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Low-Fat Spindle Cell Lipoma of Oral Cavity: Report of a Rare Case

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Abstract:

Objective(s): Spindle cell lipoma (SCL) is a rare variant of lipoma that is uncommon in oral cavity which displays as a slow-growing, painless mass. The histopathologic diagnosis is challenging especially in low-fat/fat-free subtypes, and immunohistochemical staining is of great help in differential diagnosis. The current study aimed to report a rare case of low-fat SCL. **Case:** A 38-year-old male with a relatively large lesion on buccal mucosa was referred. The interventions included excisional biopsy, H&E staining, and immunohistochemical examinations. H&E study showed a peripheral soft tissue tumor. The underlying connective tissue showed collections of fatty tissue intermixed with spindle cell proliferations. In IHC study, Spindle cells were stained strongly and diffusely for CD34, and no S100 staining was detected in neoplastic spindle cells. Toluidine blue staining showed mast cells that were detected throughout the connective tissue. These findings confirmed the diagnosis of low-fat SCL. **Conclusion:** Oral pathologists should be familiar with the clinicopathologic characteristics of this rare lesion in order to avoid misdiagnosis.

Keywords: Lipoma; Mouth; Soft Tissue Neoplasms

Introduction

Lipomas are benign mesenchymal tumors that originate from adipose tissue and most frequently arise in the trunk and extremities [1-4]. Within the oral cavity, they constitute only about 1–4% of all lipomas and account for approximately 0.1–5% of benign oral tumors [1]. Clinically, they are slow-growing, painless lesions that can develop in any region containing adipose tissue [2]. Oral lipomas have been documented in multiple areas such as the major salivary glands, buccal mucosa, lips, tongue, palate, and floor of the mouth, with the buccal mucosa and buccal vestibule being the most frequently affected areas [3].

The designation “lipoma” encompasses a spectrum of histologic subtypes such as conventional lipoma, fibrolipoma, spindle cell lipoma (SCL), angiolipoma, pleomorphic lipoma, sialolipoma, chondrolipoma, and intramuscular lipoma [1-4] These subtypes are mainly distinguished by the microscopic appearance of their tumor cells and stromal components [2]. Histopathologically, most oral cases correspond to either conventional lipoma or fibrolipoma [3], whereas the diagnosis of less common variants can be particularly challenging [2].

SCL was first characterized by Enzinger and Harvey in 1975 [5]. Typically, it presents as a well-demarcated, solitary, subcutaneous mass situated in the posterior neck, shoulder, or upper back of older men. Involvement of the oral cavity is rare—between 1984 and 2012 only 35 well-documented intraoral cases were reported [6]. Oral SCL comprises about 1.4% to 9.8% of all intraoral lipomas [7]. Histological variants of SCL include fibrous, plexiform, vascular, pseudoangiomatous, myxoid, fat-rich, and low-fat/fat-free types [2, 8-10]. Diagnosis is usually straightforward when the lesion displays its hallmark mixture of mature adipocytes and uniform CD34-positive spindle cells; however, identification becomes more difficult in low-fat or fat-depleted variants [11]. Billings and Folpe [8] defined these subtypes as those containing fewer than 5% of mature adipocytes. Because of the minimal adipocytic component and overlapping morphology, such cases may be misinterpreted as other benign or malignant soft-tissue neoplasms. Immunohistochemistry is therefore critical: diffuse CD34 positivity of spindle cells with negative S-100 expression excludes benign nerve sheath tumors, as S-100 positivity should be confined to adipocytes.

Differential diagnoses of SCL include neurofibroma, solitary fibrous tumor (SFT), well-differentiated liposarcoma/atypical lipomatous tumor (WDLS/ALT), hibernoma, angiomyxoma, angiomyofibroblastoma, and low-grade sarcoma [8,11]. Management is conservative, involving simple surgical excision, and recurrence is exceedingly uncommon [2]. This case report aimed to define a 38-year-old male with a relatively large spindle cell lipoma of the buccal mucosa. This report emphasizes the significance of careful clinicopathologic examination in order to achieve a precise diagnosis.

Case

A 38-year-old male had been referred with a relatively large painless exophytic submucosal mass on left buccal mucosa. The surface of the lesion was slightly lobulated and partially

ulcerated. It was sessile and pink with firm consistency, measuring 2.5×2×2 cm^3. Extraoral examination was unremarkable, and no cervical lymphadenopathy was detected. Patient's past medical history was clear.

Excisional biopsy was performed under local anesthesia with the provisional diagnosis of a reactive lesion such as focal fibrous hyperplasia or a benign soft tissue neoplasm such as neurofibroma or schwannoma. The gross of the lesion showed a well-circumscribed solid, white, and homogenous mass (Figure 1).

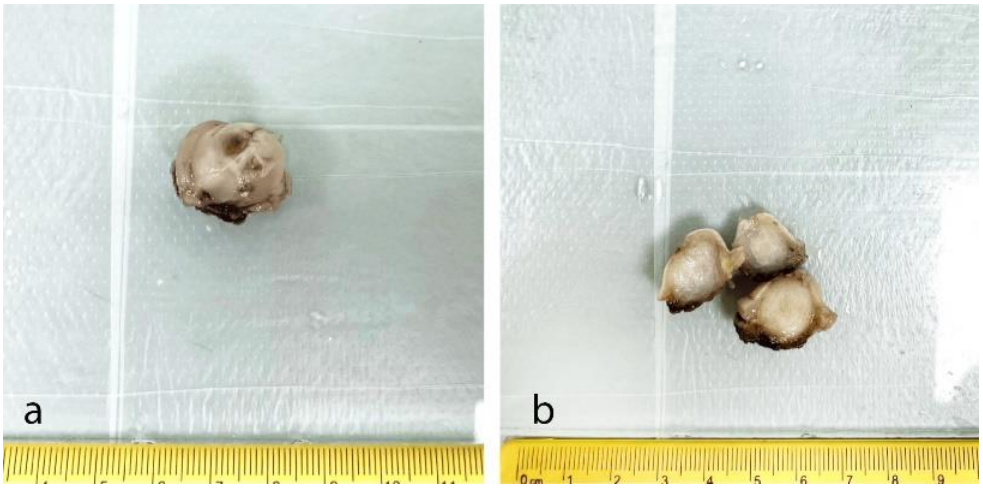

Figure 1: Gross findings of the lesion. a. A well-circumscribed solid, white, and homogenous mass with superficial ulceration. b. Cut surface consistency ranged from soft to firm.

It was processed by Hematoxylin and Eosin staining for microscopic study. Histopathologic examination exhibited a peripheral soft tissue tumor covered by focally ulcerated oral mucosa. The underlying connective tissue depicted collections of fatty tissue intermixed with spindle cell proliferations (Figure 2).

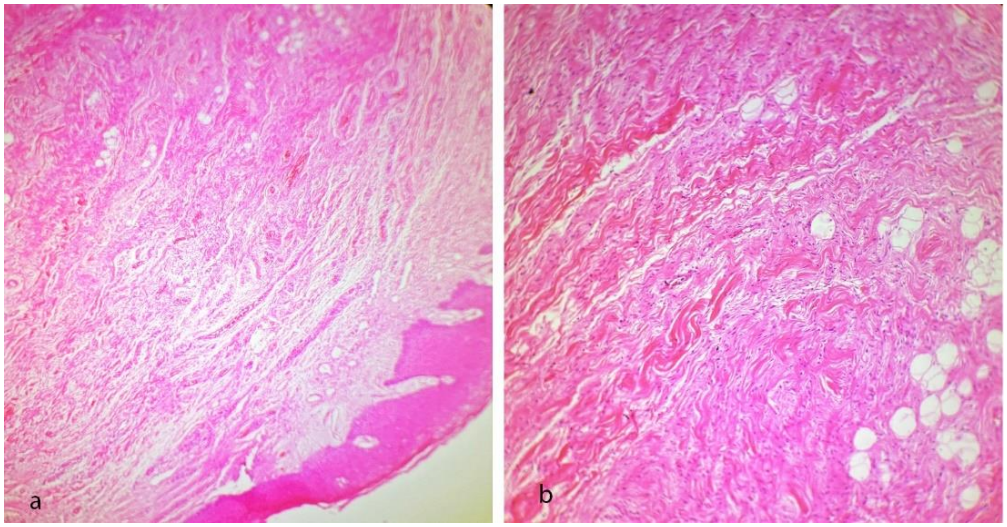

Figure 2: Photomicrograph of the lesion showing ulcerated stratified squamous epithelium along with a connective tissue with collections of fatty tissue intermixed with spindle cell proliferations (H&E, a:40X. b:100X)

The nature of the spindle cells was then determined by immunohistochemical (IHC) study. The neoplastic spindle cells were strongly and diffusely positive for CD34 and negative for S100 (Figure 3), whereas S100 positivity in adipocytes was detected (Figure 4), Toluidine blue staining was also performed, and multiple mast cells were also detected throughout the connective tissue (Figure 5). These histopathological findings confirmed the diagnosis of spindle cell lipoma (SCL). The patient remained disease-free during a 14-month follow-up period.

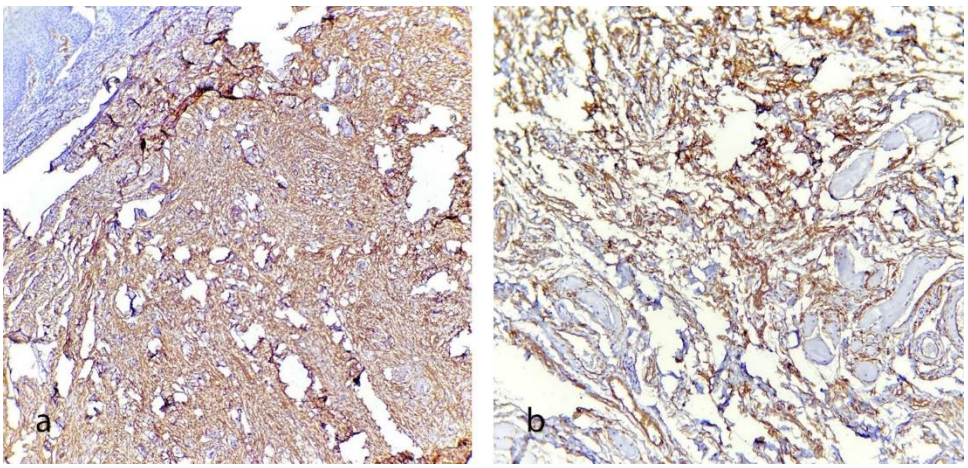

Figure 3: Photomicrograph of immune-stained lesion showing strong diffuse positivity to CD34 in spindle cells. (IHC, a:100X. b: 400X,)

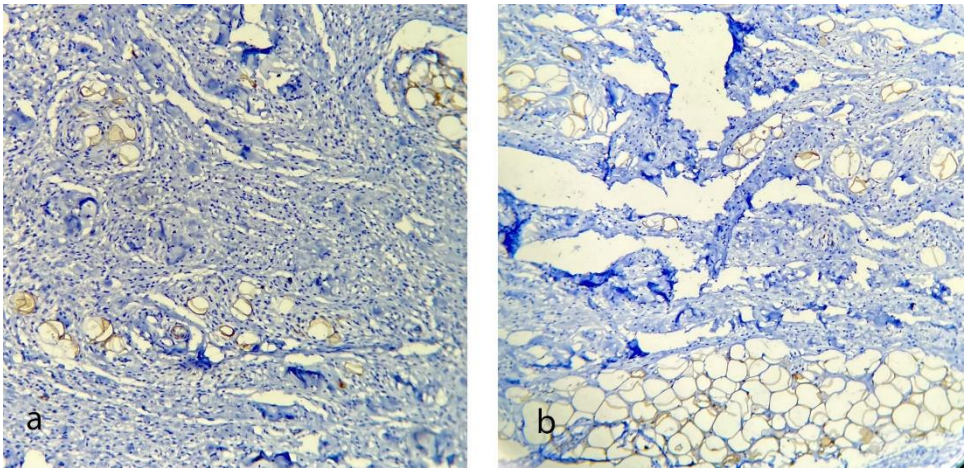

Figure 4: Photomicrograph of immune-stained lesion showing positivity to S100 confined to adipocytes (IHC, a:100X, b:400X)

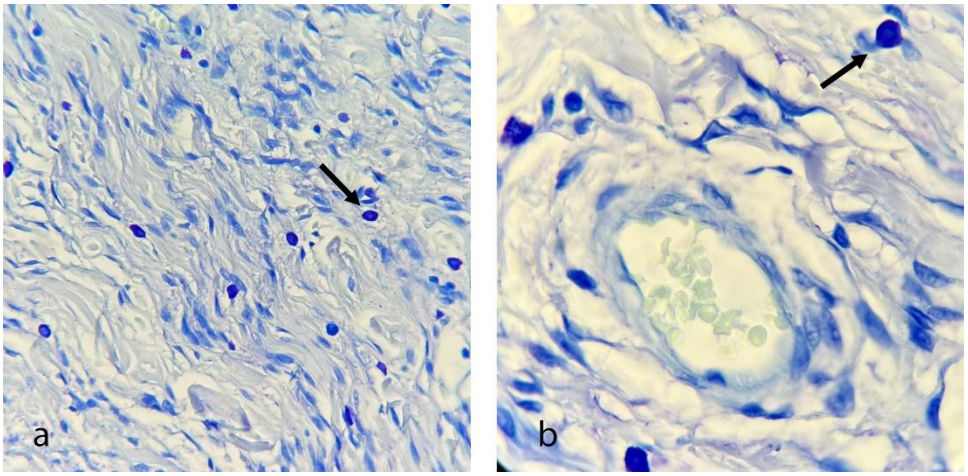

Figure 5: Photomicrograph of lesion stained by toluidine blue showing multiple mast cells throughout the lesion (TB, a:100X, b:400X)

Discussion

Spindle Cell Lipomas (SCLs) are uncommon variant of lipoma[12]. They frequently affect subcutaneous regions of the trunk and neck. SCL and pleomorphic lipoma (PL) are categorized as a single tumor; SCL/PLs account for about 1.5% of all adipocytic neoplasms and rarely occur in the oral cavity[12, 13]. In the head and neck area, SCL may arise in various intraoral and perioral sites, including the major salivary glands, lips, buccal mucosa, palate, tongue, and floor of the mouth [2]. It shows a slow-growing painless mass that does not adhere to the adjacent skin or mucosa [13]. The clinical course is usually asymptomatic unless they show a large size [4].

The present case was a 38-year-old male that had been referred with a relatively large painless exophytic submucosal mass on left buccal mucosa, who showed focal ulceration, which is probably due to its large size and secondary trauma. Lemos et al.[12] also reported a superficial ulceration of the SCL lesion. There is a male predilection for SCL with a mean age of 54 years, being uncommon in children and adolescents [2, 12]. The present patient was male and in 4th decade of life. The clinical differential diagnosis of intraoral SCL is related to the anatomic location of the lesion, where salivary gland tumors and benign mesenchymal neoplasms can be proposed [12].

Histopathologically, SCL is characterized by an intimate admixture of mature adipocytes and uniform spindle-shaped cells embedded within a collagenous and variably myxoid stroma, often containing delicate blood vessels and scattered mast cells—features that help distinguish it from other lipomatous neoplasms. Several morphologic subtypes of SCL have been reported, including fibrous, plexiform, nodular, vascular, angiomatous, pseudoangiomatous, low-fat, and fat-free variants [2, 4, 8-10]. Mitotic figures and necrosis are exceedingly rare, although occasional pleomorphic or multinucleated spindle cells may be present in limited areas [12].

The spindle cell component of SCL exhibits strong immunoreactivity for CD34, serving as a key diagnostic marker. Moreover, mast cells dispersed within the tumor stroma can be readily demonstrated using mast cell tryptase immunostaining or toluidine blue staining, both highlighting their abundance within the connective tissue. In contrast, the adipocytic elements of the lesion show positive staining for S100 protein, confirming their adipose differentiation [12]. Spindle cells in the current case also displayed a strong diffuse reaction with CD34 and positivity for S100 in adipocytes as well as MCs presence throughout the lesion's connective tissue.

Differences in fat cell size, nuclear atypia, and mitotic figures are suggestive of liposarcoma, and IHC staining is frequently positive for MDM2 and CDK4 [13]. Traumatized oral lipomas may display degenerative cellular alterations that can be misdiagnosed with a well-differentiated liposarcoma identified as an atypical lipomatous tumor [7].

Other important differential diagnoses of SCL in the oral cavity include neurofibroma, solitary fibrous tumor (SFT), and low-grade sarcoma [8, 11]. In these diagnostic challenges, pattern of S100 positivity, expression of CD34 and STAT6, and Ki-67 index are of great help, respectively.

Conservative surgical excision is the most designated treatment for oral SCL[12] as they are encapsulated lesions with good prognosis and without recurrence [13].

Conclusion

Oral SCL is a rare and histopathologically distinct variant of lipoma, and histopathologic diagnosis may be challenging. Due to the potential pitfalls, an accurate microscopic analysis of spindle cells is highly recommended for definitive diagnosis.

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