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Malignant fibrous histiocytoma of the larynx

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Abstract
Malignant fibrous histiocytomas of the upper respiratory tract are rare, aggressive mesenchymal neoplasms. We report a case of a glottic malignant fibrous histiocytoma of the larynx on a 54 year old man. Only a few have been reported in the English literature. The clinical behaviour and degree of malignancy of these tumours cannot be predicted. Wide, aggressive excision of the tumour with a margin of normal tissue appears to be the treatment of choice. About two years after total laryngectomy the patient is well and free of disease.

Introduction

Malignant fibrous histiocytoma is a tumour of mesenchymal origin, composed of several types of cells including fibroblasts, histiocyte-like cells and atypical giant cells. The term 'fibrous histiocytoma' was coined by Kauffman and Stout (1961) and O'Brien and Stout (1964) to describe a histiocytic-appearing tumour with fibrous tissue. The origin of this tumour is believed to be a pleuri-potential cell, revealing a histiocyte that, under appropriate conditions, could assume the function of a fibroblast, this accounting for the dual population of cells commonly seen in this tumour (Blitzer *et al.* 1977; Ogura *et al.*, 1980; Godoy *et al.*, 1986).

Histologically, the malignant fibrous histiocytoma is divided into five subtypes: storiform-pleomorphic, myxoid, inflammatory, giant cell, and angiomatoid (Barnes and Kanbour, 1988).

Because of this varied histological appearance and complex histopathological features, several names have been used to describe these tumours, fibrous xanthoma, xanthofibrosarcoma, dermatofibrosarcoma protuberus, xanthofibroma, nodular fibrosis.

The fibrous histiocytomas occurring in the deep soft tissues are usually malignant. Approximately 3 per cent of all lesions have occurred in the head and neck area (Ogura *et al.* 1980). In the larynx this tumour is rare. Most cases have been reported in recent years. Perhaps, because the term and entity of malignant fibrous histiocytoma has only recently been recognised, many tumours previously diagnosed as liposarcoma, fibrosarcoma, or rhabdomyosarcoma were probably examples of malignant fibrous histiocytoma. New histochemical techniques and electron microscopy lead to a more accurate histopathological diagnosis. Recurrences can be related to the inadequacy of the surgical procedure and to the size and histological grade of the neoplasm.

Case report

A 54-year-old male was admitted in December 1988 with a three month history of hoarseness for further investigation. He was a smoker of 50 cigarettes a day and heavy drinker with a past history of chronic alcoholic liver disease.

Indirect laryngoscopy showed a reddish polypoid mass in the middle third of the right vocal cord. The mobility of the vocal cords was normal. Enlarged lymph nodes could not be palpated.

X-ray of the chest, lateral X-ray of the neck and tomography of the larynx were unremarkable. A blood film and blood chemistry were also normal.

On 17 January 1989, a direct laryngoscopy and biopsy were performed. Microlaryngoscopy revealed a lesion 1.2 cm in diameter located on the right true vocal cord, extending to the anterior commissure and invading the laryngeal ventricle. There was no involvement of the posterior commissure. Several biopsies of the lesion were done. The histopathological diagnosis was: 'malignant fibrous histiocytoma'. The patient subsequently underwent a total laryngectomy. At the same surgical time primary tracheoesophageal puncture was performed. The post-operative period was normal and there were no complications. A voice prosthesis was fitted 15 days following surgery. The patient was discharged home on the 20th post-operative day. Fifteen months after surgery the patient is alive and free of disease.

Histopathology

The surgical specimen of total laryngectomy showed a pedunculated lesion measuring 1.5 × 1.2 cm, arising from the middle third of the right vocal cord and reaching the anterior commissure. The mass partially fills the laryngeal ventricle and the base of the pedicle is 7 mm wide. The remainder of the larynx was normal (Figs. 1, 2). Several sections were taken from the tumour. The cut section of the growth was whitish in colour and essentially homogeneous. Haematoxylin and eosin stains were done first, followed by special stained techniques such as reticulin, Masson trichome, avidin-biotin complex immunoperoxidase technique for keratin, AE-1/AE-3 (negative), vimentin (positive), myoglobin, actin, S-100 protein (negative) and alpha-1-antichymotrypsin; testing with carcino embryonic antigen was negative.

The non-keratinized squamous epithelium of the vocal cord was thin due to tumoral compression. The tumour was formed of cells which varied considerably in size, number, and appearance depending on the field observed. There were some areas with bundles of spindle shaped cells oriented in a pin-wheel fashion forming a storiform pattern. In other areas, accumulation of multinucleated bizarre giant cells were seen showing an osteoclast-like appearance with numerous nuclei and spacious eosinophilic cytoplasm (Fig. 3). Images of transition between both areas were observed. In the fields in which giant cells were

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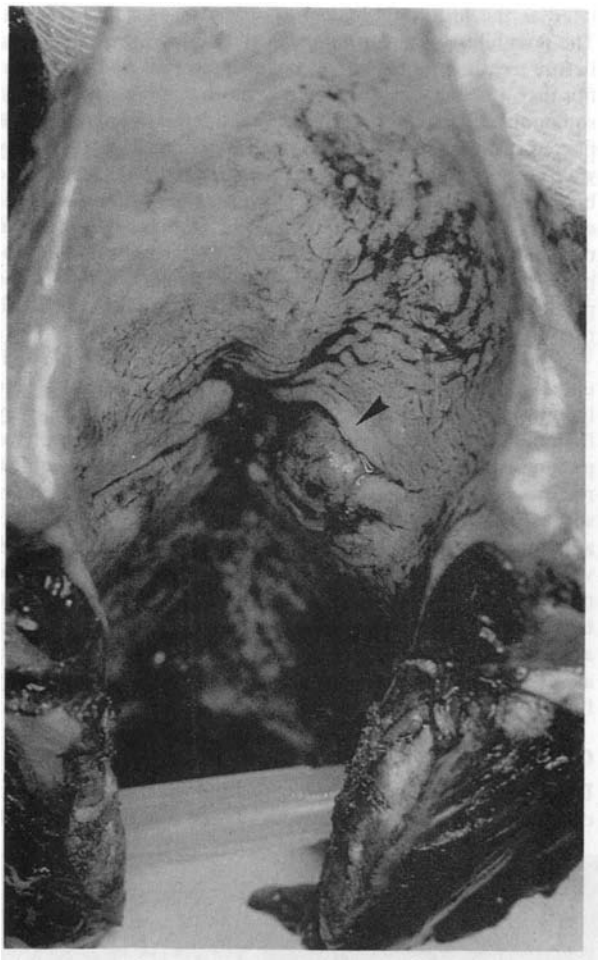

FIG. 1

Surgical specimen showing growth attached to the right vocal cord (arrow).

in predominance, a large number of congested blood vessels were seen. Small peripheral parts of the tumour showed sclerosis with thick bundles of collagen fibrils (Fig. 4). In a few peripheral areas lymphocytic infiltration, not associated with tumour necrosis, was observed.

The neoplastic cells showed considerable nuclear pleomorphism and there were numerous other atypical mitotic figures present.)

The tumour was well delimited and had not infiltrated muscle, cartilage or other adjacent structures.

The histopathological predominance of osteoclastic giant cells in the tumour suggests the diagnosis of 'malignant fibrous histiocytoma—giant cell subtype—of the larynx'.

Discussion

Malignant fibrous histiocytoma is an uncommon tumour characterized by unpredictable clinical behaviour, a high rate of recurrence, and a tendency to regional and distant metastases. Less than 20 cases of laryngeal malignant fibrous histocytoma have been reported in the literature, constituting one per cent of all malignant tumours at this site (Ferlito *et al.*, 1983). These tumours frequently go unrecognized until they reach a voluminous size. A high index of suspicion is necessary for recognizing this aggressive tumour.

Malignant fibrous histiocytoma of the larynx has an aggressive behaviour, with quick growth and a remarkable tendency to local recurrence. Cervical node and distant metastases, to organs such as the brain or lung, are also common. The prognosis is usually considered to be poor once metastases are identified (Canalis *et al.*, 1975; Ferlito *et al.*, 1983; Godoy *et al.*, 1986; Barnes and Kanbour, 1988).

The diagnosis of this tumour may be difficult, particularly when the histopathological diagnosis is made on a small biopsy specimen. Multiple biopsies are frequently required, delaying definitive diagnosis and treatment. Many times, the correct diagnosis is not established until after complete surgical excision has been performed and the pathologist has studied the entire surgical specimen (Ogura *et al.*, 1980). Ultrastructural analysis using electron microscopy, histochemical staining techniques and tissue culture methods, help in establishing the diagnosis of malignant fibrous histiocytoma. These methods are also helpful for excluding other soft tissue sarcomas.

The differential diagnosis must be made from several neoplasms, e.g. pleomorphic rhabdomyosarcoma, fibrosarcoma, spindle cell squamous carcinoma, angiosarcoma, haemangiopericytoma, pleomorphic liposarcoma and lymphomas. Special histochemical staining techniques such as Masson trichrome, PTAH, and reticulin help in differentiating these tumours, especially in the most difficult cases. Immunohistochemical techniques both confirm the diagnosis of malignant fibrous histiocytoma and help to eliminate other possibilities. It is particularly difficult to differentiate a malignant fibrous histocytoma from spindle cell carcinoma, in the absence of microinfiltration, or when the malignant fibrous histiocytoma traces a storiform pattern (Blitzer *et al.*, 1977; Ogura *et al.*, 1980; Godoy *et al.*, 1986; Barnes and Kanbour, 1988).

A review of the literature reveals that most of the cases reported until now have been found in the subglottic area (Ferlito *et al.*, 1983; Godoy *et al.*, 1986; Barnes and Kanbour, 1988). The location of the case reported here represents a very

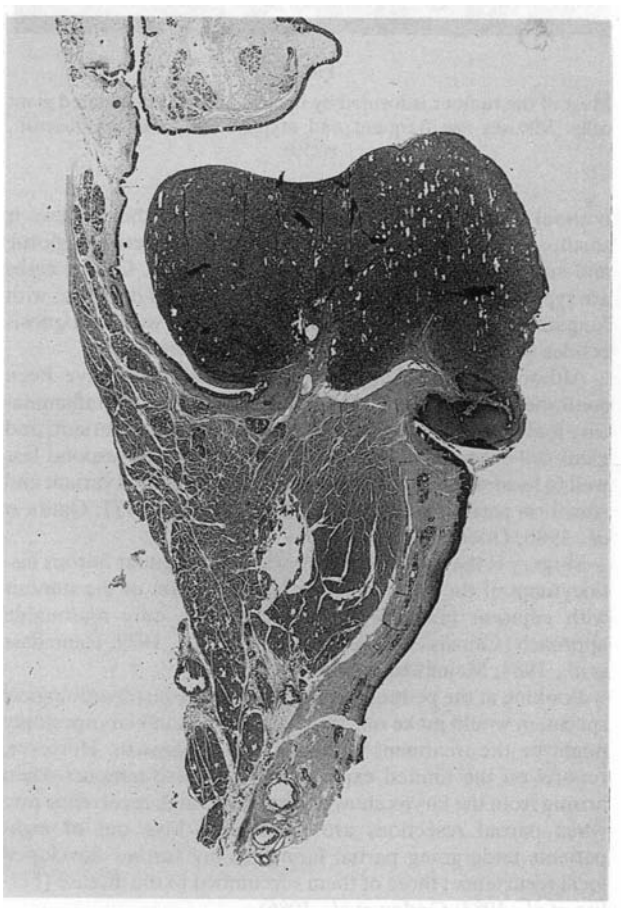

FIG. 2

Low-power photomicrograph demonstrating the polypoid neoplasm arising by a pedicle from the right vocal cord. The mass fills the laryngeal ventricle.

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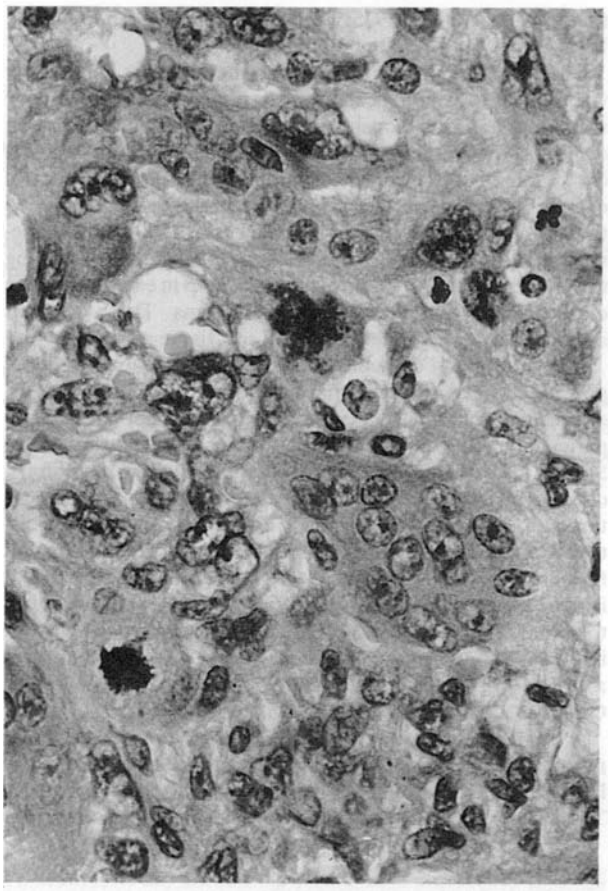

FIG. 3

Most of the tumour is formed by bizarre and multinucleated giant cells. Mitoses are frequent and atypical. (Haematoxylin-eosin, ×250).

unusual site: the vocal cord. Although the number of cases is small, several differences have been reported between glottic and subglottic malignant fibrous histiocytoma. Glottic cases are typical of older male patients, smokers and drinkers, with fungating lesions. There is a suggestion that a worse prognosis resides in the glottic group (Godoy *et al.*, 1986).

Although, no clinicopathological correlations have been confirmed for head and neck lesions, it is felt that the inflammatory malignant fibrous histiocytoma, pleomorphic variant, and giant cell subtype tend to metastasize early and respond less well to local surgical therapy alone than the myxoid variant and storiform pattern predominance (Blitzer *et al.*, 1977; Ogura *et al.*, 1980; Godoy *et al.*, 1986).

Surgery is the treatment of choice for malignant fibrous histiocytoma of the larynx. An *en bloc* resection of the tumour with adjacent musculofascial plane is the only reasonable approach (Canalis *et al.*, 1975; Kennan *et al.*, 1979; Ramadass *et al.*, 1984; Majumder *et al.*, 1989).

Looking at the pedunculated nature of the histopathological specimen would make one think whether a total laryngectomy might be the treatment for this particular growth. However, reports on the limited experience with these tumours when arising from the larynx show an extremely high recurrence rate when partial resections are performed. Five out of eight patients undergoing partial laryngectomy further developed local recurrence; three of them succumbed to the disease (Ferlito *et al.*, 1983; Godoy *et al.*, 1986).

Both indirect and direct laryngoscopy showed a bulky mass in the vocal cord apparently invading the laryngeal ventricle. The mass in the right vocal cord is also evident in the surgical specimen (Fig. 1). Clinically the lesion was not as pedunculated as the histopathological examination revealed (Fig. 2). The possibility of performing a partial resection was discussed before treating the patient. It must be stressed that malignant fibrous histiocytoma behaves more aggressively than squamous cell carcinoma. Even though few cases of malignant fibrous histiocytoma of the larynx have been reported, it may well be said that in this organ the tumour has an aggressive behaviour. Local recurrences as well as metastases to other organs or to lymph nodes are frequent. A thorough review of the literature, along with the clinical aggressiveness of the neoplasm and the information provided by the examination in this particular case decided us to select a more radical approach than that needed for a squamous cell carcinoma.

Cervical neck dissection does not appear to be indicated, unless the clinical examination is suggestive of metastatic lymph nodal involvement. Radiotherapy seems to have very limited efficacy with these tumours and should be reserved for patients with high surgical risks, patients with non-surgical recurrences, or cases with distant metastases. Some authors suggest that polychemotherapy in other soft tissue sarcomas apparently increases the disease-free period and overall survival (Blitzer *et al.*, 1977; Ogura *et al.*, 1980). However, the number of patients with malignant fibrous histiocytoma treated by chemotherapy is too small to assure the benefits of this approach in lesions located in the head and neck. In order to assess the real value of adjuvant treatments to radical surgery further studies are required.

Conclusion

Despite its location and the histopathological diagnosis, our case should have a good prognosis. This is due to: (1) its small

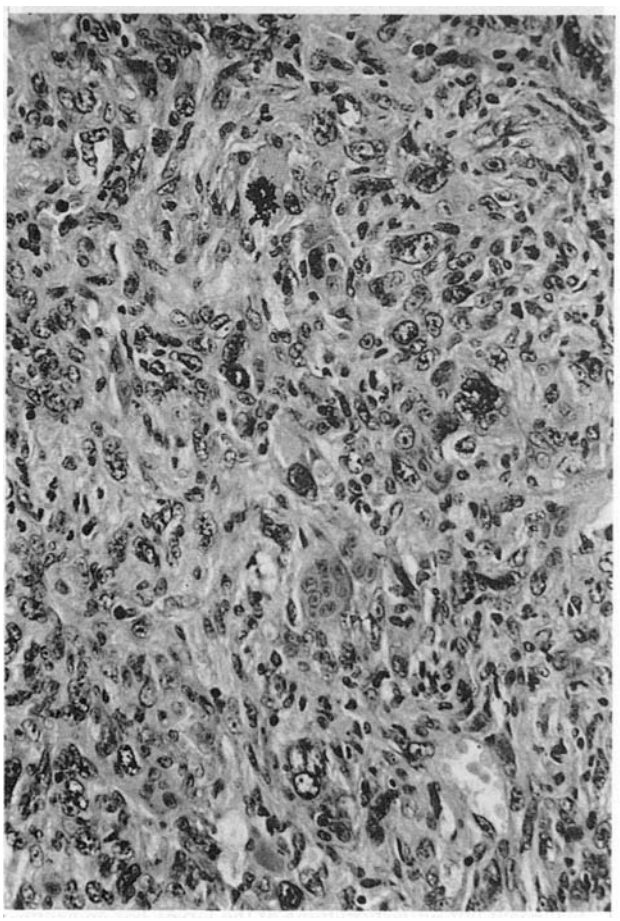

FIG. 4

Note the admixture of fusiform cells with frequent mitoses and giant cells. Bundles of immature collagen fibrils surround the cells. (Haematoxylin-eosin, ×60).

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size; (2) the absence of invasion to adjacent structures; and mainly (3) the wide margin of safety in tumour resection.

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