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CASE REPORT

Liposarcoma circumscriptum (lipoma-like) of the tongue: report of a case

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Liposarcoma, first described by Virchow in 1857, is the second most frequent sarcoma of soft tissues, although it is rare both in the head and neck and the oral cavity. Intra-orally, liposarcoma has been reported in the jaw-bones (particularly the maxilla) and the soft tissues – mainly the cheek and floor of mouth, but it is rare in the tongue. A case of well-differentiated, superficial liposarcoma circumscriptum of the tongue is reported. The authors underline the difficulties in the clinical and histopathological diagnosis, as this tumour may be confused with lipoma. In view of the indolent behaviour of this tumour type, local recurrence or metastasis rarely occur; consequently, conservative surgical therapy is advised without adjuvant chemo-radio therapy.
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Introduction

Liposarcoma is one of the most common malignant mesenchymal tumours, representing 9, 8–16% (Nikitakis *et al*, 2001) of all sarcomas of the soft tissues. In the head-neck area, the most commonly affected sites are the temporal, submental and postauricular regions and the mastoid, epiglottis, larynx and orbit.

Almost 50 cases of oral liposarcomas have been reported in the English literature. Oral liposarcomas represent 10% of all sarcomas of the maxillo-facial region (Nikitakis *et al*, 2001) and mainly involve the cheek, floor of mouth, mandible and maxilla, soft palate and lip (Favia *et al*, 2001). Almost 70% of the cases occurred in males (Nikitakis *et al*, 2001).

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Histologically, four distinct subtypes of liposarcoma are identifiable: (i) well-differentiated, (ii) myxoid, (iii) round cells, (iv) pleomorphic, which bear prognostic implications (Laurino *et al*, 2001). Well-differentiated liposarcoma represents 40–45% of all liposarcomas (Laurino *et al*, 2001), occurs with equal incidence in the retroperitoneum and limbs, followed by the paratesticular area and mediastinum, with a peak of incidence between the 5th and the 7th decade. Well-differentiated liposarcoma is subclassified in four variants: (i) adipocytic (lipoma-like), (ii) sclerosing, (iii) inflammatory, (iv) spindle-cell rich. These low-grade tumours can infiltrate adjacent tissues but have very little metastatic potential and seldom recur, mostly as a consequence of incomplete excision. Consequently, some authors employed the term 'atypical lipoma' as a synonymous of well-differentiated liposarcoma to underscore its malignant potential and possibly avoid unnecessary over-treatments (Laurino *et al*, 2001). Occasionally, well-differentiated liposarcomas may undergo de-differentiation: this may occur after repeated recurrences and is characterized by rapid growth, increased mitotic rates and cellular pleomorphism, frequently in the form of small round cell proliferation.

Our aim was to report on the clinico-pathological features of a superficial well differentiated liposarcoma of the tongue occurred in a middle-age white woman, who was treated by wide surgical excision only and who has no signs of local recurrence or metastasis at 2-year follow-up.

Case report

A 58-year-old woman presented with a painless, soft mass of the lateral border of the tongue, that had been slowly growing over the last 6 months. Her medical history and extraoral examination were unremarkable without submandibular or cervical lymphadenopathy. Intra-oral examination revealed a mobile nodular mass in the right lingual border, measuring approximately 2.5 × 1.5 cm (Figure 1). The mass was a yellow-brown

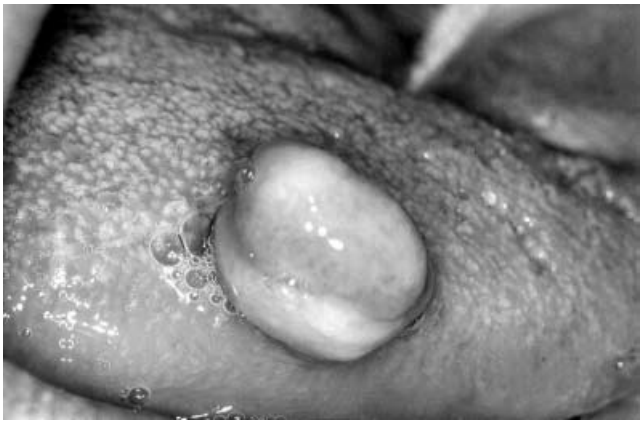

Figure 1 Clinical appearance of well-differentiated liposarcoma: a nodular, partially ulcerated exophytic mass of the right lingual border is evident

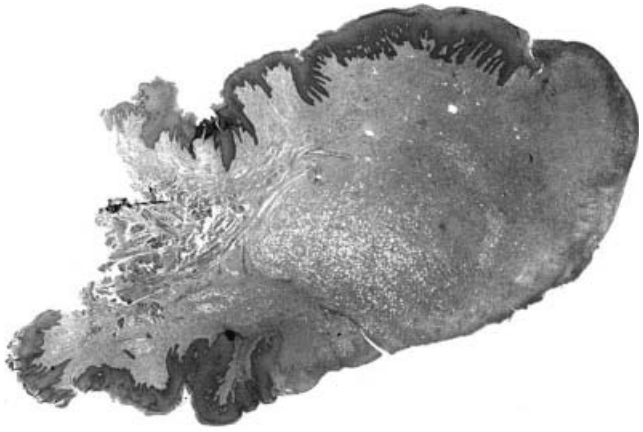

Figure 2 Low-power view of the neoplasm. The tumour extends up to the partially ulcerated epithelium and into superficial layers of the underlying muscle tissue (H & E, original magnification ×2.5)

colour, focally ulcerated and mobile on the underlaying tissues.

With the provisional clinical diagnosis of benign soft tissue tumour, the lesion was excised with a margin of normal tissues and sent for histological examination. The surgical specimen consisted of a well-demarcated mass of 3.0 × 1.5 cm, yellow on the cut surface. It was promptly fixed in neutral buffered formalin, embedded in paraffin, cut and stained with haematoxylin/eosin stained.

Panoramic histological examination showed a partly ulcerated, stratified squamous epithelium covering over 40% of the surface, and a submucosal well-demarcated mesenchymal tumour extending up to the epithelium (Figure 2).

The tumour was composed of well-differentiated lipoblasts in a pseudo-lobular arrangement, which were occasionally embedded in fibrous bundles (Figure 3), especially at the periphery of the lesion. The tumour cells showed distinct cytoplasmic multi-vacuolization and pleomorphic, peripheral and hyperchromatic nuclei with scalloped borders (Figure 4). Together with typical

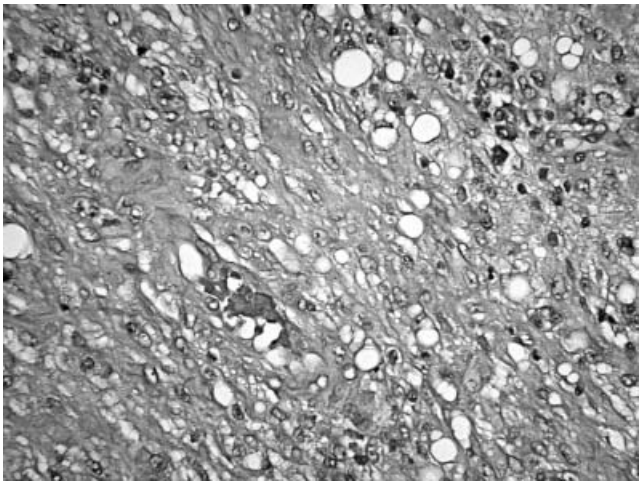

Figure 3 Well-differentiated (lipoma-like) liposarcoma is composed by variably sized mature lipocytes intermixed with lipoblasts showing intra-cytoplasmic vacuolisation (H & E, original magnification ×10)

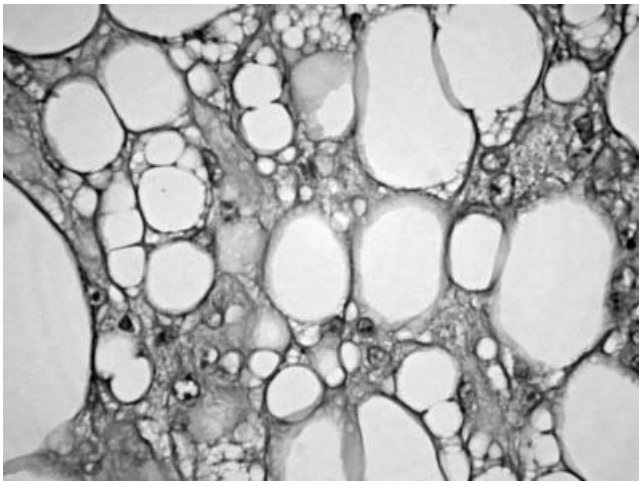

Figure 4 Atypical lipoblasts at different stages of differentiation, with characteristic multi-vacuolated cytoplasm and scalloped nuclei are easily detectable, but no mitosis are present (H & E, original magnification ×20)

lipoblasts, immature lipocytes, well-differentiated lipocytes and mature lipocytes were also detected. Mitotic figures were absent. The tumour cells extensively invaded the adjacent tissues up to the most superficial muscle layers. The surgical margins were tumour-free. After 2 years, no signs of local recurrence, regional or distant metastases are evident.

Discussion

While the cheek is the most commonly involved site by oral liposarcoma and these tumours invariably present as slowly growing lesions infiltrating the adjacent tissues (Charnock *et al*, 1991; Nakahara *et al*, 1994; Zheng and Wang, 1994), only a previous case of a well-differentiated liposarcoma has been reported in the tongue (Nunes *et al*, 2002).

As for liposarcomas of the retroperitoneum and limbs, the presumed origin of well-differentiated liposarcomas is from totipotent mesenchymal cells, as supported by the large variety of cell types detectable in the present tumour, such as lipoblasts, immature lipocytes, well-differentiated lipocytes, mature lipocytes, mulberry-cells (with round and central nuclei and numerous cytoplasmatic vacuoles) and bizarre giant cells.

Histological grading of liposarcomas is of pivotal relevance as it is directly correlated with biological behaviour and prognosis (Enzinger and Weiss, 1995), the well-differentiated forms bearing good prognosis, the myxoid and round cell variants having a more aggressive clinical course with frequent recurrences and the pleomorphic (poorly differentiated) variant being a very aggressive neoplasm that frequently metastasises. In consideration of the variable aggressiveness of distinct liposarcoma subtypes, the clinical management of these tumours vary considerably, well-differentiated liposarcomas being essentially treated by wide surgical excision with tumour-free margins, although some authors suggested the use of adjuvant radiotherapy (McCulloch *et al*, 1992). Nevertheless, the current case was apparently cured without the latter after prolonged follow-up.

Recently, some authors (Laurino *et al*, 2001) have proposed the use of a different terminology, i.e. atypical lipoma or lipoma-like liposarcoma, to designate the group of well-differentiated liposarcoma showing histological similarities with conventional mature lipoma but in which typical lipoblasts are detectable. All the above terms are interchangeable and, probably the need for such a term as atypical lipoma mostly derives from the opportunity to avoid over-treatments for tumours with attenuated clinical behaviour. In fact, recurrences and distant metastases are extremely uncommon for well-differentiated liposarcoma, the former being attributable to incomplete excision, the latter usually occurring after the tumour has become de-differentiated.

At this regard, it should be mentioned that the most important prognostic factor for well-differentiated liposarcoma is its anatomic location, superficial lesions (including the oral tissues) being considered favourable while deeply seated liposarcomas are associated with increased recurrence and metastatic rates. In addition, superficially located, well-differentiated liposarcomas should be excised with a margin of unaffected tissues for a permanent cure to be achieved and this usually requires close clinico-pathological interactions to properly identify surgical margins.

Intra-oral liposarcoma can be misdiagnosed due to its inconspicuous or ambiguous clinical and microscopic features. The clinical differential diagnosis includes reactive and neoplastic conditions (pyogenic granuloma), salivary glands neoplasms, benign or malignant mesenchymal tumours, malignant lymphoma, spindle-cell lipoma (Piattelli *et al*, 2000), myxoma, benign fat tumours (hibernoma), angiolipoma, fibrolipoma, pseudosarcomatous fasciitis, and the so-called malignant fibrous hystiocytoma (Favia *et al*, 2001; Nunes *et al*, 2002), the definitive diagnosis mostly relying on histological evaluation of the tumour. In this context, immunohistochemistry plays a marginal role as there is no specific marker for neoplastic lipoblasts. Extensive and accurate identification of the latter, in fact, still remains the most important clue for proper diagnosis.

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