

# THORAX

## Editorial

### Vocal cord dysfunction and wheezing

Most respiratory physicians have experience of treating patients whose symptoms of wheezing seem out of proportion to the pathophysiology (if any) of their asthma. These patients are difficult to treat and often continue to have severe symptoms. They frequently have side effects from their treatment, in particular iatrogenic Cushing's syndrome. Although this is acknowledged widely within the specialty it is not well documented in published papers.

Wheezing occurs in a wide range of organic lung diseases as a result of reversible and irreversible airflow limitation, localised endobronchial disease (tumour or sarcoidosis), and diffuse lung disease (pulmonary oedema or lymphangitis). The differential diagnosis of acute wheezing also includes a separate disease entity that we choose to call "vocal cord dysfunction." This condition has a psychogenic basis and distinctive features, which we believe can be used to make the diagnosis and avoid unnecessary treatment.

At a time when large efforts are being made to educate our medical colleagues, patients, and the public at large in the diagnosis and effective management of asthma, suggesting that wheezing has a psychological basis in some patients could be seen as a potentially retrograde step. It is important, however, to acknowledge the existence of "psychogenic wheeze" and consider how it may be diagnosed and treated.

Patients with vocal cord dysfunction have been reported under various names, including Munchausen's stridor,<sup>1</sup> factitious asthma,<sup>2</sup> and the more descriptive "vocal cord dysfunction presenting as asthma."<sup>3</sup> The clinical features that have been described differ to some extent, but may be divided into inspiratory vocal cord dysfunction, expiratory vocal cord dysfunction, and both. These conditions appear to form a spectrum of disease and have in common the larynx as the target organ and the absence of evidence of local organic disease. Osler clearly described patients with this condition in 1902: "Spasm of the muscles may occur with violent inspiratory efforts and great distress, and may lead to cyanosis . . . Extraordinary cries may be produced, either inspiratory or expiratory."<sup>4</sup> This illustrates the point that vocal cord dysfunction may affect any part of the respiratory cycle and may be extremely frightening to patients and their medical attendants.

#### Inspiratory vocal cord dysfunction

Inspiratory wheeze due to inappropriate adduction of the vocal cords is characteristic of this condition. Although it should be easy to distinguish stridor from asthma, a fast fourrier transform technique has shown that the sound signals of asthmatic wheeze and stridor are of a similar frequency and can be separated only by their timing in the respiratory cycle.<sup>5</sup> This explains why clinical errors may be made and large airway obstruction due to tumour or an inhaled foreign body may be mistakenly treated as asthma.<sup>6</sup> We will not discuss organic causes of upper airway obstruction, which are well reviewed elsewhere.<sup>7</sup>

A case report entitled "Munchausen's stridor" in 1974 illustrated several points that are common to the spectrum of vocal cord dysfunction.<sup>1</sup> The patient, a 33 year old woman, was admitted to hospital on 15 occasions with inspiratory wheeze precipitated by infection or emotional upset. Clinical examination showed nothing abnormal apart from tachypnoea and high pitched stridor. The attacks resolved after emergency treatment. The results of subsequent investigations, including laryngoscopy and bronchoscopy, were normal. Once the nature of the illness was recognised the patient was referred for psychiatric care and no further attacks were reported.

Subsequent reports of "non-organic acute upper airway obstruction"<sup>8</sup> and "functional upper airway obstruction"<sup>9</sup> provided more detailed physiological data. Six women aged 19-46 were described, three with a history of psychiatric illness and one with a paramedical job. They presented with stridor and dyspnoea. The patients were observed to have an increased respiratory rate at rest, whereas in normal subjects when an inspiratory resistance is applied the rate is reduced.<sup>10</sup> Arterial blood gas analysis showed respiratory alkalosis with hypoxaemia in two cases. Emergency laryngoscopy during an attack showed vocal cord adduction, which was relieved by sedation, anaesthesia, and coughing or panting. Between attacks laryngoscopy and bronchoscopy showed nothing abnormal.

Flow-volume loops were abnormal with evidence of variable extrathoracic airway obstruction, manifest as a flat inspiratory loop with an increased ratio of forced expiratory flow at 50% vital capacity to forced inspiratory flow at 50% vital capacity ( $\dot{V}_{E50}/\dot{V}_{I50}$ ) (fig 1a). Airways resistance measured during panting was normal in these

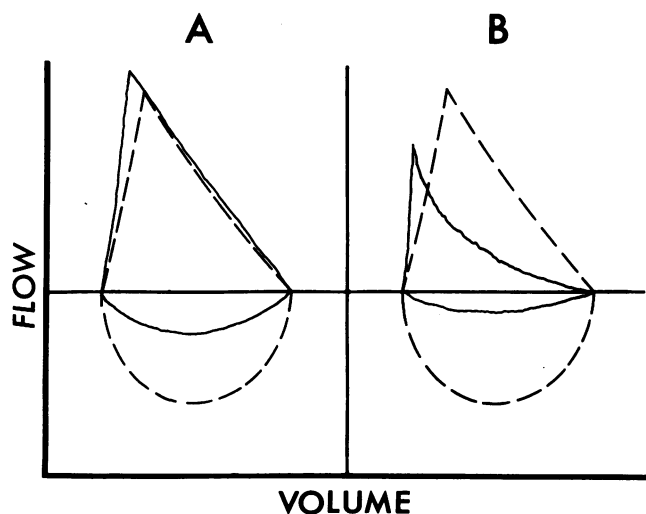


Figure 1 Schematic representation of abnormal flow-volume loops in (a) inspiratory vocal cord dysfunction and (b) inspiratory and expiratory vocal cord dysfunction. A normal flow-volume loop is represented by the broken line for comparison with the pathological recording represented by the solid line.

patients, in contrast to that of patients with organic upper airway obstruction, where airways resistance is increased.<sup>11</sup> Panting presumably causes the adducted vocal cords to relax. The combination of normal values for airways resistance and an increased  $\dot{V}_{E_{50}}/\dot{V}_{I_{50}}$  ratio is characteristic of inspiratory vocal cord dysfunction.

The consequences of inspiratory vocal cord dysfunction may be serious and all three patients described by Appelblatt and Baker<sup>9</sup> had a tracheostomy performed. Careful use of laryngoscopy and measurement of airway resistance and inspiratory and expiratory flow rates from flow-volume loops should allow physicians to make the diagnosis and avoid surgical intervention. The pathophysiology of the condition is not clear, though it has been speculated that "suggestion" mediated by the vagus may alter laryngeal tone and lower the threshold for stimuli to produce vocal cord spasm.<sup>9</sup>

### Inspiratory and expiratory vocal cord dysfunction

Patients with vocal cord dysfunction in expiration alone, or in inspiration and expiration, are often admitted to hospital and treated for acute asthma. "Factitious asthma" or "self induced wheeze"<sup>12</sup> are descriptive terms that have been used. These labels imply that the patient is trying to mislead the doctor by producing false symptoms and signs. Such deception is not necessarily conscious, so we prefer the term vocal cord dysfunction presenting as asthma.<sup>3</sup> The absence of findings compatible with asthma helps to define these patients as a group.

Patients are usually female, aged 20–40, and often have previous psychiatric illness and employment in the paramedical professions.<sup>12</sup> They often present with an apparent exacerbation of asthma and a history of many previous hospital admissions. On examination there may be inspiratory and expiratory wheeze that is loudest over the larynx and is less well transmitted to the chest wall. This physical sign is compatible with wheeze produced by apposition of the vocal cords and attenuated by transmission through the large airways and lung parenchyma.<sup>13</sup>

Arterial blood gases and the alveolar-arterial oxygen tension difference are usually normal, in contrast to those of acute asthma.<sup>14</sup> Results of lung function tests that are effort dependent may be misleading and in general show poor reproducibility. Pronounced variability in spirometric tests may in fact be a useful clue to the diagnosis (fig 2). The flow-volume loop commonly shows variable extrathoracic airway obstruction with additional expiratory airflow limitation (fig 1b). These abnormalities are attenuated by breathing a mixture of 80% helium and 20% oxygen

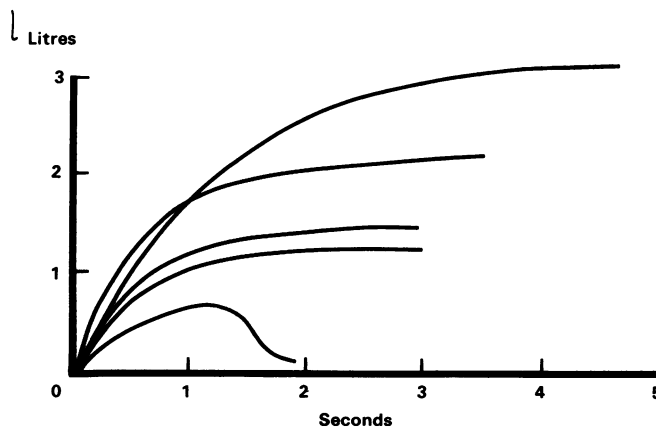


Figure 2 Series of spirometric traces from a single patient, showing the variable effort and poor reproducibility in this test in patients with vocal cord dysfunction.

(Heliox), which by increasing laminar flow through the large airways reduces wheeze and relieves symptoms. Body plethysmography shows normal values for total lung capacity and residual volume with no evidence of gas trapping, and airways resistance and conductance are normal. In contrast to patients with asthma, those with uncomplicated vocal cord dysfunction have normal bronchial hyperresponsiveness.

Laryngoscopy during wheezing shows almost complete adduction of the vocal cords throughout the respiratory cycle, with the glottis narrowed to a small posterior diamond shaped chink. This appearance may be reversed by sedation, anaesthesia, coughing, or panting. In the absence of wheeze laryngeal appearances are normal and if patients are asked to reproduce their wheeze the resulting bowing of the vocal cords is quite different from the appearance of the cords when the symptom occurs spontaneously.<sup>3</sup> There is typically little response to asthma treatment and these patients often have evidence of iatrogenic Cushing's syndrome.

### Expiratory vocal cord dysfunction

That expiratory wheeze can be voluntarily induced was first noted in 1898.<sup>15</sup> Subsequent studies, using radiographic measurements, showed that wheezing was accompanied by narrowing of the trachea and main bronchi owing to compression of the large airways by increased intrathoracic pressure. This pressure rise results from forced expiration at low lung volume and may be under voluntary control.<sup>16,17</sup>

"Emotional laryngeal wheezing"<sup>18</sup> appears to be part of the spectrum of vocal cord dysfunction, the physiological effects being due to a combination of expiratory vocal cord dysfunction and forced breathing at low lung volumes. During attacks breathing occurs close to residual volume and the expiratory flow rate reaches the maximum flow-volume envelope. Spirometric results are typically poorly reproduced, the flow-volume loop may be normal, and there is no evidence of hyperinflation on the chest radiograph. Other investigations, such as measurements of arterial blood gas tensions, airways resistance, and bronchial hyperresponsiveness, are normal, differentiating this condition from asthma. At laryngoscopy the vocal cords are adducted during expiration but the glottis widens during inspiration.

In normal individuals the size of the glottis increases during inspiration<sup>19</sup> and decreases during expiration,<sup>20</sup> its size being maximum at total lung capacity and minimum at residual volume.<sup>19,20</sup> Thus maximum expiratory flow rates at low lung volumes occur when the glottis is at its narrowest. Both the wheeze and these glottic appearances can be reproduced in normal subjects by forced expiration at low lung volume.<sup>18</sup> Continuous positive airways pressure can be used to relieve wheezing in these circumstances; it slows the expiratory flow rate, which allows the glottis to open, and increases residual volume.

### Asthma and vocal cord dysfunction

There is good evidence that glottic narrowing occurs in patients with asthma and may be an important component of airway obstruction.<sup>21,22</sup> Data collected by averaging pulmonary pressure-flow curves during tidal breathing show that the curvilinearity of the inspiratory limb in asthmatic patients is greater than in normal subjects and is corrected by breathing a mixture of 80% helium and 20% oxygen.<sup>21</sup> This finding suggests that flow regulation occurs in the larger airways and that inspiratory narrowing of the glottis and extrathoracic trachea plays a part in modulating air flow in asthma.<sup>21</sup> An extreme case of this has been

reported in an asthmatic patient with stridor in whom the glottic narrowing was thought to be involuntary.<sup>23</sup> Direct observations and measurements of the glottis in patients with airflow limitation have shown narrowing of the glottic chink on forced expiration,<sup>24</sup> and similar changes are seen in normal subjects with experimentally induced airflow obstruction.<sup>24</sup> The glottis may contribute to the control of airflow in asthma and hence effectively limit airway collapse and improve ventilation.<sup>22</sup> Other parts of the upper airway, such as the oropharynx, have been shown by fluoroscopic techniques to narrow during bronchoconstriction,<sup>25</sup> suggesting that upper airway muscles play a part in ventilatory control.

As glottic narrowing may occur for physiological reasons in patients with airflow obstruction, making a diagnosis of vocal cord dysfunction in a patient who has asthma is extremely difficult. This is exemplified by a patient of ours. A 41 year old woman with a paramedical job and an unstable family background has presented on many occasions with an acute attack of wheeze that is loudest over the larynx. During these attacks her flow-volume loop was often normal and she appeared to be breathing close to residual volume. At laryngoscopy there was vocal cord adduction during expiration. Arterial blood gas tensions during these attacks were normal. In addition, she has presented with acute attacks of asthma during which there was evidence of hypoxaemia. We have found that arterial blood gas tensions are the best basis for differentiating the two kinds of attack. Despite this policy she has been treated for acute asthma on several occasions when she has presented with vocal cord dysfunction.

### Psychiatric disorders in vocal cord dysfunction

The psychiatric diagnoses of patients with vocal cord dysfunction have been well reviewed by Martin *et al.*<sup>12</sup> They consider the condition to be a form of conversion disorder and have noted various associated psychiatric conditions, including depression, obsessive compulsive disorder, passive dependent personality, adjustment reaction, and somatisation disorder. Patients are often single women aged 20–40 who have a dependent and childlike relationship with their families and difficulty in expressing emotions such as anger, sadness, and pleasure. Martin's group believe that these patients do not knowingly control their illness for secondary gain and hence the condition should not be considered to be factitious or malingering.

The original report of Munchausen's syndrome<sup>26</sup> described patients wandering from hospital to hospital with apparently acute illnesses and a dramatic history. They appear to receive gratification by deliberately misleading doctors and hospital staff to the extent that they may submit to unnecessary and uncomfortable treatment. The use of this synonym to describe all cases of vocal cord dysfunction seems inappropriate. There is a flavour of the syndrome, however, in some of the more bizarre cases.<sup>1</sup>

Factitious illness, as typified by factitious fever,<sup>27</sup> covers a range of psychiatric disorders, from simple malingering to Munchausen's syndrome, which have in common the deliberate feigning of illness. Chronic factitious illness is often a symptom of a severe personality disorder typified by hostility, dependence, imposture, poor impulse control, and self destructive acting out.<sup>27</sup> The separation of this syndrome from hysteria presenting as conversion reactions is not straightforward, though in factitious illness there is a greater awareness of motivation and manipulation of the environment. Ultimately the exact psychiatric diagnosis may be less relevant than the treatment (if effective) and for this reason we prefer not to label the syndrome with an adjective that implies any particular underlying psycho-

pathology. It seems likely that any of the psychiatric syndromes we have described could be applicable to individual patients.

### Treatment of vocal cord dysfunction

A multidisciplinary approach, with physicians, psychiatrists, and speech therapists playing a part, is most effective in the treatment of patients with vocal cord dysfunction.<sup>3,12</sup> A useful strategy is for the physician to inform the patient that as a result of the investigations a diagnosis has been reached. The nature of vocal cord dysfunction should then be explained with the help of diagrams and video recordings of the bronchoscopic findings, if available.<sup>12</sup> Patients should be told that the triggers that bring on their symptoms are not entirely clear, but that they may be helped by speech therapy and relaxation techniques. They may be referred for psychotherapy, the aim of which is not to try to cure their psychiatric disorder but rather to allow them to retain their psychiatric symptoms without the need for vocal cord dysfunction. The basic methods used are relaxation exercises and supportive therapy. Family therapy may be important if failure of the patient to leave home and gain independence is at the centre of the psychopathology.<sup>12</sup>

The speech therapy techniques used for patients are adapted from those used for patients with other functional voice disorders, such as hoarseness, laryngitis, and vocal fatigue. The methods used include pitch change, diaphragm breathing, and reduction of extrinsic muscle tension. Patients are taught to focus attention away from the larynx and inspiration, to concentrate on active expiration using the anterior abdominal muscles, and to relax the oropharyngeal, intercostal, neck, and shoulder girdle muscles. They are then encouraged to apply these techniques when their symptoms begin.<sup>12</sup> The prognosis appears to be relatively good in inspiratory and inspiratory plus expiratory vocal cord dysfunction.<sup>3</sup> A good prognosis may be predicted by a positive reaction to the initial explanation of the diagnosis.

### Conclusion

Vocal cord dysfunction is an uncommon but important cause of wheeze. Awareness of its different manifestations should aid diagnosis and help to improve patient management by avoiding unnecessary anti-asthma treatment. Physicians should suspect vocal cord dysfunction when a patient presents with recurrent episodes of wheeze unresponsive to usual asthma treatment. Typically patients, mostly women under 40, are psychologically unstable. Useful physical signs are rapid breathing at low lung volume, absence of hyperinflation, and wheeze localised to the larynx but heard less well over the lung fields. Arterial blood gas tensions will be normal and results of spirometry poorly reproducible, and the flow-volume loop may show evidence of variable extrathoracic obstruction or may be normal. Lung function tests when feasible show normal airway resistance, and bronchial responsiveness when measured in remission is also normal. Laryngoscopy will show adduction of the vocal cords in inspiration, expiration, or both, which should be relieved by sedation. Wheeze will be relieved by breathing a helium-oxygen mixture or, in the case of expiratory vocal cord dysfunction, with continuous positive airways pressure.

In an acute attack patients should be treated for asthma unless there is incontrovertible and objective evidence that their symptoms are psychogenic. Hypoxaemia is a

particularly useful indication that asthma is likely to be genuine.

JON GOLDMAN  
MARTIN MUERS

Regional Respiratory Unit,  
Killingbeck Hospital,  
Leeds LS14 6UR

Reprint requests to Dr Goldman

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