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Pathology in Focus

Leiomyosarcoma of the tongue: a very rare tumour

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Abstract
We describe a leiomyosarcoma of the tongue in a 60-year-old man. The diagnosis was supported by immunohistochemical positivity for desmin and alpha-1 smooth muscle actin.

Key words: Tongue; Leiomyosarcoma; Immunohistochemistry; desmin; alpha-1 smooth muscle actin

Introduction

Leiomyosarcomas are very rare in the mouth (MacDonald, 1969; Haedicke and Kaban, 1988). A review of the literature revealed only one previous case in the tongue (Yannopoulos and Stout, 1962). This presented as a polypoid lesion at the tip of the tongue in an 11-month-old boy. However, the diagnosis was equivocal as it was not supported by immunohistochemistry or electron microscopy.

Case report

A 60-year-old man presented with an ulcerated mass at the tip of his tongue. This was thought to be an ulcerating squamous carcinoma on clinical examination and a small biopsy specimen was taken, measuring up to 0.4 cm. On histological examination this was found to be ulcerated squamous mucosa and although the base of the ulcer was inflamed and difficult to interpret there was no definite evidence of carcinoma. None-the-less, the lesion did appear to be malignant on clinical examination and the anterior part of the tongue was excised a few days later. On gross examination of this second specimen (Figure 1) the ulcerated region was found to overlie a tumour measuring up to 1.5 cm across. This was composed of cellular fibrous tissue with a vaguely storiform arrangement of spindle cells (Figure 2) and a moderate number of mitotic figures (6 per 10 high power fields).

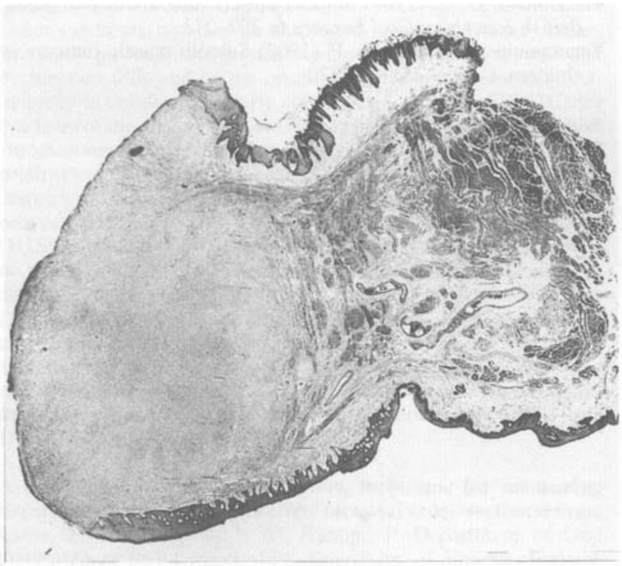

FIG. 1
A low power micrograph of the ulcerated tumour at the tip of the tongue. (H & E; ×5).

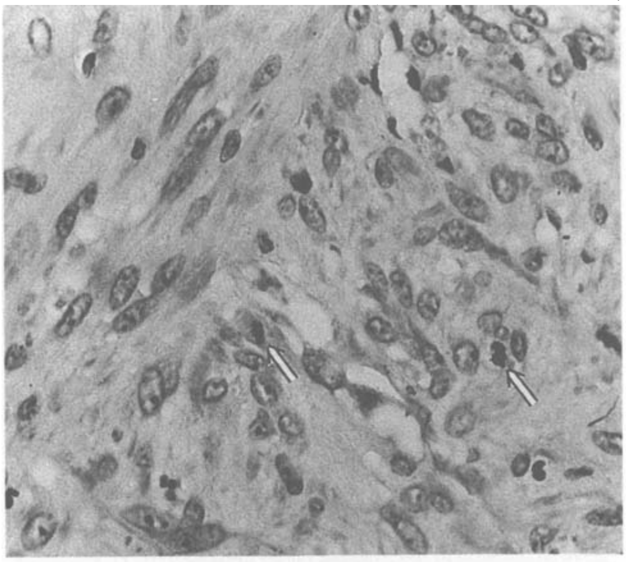

FIG. 2
A high power micrograph of the tumour showing two mitotic figures (arrowed) in this field. (H & E; ×750).

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FIG. 3

A micrograph of the tumour that has been immunostained for alpha-1 smooth muscle actin. The tumour (T) shows strong positivity as does the smooth muscle of the blood vessels (V) in the adjacent connective tissue. (Anti-alpha smooth muscle actin immunoperoxidase; ×50). The inset shows a high power view of the positive immunostaining of the tumour cells. (×250).

There was some nuclear pleomorphism but no necrosis. At its periphery the tumour infiltrated the connective tissue of the tongue but did not extend to the margins of excision. The differential diagnosis was between a spindle cell carcinoma and a low grade sarcoma. The absence of any epithelial dysplasia on the surface of the tongue along with negativity for two cytokeratin antibodies (CAM 5.2 and MNF 116) made the latter diagnosis more likely. Further immunostaining showed positivity for vimentin and desmin but not for S100 protein. On referral for an expert opinion these results were repeated but there was also positivity for alpha-1 smooth muscle actin (Figure 3). Immunostaining for desmin is accepted as being restricted to tissues showing smooth muscle or striated muscle differentiation and alpha-1 smooth muscle actin is said to be specific for smooth muscle differentiation. From these results a diagnosis of leiomyosarcoma of the tongue was made. The patient has been followed-up monthly for a year with no evidence of recurrent disease and no further treatment.

Discussion

Primary leiomyosarcomas of the tongue are very rare and we were able to find only one (equivocal) previous report (Yannopoulos and Stout, 1962). The possibility that our case was a metastatic tumour from another site must be considered but this is unlikely as no such site was identified, there was no evidence of other metastatic disease on clinical examination or chest X-ray and the patient was still healthy one year after treatment. As only one previous report is available it is difficult to predict how the tumour will behave. In the previous case there was no evidence of recurrence four years and eight months after resection. In the uterus and gastrointestinal tract, where leiomyosarcomas are less rare, complete local excision of a low grade leiomyosarcoma, eqivalent to the one that we describe here, is usually curative. However, there are exceptions in which metastatic disease occurs.

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