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Atypical Presentation of Spindle Cell Lipoma: Diagnostic Challenges and Concise Review of Literature

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Aim: This report aims to contribute to the current knowledge base by presenting a unique case of intra-oral Spindle cell lipoma (SCL) arising in an uncommon location, the buccal mucosa, and affecting a less frequent sex, female. SCL poses a significant diagnostic challenge due to its histological resemblance to malignant neoplasms. This is compounded by the scarcity of comprehensive clinical data. Consequently, we provide a concise literature review on SCL to contribute to a comprehensive understanding of its clinical features, optimal treatment approaches, and long-term outcomes.
Materials and methods: We present a clinical case of a female patient with a buccal mass. The excised soft tissue underwent histochemical and immunohistochemical staining (CD34) for confirmatory diagnosis. A comprehensive review of relevant literature on SCL was conducted, focusing on diagnostic criteria, differential diagnoses, treatment modalities, and reported patient outcomes.
Results: Here, we report a case of SCL arising in the buccal mucosa of a female patient. Histopathological examination with both histochemical and immunohistochemical staining (CD34) confirmed the diagnosis. Our review of the literature emphasizes the rarity of SCL, the diagnostic challenges it presents, and the limited understanding of its typical clinical course and prognosis.
Conclusion: This report contributes to the existing knowledge on SCL by presenting a unique case with an unusual location and affecting a less commonly reported patient sex. Further research is necessary to establish a more comprehensive understanding of SCL's clinical behavior, optimal treatment approaches, and long-term outcomes to improve patient management.

Keywords: spindle cell lipoma, CD 34, intraoral, buccal mucosa, female

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Introduction

Lipomas are benign mesenchymal tumors that are quite common. Lipoma is typically made up of mature adipose tissue exhibiting adipocytes of variable sizes. However, there are different types of lipomas that demonstrate distinct histologic characteristics, such as pleomorphic lipoma, angiolipoma, chondrolipoma, fibrolipoma, and spindle cell lipoma. In the oral cavity, lipomas are relatively uncommon and account for only 1%-4% of all intra-oral soft tissue tumors.[1]

Though lipomas are common soft tissue tumors, a rarer subtype, spindle cell lipoma (SCL), presents a significant diagnostic hurdle. These slow-growing lumps can mimic malignant tumors due to their unusual spindle-shaped cells. Unfortunately, understanding SCL is hampered by limited research. This scarcity of data makes establishing definitive diagnostic and treatment protocols challenging. Initially misclassified as a liposarcoma in 1944, SCL wasn't recognized as a distinct entity until 1975 by Enzinger and Weiss, highlighting the ongoing efforts to fully understand this unique type of lipoma.[2]

This report details a rare case of intraoral SCL affecting the buccal mucosa of a 53-year-old female patient. The buccal mucosa is an uncommon location for SCL presentation, and SCL is typically more prevalent in males.

Case presentation

53-year-old woman presented with a painless, 2-year history of a right buccal swelling opposite the lower premolars. Clinical examination revealed a 20×20 mm, exophytic mass with smooth, pink, non-ulcerated mucosa. Palpation revealed a moderately firm and mobile mass. Her past medical history was unremarkable for any significant conditions or allergies. The differential diagnosis included fibroma, lipoma, or a benign minor salivary gland due to the mass's location, clinical features, and slow growth. After obtaining written informed consent, the patient underwent an excisional biopsy under local anesthesia. Postoperative care was uneventful, and the patient was discharged.

Materials and methods

The patient signed an informed consent. This work was approved by the Institutional Review Board (FUE.REC (7)/3-2024). The entire excised mass was sent for histopathological examination. Gross examination revealed a well-circumscribed, encapsulated mass. Sections were routinely stained with hematoxylin and eosin (H&E) for histological evaluation and immunohistochemically stained with CD34 to confirm the diagnosis. (fig. 1)

Results

Microscopic examination of hematoxylin and eosin-stained sections revealed a neoplasm composed of uniformly distributed, mature adipocytes of moderate size, interspersed with rope-like collagen fibers harboring a characteristic fine vascular network. Bland spindle cells, arranged in bundles, permeated the fat cells. A well-defined fibrous capsule surrounded the neoplasm, with the subcapsular area devoid of tumor cells. Immunohistochemical staining revealed strong cytoplasmic immunopositivity for CD34 within the spindle cells. These histological features, in conjunction with the immunohistochemical findings, were diagnostic of a classical SCL. Three weeks post-operatively, the patient returned for follow-up. The buccal swelling had receded, and the overlying mucosa had healed well, with the patient maintaining normal speech and mastication function (fig. 1 and 2).

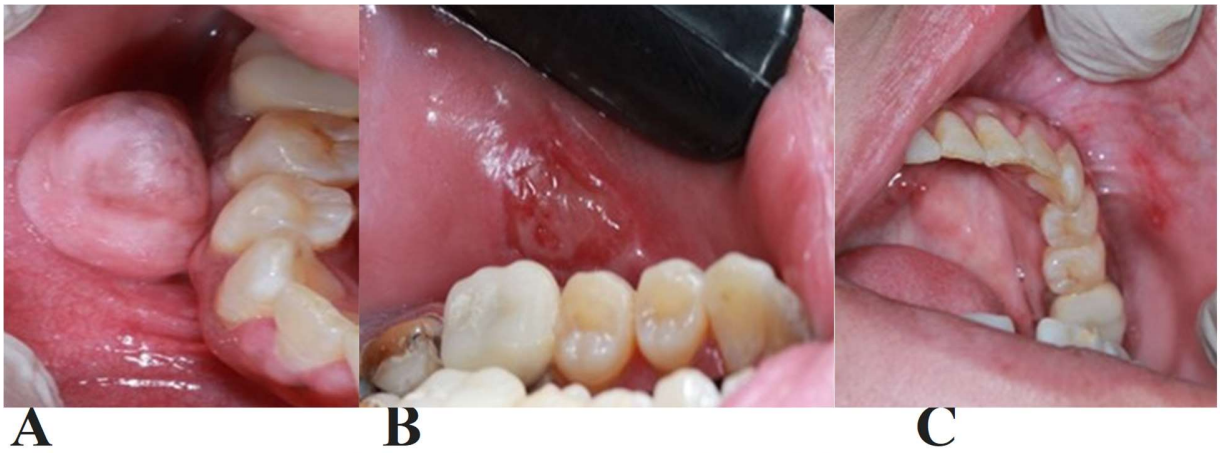

Figure 1: Intra-oral examination revealed a swelling on the right buccal mucosa (a) post-operative picture after the local excision of the spindle cell lipoma. (b) Clinical picture showing the healing process 3 weeks after the excisional biopsy (c)

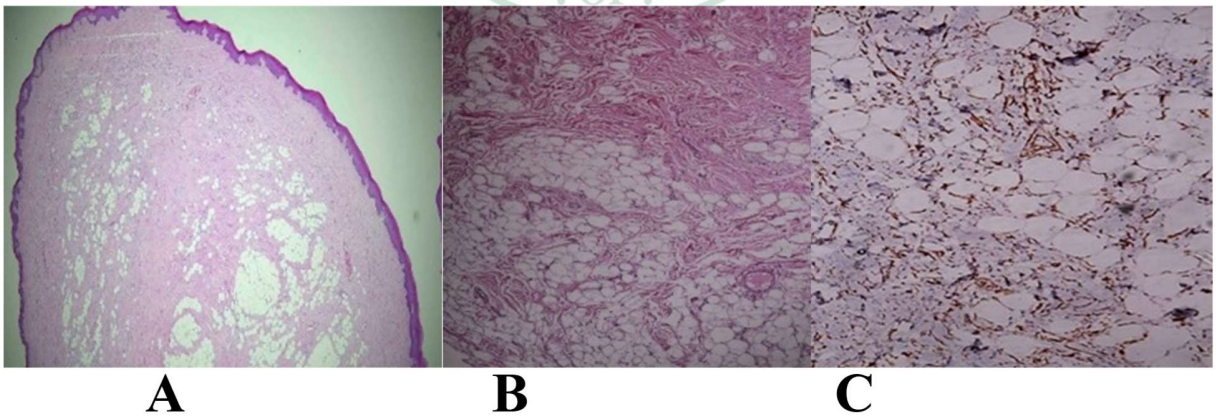

Figure 2: photomicrograph of SCL showing (a) A fibrous capsule and the subcapsular area was free of tumor cells (H&E, orig. mag. x4) (b) mature adipocytes and small uniform spindle cells (H&E, orig. mag x10) (c) spindle cells exhibiting immunopositivity (CD34, orig. magx20)

Discussion

Spindle cell lipoma (SCL) is a rare histological subtype of lipoma, first described by Enzinger and Weiss in 1975.[3] Unlike typical lipomas, which present as soft, slow-growing nodules, SCL can mimic malignant tumors due to the presence of spindle-shaped cells1. Although uncommon, SCL can occur in various intraoral locations, including the tongue, buccal mucosa, floor of the mouth, hard palate, lip, and alveolar ridge.[2]

SCL presents a unique clinicopathological entity with features overlapping those of pleomorphic lipoma. Some authors suggest they may represent a spectrum of the same disease process. Classically, SCL appears as a painless, slow-growing, mobile nodule in the subcutaneous tissue of older men, with a higher prevalence in the neck, upper back (shawl region), and less frequently in the face and oral cavity.[1,2] While SCL can occur at any age, it's most common between the fifth and seventh decades of life. Interestingly, cases involving women tend to present with lesions in less common locations and at a slightly younger age.[2] In rare instances, multiple SCL lesions may suggest a familial predisposition. [4]

SCL exhibits distinct immunohistochemical and molecular features that aid in diagnosis and differentiation from other tumors. CD34, a marker for endothelial and precursor cells, shows strong cytoplasmic expression in SCL's spindle cells. Conversely, these cells lack retinoblastoma protein expression. Desmin, a marker for muscle cells, can be expressed in a minority of cases[5]. MDM2 amplification has been identified as the most common genetic anomaly, observed in up to 80% of SCL cases. Recent advancements in molecular techniques have unveiled potential genetic drivers of SCL. MDM2 amplification, observed in up to 80% of cases, is the most frequent anomaly. MDM2 negatively regulates the p 53 tumor suppressor gene, critical for cell cycle control and apoptosis. Amplification disrupts this regulation, leading to uncontrolled cell proliferation. [6]

The precise origin of SCL's spindle cells remains unclear, although several theories exist. One hypothesis suggests their derivation from fibroblasts, which could undergo transformations leading to their spindle-shaped appearance. Alternatively, these cells might originate from immature fat cells (lipoblasts), supported by the presence of mature adipocytes interspersed within SCL. [7,8] The ability of SCL's spindle cells to differentiate into mature adipocytes under specific conditions further suggests a potential link between these cell types. Based on additional histopathological features, researchers have proposed various SCL subtypes, including classic, fibrous, myxoid, low-fat, pseudoangiomatous, and fat-rich variants. [9]

The presence of spindle cells alongside mature adipocytes is a key diagnostic feature of SCL. However, their morphology alone is not sufficient for definitive diagnosis as similar spindle cells can occur in other tumors, including liposarcoma and other soft tissue neoplasms. [5,8] While as mentioned earlier precise origin and function of spindle cells in SCL are still unclear, understanding their morphology, and immunohistochemical signatures plays a crucial role in accurate diagnosis and differentiation from malignant mimics. [10]

It is important to distinguish these tumors from other neoplasms because incorrect assessment of their benign nature can result in overtreatment. SCL can be mistaken for well-differentiated liposarcomas/atypical spindle cell lipomatous tumors (WDLS), especially when found in a deep location such as the larynx. [11] Other lipomatous or spindle cell lesions such as neurofibroma, schwannoma, angiolipoma,

solitary fibrous tumors, and dermatofibrosarcoma protuberans, share histologic similarities and may also be considered in the differential diagnosis. [12, 13] Complete surgical excision is the mainstay of treatment for SCL. This approach effectively removes the lesion while minimizing the risk of recurrence. Wide margins are not generally necessary, as SCL exhibits well-demarcated borders and minimal propensity for aggressive behavior. Minimally invasive techniques, such as liposuction or needle aspiration, are not recommended due to the possibility of incomplete removal and subsequent recurrence. [5,13,14,15]

Conclusion

SCL in this location is exceptionally rare, with few cases reported in the literature. This case report aims to contribute to the understanding of this uncommon presentation and aid in the diagnosis of similar lesions.

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Data availability

The datasets used and/or analyzed during the current study are available from the corresponding author upon reasonable request.

Ethics approval and consent to participate

Ethical approval was obtained from Future University in Egypt, faculty of oral and dental medicine Institutional Review Board (FUE.REC (7)/3-2024).

Consent for publication is not applicable.

Competing interests

The authors declare that they have no competing interests.

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