Functional stridor diagnosed by the anaesthetist

Guy Tousignant MDCM FRCP, Simcha J. Kleiman MD FRCP

While stridor is an ominous sign implying severe airway stenosis, not all stridor has an organic aetiology. We present two cases of functional stridor in which the diagnosis was made by the anaesthetist. As experts in the management of difficult airways, anaesthetists should be aware of this clinical entity. Recurrent episodes present as aphonia, dysphonia, dyspnoea, apnoea or unconsciousness. Stridor is usually inspiratory. Flow volume loops show a pattern of variable extrathoracic obstruction with diminished peak inspiratory flow. Awake fibreoptic laryngobronchoscopy reveals normal airway anatomy, intense adduction of false and true vocal cords during inspiration and normal vocal cord motion on expiration. Treatment of functional stridor is supportive. The diagnosis of functional stridor demands exclusion of life-threatening airway stenosis of organic aetiology. A high index of suspicion for this clinical entity will reduce the incidence of unnecessary interventions such as tracheal intubation and tracheostomy.

Bien que le stridor soit un signe inquiétant suggérant une sténose sévère des voies aériennes, tout stridor n'a pas nécessairement une étiologie organique. Nous présentons deux cas de stridor fonctionnel dans lesquels le diagnostic a été fait par l'anesthésiste. Comme experts de la gestion des voies aériennes problématiques, les anesthésistes devraient connaître cette entité clinique. Des épisodes répétés se présentent comme de l'aphonie, de la dysphonie, de la dyspnée, de l'apnée et de l'inconscience. Le stridor est habituellement inspiratoire. Les courbes débit-volume ont un aspect suggérant une obstruction extrathoracique variable avec un débit inspiratoire de pointe diminué. Chez le patient éveillé, la laryngobronchoscopie par fibres optiques révèle une anatomie normale des voies aériennes, une adduction intense des cordes vocales supérieures et inférieures durant l'inspiration et un mouvement normal des cordes vocales lors de l'expiration. Le traitement du stridor fonctionnel en est un de support. Le diagnostic du stridor fonctionnel demande l'exclusion d'une sténose potentiellement fatale des voies aériennes qui aurait une étiologie organique. Une forte suspicion pour cette entité clinique réduira l'incidence d'interventions inutiles comme l'intubation et la trachéostomie.

Key words
AIRWAY: obstruction, management;
LARYNX: stridor.

From the Departments of Anaesthesia, Sir Mortimer B. Davis-Jewish General Hospital, McGill University, Montreal, Canada.

Address correspondence to: Dr. Guy Tousignant, Department of Anaesthesia, Rm. A-333, Sir Mortimer B. Davis-Jewish General Hospital, 3755 Cote Ste-Catherine Road, Montreal, Quebec, Canada H3T 1E2.

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Case reports

Case 1

A 27-yr-old white man presented to the Emergency Room with shortness of breath and wheezing of two days duration. He also complained of a sore throat, hoarseness, and difficulty in swallowing. His past history included a diagnosis of asthma treated for two years with a salbutamol inhaler. Earlier that day the patient had discharged himself from another hospital against medical advice and had driven over 100 miles to reach our institution.

The patient was sitting upright in bed, breathing rapidly and using accessory muscles of respiration. Inspiratory and expiratory stridor could be heard from across the Emergency Room. On one brief occasion the consulting anaesthetist observed the patient speaking to a visitor without exhibiting stridor.

The patient was afebrile, BP 130/86 with no pulsus paradoxus, HR 88 min^{-1} and regular, RR 34 min^{-1}. Examination of the mouth was normal. The trachea was midline and stridor was best heard with the stethoscope over the larynx. Auscultation of the chest revealed air entry bilaterally with occasional expiratory wheezes.

Laboratory examination revealed no obvious derangement. Haemoglobin concentration and WBC count were normal, as were all electrolytes including serum Ca^{++}.

Arterial blood gas analysis while breathing 60% O_2 by mask showed: $PaO_2 = 279$ mmHg, $PaCO_2 = 40$ mmHg, pH = 7.40. Inspiratory and expiratory flow rates were not measured in the Emergency Room. The upright chest *x*-ray was normal except for poor inspiration.

In spite of the unremarkable physical examination of the chest, absence of pulsus paradoxus, and absence of radiographic hyperinflation, the patient was diagnosed by the Emergency physician as having status asthmaticus and upper airway obstruction of unknown aetiology. He was treated with salbutamol by nebulizer every 30 min and aminophylline *iv* (loading dose and continuous infusion). This treatment failed to improve his condition.

Soft tissue *x*-rays of the neck were done. The AP view was normal with no subglottic narrowing. There was a suggestion of an abnormal shadow below the level of the epiglottis on the lateral view, but this could not be characterized any further by the radiologist. Because of possible swelling of the airway, the patient was treated with nebulized racemic epinephrine, with no improvement.

The ENT service was consulted and informed consent obtained for direct laryngoscopy, rigid bronchoscopy and possible tracheostomy. During transfer to the Operating Room the patient's stridor became louder. He developed grunting and increasing agitation.

In the Operating Room, the patient was monitored with an ECG, automatic non-invasive blood pressure monitor and a pulse oximeter. Oxygen saturation was 99%. The patient was instructed to breathe nebulized 4% lidocaine by face mask in order to anaesthetize the airway. Sedation was accomplished with *iv* administration of droperidol 1.0 mg, and divided doses of fentanyl and diazepam to a total of 150 μg and 20 mg respectively. Sedation failed to improve the patient's stridor. The ENT surgeon performed an awake fibreoptic laryngobronchoscopy through the nose. The airway to the level of the main carina was normal except for adduction of the vocal cords during inspiration. With the patient awake the trachea was intubated under direct vision using a 6.0 mm tracheal tube. After securing the airway, anaesthesia was induced with a sleep dose of *iv* thiopentone and maintained with 60% N_2O–40% O_2 and isoflurane. Direct laryngoscopy and oesophagoscopy revealed no new findings.

At the end of the procedure, the patient emerged from anaesthesia and the trachea was extubated while the patient was awake and in the lateral position. For the first time, his breathing was quiet with no stridor. On the basis of stridor associated with paradoxical vocal cord motion, which resolved when the patient was distracted or emerging from anaesthesia, the anaesthetist proposed the diagnosis of "functional stridor."

The patient was transferred to the SICU for overnight observation. During his stay in SICU, the nurses found him to be very demanding and he threatened to discharge himself on several occasions. He developed several recurrences of stridor particularly when the nursing staff was busy with other patients. These episodes were treated with sedation.

The assessment of the consulting psychiatrist was of a narcissistic personality disorder, with poor insight and attention-seeking behaviour. As part of this picture, the patient's stridor was postulated to be an hysterical conversion reaction.

Case 2

A 23-yr-old white woman complaining of a recent onset of cough, tightness in the throat and difficulty breathing when supine was seen in consultation by the ENT service. She had noted no changes in her voice. On physical examination the patient was not in marked distress but had audible inspiratory stridor during quiet respiration. On indirect laryngoscopy the vocal cords were in the paramedian position and exhibited decreased mobility. Auscultation of the chest revealed decreased breath sounds bilaterally with occasional expiratory wheezing.

During the course of indirect laryngoscopy of the airway, stridor increased. The patient developed tracheal tugging and intercostal retractions. Her apparent clinical condition deteriorated to the point where the otolaryngologist performing the indirect laryngoscopy called the hospital cardiac arrest team. She was given oxygen by mask and was at no time cyanotic. Stridor improved after *iv* injection of lorazepam 1 mg. No other diagnostic studies, including flow volume loops, were performed. A working diagnosis of vocal cord paralysis was made. Although it was noted that a "psychosomatic" aetiology could not be ruled out, two ENT surgeons agreed to perform a tracheostomy and to evaluate the problem further.

The patient presented to the OR with severe stridor and respiratory distress, using all accessory muscles of respiration. After induction of general anaesthesia with thiopentone and paralysis with succinylcholine, the trachea was intubated with no difficulty. A tracheostomy was performed and a #6.0 Shiley tracheostomy tube was inserted. At the conclusion of the procedure vocal cord mobility was noted to be normal during emergence from general anaesthesia.

On the first postoperative day, awake flexible fibreoptic laryngoscopy demonstrated good vocal cord motion immediately before and after coughing. On this basis it was decided that the aetiology of her stridor was most likely "psychosomatic." In the postoperative period complaints of shortness of breath were treated with reassurance. A psychiatric assessment proposed the diagnosis of a conversion disorder and a possible masked depression. She was treated with relaxation therapy. Before discharge from hospital, it was established that she could tolerate "corking" of her tracheostomy without developing respiratory difficulties. She was followed as an out-patient by the ENT and psychiatry services. Her tracheostomy cannula

was removed five months after surgery and the wound was allowed to close.

Fifteen months after her initial tracheostomy, the patient was again brought to the Operating Room for suspension laryngoscopy under "general anaesthesia without paralysis" because of recurrent stridor. The possibility of performing another tracheostomy was raised. After discussion with the otolaryngologist, it was decided that the patient should have an awake flexible fibreoptic laryngobronchoscopy by the anaesthetist. Her vocal cords were seen to be abducted during quiet respiration. When asked to inspire deeply, the vocal cords moved toward the midline and the patient became extremely stridorous. She was treated with midazolam, 1 mg *iv*. Following sedation, the vocal cords were seen to be moving normally especially when her attention was distracted from her breathing. The diagnosis of "functional stridor" of non-organic aetiology was made. The patient did not undergo tracheostomy. She continues to be seen as an outpatient by the ENT and psychiatry services.

Discussion

Anaesthetists are often consulted in the management of patients presenting with stridor. "Functional stridor" should be borne in mind as part of the differential diagnosis when approaching the stridorous patient.

To the best of our knowledge there is only one suggestion in the anaesthesia literature of a syndrome of upper airway obstruction lacking an organic aetiology.[1] Over the years, this syndrome has been given many names in the otolaryngology, internal medicine and respirology literature. It has been called croup (false, spasmotic,[2] hysteric[3]), spasm[4] (laryngeal, glottic), emotional laryngismus stridulus,[5] psychogenic upper airway obstruction,[6] functional upper airway obstruction,[7] non-organic upper airway obstruction,[8] Munchausen's stridor,[9] factitious asthma,[10] emotional laryngeal wheezing,[11] as well as paradoxical vocal cord motion.[12]

The earliest descriptions of this clinical problem date back to the 19th century.[3] Most references are to young women with frequent attacks of "asthma." These descriptions often include emotional disturbances such as infantile behaviour, immature personality, anxiety states and conversion reactions. Attacks tend to be recurrent, often with a psychological precipitant. The presentation may include aphonia, dysphonia, apnoea, or unconsciousness. Stridor is usually inspiratory or both inspiratory and expiratory. Rarely, pure expiratory stridor occurs. If these patients can be distracted, they will often be able to speak in long sentences, as was the case in the first case report. Asking the patient to cough will usually elicit normal respiration before and after coughing.[9]

On physical examination these patients are usually afebrile and tachypnoeic. While they do not have the frightened look characteristic of patients with airway obstruction, they frequently use accessory muscles of respiration. Heart rate and blood pressure are normal, with no pulsus paradoxus. On auscultation the chest is typically clear. Stridor is best heard over the larynx. Laboratory examination including complete blood count, biochemical analysis, including serum Ca^{++} concentration, is normal. Arterial blood gas analysis is either normal or shows a respiratory alkalosis. Some patients have exhibited severe hypoxaemia ($PaO_2 < 50$ mmHg).[7]

Cormier studied pulmonary function in three patients with upper airway obstruction in which no organic cause could be identified.[8] Flow volume loops showed a pattern of severe variable extrathoracic obstruction with a decreased peak inspiratory flow. As well, there was an increased ratio of the forced expiratory flow at 50% vital capacity to the forced inspiratory flow at the same lung volume. Airway resistance was normal in the two patients in whom it was measured. Cormier concluded that this clinical entity could be identified easily by the marked discrepancy between inspiratory flow limitation and airway resistance.

The syndrome has also been mistaken for bronchial asthma.[6,10,13] However, many salient features of asthma are absent. Patients with functional stridor have no pulsus paradoxus, no elevated alveolar-arterial oxygen tension difference and no decrease in expiratory flow rates. In addition, there is no hyperinflation on chest *x*-ray and bronchial hyperreactivity cannot be demonstrated.[13] Simple spirometry will show normal expiratory flows and volumes.[10] Radiographic views of the soft tissues of the neck are normal.

The gold standard for diagnosis remains endoscopy, which will reveal normal airway anatomy. Whereas the usual pattern of vocal cord motion involves wide abduction of the cords on inspiration, these patients demonstrate intense adduction of false and true vocal cords during inspiration and normal vocal cord motion on expiration.[2,7,12,13] This abnormality may be demonstrated to resolve by sedating or distracting the patient, while the vocal cords are observed with a flexible fibreoptic bronchoscope. With this approach, should an organic cause of stridor be demonstrated, it can be assessed and perhaps even managed by tracheal intubation using the bronchoscope (Figure). We would discourage the approach of induction of anaesthesia and paralysis before definitive diagnosis, as was seen in our second case.

Once diagnosed, treatment of functional stridor consists of supportive therapy, psychotherapy and speech therapy. Administration of placebo or sedatives may help to control symptoms in the acute episode.

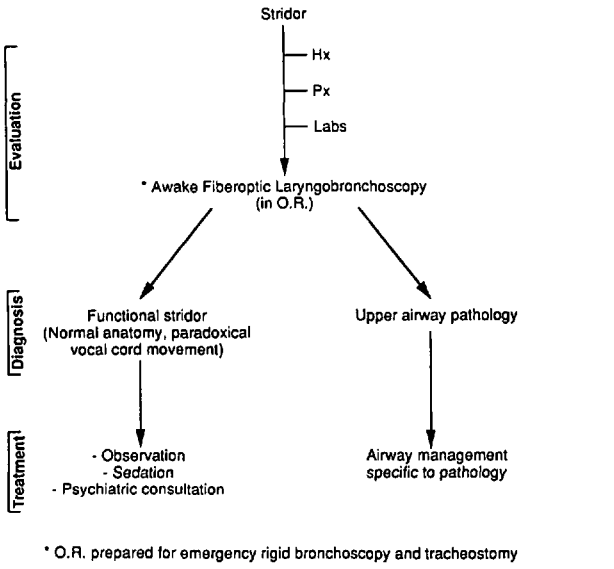

FIGURE Suggested algorithm for the management of a stridulous patient.

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