

## Malignant fibrous histiocytoma of the tongue

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### Abstract

A case of malignant fibrous histiocytoma, which developed in the tongue of a 16-year-old girl is presented. Surgical excision with, or without, radical neck dissection is the treatment of choice of this type of lesion.

**Key words:** Sarcoma; Histiocytoma, Fibrous; Tongue

### Introduction

Malignant fibrous histiocytoma (MFH) is a high grade and aggressive sarcoma originally described by O'Brien and Stout in 1964.<sup>1</sup> It is now considered the most common soft tissue sarcoma in adults.<sup>2</sup> Although MFH commonly develops in the extremities and retroperitoneum, cases in essentially all regions of the body including the head and neck have been reported. It has been estimated that approximately one to three per cent of these tumours occur in the head and neck region.<sup>3</sup> Only two cases with the tongue as a primary site of MFH have been previously reported.<sup>4,5</sup> The present report describes a further case.

### Case report

A 16-year-old Chinese girl was referred to the Kaohsiung Medical University Hospital due to a rapidly enlarging painless swelling in the left posterior dorsal tongue of five days duration (Figure 1). The lesion was neither numb nor tender and was aspirated by a local practitioner but no purulent exudate could be drawn. Her general medical history was unremarkable. Clinical examination revealed a well-defined swelling about 2 × 2.5 cm in diameter, covered with normal appearing mucosa except for a small wound due to aspiration. The swelling was rubbery in consistency and fixed to the underlying connective

tissue. No limitation of tongue movement was noted. No regional lymphadenopathy was found. A clinical impression of mesenchymal tumour was made, however, due to the rapid enlargement of the swelling, malignancy was suspected. The lesion was biopsied and suggested microscopically to be giant cell type MFH. She was scheduled for surgical resection of the tumour. A hemiglossectomy with modified radical neck dissection was performed. The margins of the tumour and the neck lymph nodes in the resected specimens were negative for tumour on analysis of frozen sections. The cut surface of the surgical specimen showed a well-delineated tumour surrounded by greyish-white lingual tissue. Histological examination revealed a predominance of multinucleated giant cells, intermixed with fibroblast-like and histiocyte-like cells (Figure 2), displaying prominent pleomorphism and hyperchromatism (Figure 3). Immunocytochemically, many of the pleomorphic polygonal cells stained positively with  $\alpha$ -1-antichymotrypsin but not for keratin, desmin and S-100. Therefore, consistent with the results of the incisional biopsy, a diagnosis of a giant cell type MFH in the tongue



FIG. 1  
Swelling over the left dorsal tongue.

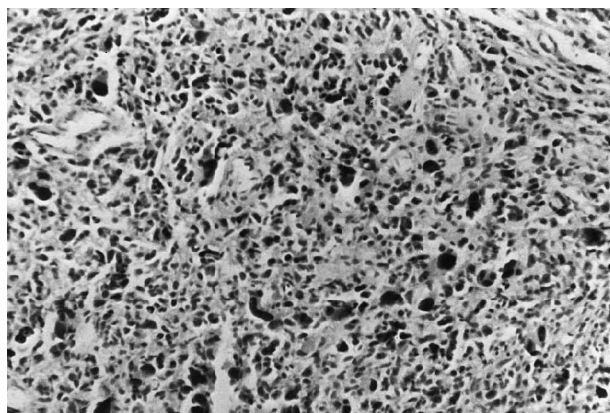


FIG. 2  
Histological examination revealed a predominance of giant cells, intermixed with fibroblast-like and histiocyte-like cells (H & E; ×40).

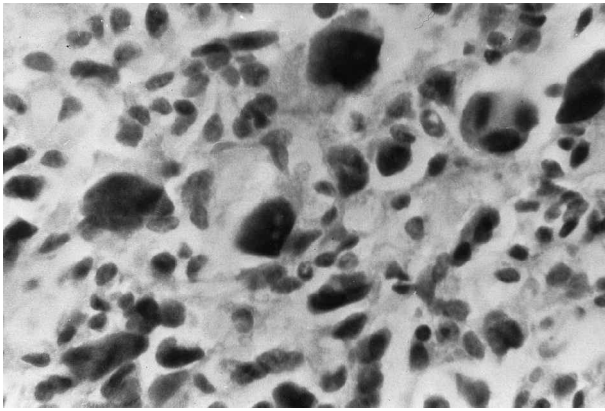


FIG. 3

The giant cells displayed prominent pleomorphism and hyperchromatism (H & E;  $\times 100$ ).

was made. She recovered well post-operatively and remained free of disease at three years follow-up.

### Discussion

Although MFH has been accepted as a distinct clinicopathologic entity, the exact histogenesis of this sarcoma remains uncertain. The majority of investigators have favoured primitive mesenchymal cells such as fibroblasts and histiocyte-like cells as the origin of these tumours.<sup>6</sup> For this case, the histiocyte-like giant cells displayed  $\alpha$ -1-antichymotrypsin staining while keratin, desmin and S-100 were all negative. Thus, the diagnosis of MFH was confirmed by ruling out other sarcomas, myogenic tumour, sarcomatoid carcinoma or melanoma.

Most reported cases of MFH in the oral region have occurred in bone.<sup>7</sup> Excluding cases of post-radiation MFH and cases with the pre-existence of other primary malignancy, only eight cases of MFH arising *de novo* in the oral cavity as solitary soft tissue lesions have been previously described.<sup>1,4,5,8-11</sup> The clinicopathological features of these cases (including the present lesion) are summarized in Table I.

The age of these nine patients at the time of initial treatment ranged from 16 to 73 years with an average of 46 years. The current lesion represents the first such case reported in adolescence. The majority of the patients in this series of MFH were female and the overall male-female ratio was 1:2. The duration of symptoms prior to treatment ranged from five days to two years in the seven patients of this series for whom it was reported.<sup>5,8-11</sup> The most common complaint was of a painless enlarging mass, however, in the case reported by Bailey,<sup>8</sup> a very tender, hot and painful lesion was noted. Only two cases in this series (the present case and that reported by Barnes and Kanbour<sup>4</sup>), presented with a rapidly enlarging swelling. At the time of initial treatment, the size of the tumours in this series ranged from 2 to 9 cm with an average size of approximately 5 cm.

As can be seen from Table I, the primary treatment of choice in this series of MFH patients was surgical excision with, or without, radical neck dissection. Except for the patient in the case reported by Bailey,<sup>8</sup> who was observed for only a short period, the follow-up period of cases in this series ranged from 18 to 24 months. All patients were alive within 18 months after initial treatment, and only one patient had died of the disease at three years after primary treatment.<sup>10</sup> Furthermore, only two patients had recurrence of the disease, both of which had metastasis to the lung.<sup>9,10</sup> It has been suggested that from 25 to 35 per cent of patients with MFH of the head and neck will develop metastasis, most often to the lung.<sup>8</sup> This finding suggests that the prognosis of patients with solitary oral primary MFH may be good.

Microscopically, MFH can be classified into five types: pleomorphic-storiform, giant cell, inflammatory, angiomatoid and myxoid.<sup>10</sup> Of the nine cases of MFH in this series, only storiform-pleomorphic (six patients<sup>4,5,8,9,11</sup>) and giant cell types<sup>1,10</sup> were noted. The giant cell type MFH has been thought to carry the worse prognosis,<sup>10</sup> however, all three cases of giant cell MFH in this series had no evidence of recurrence at a minimum of 18 months follow-up after treatment.

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TABLE I  
REPORTED CASES OF PRIMARY MALIGNANT FIBROUS HISTIOCYTOMA ARISING SOLITARILY IN THE ORAL CAVITY

Authors	Location	Age (yr)/sex	Histology	Symptoms	Period before recurrence	Initial treatment	Follow-up
O'Brien and Stout <sup>1</sup>	Buccal mucosa	50/female	Giant cell	Not reported	No	Excision	NED (24 months)
Brandenburg and Frank <sup>9</sup>	Lower lip	44/male	Storiform-pleomorphic	Not reported	Yes and lung metastasis	Excision	LWD (48 months)
DiLascio and Devlin <sup>11</sup>	Left cheek	20/female	Storiform-pleomorphic	8 months	No	Excision	NED (18 months)
Bailey <sup>8</sup>	Upper lip	54/female	Storiform-pleomorphic	1 month	No	Excision + radiotherapy	NED
Manni <i>et al.</i> <sup>4</sup>	Left tongue	61/male	Storiform-pleomorphic	6 months	No	Excision + M RND	NED (24 months)
Bras <i>et al.</i> <sup>10</sup>	Lower gingiva	73/female	Giant cell	6 months	Yes and lung metastasis	RND	DOD (36 months)
Barnes and Kanbour <sup>4</sup>	Right hard palate	73/female	Storiform-pleomorphic	1 month	No	Excision + RND	NED (48 months)
Barnes and Kanbour <sup>4</sup>	Right tongue	21/female	Storiform-pleomorphic	12 months	No	Excision + M RND	NED (37 months)
Chen <i>et al.</i> (Present case)	Left tongue	16/female	Giant cell	5 days	No	Excision + M RND	NED (36 months)

RND = Radical neck dissection; M RND - Modified radical neck dissection; NED = No evidence of disease; LWD = Living with disease; DOD = Died of disease

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