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Solitary fibrous tumour arising from sublingual gland: report of a case

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Abstract
A solitary fibrous tumour is a pleural tumour which may rarely be detected at non-pleural sites. In this report, the case of a solitary fibrous tumour arising from the sublingual salivary gland is described.

Key words: Salivary gland neoplasms; Pleural neoplasms, solitary fibrous tumour

Introduction

Mesenchymal salivary gland tumours are rare. Myoepitheliomas, vascular tumours, lipomas and schwannomas are among those well known. In this report a case of solitary fibrous tumour (SFT) arising from the sublingual salivary gland is described. SFT is a distinctive mesenchymal tumour of the pleura which is also known as solitary fibrous mesothelioma (Gary and Rosai, 1989). Histologically similar tumours have been described extrapleurally in the pericardium (Dalton *et al.*, 1979), peritoneum (El-Naggar *et al.*, 1989), mediastinum (Gary and Rosai, 1989), lung (Yousem and Flynn, 1988), liver (Gary and Rosai, 1989), thyroid (Gianluca *et al.*, 1993), nasal cavities (Zukerberg *et al.*, 1991), nasopharynx (Hybels and El-Naggar, 1993) and paranasal sinuses (Zukerberg *et al.*, 1991). Evidence of a fibroblastic origin for this benign tumour of adulthood has been provided by immunohistochemical studies (Gary and Rosai, 1989). This report discusses the SFT which should be considered in the differential diagnosis of salivary gland tumour.

Case report

A 50-year-old white man presented with a 12-year history of a painless, slowly-growing sublingual mass in the region of the right sublingual gland. The lesion appeared well defined, firm and solid. The overlying mucosa was normal. On the basis of the clinical data a presumptive clinical diagnosis of 'neoplastic salivary gland lesion probably a pleomorphic adenoma' was made. Fine needle aspiration cytology of the mass was hypocellular, and consisted of a few clusters of spindle–ovoid cells consistent with either a mesenchymal or a myoepithelial tumour. The tumour was easily excised intraorally under general anaesthesia. Grossly the tumour was well encapsulated and measured 3 × 2.5 × 2 cm. The cut surface was firm and white–grey in colour.

Microscopically the lesion was bordered by a fibrous capsule and had alternating hypercellular and hypocellular areas (Figure 1). Hypercellular storiform configuration and branching network of vascular channels forming haemangiopericytoma-like areas (Figure 2) alternated with hyalinized and less cellular areas (Figure 3). Remnants of dilated salivary gland ducts with peripheral hyalinization involved by the tumour were noticed (Figure 4). The constituent cells were spindle-shaped with round, oval or

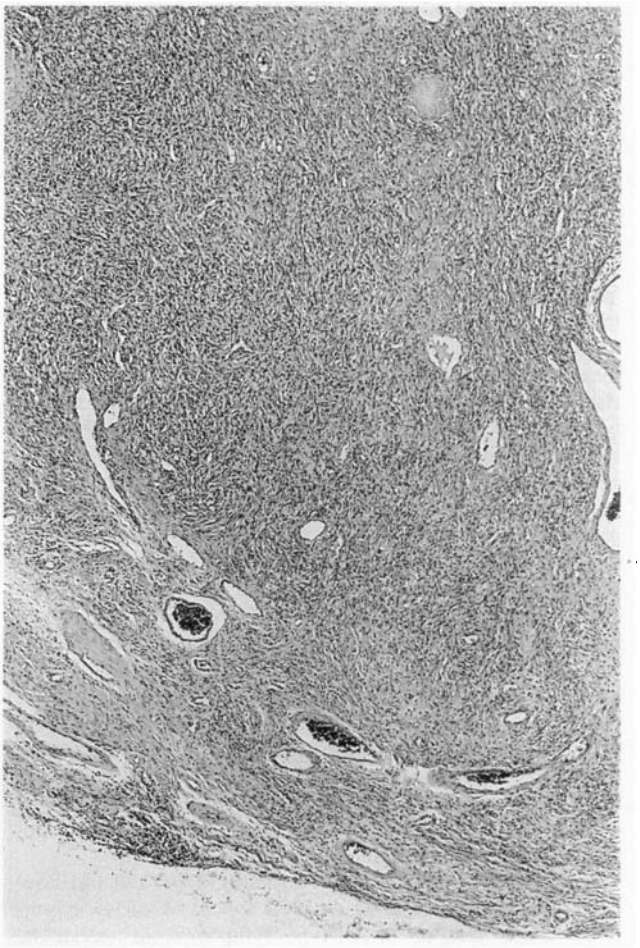

Fig. 1
Well demarcated spindle cell tumour. (H&E; ×50).

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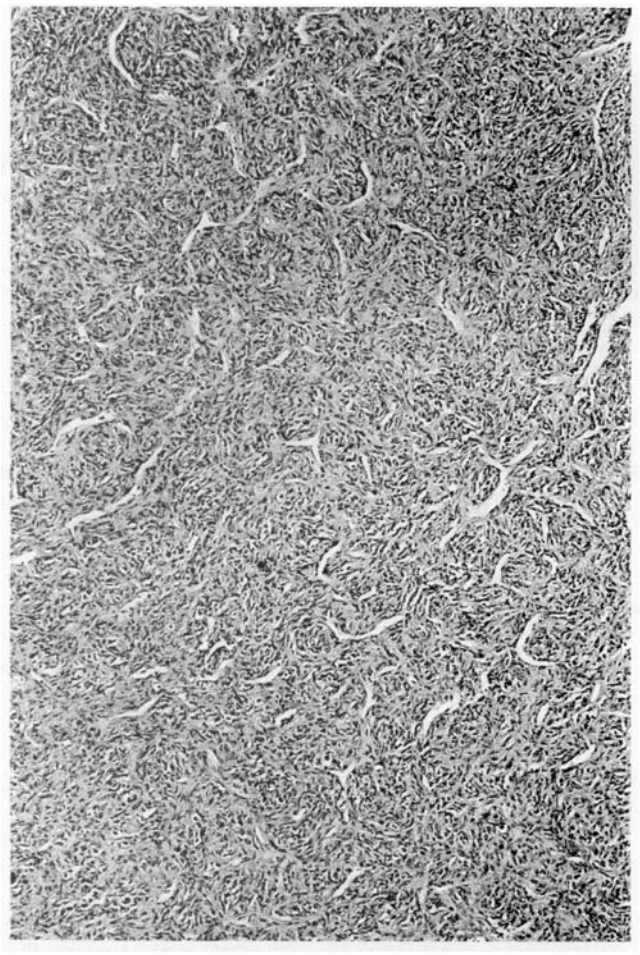

FIG. 2
Vascular pattern of SFT reminiscent of haemangiopericytoma. (H&E; ×100).

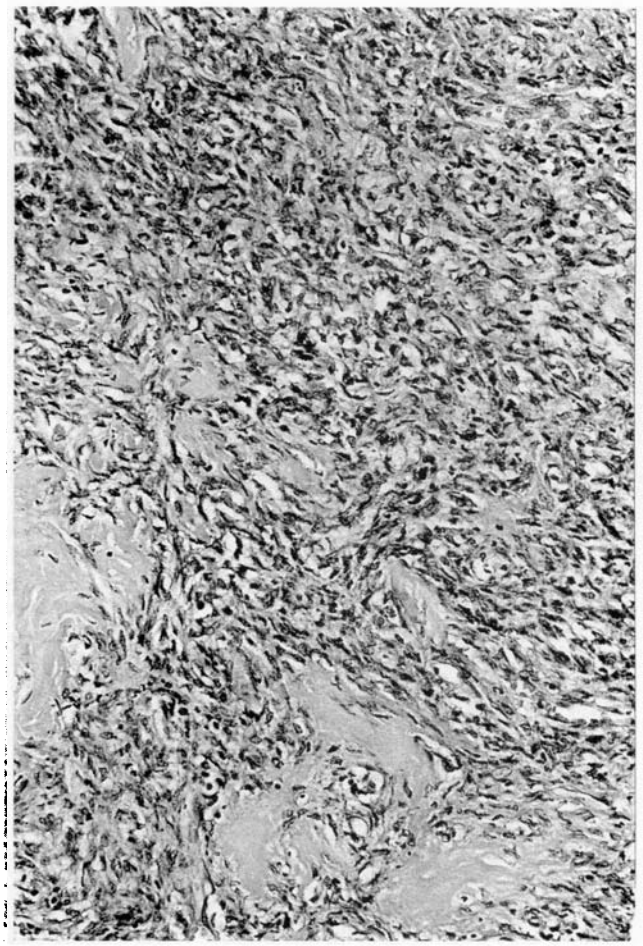

FIG. 3
Hypercellular areas alternating with bands of hyalinized collagen. (H&E; ×200).

elongated nuclei without prominent nucleoli. The cytoplasm were eosinophilic without distinct margins (Figure 5). Scattered lymphocytes were present. A few mitoses were noticed. There was no evidence of necrosis or invasion of surrounding structures. Deparaffinized sections were immunostained with antibodies to keratin (AE1 and AE3; San Ramon, USA), epithelial membrane antigen (EMA) (Biogenex, San Ramon, USA), vimentin (Biogenex, San Ramon, USA), S-100 protein (Biogenex, San Ramon, USA), desmin (Dakopatts, Denmark), Q-bend/10 (Biogenex, San Ramon, USA).

Immunohistochemical studies showed positivity of tumour cells for vimentin only. No staining for desmin, S-100 protein, keratins and EMA was seen. On the other hand, EMA was positive in the entrapped salivary gland duct lumens, keratin in the duct cells and Q-bend in vascular endothelia.

Discussion

Solitary fibrous tumours (SFT), when seen in unusual locations, may pose some diagnostic difficulties. SFT arising in a salivary gland must be considered in the differential diagnosis of mesenchymal tumours of salivary glands like haemangiopericytoma, benign and malignant fibrous histiocytoma, schwannoma, synovial sarcoma, myoepithelioma, fibroma, fibrosarcoma, pleomorphic lipoma, spindle cell lipoma, liposarcoma, neurofibroma and malignant schwannoma (Dalton *et al.*, 1979; Yousem and Flynn, 1988; El-Naggar *et al.*, 1989; Gary and Rosai, 1989; Zukerberg *et al.*, 1991; Gianluca *et al.*, 1993; Hybels and El-Naggar, 1993).

In the present case, haemangiopericytoma, synovial sarcoma and myoepithelioma have been considered in the differential diagnosis. Haphazard or patternless arrangement of the tumour cells was the main microscopic feature of SFT. Vascular areas reminiscent of haemangiopericytoma were present only focally and alternated with largely collagenized hypocellular areas. Remnants of the salivary gland ducts entrapped within the tumour may be misinterpreted as a component of biphasic synovial sarcoma. Peripheral hyalinization, positive cytoplasmic keratin and luminal EMA positivity of these ductal structures associated with the negative keratin and EMA immunostaining of the tumour cells rule out the possibility of synovial sarcoma. Spindle-cell type myoepithelioma may be confused with lesions of fibroblasts. However, lack of S-100 protein expression in the tumour cells of the present case is contrary to a myoepithelial origin. The clinical and histological characteristics of our case suggest that the SFT arises within the salivary gland taking its origin from the local fibroblasts. Our immunohistochemical staining showed vimentin positivity of the tumour cells and this result agrees with the immunophenotypic patterns described by some authors who propose a fibroblastic origin for SFT (El-Naggar *et al.*, 1989; Gary and Rosai, 1989). Although some pleural SFTs, containing atypical cells with mitotic activity, and having a diameter of more than 10 cm behave in an aggressive fashion (Gary and Rosai, 1989), at non-pleural sites their behaviour has usually been benign (Zukerberg *et al.*, 1991; Gianluca *et al.*, 1993; Hybels and El-Naggar, 1993), and local excision is the treatment of choice (Hybels and El-Naggar, 1993). Our case was alive and well six months after total excision.

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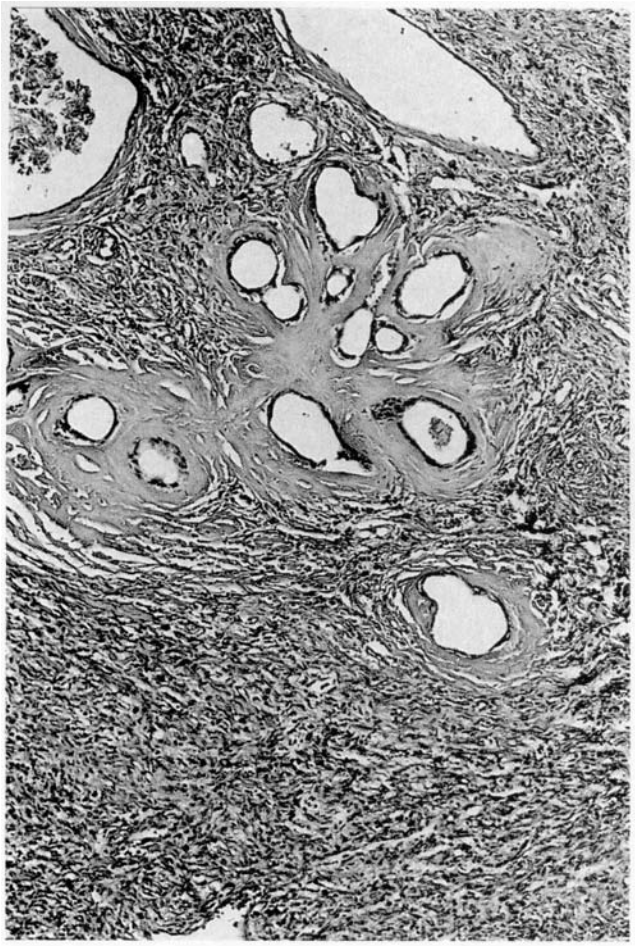

FIG. 4

Spindle cell tumour entrapping the remnants of salivary gland ducts with peripheral hyalinization. (H&E; ×200).

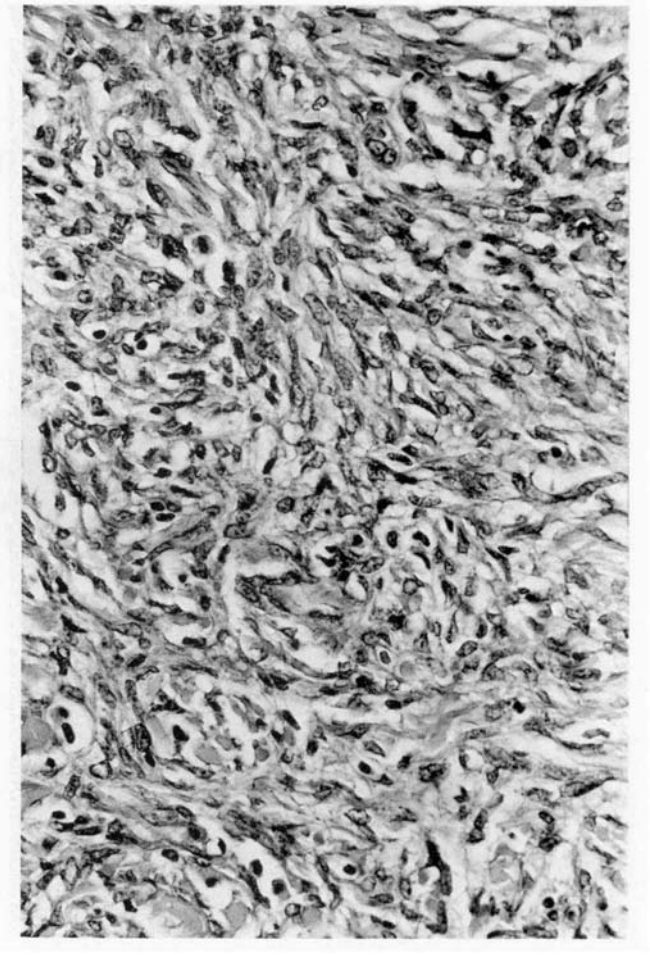

FIG. 5

The tumour is composed of cells with oval to spindle-shaped nuclei and indistinct cells borders. (H&E; ×400).

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References

Dalton, W. T., Zolliker, A. S., McCaughey, W. I. E. Jacques, J. Karnerstein, M. (1979) Localized primary tumor of the pleura: an analysis of 40 cases. *Cancer* **44:** 1465–1475.

El-Naggar, A. K., Ro, J. Y., Ayala, A. G., Ward, R. M., Ordonez, N. G. (1989) Localized fibrous tumor of the serozal cavities, immunohistochemical, electron microscopic and flow-cytometric DNA study. *American Journal of Clinical Pathology* **92:** 561–565.

Gary, B. W., Rosai, J. (1989) Solitary fibrous tumor of the mediastinum. A report of 14 cases. *American Journal of Surgical Pathology* **13:** 547–557.

Gianluca, T., Clara, S., John, N., Maria, K. T., Manuel, S. S. (1993) Solitary fibrous tumor of the thyroid: clinicopathological, immunohistochemical and ultrastructural study of three cases. *Virchows Archiv A: Pathological Anatomy and Histopathology* **422:** 491–497.

Hybels, R. D., El-Naggar, A. K. (1993) Solitary fibrous tumor. *Annals of Oto-Rhino-Laryngology* **102:** 74–76.

Yousem, S. A., Flynn, S. D. (1988) Intrapulmonary localized fibrous tumor: intraparenchymal so-called fibrous mesothelioma. *American Journal of Clinical Pathology* **89:** 365–369.

Zukerberg, L. R., Rosenberg, A. E., Randolph, G., Pilch, B. Z., Goodman, M. L. (1991) Solitary fibrous tumor of the nasal cavity and paranasal sinuses. *American Journal of Surgical Pathology* **15:** 126–130.

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