

Case Report

Vocal cord dysfunction in three children - misdiagnosis of bronchial asthma?

Niggemann B, Paul K, Keitzer R, Wahn U. Vocal cord dysfunction in three children - misdiagnosis of bronchial asthma?
Pediatr Allergy Immunol 1998; 9: 97–100. © Munksgaard 1998.

Vocal cord dysfunction (VCD) is a paradoxical function of the vocal cords, leading to intermittent predominantly inspiratory dyspnea, but with no response to bronchodilator and anti-inflammatory drug therapy. We report on three children with VCD: 1) A 12-year old boy, who was treated for many years for bronchial asthma and who presented with inspiratory dyspnea and a functional reduction of the inspiratory and expiratory flow-volume curve, 2) a 13-year old girl who was also treated for bronchial asthma on a long-term basis and in whom the paradoxical vocal cord movement could be demonstrated by laryngoscopy, and 3) a 17-year old girl who, besides clinical symptoms of bronchial asthma in her anamnesis, suffered from an intermittent severe inspiratory dyspnea, refractory to bronchodilator treatment. Laryngoscopy proved the diagnosis of VCD. No patient showed a deterioration on discontinuation of their antiasthmatic therapy. VCD is best diagnosed by assessment of the vocal cords during laryngoscopy. The following therapeutic measures are helpful: 1) Demonstration of diagnosis (e.g. videodocumentation of laryngoscopy) and reassurance of patients and parents, 2) speech therapy, and 3) psychological intervention and/or psychotherapy. Our three cases point to a differential diagnosis of recurrent dyspnea in children and adolescents which may be overlooked. It is important to question earlier diagnoses, and to objectively evaluate the type of dyspnea.

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Key words: adolescent; asthma; children; vocal cord dysfunction

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Accepted 25 March 1998

Introduction

During normal and quiet breathing, inspiration is accompanied by widening of the glottis, whereas during expiration there is a narrowing of the vocal chink by adduction of the vocal cords. In recent years, a number of cases have been reported of a paradoxical function of the vocal cords leading to dyspnea in adults (1, 2, 3, 4), in children/adolescents (5, 6, 7, 8, 9, 10, 11) and even in infants (12, 13). This is now called Vocal Cord Dysfunction (VCD). We report on three children with a vocal cord dysfunction who had all been treated previously for bronchial asthma.

Case reports

Case 1

A 12-year old corpulent boy (with a history of several croup-episodes) was treated for many years for

bronchial asthma with cromolyn, beta-adrenergic substances, theophylline, adrenaline, and inhaled steroids. No relatives had allergic asthma, and he was found not to be sensitized to common inhaled or food allergens by means of specific IgE in serum and Skin Prick Test.

From time to time he presented with intermittent inspiratory dyspnea. Multiple lung function tests showed variable reductions of the inspiratory and expiratory loop of the flow-volume-curve next to completely normal findings (Fig. 1). A closer look into the patients records from several years ago revealed that there has been already an inspiratory and expiratory stridor. After a negative non-specific provocation with histamine, his antiasthmatic medication was discontinued. No deterioration was noted, lung function tests remained uneventful. Unfortunately, no laryngoscopy could be performed during an episode. Psychological therapy was initiated because evaluation by a psychologist revealed severe school anxiety.

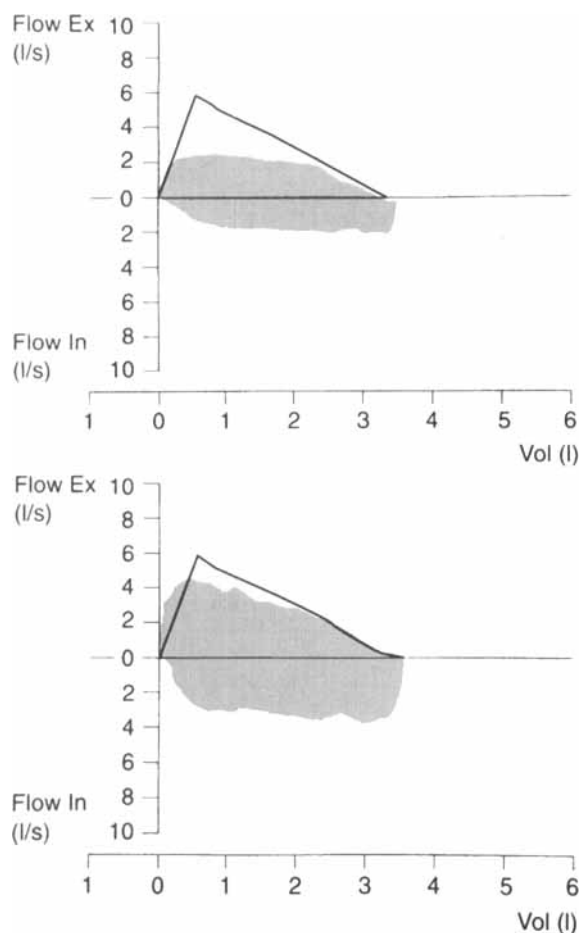


Fig. 1. Flow-volume-curve of patient 1 with (A) inspiratory and expiratory flow limitation during an attack and (B) normal flow-volume curve during interval. Black line represents predicted normal expiratory curve.

Case 2

This 13-year old girl was also treated for many years for bronchial asthma with cromolyn, salbutamol and inhaled steroids. Allergy tests showed no atopic disposition. Lung function tests in symptom-free intervals were normal. In-patient observation in our clinic showed the predominantly inspiratory character of her dyspnea. During one of these episodes, a laryngoscopy performed by a flexible bronchoscope showed a paradoxical function of her vocal cords. The antiasthmatic therapy was discontinued without any problems.

Case 3

Besides a history of clinical symptoms of asthma, the 17-year old girl suffered from an intermittent, severe inspiratory dyspnea which was refractory to bronchodilator treatment. In the lung function testing, we found a variable, sometimes combined ob-

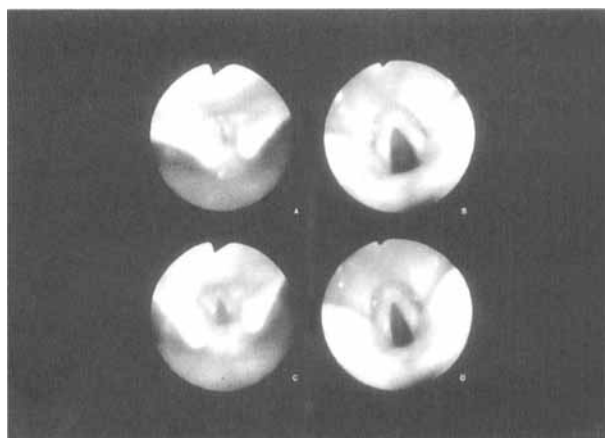


Fig. 2. Laryngoscopy of patient 2 during an episode: (A) Inspiration with almost complete closure of vocal cords and (B) Expiration with slight widening of vocal cords. Normal situation after sedation, with (C) wide vocal chink during inspiration, (D) a slightly narrower vocal chink during expiration.

structive ventilatory disorder of the upper and lower airways. Non-specific bronchial provocation test with histamine did not show signs of a significant bronchial hyperreactivity ($PC_{20} FEV_{1} \geq 8 \text{ mg/ml}$ histamine). Allergy tests remained negative.

Especially impressive was the complete lack of response to bronchodilator and adrenaline treatment during dyspnea attacks. However, an intravenous application of 2.5 mg midazolam brought immediate and complete remission of her dramatic symptoms within a few minutes. A laryngo-bronchoscopy in a phase of mild symptoms led to the diagnosis of VCD (Fig. 2). This patient also presented other non-specific symptoms including some of gastrointestinal origin, and was treated in the psychosomatic department for many weeks. All symptoms disappeared completely.

Discussion

Severe dyspnea may have several origins. We report on three children with an anamnesis of bronchial asthma which turned out to be VCD. One triggerfactor for eliciting symptoms of VCD is psychological stress, e.g. anxiety over an impending parental divorce, parental-child conflict, anxiety about parental expectations of scholastic performance (7). Other eliciting factors include physical exercise and hyperventilation.

VCD has been described in adults and in children. While VCD mostly occurs in females aged 20 to 40 years (14), we report on three children/adolescents, two of which were girls. The ages of our patients (12, 13 and 17 years) is in accordance with other reports on children 10 to 15 years old (7).

During an acute episode of VCD there may be severe respiratory distress, tachycardia, and accessory

Table 1. Differential diagnosis of VCD and bronchial asthma

	VCD	Bronchial asthma
Clinical symptoms	recurrent dyspnea	recurrent dyspnea
Dyspnea	mainly inspiratory	mainly expiratory
Lung function	mainly inspiratory obstructive ventilatory disorder	peripheral and central obstructive expiratory ventilatory disorder
Laryngoscopy (during episode)	paradoxical vocal cord function	regular vocal cord function
Blood gas tensions	normal or hyperventilation	hypoxaemia
Improvement on adrenaline	—	(+)
Improvement on salbutamol	—	+
Improvement of steroids	—	+
Improvement on sedation	+	—

muscle use. Chest auscultation reveals inspiratory and expiratory wheezing, not necessarily typical stridor. The character of the inspiratory noise caused by pathological adduction of the vocal cords is similar to the wheezing of the asthmatic patient, which may be one reason why VCD is often overlooked - even in hospitals (7). The duration of attacks vary from a few minutes to days.

From our cases and the literature, the following factors should lead to diagnosis of VCD:

1. The dyspnea is predominantly inspiratory (in the absence of upper respiratory tract infections).
2. No (or marginal) response to bronchodilator therapy.
3. Variable flattening of the inspiratory limb of the flow-volume curve suggestive of extrathoracic airflow limitation.
4. Respiratory cycle dependent assessment of the vocal cords during laryngoscopy under local anaesthesia.
5. No deterioration after discontinuation of anti-asthmatic therapy.

Besides laryngoscopy, other measures for directly assessing dysfunction of the vocal cords have been reported in the literature, such as computerized laryngeal ultrasound investigations, which have the advantage of being non-invasive, painless and widely accepted (14). However, this technique requires experience which cannot be obtained in most centres, due to the low numbers of these patients.

A subgroup of asthmatic patients may have a clinically relevant component of upper respiratory obstruction contributing to their symptoms (1). However, in our three patients no deterioration was noted after discontinuation of their antiasthmatic

maintenance drug therapy and the diagnosis of bronchial asthma was discarded in all cases. Furthermore, all patients remained symptom-free during the following months. It is therefore important to distinguish between bronchial asthma and VCD. Table 1 summarizes some differentiating diagnostic criteria.

One of the differentiating factors between asthma and VCD has been the absence of nocturnal awakening: however, in a recent case report, four patients showed nocturnal awakening as part of their symptom complex (15).

VCD has most often been misdiagnosed as bronchial asthma (15, 16) and in some cases there seems to be a coincidence of the two (1). In our three patients the diagnosis of bronchial asthma could be discarded. Other diseases have also been associated with VCD, such as cystic fibrosis (17), gastroesophageal reflux (13) and multiple chemical sensitivity (18). By correctly identifying patients with VCD, unnecessary medication, intubation or even tracheostomy can be avoided (1).

Whereas acute attacks may be treated by mild sedation under intensive care, for the long-term, a multidisciplinary approach is needed with the following measures:

1. Demonstration of diagnosis (e.g. video-documentation of laryngoscopy) and reassurance of patients and parents.
2. Speech therapy
3. Relaxation techniques.
4. Psychological intervention and/or psychotherapy.

As in other case reports (18), conversion disorders may play an important role, such as in our patient 3, so that long-term psychiatric therapy may be necessary.

In conclusion, our three cases and the literature indicate that VCD is more common than is generally appreciated. It represents a potentially overlooked differential diagnosis of recurrent dyspnea in children and adolescents. In order to avoid mistreatment it is important to question past diagnoses, and to characterize the type of dyspnea.

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