An Uncommon Cause of Stridor
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Abstract
An adult male patient presented to an Accident and Emergency Department with stridor, and was diagnosed clinically to have infective exacerbation of chronic obstructive pulmonary disease. A CT scan of his neck revealed a large mass occluding the laryngeal airway. A biopsy and subsequent histopathological analysis of this mass revealed no evidence of a malignancy. Despite this, a laryngectomy was undertaken and a large verrucous squamous cell carcinoma was confirmed.

CASE REPORT
A 59 year old Caucasian male attended the Accident and Emergency Department at the University of Wales Hospital, Cardiff, complaining of severe and worsening shortness of breath, with a three month history of a chronic non productive cough and loss of appetite.

One week prior to his emergency admission, he had been reviewed by his general medical practitioner regarding an acute onset of a productive cough which had resulted in the prescription of a course of amoxicillin.

Despite an unremarkable medical history, he had smoked 20 cigarettes a day for most of his adult life, but had given up 4 months previously. The patient had also been suffering from hoarseness of the voice for 4 years, but this had not been investigated.

On examination, the patient was tachypnoiec with stridor, and had such difficultly speaking that he was unable to finish sentences. Auscultation revealed poor air entry bilaterally with diffuse wheezes. Cardiovascular, abdominal and neurological examination was unremarkable.

His initial oxygen saturation was 91% on air. Repeated arterial blood gas analysis displayed a pattern of deteriorating respiratory acidosis. His white blood cell was elevated at 13.1x109/l and his CRP was 64mg/l. The remaining haematological investigations were within normal range. A chest radiograph showed opacification peripherally in the left mid zone. A CT pulmonary angiogram showed infective consolidation of left upper and lower lobes and small airway disease of both lung bases, but no evidence of pulmonary embolism or malignancy.

These clinical and radiological findings led to a diagnosis of infective exacerbation of chronic obstructive pulmonary disease.

The patient deteriorated and was transferred to the Intensive Care Department where he was subsequently intubated. Intubation itself was straightforward. At this point, in light of the history of hoarseness of voice, an opinion was sought from the Ear Nose and Throat Department. As part of their assessment a CT scan of the neck was requested.

A CT scan was undertaken using Niopam 300mg/l contrast given according to the 120/120 technique. This revealed an 11cm soft tissue mass extending from the nasopharynx to below the thyroid isthmus (Figures 3 and 4). This mass compromised the patient’s airway for all of its length and abutted the lateral and posterior walls of the pharynx. The loss of a clear tissue plane between the mass and the posterior oropharynx strongly suggested involvement (Figure 1). The soft tissue lesion extended posteriorly around the greater horn of the hyoid bone. There was also involvement of the strap muscles of the neck (Figure 2), as well as subcutaneous striations. The thyroid and cricoid cartilages showed altered texture suggesting involvement, and both the nasogastric and endotracheal tubes were displaced by the mass. No pathological cervical lymphadenopathy was observed, although ultrasound examination of the neck revealed a prominent, right side level II reactive node.

In light of the clinical presentation, the radiological diagnosis was more suggestive of an infective aetiology, although malignancy could not be excluded.
The patient underwent a tracheostomy and panendoscopy. Biopsies from the larynx, naso and oropharynx, revealed no infective elements or dysplasia, and no tissue diagnostic of malignancy, though some fragments of tissue showed papilloma-like architecture.

The clinical and radiological findings were discussed extensively at both local and regional Head and Neck Multi Disciplinary Meetings, and a laryngectomy was advised and undertaken. Histological assessment of the operative specimen revealed a completely excised verrucous squamous cell carcinoma of the larynx. The patient underwent a course of radiotherapy.

At a review appointment 8 months after laryngectomy the patient complained of increasing malaise and weight loss. Although a CT of the neck showed no disease recurrence, a chest radiograph showed bilateral cavitating mass lesions, consistent with metastases.

The patient was subsequently referred for palliative care.

**Figure 1**
Figure 1: Post contrast axial CT at level of oropharynx. A soft tissue mass completely occludes the airway. The lesion abuts both the endotracheal and nasogastric tubes

**Figure 2**
Figure 2: Post contrast axial CT at the level of the thyroid cartilage. The soft tissue mass continues to occlude the airway. Involvement of the strap muscles of the neck is also seen

**Figure 3**
Figure 3: Post contrast axial CT at the level of the thyroid. The inferior border of the soft tissue lesion extends into the subglottic region
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Figure 4
Figure 4: Post contrast sagittal CT displaying a soft tissue mass occluding the naso, oro and hypopharynx. The endotracheal and nasogastric tubes are also seen.

Figure 5
Figure 5: Chest radiograph of patient 3 months post completion of surgery and radiotherapy. Multiple cavitating pulmonary lesions seen. These appearances are strongly suggestive of metastases.

DISCUSSION
Verrucous squamous cell carcinoma (VSCC) of the upper aerodigestive tract is not uncommon, but is rare in the larynx, accounting for 1-3% of laryngeal carcinomas (1). These tumours may be confused for a benign condition, and if left untreated can become locally aggressive. It is most prevalent in smokers and males. The most common presentation is that of hoarseness of voice and dysphagia (1). This patient presented with stridor and respiratory failure necessitating intubation, something which the authors believe has not been reported in the medical literature before.

In this case, an initial clinical diagnosis of infective exacerbation of COPD was made and supported by the findings of the chest radiograph and CT pulmonary angiogram. The subsequent CT neck findings, combined with the clinical presentation, were more suggestive of an infective cause of neck swelling, although malignancy had been considered. The final histology was surprising. VSCC can be difficult to diagnose both clinically and histopathologically. Indeed, a study by Orvidas et al (1) found that 52% of patients with malignant VSCC had initial benign histopathology. The difficulty in obtaining a confirmed histological diagnosis has implications in how quickly the disease can be treated.

Conventional treatment involves surgical resection. The use of radiotherapy has been widely discussed in the literature, with concern raised over the association with increased recurrence rates (2), the incidence of regional metastasis (3) and the risk of anaplastic transformation (2).

Recurrence of VSCC is rare (1). According to Ferlito and Recher (10), there are no reported cases of pure verrucous carcinoma metastasizing. In light of the unusual behaviour of this tumour, the pathology was reviewed. Histology did not rule out a hybrid VSCC. This is relevant as hybrid varieties can behave more aggressively than pure verrucous carcinoma (1). Whether radiotherapy could have contributed to this lesion metastasizing is controversial. Despite the findings of Edstrom et al (3), a study by Huang et al (11) suggests that this treatment modality has no effect on incidence of metastasis. Full discussion of this is not within the scope of this case report.

The initial histopathology results of the biopsies taken from this patient revealed papilloma-like growths, raising the possibility of human papilloma virus (HPV). HPV, commonly seen in cervical cancer, has been linked with head and neck cancer for over 20 years (4). A study by Termine et al showed 34.5% of head and neck squamous cell
carcinomas to be linked with HPV (5). Of the numerous HPV types, numbers 16 and 18 are most often associated with head and neck malignancy, although types 6 and 11 have been linked with verrucous tumours (6). Confirmation of presence of HPV is ascertained via p16 immunochemistry, polymerase chain reaction and in situ hybridisation tests. Cigarette smoking can reduce p16 expression and so affecting reliability of results (7). These tests were not undertaken in this case, because the patient had only recently stopped smoking. The diagnosis of HPV related head and neck cancer is important as it tends to respond better to chemotherapy (8), rather than surgery, and it is associated with improved 2 year survival rates (9).

CONCLUSION
This case discusses an unusual cause of stridor in a patient attending an Accident and Emergency Department. Although the authors are not suggesting including verrucous carcinoma in the differential diagnosis of all patients with stridor, this report highlights the advantages of exploring all aspects of the patient’s history, including, as in this case, those mentioned in passing.

References
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