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Case Report

Primary gingival leiomyosarcoma

A clinicopathological study of 1 case with prolonged survival

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Abstract

Background: Leiomyosarcoma is a relatively uncommon mesenchymal tumor that exhibits smooth-muscle differentiation. Only 3 to 10% of leiomyosarcomas arise in the head and neck, the nose and paranasal sinuses, skin and subcutaneous tissue and cervical esophagus being the most common localizations. Most leiomyosarcomas involving the oral tissues primarily affect the maxillary sinus, the maxillary or mandibular bone. A review of the English-language literature since 1908 revealed 30 reported cases of primary leiomyosarcoma of the oral mucosa and soft tissues.

Material and Methods: We report on a case of gingival leiomyosarcoma, arising in a 31-year-old female and involving the upper alveolar mucosa. Following the diagnosis of malignant neoplasm on frozen sections and an en-block resection, the tumour was formalin-fixed and paraffin embedded for histological and immunohistochemical examination.

Results: Microscopically, the tumor was composed of interlacing fascicles of spindle-shaped cells with elongated, blunt-ended nuclei and eosinophilic cytoplasm, containing PAS-positive granules. Mitoses, both typical and atypical, and scattered necrotic foci were present. Consistent desmin, muscle specific and α-smooth muscle-specific, and vimentin immunoreactivity was demonstrated in the tumor cells. The patient is alive and free of disease at a 7-year follow-up.

Conclusions: Intra-oral leiomyosarcomas are exceptionally rare. Accurate diagnosis and treatment is largely based on the careful search of clinical signs indicative of malignancy (e.g., neoplastic bone destruction, wide invasion of adjacent tissues) and intra-operative (frozen sections) examination of the lesion. Though the case reported herein showed an attenuated clinical behavior, prolonged follow-up is mandatory in view of possible tumor relapse.

Key words: leiomyosarcoma; oral mucosa; oral neoplasms; soft tissue tumors; oral sarcoma

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Leiomyosarcoma (LMS) is a relatively uncommon mesenchymal tumor that exhibits smooth-muscle differentiation (Ezinger & Weiss 1988). Primary lesions are usually found in the soft tissues or in a visceral location, including the gastrointestinal, urinary and female genital tracts (Kissane 1990). Only 3 to 10% of LMSs arise in the head and neck, the nose and paranasal sinuses, skin and subcutaneous tissue, the cervical esophagus being the most commonly affected sites (Piattelli & Artese 1995).

The occurrence of LMS in an intra-oral location is very rare: a review of the English-language literature revealed only 30 cases primarily affecting the oral mucosa or soft tissues (Table 1). The rare occurrence of LMS in the oral cavity has been correlated with the paucity of smooth muscle structures in this location, as compared with their abundance in other sites (Takagi & Ishikawa 1972).

Table 1. Clinico-pathological features of the intra-oral leiomyosarcomas reported in the English literature

Authors	Year	Sex	Age (years)	Site	Treatment	Recurrence	Metastasis	Follow-up	Result
Greever et al., Stout & Hill**	1949	F	61	cheek	surgery	yes	yes	6 years	died
Yannopoulos & Stout	1962	M	1	tongue	surgery	–	–	56 months	alive
O'Day et al.	1964	M	3	floor of mouth	surgery	yes	yes	22 months	died
O'Day et al.	1964	M	9	soft palate	surgery	yes (R)	–	30 months	died
O'Day et al.	1964	F	19	floor of the mouth	surgery	–	Yes	11 years	alive
Botting et al.	1965	M	8.5	soft palate	surgery+R	yes	yes	39 months	died
Goldberg et al.	1970	M	54	tongue	surgery	–	–	30 months	alive
Mindell et al.	1975	M	59	tongue	surgery	–	–	3 years	alive
Brandjord et al.	1977	F	63	floor of the mouth	surgery	–	yes	27 months	alive
Farman & Kay	1977	F	88	cheek	surgery	–	–	6 months	alive
Wong et al.	1978	M	25	soft palate	surgery	yes	–	1.5 years	died
Robinson et al.	1978	M	88	cheek	surgery	yes	–	5 years	alive
Weitzner	1980	F	39	gingiva (maxillary)	surgery	–	–	34 months	alive
Lack	1986	F	2,5	tongue (base)	surgery+chem.	–	–	4 years	alive
Poon et al.	1987	M	23	gingiva (mandibular)	surgery	–	–	8 months	alive
Kawakami et al.	1987	F	62	gingiva (maxillary)	R+C+S	–	–	5 years	alive
Piattelli & Piattelli	1991	F	78	lip (superior)	surgery	–	–	30 months	alive
Piattelli & Piattelli	1991	F	71	lip (inferior)	surgery	n.r.	–	n.r.	n.r.
Swanson et al.	1991	F	15	lip	surgery	–	–	1 years	alive
Sozery et al.	1992	F	64	gingiva (mandibular)	surgery+C	–	–	1 years	alive
Freedman et al.	1993	F	27	gingiva (maxillary)	S+R	–	yes	21 months	died
Schenberg et al.	1993	M	42	cheek	surgery	–	–	32 months	alive
Schenberg et al.	1993	M	38	cheek	surgery	–	–	13 months	alive
Aydin & Dreyer	1994	F	70	tongue (base)	radiotherapy	–	–	1.5 years	alive
Banuls et al.	1994	F	64	lip (upper)	surgery	–	–	–	died
Mayall et al.	1994	M	60	tongue	surgery	–	–	1 year	alive
Piattelli & Artese	1995	F	80	tongue	no treatment	n.r.	n.r.	n.r.	n.r.
Izumi et al.	1995	M	70	gingiva (maxillary)	surgery	–	–	22 months	alive
Mesquita et al.	1998	F	23	buccal mucosa	surgery	–	–	2 years	alive
Goldschmidt et al.	1999	F	24	gingiva (mandibular)	surgery	–	–	10 years	alive

N.B.: * Zieler in 1908 and Hayn in 1911 described the same case; ** Greever et al. in 1949 and Stout & Hill in 1958 described the same case S=re-excision, R=radiotherapy, C=chemotherapy, n.r.=not reported.

LMSs occurring in regions with scanty or absent smooth muscle structures, such as the jawbones and oral tissues possibly arise from the tunica media of blood vessels (Fu & Perzin 1975), *erectores pilorum* (Stout & Hill 1958), circumvallate papillae (Phillips & Brown 1971), myoepithelial cells (Miles & Waterhouse 1962), or from pluripotential, undifferentiated, mesenchymal cells (Izumi et al. 1995, Kratochvil et al. 1982, Martin-Hirsch et al. 1991, Sanerkin 1979).

LMSs usually show distinctive morphologic and architectural features: they are composed of spindle-shaped cells with elongated nuclei and eosinophilic cytoplasm, sometimes showing longitudinal fibrils. More rarely, these tumors consist of rounded epithelioid cells with eosinophilic or clear cytoplasm (epithelioid leiomyosarcoma) (Neville et al. 1995). The presence of frequent and atypical mitotic figures and necrotic foci indicates aggressive behavior.

The prognosis of LMS is usually poor, due to high recurrence and metastatic rates.

This study reports on a case of primary gingival LMS and on its morphologic and immunophenotypic features. Furthermore, an extensive literature review was carried out to compare the clinico-pathologic features of the current case with those previously reported, and to possibly explain the prolonged survival observed in our patient.

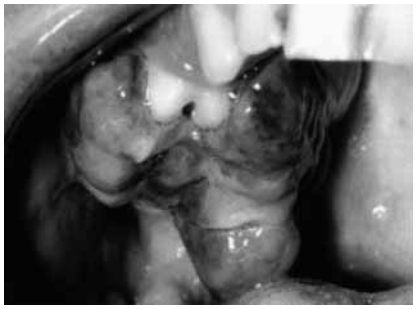

Fig. 1. Vestibular and palatal swelling in the right maxillary premolar region. The nodule was slightly tender to palpation and fixed to adjacent tissues, reddish in color and measured 5 cm in maximum diameter. The neoplasm shows occlusal impressions of the opponent teeth.

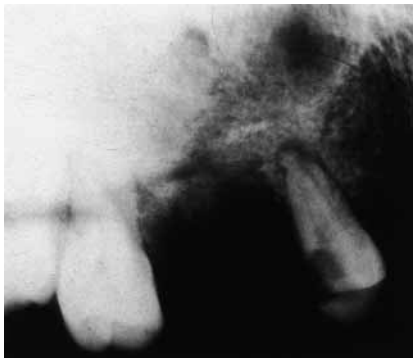

Fig. 2. Intra-oral radiograph demonstrating ill-defined bone resorption mesially and distally to the 2nd premolar. The latter also shows penetrating caries with periapical radiolucency.

Table 2. List of the antibodies used to immunocharacterize gingival leiomyosarcoma. All of them are monoclonal, with the exception of anti-S-100 protein, which is rabbit polyclonal, and were purchased from Dako (Dako Italia SpA, Milan, Italy)

Antigen detected	Clone	Dilution	Positive controls	Results
actin (α-smooth muscle)	*1A4*	1:120	leiomyoma	+++
actin (muscle-specific)	*HHF 35*	1:150	leiomyoma	+++
CD 30 (Ki-1)	*Ber H2*	1:15	anaplastic lymphoma	−
CD 31	*JC/70A*	1:10	hemangioma	−
CD 34	*QBend 10*	1:50	hemangioma	−
CD 45	*2B11*	1:100	lymph node	−
CD 99	*12E7*	1:120	Ewing's sarcoma	−
cytokeratins	*MNF 116+AE 1/AE 3*	1:50+1:100	breast carcinoma	−
desmin	*DE-R-11*	1:50	striated muscle	++
GFAP	*DP 46,10*	1:120	astrocytoma	−
melanoma Ag	*HMB 45*	1:200	melanoma	−
S-100 protein	−	1:2000	melanoma	−
vimentin	*V9*	1:10	leiomyoma	+++

Case description and results

A 31 year-old female was referred to the Department of Dental Sciences, Center for the Study of Oral Tumors of the University of Bari, for a maxillary swelling of 3 months duration. Clinical examination disclosed a reddish lump of soft consistency, 5 cm in maximum diameter, in the right vestibular, maxillary, premolar region (Fig. 1). It was slightly tender to palpation and fixed to the surrounding tissues.

An intra-oral radiograph demonstrated a periodontal and periapical lesion, associated with bone destruction, in the 14–17 region (Fig. 2). There was no regional lymphadenopathy, the remaining clinical findings being unremarkable.

Based on these features, the diagnosis of a malignant neoplasm was postulated and a segmental (13–18) maxillary resection was performed. Intra-operative examination on frozen sections confirmed the diagnosis of malignant spindle cell neoplasm with sarcomatous features. The post-operative course was uneventful.

The patient is currently monitored every 6 months and is disease-free, 7 years after the original diagnosis.

Gross findings

On gross examination, the surgical sample consisted of a soft, solid white-pinkish mass, with slightly irregular edges. Furthermore, macroscopic invasion of the adjacent maxillary alveolar bone was evident.

Histopathology

The surgical samples were promptly fixed in 10% neutral buffered formalin for 24 h and embedded in paraffin. 5-μm thick sections were cut and stained with hematoxylin and eosin (H&E), PAS and Gomori's reticulin stain. Consecutive sections were employed for the immunohistochemical study, which was performed by a standard avidin-biotin/peroxidase (ABC) technique, using the antibodies listed in Table 2. Sections to be stained for muscle-specific, α-smooth-muscle actin, cytokeratins, S-100 protein and vimentin, were subjected to 3×5 min cycles of microwave irradiation, after immersion in 0.10 M citrate buffer, pH 6.0, in a microwave oven operating at 750 W. Adequate positive (listed in Table 2) and negative controls (obtained by substituting the primary antibodies with normal mouse or rabbit serum) were included in the procedure.

Histologically, the neoplasm consisted of intersecting bundles of spindle-shaped cells irregularly infiltrating the

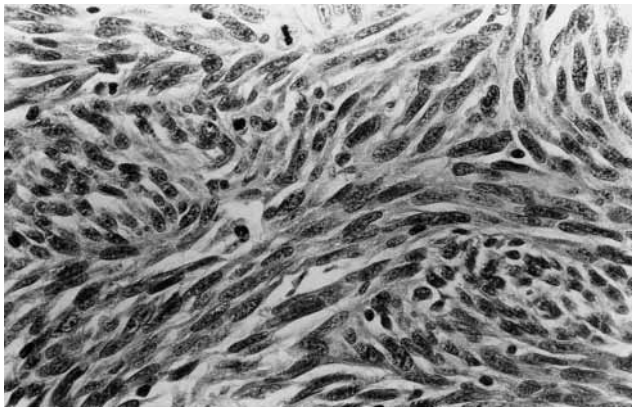

Fig. 3. The tumor was composed of spindle-shaped cells, with blunt-ended nuclei, arranged in dense interlacing fascicles. Several mitotic figures are easily detectable (H&E, 250×).

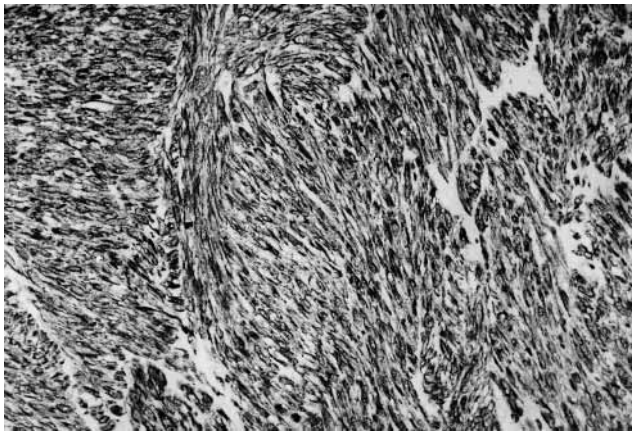

Fig. 4. Consistent α-smooth muscle actin immunoreactivity is evident in most tumor cells (ABC anti-α-smooth muscle actin, 250×).

Table 3. Distribution according to site of previously reported intra-oral leiomyosarcomas

Site	# cases	(%)
tongue	7	23.3
gingiva	7	23.3
cheek	6	20
lip	4	13.4
floor of the mouth	3	10
soft palate	3	10
total	30	100

oral mucosa and the adjacent alveolar bone. Nevertheless, the surgical margins were devoid of neoplastic infiltration. Frequently, the bundles of neoplastic cells were perpendicularly oriented with each other, and merged into the blood vessels wall.

The spindle-shaped tumor cells had blunt-ended, elongated nuclei that showed rounded profiles in transversally cut fasicles (Fig. 3). Hyperchromatic and pleomorphic nuclei were frequently detectable. The tumor cells also showed prominent eosinophilic cytoplasm, with occasional paranuclear vacuolization, and contained occasional PAS-positive granules. Cross striation was never evident. Mitotic figures, both typical and atypical, were frequently detected (12–26/10 HPF). Necrotic foci were scattered throughout the tumor.

Immunocytochemistry

The tumor cells displayed consistent muscle-specific and α-smooth-muscle actin, desmin and vimentin immunoreactivity (Fig. 4), whereas all other antigens tested were not detectable.

Based on the above features, the diagnosis of grade 2 (Coindre et al. 1988) leiomyosarcoma was formulated.

Discussion

Intra-oral LMS's most commonly affect middle-aged and older adults (mean age: 44.3 years, range: 1–88) (Table 2), with a slight female predominance (53.4%). The distribution according to the site of presentation of previously reported intra-oral LMSs is detailed in Table 3. The most common symptom at presentation was the presence of a painless/painful nodule (Schenberg et al. 1993). Surgical resection, including adequate margins of uninvolved tissue, was the elective treatment in 24 (77.4%) cases, while combinations of surgery and radio/chemotherapy were administered in 5 (16.1%) cases. 1 case (3.2%) was treated by radiation therapy alone and another patient (3.2%) refused any treatment. Local recurrence occurred in 6 (20.6%) of 29 patients with reported follow-up, at different time intervals (range: 6 months–10 years). Distant metastases (pulmonary or hepatic) developed in 6 (20%) of 30 adequately documented cases, whereas metastatic deposits in the regional lymph nodes were not documented. Overall, 7 (23.3%) of 30 patients died as direct consequence of their tumor.

The case reported here does not differ substantially in its clinical presentation from the other documented cases of intra-oral LMSs, with the exception of the younger age at presentation, and of the relatively indolent clinical course.

Histopathologically, the current case and the previously reported ones displayed typical morphological and architectural features of LMS (Izumi et al. 1995). Nevertheless, a definitive diagnosis may be difficult to achieve on frozen and permanent sections, immunohistochemistry being quite often necessary to ascertain the smooth muscle cell origin of the neoplasm.

In fact, several different tumors with prominent spindle cell component may be difficult to differentiate from LMS based on morphology alone, including:

1. spindle cell carcinoma, that usually shows cytokeratins immunoreactivity;
2. malignant myoepithelioma (myoepithelial carcinoma), showing cytokeratins and actin immunoreactivity;
3. malignant peripheral nerve sheath tumors, that exhibit S-100 protein and vimentin positivity;
4. malignant melanoma, showing S-100 protein and HMB 45 immunoreactivity;
5. anaplastic lymphoma, displaying CD 45 and CD 30 immunoreactivity;
6. solitary fibrous tumor, demonstrating CD 34 and vimentin positivity along with focal actin and desmin immunoreactivity.

The prognosis of intra-oral LMS is uncertain: many reported intra-oral LMS's recurred or metastasized repeatedly. Local recurrence occurred in 20.6% of patients and distant metastases (pulmonary or hepatic) developed in 20%. The variable outcome of LMS's may be related to different therapeutic approaches: excision, enucleation and curettage or radical surgery have been advocated as treatment options (Fields & Helwig 1981, Martis 1978, Wile et al. 1981). However, complete surgical removal by conservative treatment can be difficult in some instances (e.g., involvement of the mandibular condyle or maxillary sinus). This may account for the high mortality rates detected in the current review (25%). The patient reported here showed clinical signs suggestive of malignancy, such as radiologically-detectable bone destruction. Therefore, radical surgery was promptly planned and performed after intra-operative histopathological confirmation of the malignant nature of the lesion. This may account for the relatively favorable prognosis of this case.

Radiotherapy can be used as adjunctive treatment, postoperatively (Schenberg et al. 1993), even if some authors reported that it does not influence recurrence-free or overall survival rates for intra-oral LMSs (Fields & Helwig 1981, Fu & Perzin 1975, Kuruvilla et al. 1990, Wile et al. 1981). Chemotherapy seems ineffective and should be reserved for the palliation of patients with metastatic disease or inoperable tumors (Chang et al. 1989, Fields & Helwig 1981, Josephson et al. 1985).

In view of the indolent course of some of these neoplasms and of possible late recurrences or metastases, prolonged or indefinite follow-up has been suggested (Izumi et al. 1995). The patient should be closely followed at least during the 1st 5 years, when LMS's are more likely to recur or metastasize (Izumi et al. 1995, Schenberg et al. 1993).

In conclusion, the diagnosis of leiomyosarcoma depends on accurate morphological and immunohistochemical characterization, aimed at ascertaining the smooth muscle nature of any intra-oral tumor with prominent spindle-cell features. The prognosis of intra-oral LMS is highly variable and seems to largely depend on the possibility of curative radical excision of the tumor. In this view, careful search of clinical signs indicative of malignancy (e.g., neoplastic bone destruction, wide invasion of adjacent tissues) should be performed. When in doubt, intra-operative examination of the lesion on frozen sections may allow an accurate diagnosis and adequate surgical procedures. Nevertheless, prolonged survival seems possible, even with conservative surgery, but strict follow-up is mandatory for prolonged periods of

time, in view of possible late recurrences or metastases.

Zusammenfassung

Primäres gingivales Leiomyosarkom: eine klinikopathologische Studie von einem Beispiel mit prolongiertem Überleben

Hintergrund: Das Leiomyosarkom ist ein relativ unbekannter mesenchymaler Tumor, der die Differentiation der glatten Muskeln betrifft. Nur 3 bis 10% der Leiomyosarkome kommen im Kopf und Nacken vor. Die Nase und die paranasalen Sinus, die Haut und das subkutane Gewebe und der zervikale Ösophagus sind die häufigsten Lokalisationen. Häufig sind die oralen Gewebe durch Leiomyosarkome involviert, primär sind die maxillären Sinus betroffen, der maxilläre oder der mandibuläre Knochen. Eine Übersicht der englischsprachigen Literatur seit 1908 erbrachte 30 berichtete Beispiele des primären Leiomyosarkoms der oralen Mukosa und des weichen Gewebes.

Material und Methoden: Wir berichten über einen Fall eines gingivalen Leiomyosarkoms bei einer 31jährigen Frau, wo die obere alveoläre Mukosa einbezogen war. Nach der Diagnose des malignen Neoplasmas mit Gefrierschnitten und einer en-block Resektion wurde der Tumor für die histologische und immunohistochemische Untersuchung mit Formalin fixiert und in Paraffin eingebettet.

Ergebnisse: Der Tumor war mikroskopisch mit verflochtenen Faszikeln von spindelförmigen Zellen mit elongierten, stumpf endenden Kernen und eosinophilem Cytoplasma, was PAS positive Granula enthielt, zusammengesetzt. Mitosen, sowohl typisch als auch untypisch, und verstreute nekrotische Herde waren vorhanden. Einheitliches Desmin, für Muskeln und α-glatte Muskeln spezifisch, und Vimentin Immunreaktivität wurde in den Tumorzellen gezeigt. Die Patientin ist am Leben und frei von der Erkrankung seit 7 Jahren.

Schlussfolgerung: Intraorale Leiomyosarkome sind äußerst selten. Die richtige Diagnose und Therapie basiert auf einer sorgfältigen Suche von klinische Zeichen der Malignität (z.B. neoplastische Knochendestruktion, breite Invasion der angrenzenden Gewebe) und intraoperativer (Gefrierschnitte) Überprüfung der Läsion. So zeigt der hier berichtete Fall eine abgeschwächte klinische Erscheinung, die prolongierte Beobachtung nach der Therapie ist obligatorisch im Blick auf mögliche Tumorrückfälle.

Résumé

Leiomyosarcome gingival primaire: une étude clinicopathologique d'un cas avec survie prolongée

Origine: Le leiomyosarcome est une relativement peu commune tumeur mésenchymateuse qui présente une différentiation des muscles lisses. Seuls 3 à 10% des leiomyosarcomes surviennent sur la tête et le cou, le nez et les sinus paranasaux, la peau et les tissus sous-cutanés et l'œsophage cervical étant les localisations les plus fréquentes, la plupart des leiomyosarcomes impliquant les tissus oraux affectent d'abord les sinus maxillaires, l'os maxillaire ou mandibulaire. Une revue de la littérature anglaise depuis 1908 montre 30 cas rapportés de leiomyosarcome primaire des muqueuses orales et des tissus mous.

Matériaux et méthodes: Nous rapportons le cas d'un leiomyosarcome gingival, survenant chez une femme de 31 ans et impliquant la muqueuse alvéolaire supérieure. Suite au diagnostic de néoplasme malin sur des coupes congelées, et une resection en bloc, la tumeur a été fixée à la formaline et enchassée dans de la paraffine pour examens histologiques et immunohistochimiques.

Résultats. Au microscope, la tumeur était composée de fascicules entrelacés de cellules en forme de fuseau avec un noyau allongé et émoussé et un cytoplasme eosinophile contenant des granules PAS positives. Les mitoses, à la fois typiques et atypiques, et des foyers nécrotiques eparpillés étaient visibles. De la desmine, spécifique des muscles et des muscles lisse α et une immunoréactivité à la vimentine furent mis en évidence dans les cellules tumorales. La patiente est toujours vivante et débarassée de sa maladie 7 ans plus tard.

Conclusions: Le leiomyosarcome intra-oral est exceptionnellement rare. Le diagnostique précis et le traitement reposent sur la recherche soigneuse des signes cliniques indiquant une malignité (par exemple, la destruction néoplastique de l'os, une large invasion des tissus adjacents) et sur l'examen per-opératoire de la lésion. Bien que le cas rapporté ici montre un comportement clinique atténué, un suivi prolongé est obligatoire pour discerner les possibles rechutes de la tumeur.

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