Pleomorphic Lipoma of the Tongue as Potential Mimic of Liposarcoma

We herein report a rare case of pleomorphic lipoma of the tongue with a review of world literature. A 44-year-old woman presented with a nodule of the tongue that had been present for over three years. Clinical examination revealed a yellowish sub-mucosal lesion, measuring 3 cm in maximum diameter, protruding from lingual surface. A first biopsy showed a lipomatous tumour composed of mature adipocytes intermingled with myxoid areas composed of spindle uniform in size and shape and multinucleated floret-like giant cells. Spindle and giant cells were positive for CD34. A diagnosis of pleomorphic lipoma was made. In view of the benign nature of this mass, it was de-bulked rather than completely excised in order to preserve swallowing function.

KEYWORDS: Differential diagnosis, immunohistochemistry, pleomorphic lipoma

INTRODUCTION

Pleomorphic lipoma (PL) is a rare variant of lipoma on a continuum with spindle lipoma. Microscopically, these tumours are composed of mature fat cells mixed with bland spindle cells, hyperchromatic round cells, and multinucleated giant cells.[1] The vast majority of tumours are located in the subcutaneous tissue of the posterior neck and upper back.[1] PL of the tongue is a very rare and benign tumour. To best of our knowledge, this is the third cases reported in international literature.[2,3] A final diagnosis may be difficult because of the prevalence of spindle and multinucleated pleomorphic cells and the presence of abundant myxoid stroma. We add a new illustrative case to the 2 cases of PL of tongue reported in literature.

CASE REPORT

A 44-year-old woman presented with a slow-growing, painless nodule of the tongue with difficulty in deglutition. Clinical examination revealed a soft, sub-mucosal nodule, measuring approximately 3 cm in maximum diameter, covered by normal mucosa. A surgical excision was performed. Grossly, the mass was poor circumscribed and lipomatous in appearance with mucoid areas. Histological examination showed mature adipocytes, diffusely infiltrating striated muscle fibres, [Figure 1] and abundant mucoid stroma [Figure 2]. In this myxoid background were present bland-appearing spindle cells and multinucleated giant cells with radially arranged nuclei in a "floret-like" pattern [Figure 3]. Some thick rope-like collagen bundles were also present. The spindle and giant cells were strongly positive for CD34 [Figure 4] and negative for S100. A fluorescence *in situ* hybridization (FISH) was performed on formalin-fixed, paraffin-embedded tissue sections using MDM2 (12q15) dual-colour probe (Vysis, Downers Grove, IL, USA). No amplification of the MDM2 gene was disclosed. A final diagnosis of pleomorphic lipoma of the tongue was made. The post-operative course was uneventful; after 1 year, the patient was free of disease although the tumour was present at the margins of the excised specimen and is currently on follow-up.

Access this article online

Quick Response Code:

Website: www.jcasonline.com

DOI: 10.4103/0974-2077.110101

DISCUSSION

Lipomas are the most common mesenchymal tumours of soft tissue but are uncommon in the oral and

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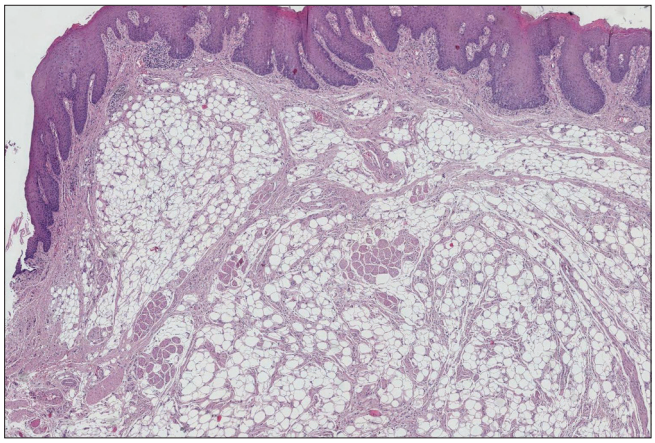

Figure 1: The mass was poor circumscribed and lipomatous in appearance with mature adipocytes, diffusely infiltrating striated muscle fibers (H and E, ×10)

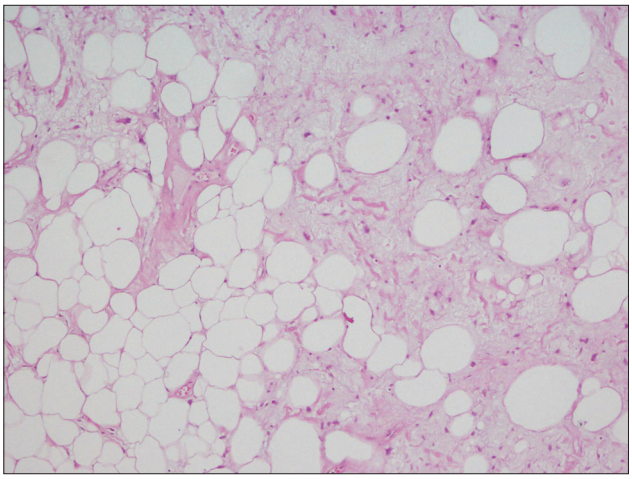

Figure 2: Mature adipose tissue was admixed with abundant mucoid stroma (H and E, ×20)

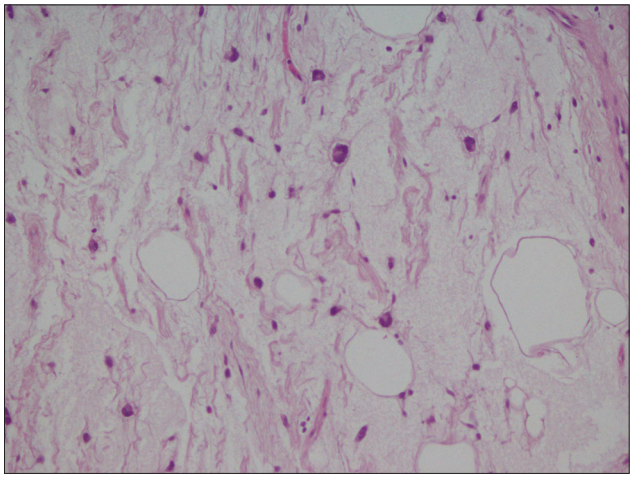

Figure 3: Mucoid areas showed bland-appearing spindle cells and multinucleated giant cells with radially arranged nuclei in a "floret-like" pattern (H and E, ×20)

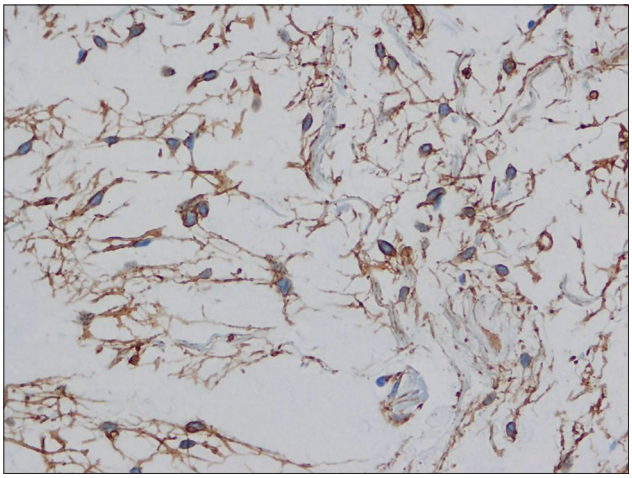

Figure 4: The spindle and giant cells were strongly positive for CD34 (immunoperoxydase, ×40)

maxillofacial region.[4] Classic lipoma is the most frequent type in these sites, but other variants have been described including some cases of atypical lipomatous tumour (ALT)/well-differentiated liposarcoma (WDL).[5]

PL is a distinctive variant of lipoma originally described by Enzinger, characterized by replacement of mature fat by bundle of spindle cells, hyperchromatic round cells and giant cells with floret-like nuclei admixed with dense collagen fibres or within a myxoid stroma.[1] Exceptionally, PL have been described in the tongue; to best of our knowledge, only two case have been reported in literature.[2,3] The different proportion of fat cells, spindle cells, collagen fibres, and mucoid substance confers to PL a variable macroscopic and microscopic appearance that can hamper the diagnosis. Neurofibroma, ancient schwannoma, myxoma, and nodular fasciitis are the lesions that most frequently showed cellular features that overlap with PL. The most difficult differential diagnosis is with ALT/well-differentiated liposarcoma (WDL), and with pleomorphic liposarcoma.[6] The presence of infiltrative margins, the identification of "bizarre" multinucleated giant cells with hyperchromatic nuclei, and some lipoblast-like cells may suggest a diagnosis of malignancy. Moreover, the floret-like cells of PL are not pathognomonic but may be present also in liposarcomas. Unlike the latter, the former lacks of true lipoblasts, cellular pleomorphism, marked vascularisation, mitotic activity, and show an intense immunoreactivity for CD34. In difficult cases, the combination of immunohistochemistry and (FISH) that shows the amplification of MDM2 and CDK4 genes may play a role in differential diagnosis with benign adipose tumours. Despite the worrisome morphological features, PL is benign, rarely recur, and a possible malignant transformation is not described in literature. In conclusion, PL is a benign lesion that

rarely can arise in unusual site as the tongue and is cured by complete local excision. The cytological features and the immunoreactivity for CD34 may suggest a PL addressing a proper surgical procedure.

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How to cite this article: D'Antonio A, Locatelli G, Liguori G, Addesso M. Pleomorphic lipoma of the tongue as potential mimic of liposarcoma. J Cutan Aesthet Surg 2013;6:51-3.

Source of Support: Nil. **Conflict of Interest:** None declared.