The Journal of Laryngology & Otology
October 2001, Vol. **115,** pp. 856–858

Inflammatory myofibroblastic tumour of the larynx

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Abstract
A case of inflammatory myofibroblastic tumour of the larynx in a 74-year old man is reported. The lesion presented as a polypoid tumour arising in the left true vocal fold. The patient died of non-tumour-related causes and the autopsy showed persistence of the laryngeal tumour. Multicentricity and distant metastases were not detected. The literature about this entity and its differential diagnosis is reviewed and briefly commented upon.

Key words: Laryngeal Neoplasms; Neoplasms; Muscle Tissue

Introduction

Inflammatory myofibroblastic tumour (IMT) is a rare fibro-inflammatory space-occupying lesion whose exact nature is only beginning to be elucidated.[1] Lung, liver, gastrointestinal tract (omentum and mesentery) and central nervous system are the organs most commonly affected. This tumour rarely affects the head and neck, if it does the upper aerodigestive tract and orbit are the most commonly involved sites.[2,3] To date, only a few cases have been reported in the larynx,[4,5] and some of them have been referred to in the literature as nodular fasciitis.[6]

We present an IMT arising in the left true vocal fold in a 74-year-old man. The lesion was incompletely excised with conservative surgery. The patient died of cardiac failure two months later. The autopsy confirmed both the cause of death and the initial diagnosis of the laryngeal tumour. Multifocality, locoregional and distant metastases were not demonstrated.

Case report

Clinical data

A 74-year-old man treated with haemodialysis due to chronic renal failure presented with hoarseness, foreign body sensation, and severe dyspnoea. Symptoms had started several months before and increased slowly but relentlessly. Physical examination revealed a tumour on the left vocal fold that nearly completely occluded the airway. Computed tomography (CT) scans did not show associated locoregional lymphadenopathies (Figure 1(a)). The precarious medical situation of the patient predicated avoidance of general anaesthesia and radical surgery. For this reason, only tracheotomy and several partial resections of the tumour were performed. The specimens were histologically diagnosed as 'congruent with inflammatory myofibroblastic tumour'. Two months after the last local resection, the patient died of cardiac failure. A complete postmortem was performed. No previous history of local trauma was recorded in the clinical records.

Pathological findings

Grossly, the tumour was firmly attached to the left true vocal fold and presented as a polypoid shape, measuring 3.5 cm in diameter. On cut section the tumour was fibrous and non-invasive. (Figure 1(b)).

Microscopically, the tumour was covered by benign squamous epithelium with focal keratosis and superficial erosions. The tumour itself was composed of spindle or stellated cells with some nuclear irregularities, nucleoli, and basophilic cytoplasm (Figure 2(a)). Mitotic count displayed 2–3 mitoses/10 high power fields. The interspersed stroma was heavily hyalinized, sometimes myxoid, and contained blood vessels and chronic inflammatory infiltrates. Discrete zonation/maturation areas were seen, the more cellular areas being located under the surface epithelium.

Immunohistochemically, proliferating cells disclosed intense positivity with vimentin and weak positivity with actins (muscle-specific and smooth muscle-specific). S-100 protein desmin, CD34 and keratins (AE1-AE3) were consistently negative (Figure 2(b)). The postmortem also demonstrated absence of both tumour multifocality and metastatic seeding.

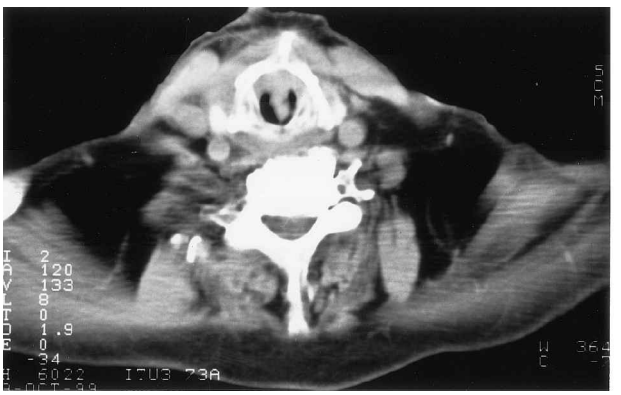

Fig. 1 (a)
CT scan showing a large intralaryngeal polypoid mass.

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Accepted for publication: 26 April 2001.

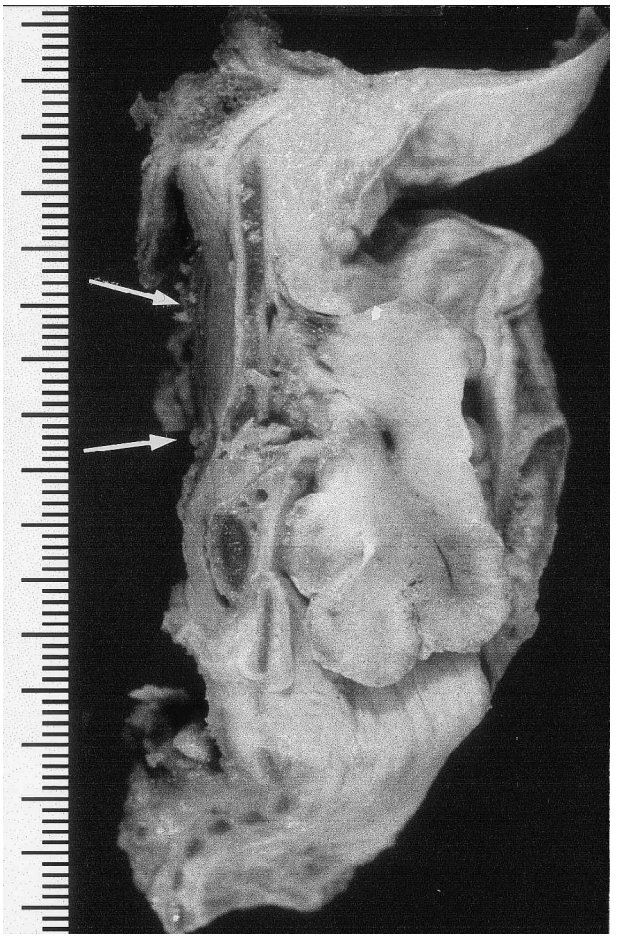

FIG. 1 (b)

Macroscopic midsagittal section revealing a polypoid tumour that arises in the left true fold. Note the lack of laryngeal wall infiltration at the origin (arrows).

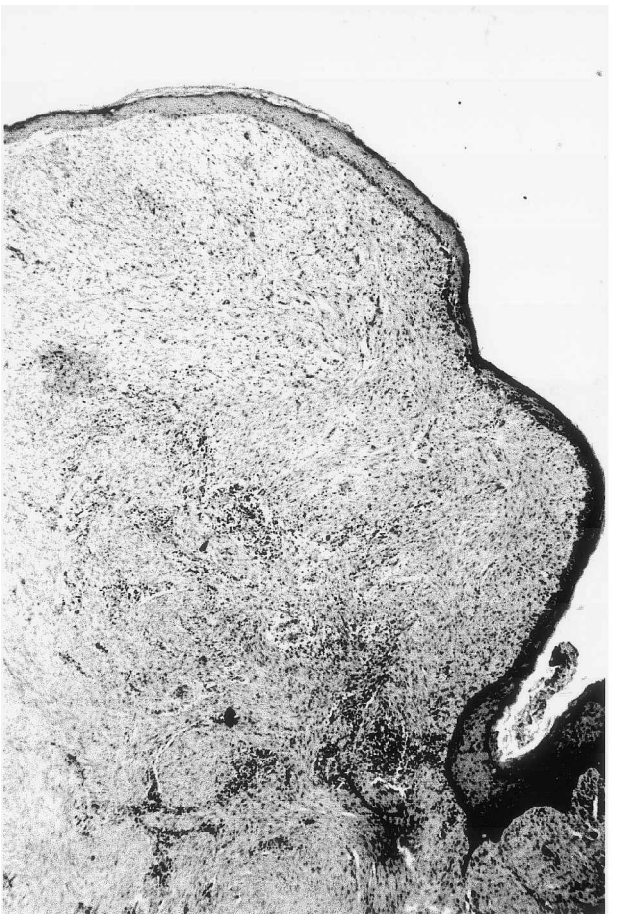

FIG. 2 (a)

A low power view of the tumour displays a spindle-cell proliferation with focal storiform pattern and myxoid areas growing under a benign surface epithelium (H&E; 30).

Discussion

IMT is a confusing and ubiquitous entity of unknown aetiology that has received several names in the literature, i.e., inflammatory pseudotumour, plasma cell granuloma, and pseudosarcomatous myofibroblastic lesion. For decades, the entity was well recognized in the lung, where it pursues a benign course. However, when occurring in extrapulmonary sites, IMT is more unpredictable with some documented cases presenting local aggressiveness and even metastasis.[2] In an attempt to organize this heterogeneous group of lesions, Batsakis *et al.*[2] considered two different types of lesion within this generic term: inflammatory/reparative and myofibroblastic proliferations. The classic plasma cell granuloma of the lung may be included within the first group. The latter group are the main concern for pathologists because they may present with either benign, locally aggressive, or even with overtly malignant behaviour, and the evolution in every case cannot always be predicted with certainty on histological grounds.

IMT has been described recently in the larynx,[4,5] although by 1984 some authors[6] had already identified a case of laryngeal nodular fasciitis histologically similar to IMT. Most of the reported cases are polypoid masses that arise in the true folds of the larynx in males. Frequent symptoms are hoarseness, foreign body sensation, dyspnoea and stridor. Surgical excision with free margins is the treatment of choice.

The tumour is composed of spindle and stellate cells lying in a hyalinized stroma with some myxoid-appearing areas. Usually, vimentin and actin show overtly positive immunostaining. The case we present recapitulates the previously reported morphological features of IMT. Similarly to cases reported by Wenig *et al.*[4] and Corsi *et al.*,[7] the tumour recurred locally and even persisted as seen at postmortem, but did not display metastatic dissemination.

In the clinical context of a polypoid endolaryngeal mass that may show some degree of anisomorphism, the main differential diagnosis is spindle-cell squamous cell carcinoma. Usually, the first contact that pathologists have with this entity is through an endoscopic biopsy. In such small tissue fragments this differential diagnosis remains a challenge[1,4]. In fact, cell pleomorphism and keratin positivity, that has been occasionally reported[1] , may favour, erroneously, a diagnosis of spindle cell carcinoma. Histological clues that support the diagnosis of IMT are: proliferating cells within a loose stroma, cells haphazardly arranged without definite nests or bundles, absence of true infiltrating growth, absence of true atypia and atypical mitoses, and also lack of dysplastic changes in the overlying epithelium.[4] Other less common malignant neoplasms that must be ruled out are the wide spectrum of sarcomas, i.e., malignant fibrous histiocytoma and leiomyosarcoma.

In spite of the fact that IMT is a well-known morphological entity, its exact nature and behaviour must still be delineated. Most probably, it should be

https://doi.org/10.1258/0022215011909189 Published online by Cambridge University Press

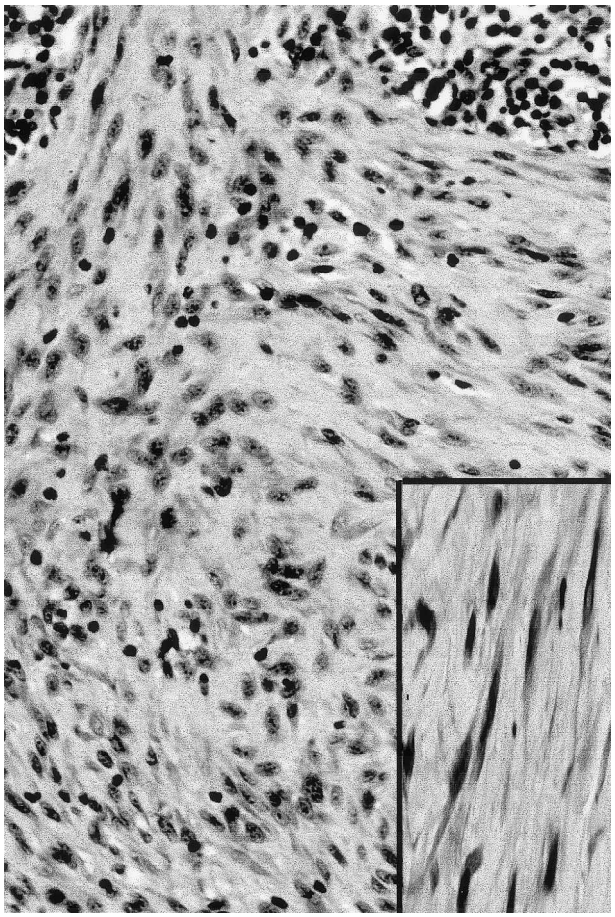

FIG. 2 (b)

Positive immunoreaction of proliferating cells with muscle specific actin (H&E; ×140) Inset: Cellular detail shows benign elongated elements with hyperchromatic nuclei and fibrillar cytoplasm.

considered as a stromal tumour with undetermined or low biological aggressiveness[3] related to inflammatory fibrosarcoma.[1,8]

References

1 Brandwein MS, Kapadia SB, Gnepp DR. Inflammatory myofibroblastic tumor (inflammatory pseudotumor, plasma cell granuloma). In: Gnepp DR. ed. *Diagnostic Surgical Pathology of the Head and Neck.* Philadelphia: W.B. Saunders Co., 2001:287–8

2 Batsakis JG, Luna MA, El-Naggar AK, Goepfert H. Inflammatory pseudotumor. What is it? How does it behave? *Ann Otol Rhinol Laryngol* 1995;**104**:329–31

3 Coffin CM, Humphrey PA, Dehner LP. Extrapulmonary inflammatory myofibroblastic tumour: A clinical and pathological survey. *Sem Diagn Pathol* 1998;**15**:85–101

4 Wenig BM, Devaney K, Bisceglia M. Inflammatory myofibroblastic tumour of the larynx. A clinicopathologic study of eight cases simulating a malignant spindle cell neoplasm. *Cancer* 1995;**76**:2217–29

5 Kendall CH, Johnston MN. Pseudomalignant laryngeal nodule (inflammatory myofibroblastic tumour) *Histopathology* 1998;**32**:286–7

6 Jones SR, Myers EN, Barnes L. Benign neoplasms of the larynx. *Otolaryngol Clin North Am* 1984;**17**:151–78

7 Corsi A, Ciofalo A, Leonardi M, Zambetti G, Bosman C. Recurrent inflammatory myofibroblastic tumour of the glottis mimicking malignancy. *Am J Otolaryngol* 1997;**18**:121–6

8 Meis JM, Enzinger FM. Inflammatory fibrosarcoma of the mesentery and retroperitoneum. A tumor closely simulating inflammatory pseudotumor. *Am J Surg Pathol* 1991;**15**:1146–56

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Dr C. Ereño takes responsibility for the integrity of the content of the paper.
Competing interests: None declared