryngoscopy in symptomatic patients is rapid, safe, and informative in most of the cases. Patients with exertional VCD have paradoxical vocal cord adduction on inspiration while abduction during expiration is preserved. In the other cases, patients with VCD have adduction during both inspiration and expiration. These patients can present an acute onset of severe respiratory symptoms that can result in emergency intervention with endotracheal intubation or tracheotomy.<sup>8</sup> A minority of patients (11%) may have expiratory VCD with only the expiratory adduction, as reported in the case series of VCD by Newman *et al.*<sup>9</sup>

Unfortunately the diagnosis of VCD can be difficult and problematic, since a definitive diagnosis of VCD is made only by visualization of adducted cords during an acute episode using a flexible nasopharyngeal fiber-optic laryngoscopy, but it is extremely difficult to reproduce an attack in a controlled setting. For these reasons, VCD can be misdiagnosed with other causes of upper airway obstruction and asthma.<sup>4,10</sup> Nasopharyngeal fiber-optic laryngoscopy and spirometry are useful in the differential diagnosis to avoid unnecessary treatment and iatrogenic morbidity.

VCD has also been seen in patients with Chiari 1 malformations. In a review of 31 young children with Chiari 1 malformations, VCD was seen in 26%, all under the age of 3 years.<sup>11</sup> Since VCD is atypical in children, an alternative diagnosis must be always considered. In these patients head magnetic resonance imaging demonstrates the herniation of the cerebellar tonsil below the foramen magnum consistent with a Chiari 1 malformation, and decompressive neurosurgery solves the respiratory symptoms. Other alternative diagnosis can include laryngomalacia,<sup>12</sup> subglottic stenosis,<sup>3</sup> vocal cord paresis or paralysis.<sup>13</sup> In adults, but not in children, chronic refractory cough and laryngopharyngeal reflux have been described in association with paradoxical vocal fold movement disorder.<sup>14</sup> In these patients respiratory retraining therapy may represent an effective therapy for cough in the absence of relief from standard management of laryngopharyngeal reflux.

Considering the pathophysiology of VCD, a strong correlation with underlying psychiatric disorders has been reported<sup>15,16</sup> underlying the importance of a psychiatric consultation for further ther-

apy and consideration of underlying somatoform disorders. Stress and emotions, as well as times of increased panic or anxiety, have been suggested as triggers for VCD.<sup>14,17</sup>

Once the diagnosis of VCD is confirmed, the first step is to reassure patients that the condition is benign and self-limited. Exertional VCD can be prevented by an anticholinergic aerosol administered prior to exercise.<sup>3</sup> In these patients symptoms generally subsided with a decrease or cessation of exercise. While the use of medications can be attempted, effective long-term therapy requires in all the patients with VCD psychological treatments, including speech therapy,<sup>18</sup> relaxation, biofeedback,<sup>19</sup> hypnosis<sup>20</sup> and breathing techniques are interventions for VCD proposed to control symptoms and reduce relapses. A psychiatric consultation for consideration of underlying somatoform disorders and appropriate therapy should be considered.

Asthma medications, such as inhaled bronchodilators and corticosteroids, should be used only if the diagnosis is unclear, but patients may have minimal response to them. Sedation with benzodiazepines can be used successfully, particularly in patients with anxiety,<sup>21</sup> as reported in our case. A recent systematic literature review by Slinger *et al.* no longer recommends the use of Heliox even the clear clinical rationale, in absence of strong evidence relating to the effectiveness of Heliox for VCD.<sup>22</sup> Recently, Fowler *et al.* have proposed a 12-item questionnaire, called the VCDQ, as a valid tool for symptom monitoring in patients with VCD.<sup>23</sup> This scoring system must be studied in large populations, but it can be reasonably considered a novel way to assess severity of disease and monitor response to therapy in patients with VCD. As reported in a systematic review by Mahoney *et al.*, glottic airway and respiratory retraining, botulinum toxin injections, low-dose amitriptyline and inspiratory muscle strength training devices have been associated with symptom reduction in adults and adolescents with VCD, but limited objective data exist to support the effectiveness of these interventions, and robust controlled trials are needed in this area.<sup>24</sup>

## Conclusions

VCD is a debilitating condition that affects both the general health and the quality of life of the patients. VCD often presents as stridorous breathing, which may be misdiagnosed as asthma, leading to unnecessary treatment and iatrogenic morbidity. Despite the typically benign and self-limited episodes, VCD can lead to severe symptoms, the impression of impending respiratory failure, and even emergent intubation or tracheostomy. Definitive diagnosis of VCD is made by the visualization of adducted cords during an acute episode using a flexible transnasal fiber-optic laryngoscopy. An association with psychogenic factors has been reported, and a higher incidence of anxiety-related illness has been demonstrated in patients with VCD. Optimal management of a patient with VCD should always require a multidisciplinary approach involving several specialists, *i.e.*, the otolaryngologist, pulmonologist, allergist, gastroenterologist, neurologist, speech pathologist, and psychiatrist or psychologist.<sup>25</sup>

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