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Leiomyosarcoma of the parotid gland

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Abstract
A 36-year-old patient with a primary leiomyosarcoma of the left parotid gland is presented. To our knowledge this is only the second case of a parotid leiomyosarcoma in the world according to the literature review.

Key words: Leiomyosarcoma; Parotid neoplasms

Introduction

Leiomyosarcoma is a malignant tumour of smooth muscle origin which is rarely seen in the head and neck region (Sözeri *et al.*, 1992; Rowe-Jones *et al.*, 1994; Zbären and Ruchti, 1994). Smooth muscle is seldom encountered in the head and neck region; mainly in the walls of blood vessels and in the erector pili musculature of the skin. Sandhyamani *et al.* (1983) reported the first leiomyosarcoma in the parotid region.

Case report

A 36-year-old male was admitted to our clinic in September 1994, with a one year history of a left-sided mass, the size of a nut, in front of his pinna which had showed abrupt growth and pain four weeks prior to consultation.

Physical examination revealed a firm, mobile, painless mass 3 × 4 cm in size in the left parotid region (located inferomedially) which caused no change in the overlying skin. No palpable neck nodes were present. The ultrasonographic evaluation demonstrated a hypoechoic lesion. Fine needle aspiration biopsy reported it as benign, and scintigraphic imaging was normal.

Superficial parotidectomy was performed in October 1994. Excessive bleeding was present during the parotid dissection. The operation was terminated as the frozen section was reported as benign. However, the histopathological evaluation of the specimen identified the lesion as a leiomyosarcoma. The patient was prepared for a second operation, total parotidectomy. No pathology was observed after bone scanning the whole body. The ultrasonographic examination of the abdomen was also within normal limits. After the injection of Gd-DTPA, Magnetic resonance imaging (MRI) intravenously revealed fibrosis in the posterior part of the left parotid region. Computed tomography (CT) of the thorax was normal. Total parotidectomy preserving the facial nerve was performed on the patient in whom no distant metastases were found. In the thoracic, and cervical CT scans which were repeated post-operatively, no residues nor recurrences were encountered.

In the light of the oncology consultation two courses of chemotherapy were given to the patient to whom radiotherapy has been recommended also. Radiotherapy was started after 2.5 months. The patient quit radiotherapy because of psychological problems caused by the extensive aphthous stomatitis which occurred by the fifteenth day of radiotherapy.

In the follow-up of the patient, he was lost between the third and the eighth months after the operation and then presented in the ninth month with a firm lymphadenopathy 4 × 6 cm in size located at the superior portion of the preauricular region. CT scan of the thorax, neck and the parotid region was still normal.

In August 1995, the mass was removed together with the skin overlying it. Although the tumour had not invaded the zygoma nor the mandible, the periosteum was included in the specimen. The facial nerve and its branches were attacked by the tumour. The tumour was widely excised including the facial nerve, leaving safe margins. The diagnosis of leiomyosarcoma was confirmed by histopathology. Radical neck dissection on the left side was performed in September 1995, due to the lymphadenopathy in the upper jugular chain.

Pathology

Microscopically, the biopsy material was neoplastic and was made up of spindle-shaped cells arranged in interweaving long fascicles (Figures 1 and 2). The lesion was highly cellular. The nuclei were rod-shaped, some of them were hyperchromatic and moderately polymorphic. The cytoplasms were eosinophilic. On one side of the preparation the tumour's infiltration of the salivary gland was striking. More than five mitotic figures were counted per 10 high power fields (Figure 3). Necrosis was not present. Cells were stained heavily with Masson's trichrome. Using reticulum-specific stains, the thin reticulum fibres enveloping the cells individually were seen. Immunohistochemical preparations showed strong positive staining against antibodies to vimentin and smooth muscle-specific actin, and negative staining against antibodies to S100 and desmin.

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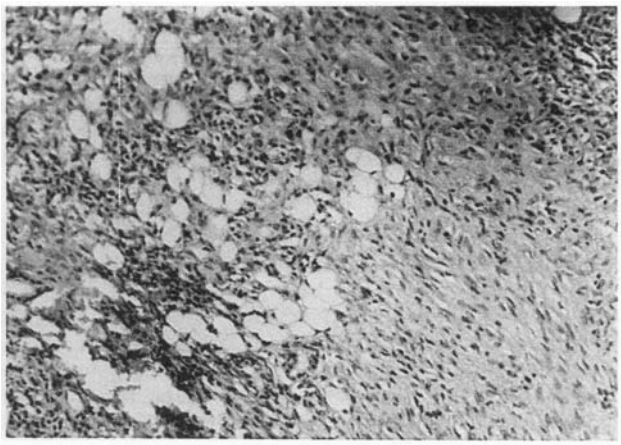

FIG. 1
Salivary gland excretory duct and the tumour site consisting of spindle cells (H & E; × 42).

In the light of all these findings the case was evaluated as leiomyosarcoma (Grade 1).

Discussion

When Seifert and Oehne (1986) analysed 167 mesenchymal (non-epithelial) salivary gland tumours, they found that 90 per cent of them were in the parotid gland and 10 per cent were in the submandibular gland. Yet, 90 per cent of these tumours were benign and 10 per cent were sarcomas. Of the 17 patients with sarcomas, five were malignant fibrous histiocytoma, five were malignant schwannoma, four were embryonic rhabdomyosarcoma and the remaining three were myxoid liposarcoma, leiomyosarcoma and malignant haemangioendothelioma. In the classification of these tumours immunohistochemical studies constituted a very important diagnostic tool. According to the literature research our case is the second leiomyosarcoma to be found in the parotid gland and in immunohistochemical staining it was vimentin (+), actin specific to smooth muscle (+), desmin (−) and S100 (−).

Leiomyosarcomas make up seven per cent of all soft tissue sarcomas (Schenberg *et al.*, 1993). Approximately 40 cases of leiomyosarcomas have been presented in the head and neck region. It has been pointed out that it is mostly encountered in the sinonasal tract, scalp and in other superficial soft tissues (Mindell *et al.*, 1975; Josephson *et al.*, 1985; Kuruvilla *et al.*, 1990). Sandhyamani *et al.* (1983) described leiomyosarcoma of the parotid gland for the first time, and noted that it recurred three times and metastasized to one of the adjacent lymph nodes. In our case metastasis to the superior preauricular lymph node was seen nine months after the operation, while metastasis to the jugulodigastric lymph node was seen 11 months after the operation.

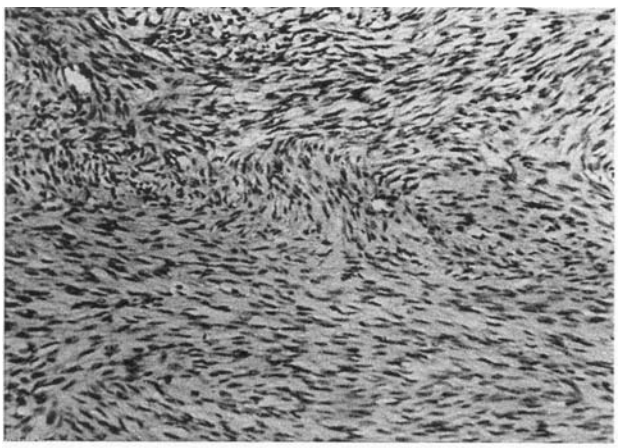

FIG. 2
Intersecting fascicles of spindle cells (H & E; × 120).

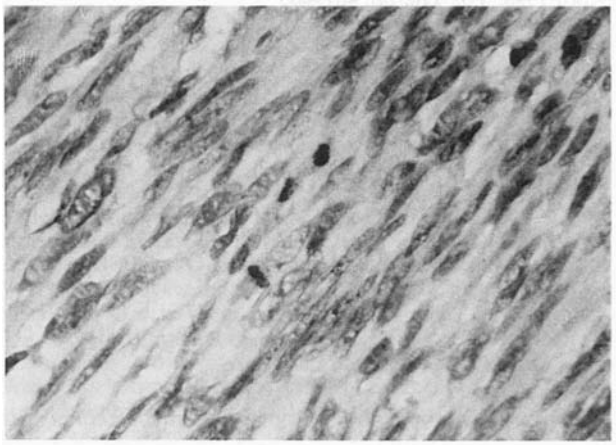

FIG. 3
Neoplastic spindle cells showing mitotic activity (H & E; × 240).

Symptoms of leiomyosarcoma arising in the head and neck region are usually nonspecific. The most significant symptom is the mass. The diagnosis is made by means of histopathological and immunohistochemical studies (Mindell *et al.*, 1975).

The lesion is smooth, circular and stable in general. Haemorrhage and necrosis may be seen in larger lesions (Mindell *et al.*, 1975). In our case, haemorrhage during the operation and focal areas of necrosis were encountered.

Other spindle cell tumours, especially the tumours of the neural sheath and malignant melanoma of spindle-cell type must be included in the differential diagnosis. Tumours of the neural sheath must be excluded based on the nuclear features of tumour cells, failure to demonstrate areas as Antoni A and Antoni B and the absence of immunoreactivity to S100 in immunohistochemical studies.

It is distinguished from malignant melanoma of spindle-cell type by the lack of pigment and the large eosinophilic nuclei in the cells. Moreover, malignant melanomas are more polymorphic and immunoreactive to S100.

Wide local excision and radical neck dissection in patients with palpable neck nodes are recommended for treatment (Sözeri *et al.*, 1992; Schenberg *et al.*, 1993; Rowe-Jones *et al.*, 1994). We performed total parotidectomy on our case, and carried out a radical neck dissection 11 months later, when the patient presented with a palpable neck node.

Leiomyosarcoma is not a lesion with a high metastatic potential. If it occurs, it typically metastasizes haematogenously (Zbären and Ruchti, 1994). Of the 30 reported sinonasal tract leiomyosarcomas, only five had metastases (Kuruvilla *et al.*, 1990). In the follow-up of four oral cavity leiomyosarcomas local recurrence was reported as 36 per cent and distant metastasis, mainly to the lung, was found to be 39 per cent (Schenberg *et al.*, 1993). Besides surgery, chemotherapy and radiotherapy are available for treatment. The tumour response to radiotherapy is poor (Zbären and Ruchti, 1994).

The prognosis is poor. It has been found that 50 per cent of the cases with sinonasal tract leiomyosarcomas were dead within one year of diagnosis (Josephson *et al.*, 1985).

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It is reported that the survival rates are better in the gastrointestinal tract and skin (Zbären and Ruchti, 1994).

References

Josephson, R. L., Blair, R. L., Bedard, Y. C. (1985) Leiomyosarcoma of the nose and paranasal sinuses. *Otolaryngology – Head and Neck Surgery* **93:** 270–274.

Kuruvilla, A., Wenig, B. M., Humphrey, D. M., Heffner, D. K. (1990) Leiomyosarcoma of the sinonasal tract. A clinicopathologic study of nine cases. *Archives of Otolaryngology, Head and Neck Surgery* **116:** 1278–1286.

Mindell, R. S., Calcaterra, T. C., Ward, P. H. (1975) Leiomyosarcoma of the head and neck: A review of the literature and report of two cases. *Laryngoscope* **85:** 904–910.

Rowe-Jones, J. M., Solomons, N. B., Ratcliffe, N. A. (1994) Leiomyosarcoma of the larynx. *Journal of Laryngology and Otology* **108:** 359–362.

Sandhyamani, S., Mahapatra, A. K., Kapur, B. M. (1983) Leiomyosarcoma of the parotid gland. *Australia New Zealand Journal of Surgery* **53:** 179–181.

Schenberg, M. E., Slootweg, P. T., Koole, R. (1993) Leiomyosarcomas of the oral cavity. Report of four cases and review of the literature. *Journal of Craniomaxillofacial Surgery* **21:** 342–347.

Seifert, G., Oehne, H. (1986) Die mesenchymalen (nicht-epithelialen) speicheldrüsetumoren. Analyse von 167 tumorfällen des speicheldrüsen-registers. *Laryngology Rhinology* **5:** 485–491.

Sözeri, B., Önerci, M., Hoşal, S., Ruacan, S. (1992) Primary gingival leiomyosarcoma. *Journal of Laryngology and Otology* **10:** 46–47.

Zbären, P., Ruchti, C. (1994) Leiomyosarcoma of the middle ear and temporal bone. *Annals of Otology, Rhinology and Laryngology* **103:** 537–541.

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