Acute Airway Obstruction Secondary to Vocal Fold Heterotopic Ossification

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Abstract A 29-year old man of Eritrean origin presented with acute stridor and respiratory distress on a background 1 year history of progressive breathing difficulty and worsening inspiratory stridor. Fibreoptic laryngoscopy revealed an indeterminate swelling of the left vocal fold leaving no clear airway visible. The patient refused surgical tracheostomy. Microlaryngoscopy revealed a hard, calcified mass arising from the left cord preventing intubation. Histological analysis after excision revealed features consistent with heterotopic ossification. At 4 months repeat microlaryngoscopy was performed revealing normal appearance of the larynx and subglottis. Heterotopic ossification in the larynx is a very rare condition that presents a diagnostic and therapeutic challenge. In the first documented case in the larynx, we describe how the disease caused life threatening airway obstruction, but was managed in a way that led to preservation of laryngeal function and complete resolution of the condition.

Keywords Airway obstruction · Heterotopic ossification · Larynx · Myositis ossificans · Tuberculosis · Osteoma

Introduction

Airway obstruction is a life threatening emergency necessitating prompt management to obtain a secure airway. Its causes vary from accidental foreign body inhalation, to malignant upper airway tumours. Malignant bone forming lesions of the larynx, such as osteosarcoma, have been described but they are extremely rare (<20 cases in the world literature), and benign bone forming lesions are rarer still. We present a case of acute airway obstruction secondary to an ossified vocal fold.

Case Report

A 29-year old man of Eritrean origin presented with acute stridor and respiratory distress on a background 1 year history of breathing difficulty and worsening inspiratory stridor. Following stabilisation of his initial condition with intravenous steroids and adrenaline nebulisers, fibreoptic laryngoscopy was performed revealing an indeterminate swelling of the left vocal fold leaving no clear airway visible. The patient refused surgical tracheostomy and was transferred to the National Centre for Airway Reconstruction for further management.

Four years previously, the patient had presented with a chronic suppurative abscess of the left arm with a histological diagnosis of tuberculosis. Only 2 months of chemotherapy were completed before the patient was lost to follow-up. There was no other significant past medical history.

Microlaryngoscopy revealed a hard, calcified mass arising from the left cord preventing intubation, or rigid endoscopic assessment of the upper trachea (Fig. 1). To secure the airway the mass was excised via carbon dioxide

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Fig. 1 Rigid laryngoscopic view of left vocal fold mass

Fig. 4 Rigid endoscopic view 4 months post laser treatment

Fig. 2 Rigid endoscopic view during CO₂ laser

Fig. 3 Axial CT scan showing calcified left vocal fold extending into the subglottis

laser under supraglottic jet ventilation. Significant laser flaring during this procedure suggested heavy calcification (Fig. 2). Bronchial aspirates taken at the time were positive for *Klebsiella oxytoca* and *Enterobacter cloacae*. Blood

tests for white cell count, calcium, parathyroid hormone, ANA and ANCA were all within normal limits.

Subsequent computed tomography (CT) of the neck and chest revealed calcified thickening of the proximal upper thoracic trachea and nodular infiltration at the left lower lobe of the lungs. There was also visible calcification of the left vocal fold (Fig. 3).

At 2 months, rigid bronchoscopy was performed. Bronchial washings were positive for *Staphylococcus aureus* and respiratory flora only. At 4 months repeat microlaryngoscopy was performed revealing normal appearance of the larynx and subglottis (Fig. 4). Samples were obtained for histology and microbiology.

Histopathology revealed a thin layer of squamous epithelium (vocal fold epithelium), overlying a mixture of remodelled lamellar and woven bone within the superficial stroma. There was no cartilage, osteoid, or dystrophic calcification present in the sample (Fig. 5a, b). In the context of its location this was considered in keeping with heterotopic ossification.

Discussion

Heterotopic ossification (HO) is the process of bone formation outside of the skeleton, within the soft tissues. Previously synonymous with myositis ossificans, this association fell out of favour after descriptions of the process occurring outside muscular tissue. It can be classified into congenital or acquired with the latter being by far the most common. The autosomal dominant hereditary form (myositis ossificans progressiva) carries an incidence of only 1 per 2 million, and results in severely impaired joint mobility and ankylosis by early adulthood [1]. The acquired form is caused either by direct musculoskeletal trauma [2] (be it accidental or surgical) or via neurogenic injury (spinal cord or central nervous system injury) [3]. The incidence of the acquired variant depends on the

Fig. 5 **a** HE $\times 25$ Low power examination reveals a squamous epithelium overlying interconnected trabeculae of lamellar and woven bone. **b** HE $\times 100$ High power magnification demonstrating viable lamellar bone with active remodelling (osteoblasts *upwards arrow* and osteoclast *rightwards arrow*) on the surface

aetiology but can be as much as 53 % after hip arthroplasty [1], although this form does not normally cause significant symptoms. Most of the available literature describes its occurrence after long bone fractures or surgical joint replacements. There have, however, been cases described where the site was more removed from the skeleton; for example, in midline abdominal incisions and within the gastrointestinal tract [4, 5]. Although the exact aetiology is still poorly understood, it is thought that mesenchymal cell transformation to osteogenic cells within connective tissue septa is a possible trigger [1]. However, it has not previously been described within the laryngeal soft tissues.

There have been four reports of bone formation within the larynx but this has been in the context of an osteoma arising from ossified laryngeal cartilage [6–9]. Osteomas themselves are not uncommon in otorhinolaryngology, frequently arising from the skull, facial bones and paranasal sinuses. Central and peripheral variants arise from a proliferation of cancellous bone in the endosteum or

periosteum, respectively [9]. The extra-skeletal variant usually develops within muscle, but remains in contact with the underlying bone either by a broad base or a stalk, and therefore is not found freely within the soft tissues [10]. This distinction is important in our case as lamellar and woven bone was found within the superficial stroma and not in association with ossified cartilage. This therefore excluded the main differentials of osteoma and osteosarcoma.

This is also the first reported case of acute airway obstruction secondary to a benign bone-forming lesion precluding intubation. As the patient declined a tracheostomy, his treatment required rapid microlaryngoscopic assessment and CO₂ laser excision. As ventilation was sufficient via supraglottic jetting, the need for a tracheostomy was ultimately avoided. This approach allowed for direct instrumentation of the mass and laryngeal function was preserved.

The CT findings also raised the possibility of pulmonary tuberculosis as the precipitant for this ossification. Diffuse pulmonary ossification has been reported before in pulmonary tuberculosis and the case involved had also failed to complete the full course of antibiotic treatment [11, 12]. However, multiple cultures, bronchial washings and blood tests failed to yield any acid fast bacilli, despite the patient's previous failure to complete antibiotic therapy. This excluded tuberculosis as a cause. The initial culture of *K. oxytoca* and *E. cloacae* was thought to be due to obstruction of the normal airway clearance pathways, and not causal.

Conclusion

Heterotopic ossification in the larynx presents a significant diagnostic and therapeutic challenge. We describe the first known case of its kind and a management that resulted both in preservation of laryngeal function, and a rapid recovery with return to normal life for the patient.

Conflict of interest None.

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