Three Cases of Paradoxical Vocal Cord Adduction Followed Up Over a 10-Year Period*

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Paradoxical vocal cord adduction (PVCA) is a rare disorder that may present with symptoms similar to asthma. Incorrect diagnosis may result in patients being unwittingly treated with prolonged high doses of antiasthma medication. PVCA probably forms part of a spectrum of uncommon and complex breathing disorders related to laryngeal dysfunction. Herein, we describe three cases of PVCA that illustrate the spectrum of clinical and physiologic presentation and the long-term natural history over a 10-year follow-up period. We conclude that PVCA, contrary to previous reports, is not always a benign condition; it may feature marked hypoxemia, fail to respond to previously advocated therapeutic strategies, and can persist on a long-term basis.

Paradoxical vocal cord adduction (PVCA) is an uncommon but important cause of wheeze. It can be mistakenly diagnosed as asthma, and patients are difficult to treat with severe symptoms often persisting. Patients frequently develop side effects of prolonged antiasthma medication and in rare cases, tracheostomy has been required to control symptoms. PVCA may be inspiratory or expiratory. Typically the patient is female, younger than 40 years old, and a health care worker. Arterial blood gas tensions are usually normal and spirometry readings are often poorly reproducible. Airway reactivity measurements, if feasible, are normal. Examination of vocal cords shows adduction of the cords on either inspiration or expiration which may be relieved by sedation. Little is recorded, however, in respect to the long-term natural history of this condition. Herein, we present three patients with vocal cord dysfunction who demonstrate the spectrum of clinical presentation and the difficulty in clinical management over a 10-year period.

Case Reports

Case 1
A 23-year-old female nurse presented in 1980 with episodes of wheeze and cough (precipitated by dust and exercise), early morning shortness of breath, and marked inspiratory stridor. She had a history of childhood asthma but had been asymptomatic for many years. Peak expiratory flow rate (PEFR) measurements during attacks were consistent with variable airflow obstruction with evening measurements of 500 L/min falling to 100 L/min in the early morning during attacks. Measurements of forced expiratory volume in 1 s (FEV1) of 3.9 L and forced vital capacity (FVC) of 3.99 between attacks fell to 2.35 L and 2.40 L, respectively, during attacks. These episodes were associated with normocapnic hypoxemia (eg, PaO2 6.1 kPa; PaCO2 4.9 kPa). She had positive skin responses to extracts of common allergens. Over the following 2 years, she required 17 admissions to hospital often to intensive care. Despite therapy with oral steroids, β-agonists, anticholinergics, and theophyllines, her condition deteriorated. Flow volume loop measurements indicated severe inspiratory extrathoracic upper airway obstruction. Direct laryngoscopy confirmed gross inspiratory adduction of the vocal cords. Results of neuropsychiatric evaluation were normal. The patient did not respond to lengthy courses of psychotherapy, speech, or hypnotherapy. A permanent tracheostomy was fashioned with resultant dramatic improvement in symptoms with no further hospital admission. During the past 10 years, she has suffered from recurrent chest infections, exercise-induced wheeze, and has required refashioning of her tracheostomy. She continues to require antiasthma medication for control of symptoms and still demonstrates adduction of her cords on inspiration.

Case 2
In 1981, a 24-year-old medical secretary presented with a history of recurrent episodes of wheeze, stridor, and shortness of breath. The PEFR measurements varied from 450 L/min to 90 L/min during attacks. She had positive skin tests to extracts of common allergens and allergic asthma had been diagnosed at age 10 years. Over the following 2 years, she required 10 admissions to hospital. These episodes were associated with normocapnic hypoxemia (eg, PaO2 5.18 kPa; PaCO2 4.67). In spite of increasing antiasthma therapy, her condition continued to deteriorate. Flow volume loop measurements demonstrated marked obstruction of inspiratory flow (Fig 1) and direct laryngoscopy showed marked adduction of the vocal cords during inspiration. This patient did not respond to prolonged psychotherapy or speech therapy and developed signs of steroid toxicity. Ten years later, she continues to require antiasthma therapy, including oral steroids for maximal control of symptoms. Evaluation of her upper airway in 1991 showed persistent marked inspiratory adduction of her vocal cords.

Case 3
A 33-year-old nurse-midwife presented in 1982 with episodic wheeze, shortness of breath, and stridor. She had presented to another hospital with similar symptoms in 1974 and was treated with maximal intravenous therapy without improvement. Measurements of airway caliper were considered unreliable and poorly reproducible. Laryngoscopy at that time showed normal findings. An exercise test did not induce symptoms of asthma. Her symptoms were considered to have a psychosomatic basis and treatment with

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her medication was withdrawn. However, she continued to have recurrent episodes of dyspnea, stridor, and wheeze and recommenced on a regimen of antiasthma therapy, including oral steroids. In 1977, she was admitted to intensive care elsewhere with apnea during an attack. In 1984, direct laryngoscopy demonstrated markedly inappropriate inspiratory adduction of the vocal cords during an episode of stridor. During attacks, she was noted to have normocapnic hypoxemia (eg, PaO2, 8.45 kPa; PaCO2, 4.35). The patient did not respond to prolonged psychotherapy, speech therapy or biofeedback therapy. Partial success in the management of her stridor was achieved through hypnotherapy, but she continues to require antiasthma therapy on an intermittent basis for control of symptoms 10 years after initial diagnosis of PVCA.

**DISCUSSION**

Paradoxical vocal cord adduction is an extremely uncommon condition and is due to inappropriate adduction of otherwise normal vocal cords. Upper airway obstruction without organic abnormalities was first described by Patterson et al in 1974. Although functional upper airway obstruction is usually due to PVCA, recently functional pharyngeal constriction and abnormal motion of the arytenoid region have been associated with upper airway symptoms. Functional upper airways obstruction presenting as asthma has been described, but it may also confuse the treatment of patients with asthma.

While these three patients show several of the classic features of PVCA, they also demonstrate a much wider spectrum of clinical and physiologic presentation than heretofore reported. Additionally, they provide insight into the long-term natural history of this condition that has not been recorded previously (to our knowledge). All three patients were women, younger than 40 years, who worked in health-care-related jobs, a combination of characteristics noted in many previous case reports. In contrast, one of the striking features of these three patients was that symptomatic episodes were associated with demonstrable hypoxemia and normocapnia. Although hypoxemia has been reported in patients with PVCA, it is considered extremely uncommon. Indeed, it has even been suggested that hypoxemia reliably differentiates between episodes of “true” asthma and PVCA. Our experience clearly demonstrates that this is not the case. In two of the three patients who also had previous histories of childhood asthma, we cannot be certain that PVCA occurred independently as an isolated phenomenon or whether there was coincidental active asthma. However, tracheostomy resulted in dramatic improvement in symptoms in one of our patients while her symptoms of mild asthma persisted, indicating that most of her airway obstruction was due to PVCA. It is noteworthy that our third patient, who had no history of childhood asthma, also demonstrated hypoxemia during symptomatic episodes, indicating that isolated PVCA without asthma can cause hypoxemia. Although minor glottic narrowing may occur in patients with asthma and in experimentally induced bronchospasm in normal subjects, it is highly unlikely that such a combination could account for the major dominant upper airway obstruction in our patients. In the recent report of a patient with asthma associated with inspiratory stridor due to pharyngeal constriction, there was no associated hypoxemia. We must conclude therefore that in our three patients, it is most likely that hypoxemia was preponderantly due to PVCA. This emphasizes that arterial hypoxemia cannot be used safely to distinguish attacks of asthma from those due to PVCA.

PVCA is considered to be a hysterical conversion reaction usually responsive to a range of psychological therapies. Speech therapy involving relaxation techniques and training of vocal cords has been successfully used with psychiatric or psychological support for more resistant cases. Unfortunately, this was not the case in our patients where none responded to prolonged courses of psychotherapy and speech therapy and just one of the three showed a partial response to hypnotherapy. Indeed, failure of such therapy dictated a tracheostomy in one of our patients, a procedure that dramatically alleviated her upper airway obstruction and effectively halted a pattern of repetitive prolonged hospital admissions and resulted in a reversal of previously gross steroid toxicity.

We believe that these three cases demonstrate that PVCA is not always a benign condition, that it may occur concurrently in patients with asthma, that it may induce significant hypoxemia, and that a wide range of therapeutic options, including tracheostomy, may be required to achieve maximal control of symptoms. Finally, the follow-up of these patients over a
10-year period indicated persistence of vocal cord dysfunction, establishing that in some patients at least, it may remain on a long-term basis.

REFERENCES