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Atypical lipoma of the tongue

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Abstract
We report an atypical lipoma arising in the tongue of a 43-year-old man who presented with an indolent dorsal lingual swelling. Atypical lipomas contain multivacoulated lipoblasts, which distinguishes them from benign lipomas. The superficial location in this case distinguishes this tumour from well-differentiated liposarcoma, which is biologically similar in lacking the propensity for metastasis. The superficial location of atypical lipoma allows a complete resection, which is often not possible for the deep-seated counterpart. Atypical lipoma and well-differentiated liposarcoma, if left *in situ*, may undergo transition to de-differentiated liposarcoma. Atypical lipoma should be completely excised with a cuff of normal tissue in order to prevent repeated local recurrence and the possibility of de-differentiation.

Key words: Lipoma; Liposarcoma; Tongue Neoplasms

Case report

A 43-year-old Caucasian male was referred by his general practitioner to the ORL/HNS Department of Warwick Hospital with a six-year history of an indolent swelling of the right anterolateral border of his tongue. There had been a gradual enlargement of the swelling giving rise to local irritation against the teeth. There was no bleeding or discharge and there were no symptoms referable to the upper aerodigestive tract. He was otherwise well. He was a non-smoker and did not drink alcohol.

Physical examination revealed an 8 mm, firm, ovoid, superficial, non-ulcerated lesion of the right dorsal/anterolateral lingual border, adjacent to the second right lower premolar tooth.

In view of the long-standing history of dental trauma the working diagnosis was a traumatic sessile fibro-epithelial polyp. Excision biopsy was performed under local anaesthesia; the swelling was excised with the overlying mucosa and taken down to the lingual musculature.

Pathology

The specimen was a dome-shaped piece of tissue 1 cm in maximum diameter covered by mucosa (Figure 1). On microscopic examination the overlying squamous epithelium was normal. In the underlying stroma there was soft tissue swelling composed of lipocytic cells, which showed variation in size. Numerous cells were present in which the fat vacuoles indented mildly hyperchromatic nuclei, typical of lipoblasts (Figure 2). Mitotic figures were absent and there was no evidence of necrosis. In the periphery of the specimen ordinary lipocytes were evident in the sub-epithelial stroma. The appearances of a well-differentiated lipomatous lesion containing lipoblasts were diagnostic of an atypical lipoma. The lesion was close to the margins of excision.

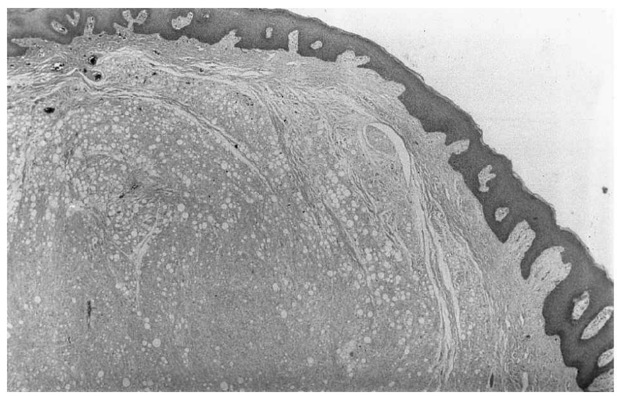

Fig. 1

Low-power view of excised lingual mass; note intact mucosal surface and multiple vacuoles in underlying stroma (H & E; ×2.5).

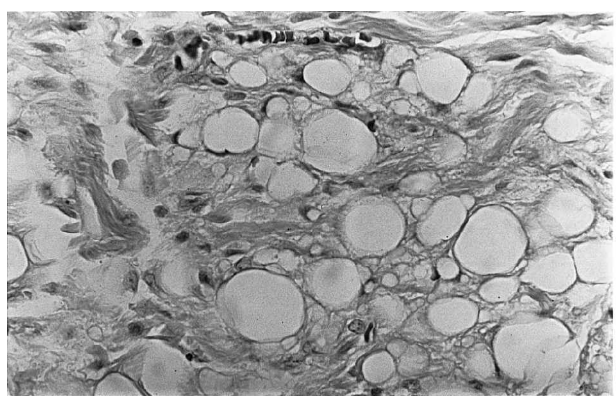

Fig. 2

Higher-powered view of same lingual mass: there are multiple lipocytic cells of varying size separating striated muscle bundles. Note the peripheral nuclei, which are indented by intracellular fat vacuoles (H & E; ×40)

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Outcome

A wide local excision was performed under general anaesthetic a month after the patient's initial procedure. There is no evidence of local recurrence at 10 months' follow-up.

Discussion

Lipomas account for nearly one half of all soft tissue tumours.[1] The most frequent sites of origin are the trunk and extremities with only 2.2 to 4.4 per cent arising within the oral cavity.[2] Lipomas account for 0.3 per cent of all lingual tumours.[3]

The first reported case of a lingual lipoma was credited to Barling in 1858.[4] Eighty per cent of lingual lipomatous tumours are benign lipomas, while almost 20 per cent contain fibrous elements (fibrolipoma) or vascular channels (angiolipoma) while other variants are rare.[4] Up to 1986, only 100 cases of lipomas arising in the tongue were reported.

Lingual lipomas are usually well-circumscribed, submucosal, less than 1 cm in size and located on the lateral edge of the anterior two-thirds of the lingual surface.[4] There is a male predominance and local discomfort is the common mode of presentation.[4] Clinically the differential diagnosis includes schwannoma, cysts, minor salivary gland adenoma and lingual thyroid.

Liposarcoma accounts for about 15 per cent of all soft tissue tumours.[5] This tumour was first described by Virchow in 1857.[6] The hallmark of liposarcoma is the lipoblast, which is an atypical cell nucleus indented by cytoplasmic fat-containing vacuoles.[1] Liposarcomas are currently classified according the histological features as either well-differentiated/atypical lipoma, myxoid, round cell, pleomorphic or de-differentiated.[7]

Enterline *et al.* were the first authors to suggest a correlation between histological pattern and biological behaviour in liposarcomas.[8] In their paper, they recognized the 'lipoma-like' group as a distinct subset; these well-differentiated neoplasms tended to recur locally, but did not metastasize. This group of tumours was designated 'well differentiated liposarcoma' by Enzinger and Winslow, who further subdivided it into lipoma-like and sclerosing types.[9] Evans suggested a distinction between WDLs occurring superficially and those arising in deep soft tissue and the retroperitoneum;[10] the term *atypical lipoma* is now reserved for the former group. This was based on the observation that local recurrence rate and overall prognosis, due to the superficial location and hence earlier presentation, is better for atypical lympomas than for well-differentiated liposarcomas.

Well-differentiated liposarcomas and atypical lipomas are characterized microscopically by highly differentiated lipocytic cells that resemble normal lipocytes and lack mitotic activity or necrosis. Lipoblasts may be scanty or plentiful but are required to make the histological diagnosis. The presence of chromosomal alterations in atypical lipoma/well-differentiation liposarcoma, most significantly the presence of supernumerary ring or giant marker chromosomes, may be used to cytogenetically differentiate well-differentiated liposarcoma from other lipomatous tumours.[11]

Well-differentiated liposarcoma/atypical lipomas have the tendency to repeated local recurrence; however, they do not possess the capacity for metastasis.[11] Morbidity and mortality are related to the local tumour recurrence rate, which is inversely proportional to completeness of surgical excision. Prognosis is also adversely affected by the propensity of atypical lipomas and well-differentiated liposarcomas to undergo de-differentiation to high-grade sarcoma. De-differentiated liposarcomas are locally aggressive and have the capacity for distant metastasis. De-differentiation appears to be a time-dependent phenomenon being most frequent in deeply located regions where the likelihood of clinical persistence of disease is higher due to incomplete excision.[12]

Up to 1994 there were only 83 reported cases of head and neck liposarcomas.[13] Most were reported prior to Evans' description of the topographic classification of well-differentiated liposarcomas, and there is a possibility that some tumours may have been misdiagnosed. The first reported case of liposarcoma arising in the tongue was published in 1976; the authors reported this case as a well-differentiated liposarcoma.[6] Stewart *et al.* recommended that head and neck well-differentiated liposarcoma in easily accessible areas such as tongue and parotid be classified as atypical lipomas; similar tumours arising in the pharynx, larynx, and deep cervical tissues should be designated as liposarcoma because of the relative difficulty in achieving complete excision.[13]

Our case was diagnosed according to the histological characteristics specified as pathognomonic for atypical lipoma by Weiss and Sobin.[7] Cytogenetic markers were not employed for confirmation. To our knowledge, this case is the fourth reported example of a lingual atypical lipoma, and the first from the British Isles.

The recommended management of head and neck atypical lipomas is wide local excision, with post-operative radiotherapy being reserved for selected cases where adequate resection margins are doubtful or technically difficult to achieve.[6,13,14] The recurrence rate for head and neck atypical lipoma after surgical removal is quoted as 30 per cent;[13] 10-year recurrence rates of eight per cent in cases of atypical lipoma/well-differentiated liposarcoma treated empirically by a combination of surgery and radiotherapy have been reported.[14] Block dissection of first-echelon lymph glands is not necessary for atypical lipoma/well-differentiated liposarcoma because regional metastasis does not occur in the absence of de-differentiation.[13]

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