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Functional laryngeal dyskinesia: an important cause of stridor

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Abstract
Functional laryngeal dyskinesia is a disorder that presents with stridor. Failure to recognize its features can result in inappropriate investigation and treatment for a condition that has a psychogenic origin. The key feature in diagnosis is paradoxical vocal fold adduction on inspiration, as seen on fibre-optic nasendoscopy. This phenomenon together with the associated stridor may disappear after distraction techniques or when the patient is asleep. We present five such cases which presented to hospitals in south west London over a 12 month period.

Key words: Larynx; Dyskinesia; Stridor

Introduction

The term functional laryngeal dyskinesia (FLD) was coined by Ferris *et al.*[1] in 1998. Prior to this there have been many publications describing upper airway obstruction and stridor without an underlying physical cause. However, only a few of these cases have paradoxical vocal fold movements documented. In fact, episodic vocal fold dysfunction simulating asthma was first reported in 1842 with the name of 'hysteric croup'.[2] Since this time other cases have been described as psychogenic stridor, psychogenic asthma, Munchhausen's stridor, functional laryngeal obstruction and laryngeal asthma. The term functional laryngeal dyskinesia appears to be the most appropriate term for a condition, which involves an abnormality of laryngeal movements from an underlying functional cause. The functional aetiology is supported by the demonstration of reversibility of airway distress by distraction techniques in the short term and psychotherapy and biofeedback techniques in the long term. We believe that FLD is an important cause of upper airway obstruction and should be remembered in the differential diagnosis of stridor. If the diagnosis is not made, invasive investigation and treatment including tracheostomy may occur, thus enhancing the secondary functional gain for the patient.

Case reports

Case 1

A 22-year-old female nurse presented with a two-week history of non-productive cough followed by one week of persistent inspiratory stridor. There had been two episodes of minor haemoptysis but no chest pains. She smoked 10 cigarettes per day. On examination there was audible inspiration stridor. Chest and lateral neck X-rays and blood gas analysis were normal. Fibre-optic nasendoscopy revealed mild supraglottic erythema and paradoxical adduction of both vocal folds on inspiration. The patient was admitted for observation of the airway and given intravenous cefuroxime. The stridor temporarily settled whilst the patient was asleep on the ward. Over the next three days her symptoms settled completely and she was discharged after normal vocal fold movements were noted on nasendoscopy.

The patient was reviewed in the speech therapy department three months later. Mild inspiratory stridor was again present but this disappeared when the patient laughed. The stridor was also eliminated with a 'sh' sound on inspiration. A course of speech therapy was arranged and there have been no further episodes requiring hospital admission.

Case 2

A 14-year-old student presented to the Accident and Emergency (A&E) department with persistent noisy breathing and shortness of breath, that had developed after a run. There was a history of asthma but she was not on regular medication. On examination there was mild inspiratory stridor but the patient was not distressed. The chest was normal and the peak flow measurement was within the normal range. After a brief period of observation in A&E the stridor settled and the patient was discharged home. Nasendoscopy was not performed at this attendance.

One month later she represented to the A&E department with noisy breathing following swimming. She also complained of a mild sore throat. On examination there was once again mild inspiratory stridor, but no abnormality in the chest. Fibre-optic nasendoscopy revealed paradoxical adduction of the vocal folds on inspiration. She was therefore admitted for observation but the stridor settled overnight and she was allowed home the following day.

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Case 3

A 77-year-old female retired radiological clerk presented with a six-month history of recurrent episodes of noisy breathing. This followed an upper respiratory tract infection. She had previously undergone a thyroidectomy, but there had been no alteration in vocal function following this. At this particular presentation the noisy breathing had been present for several hours and so she attended the A&E department. On examination there was mild biphasic stridor with no abnormality of the chest. Fibre-optic nasendoscopy revealed paradoxical adduction of the vocal folds on inspiration. She was admitted for observation and given intravenous steroids. The stridor settled during the next few hours and the patient was discharged the following day. A course of speech therapy was arranged and after two sessions the intermittent biphasic stridor settled.

Case 4

A 54-year-old woman presented to the A&E department with a four-hour history of increasing shortness of breath and stridor. In addition she had developed a sore throat and cough two days previously. Three months prior to this episode she had been admitted with the same problem to another hospital and been diagnosed with epiglottitis. On examination she had marked inspiratory stridor but no abnormality in the chest. Fibre-optic nasendoscopy revealed erythema of the hypopharynx and larynx. There was paradoxical vocal fold adduction on inspiration. X-rays of the chest and lateral neck were normal. She was admitted for observation and given intravenous hydrocortiscone and antibiotics. During periods when the patient was asleep, the stridor seemed to settle. Her condition improved over the next 48 hours with the stridor gradually ceasing. Repeat nasendoscopy revealed that the vocal fold movements had returned to normal and she was discharged home.

Case 5

A 29-year-old daughter of a psychiatrist from the USA presented to the A&E department with marked inspiratory stridor associated with a weak voice, that had been present for six hours. Nasendoscopy revealed paradoxical movement of the vocal folds with respiration. She gave a history of childhood asthma but on six occasions during her lifetime she had been intubated due to respiratory difficulties. In 1993 a diagnosis of 'functional stridor' was made and she was taking regular diazepam 5 mg tds. After some reassurance and the administration of further diazepam the stridor settled and she was discharged home. Similar episodes occurred on the following two nights, that again settled spontaneously.

Discussion

FLD is an important cause of stridor. Patients presenting with this condition tend to be young females. In our series all the patients were female with a mean age of 39.2 years. Most patients tend to be from a high socioeconomic group and are well educated. There often seems to be an affiliation with the medical profession.[3] In the series described, patients included a nurse, a retired radiological clerk and the daughter of a psychiatrist. In many cases there is often a precipitating psychological or psychosocial event that can be identified.

Other important factors in FLD include the observation that the voice is nearly always normal and despite the apparent airway compromise, sustained speech is possible. In addition, many patients have been previously diagnosed with asthma and are currently taking regular medication. In some, genuine asthma is present but in others the diagnosis is erroneous and has been confused with FLD per se. Some patients have recently suffered with an upper respiratory tract infection. Similarly gastroesophageal reflux[4] and recent exposure to cold air, particularly during exercise, may have a possible link.[5] It, therefore, seems possible that laryngeal inflammation could be an initiating factor. Maschka *et al.*[6] have devised an interesting classification scheme that divides paradoxical vocal fold movement into organic and non-organic aetiologies.[6]

Normal respiratory function and vocal fold movement are co-ordinated in the medulla, that contains aggregates of respiratory neurones including the dorsal and ventral respiratory groups. The dorsal respiratory group is part of the nucleus of the tractus solitarius, from which it receives afferent information from respiratory-related mechanoreceptors and chemoreceptors. The ventral respiratory group is located rostrally in the nucleus ambiguus and caudally in the nucleus retroambiguus. The nucleus ambiguus contains not only inspiratory neurones, but also motor neurones to the laryngeal muscles. In addition there are parasympathetic neurones to the bronchioles and heart. The neurones to the intrinsic muscles of the larynx travel from the nucleus ambiguus in the vagus nerve. Movements of the vocal folds in respiration and indeed in phonation, are coordinated by these respiratory centres.

Previous studies of upper airway neural reflexes, have shown that stimulation of receptors in the larynx may induce either cough or vocal fold adduction and apnoea.[7] In trials on animals, single shock stimulation evoked a simple closure of the larynx. Repetitive stimulation of the superior laryngeal nerve produced excitatory laryngeal adduction and laryngospasm, which persisted beyond stimulation.[8] Reflex laryngeal closure with involuntary induced spasms of the laryngeal adductor muscles is normally inhibited by higher cerebral function. If the inhibitory process is reduced, glottic closure is facilitated. A possible explanation for the loss of control of laryngeal function, would be the fixation on the frightening experience of airway obstruction caused by the reflex in the acute phase of laryngeal irritation. Subsequently a learned, inappropriately used mechanism, causing spasm in the organ of fixation develops due to behavioural conditioning. Therefore recurrent episodes of vocal fold adduction may follow if throat irritation is experienced again. Many patients do have repeated visits to hospital including admission with this problem. In the series by Ferris *et al.*,[1] patients were hospitalized a mean of three times before a definitive diagnosis was made.

The key to examination of patients with FLD is the observation of stridor without other physiological evidence of airway compromise. Thus the pulse and blood pressure are usually normal and there is no evidence of peripheral or central cyanosis. The stridor should become less intense during sleep and if distraction techniques are used. Fibre-optic nasendoscopy reveals paradoxical vocal fold adduction on inspiration. It is useful to elicit vocal fold abduction, even if momentary and during expiration, to exclude vocal fold paralysis or fixation. Vocal fold abduction during inspiration will reassure the surgeon that the airway is safe and can be elicited by using a number of techniques. Firstly the larynx should be visualized in all phases of respiration for several minutes and the effects of coughing and phonation observed. If vocal fold abduction still cannot be elicited during inspiration, various distraction techniques can be employed. The patient can be asked to count backwards, from 100 in sevens or asked to hold their breath and slowly count to 20. If these measures are unsuccessful a 'sniff test'

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in which the patient is instructed to rapidly sniff in through the nose may prove effective.[1] In addition to the detailed examination described above, some routine investigations may be arranged. Blood gas analysis is usually normal. Hypoxia, hypercarbia and respiratory acidosis indicative of respiratory decompensation should not be encountered with FLD. The chest X-ray should be normal and respiratory function tests do not produce any characteristic pattern.

The treatment of FLD should be calming reassurance together with an explanation of airway physiology to the patient. Accurate diagnosis helps to avoid invasive treatment, which can enhance the secondary functional gain. In the series of 10 patients reported by Ferris *et al*.,[1] four were intubated and two received tracheostomies. The importance of making the correct diagnosis cannot be overemphasized therefore. Three of the five patients in this series had a preceding history of upper respiratory tract infection. With this in mind, the role of antibiotics and both nebulized and systemic steroids although not proven, may be justified. Sedatives may be administered if the clinician is confident of the diagnosis and the patient is anxious. In general however, one should remember the functional nature of the aetiology and treatment should be directed in this regard. After the acute episode has subsided referral to a psychiatrist or psychotherapist is advised for further treatment and prevention of repeat episodes. Various underlying psychiatric disorders have been discovered including conversion disorder, factitious disorder, hysteria and depression. Some authors have also advocated speech therapy as a useful therapeutic option.

Conclusion

Functional laryngeal dyskinesia is an important cause of stridor. The key feature in diagnosis is paradoxical vocal fold adduction on inspiration, which may subside with distraction techniques. Treatment should be conservative rather than invasive.

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