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Primary gingival leiomyosarcoma

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Abstract
A case of primary gingival leiomyosarcoma in a 64-year-old woman is presented. The treatment modalities are discussed and the previous literature is reviewed.

Introduction

Leiomyosarcoma (LS), a malignant smooth-muscle tumour, is common in the uterus and gastrointestinal tract, because of a preponderance of smooth muscle in these organs. However, it is an extremely rare lesion in the oral cavity. Only four primary gingival leiomyosarcomas (PGLS) were encountered in the literature.

Case report

A 64-year-old woman was referred to our clinic on November 11, 1989, because of a painless gingival swelling behind her left lower second premolar tooth for about two months. She was operated by a dental surgeon with the suspicion of Reparative Giant Cell Granuloma, but the lesion was diagnosed as a leiomyosarcoma and it involved the deep surgical margin. Two weeks after surgery a new swelling appeared at the previous site. Examination revealed a rubbery firm, painless, 0.5 × 1.0 cm mass with a smooth surface 3–4 mm behind the left lower second premolar tooth (Fig. 1).

The first and second lower molar teeth had previously been extracted. No lymphadenopathy was palpable. A chest X-ray, skeletal survey, gastrointestinal tract series including barium enema radiographic examinations, Tc^{99} whole body scan and abdominal ultrasound revealed no pathological findings. All the laboratory data were within normal limits.

Wide surgical excision and mandibular curettage was performed under general anaesthesia on December 7, 1989. The surgical specimen included the mandibular periosteum from the second premolar tooth to the retromolar region. Post-operative adjuvant chemotherapy was given and no recurrence has been observed for the first 12 month period.

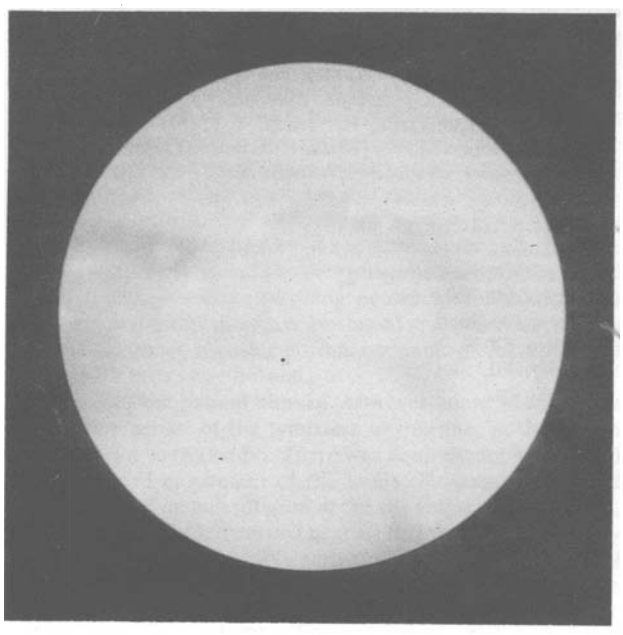

FIG. 1
The photograph of the lesion in the oral cavity.

Histopathological findings

Histopathological examination of the tissue revealed solid nodular tumour beneath the intact surface squamous epithe-

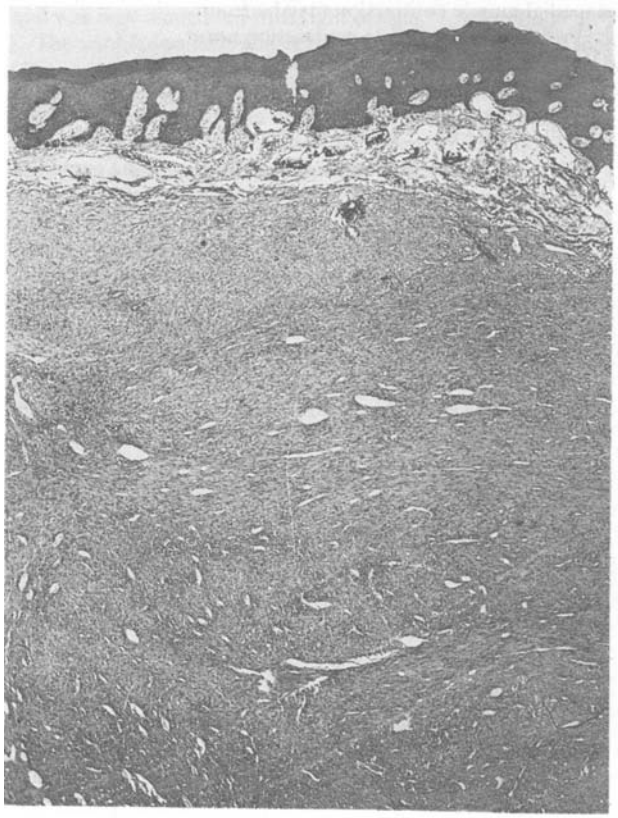

FIG. 2
Histological section of tumour beneath the intact surface epithelium. The tumour is cellular with crossing bundles of neoplastic smooth muscle tissue. H&E ×115.

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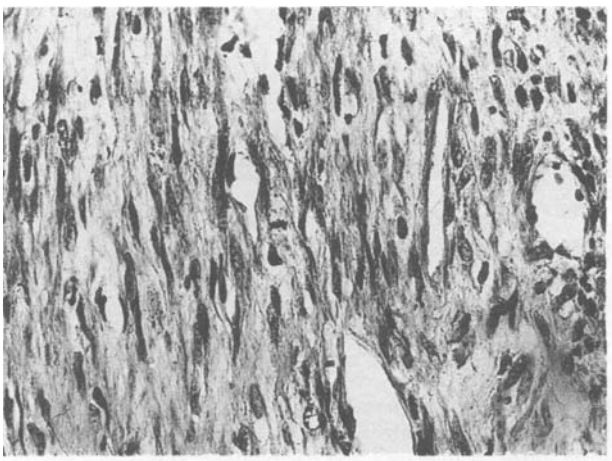

FIG. 3

Higher magnification reveals pleomorphic fusiform tumour cells with irregular chromatin. Occasional mitosis are noted. H&E ×725.

lium (Fig. 2). The tumour was uncapsulated and extended irregularly into the stroma as well as into the minor salivary glands. The tumour cells had fusiform nuclei and prominent nucleoli with elongated eosinophilic cytoplasm in haematoxylin-eosin stained sections (Fig. 3). These cytoplasmic processes gave characteristic staining reactions for muscle fibres with Masson's Trichrome and Van Gieson stains. Immunoperoxidase staining was performed using an avidin-biotin-complex method with antibodies from DAKOPATTS. Tumour cells demonstrated intra-cytoplasmic staining for despin in many fields. (Fig. 4). No staining for neuron-specific enolase, S-100 protein and cytokeratin (CAM 5.2) was observed. Mitoses were quite frequent and two mitotic figures per ten high power (40×) microscopic fields were counted.

Discussion

Smooth muscle tumours occur frquently in the uterus, gastrointestinal tract, skin and subcutaneous tissue. This can be explained on the basis of abundance of smooth muscle in these areas. Fields and Helwig (1981) pointed out leiomyosarcomas are extremely rare in the oral cavity and whenever they do occur it is difficult to determine whether they develop through malignant transformation of a leiomyoma or *de novo*. Many authors (Charlton, 1964; Cheek and Nickey, 1965; Garret, 1969; Mindell *et al.*, 1975; Poon *et al.*, 1987) also reported that leiomyosarcomas are extremely rare tumours of the oral cavity particularly in the gingiva. Probably it is related to the general absence of smooth muscle in this region, except in blood vessels, the circumvallate papillae of the tongue and occasionally aberrant primitive mesenchymal tissues. Ohashi *et al.* (1984) stated that smooth muscle tumours located in nose, paranasal sinuses, and nasopharynx, may originate from the myoepithelial cells in the submucosal glands. Nevertheless the exact source of the smooth muscle has not been determined. In our case, the tumour occurred far away from the cheek, and the circumvallate papillae of the tongue, and minor salivary glands which were not found within the gingival tissue. It is suspected that the tumour might have been derived from the primitive mesenchyme or the smooth muscle element of the blood vessels. According to Josephson *et al.* (1980) and Nishi *et al.* (1987), leiomyosarcomas recur and metastasize in about 40 to 50 per cent of the cases with regard their behaviour. Regional lymphatic involvement is common. Martis (1978), Poon *et al.* (1987), and Weitzner (1980) found that the lungs and liver are the common sites of haematogenous meatastases. A mortality rate of 40 per cent has also been noted in reported cases. Since, the number of reported cases of leiomyosarcoma in the oral cavity is very few, no definite conclusions concerning their behaviour and prognosis can be drawn except for those patients with distant metastases.

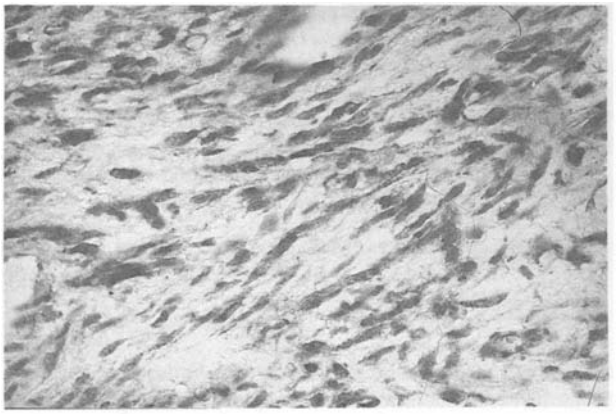

FIG. 4

Immunoperoxidase staining for despin. Note dense granular intracytoplasmic staining of fusiform tumour cells. Avidin-biotin-complex and diaminobenzidine. ×725.

Kratochvil *et al.* (1982) and Weitzner (1980) advised wide surgical excision with radical neck dissection if palpable nodes are present. Prophylactic neck dissection is not recommended. Long-term follow-up is mandatory because of the high rate of local recurrence and distant metastases.

Leiomyosarcoma is regarded as resistant to radiotherapy (Kahn and Koral, 1989). Ohashi *et al.* (1984) suggest that adjunctive chemotherapy might be useful to enhance the effect of surgery for oral leiomyosarcoma.

We performed only wide local excision without neck dissection as there were no palpable nodes in the neck. The patient was not given radiotherapy. Following surgery we gave adjunctive chemotherapy.

As a result it can be said that early wide surgical resection is the most effective means of therapy for both primary and recurrent leiomyosarcoma of the head and neck region. Radical neck dissection is reserved for regional lymph node metastases.

Conclusion

A case of leiomyosarcoma on the gingiva is presented. The most common encountered symptom at the early stage of gingival leiomyosarcoma is a slowly enlarging, non-ulcerated and painless mass. It shows a high incidence of local recurrence and metastasis, and has a poor prognosis. Early wide surgical excision plays the most effective role in the treatment. Adjunctive chemotherapy might be helpful after surgery.

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