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Case Report

Intraoral myxoid fibrolipoma with chondroid areas: Diversified differentiation or a mere coincidence?

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Abstract

Background: Oral mesenchymal tumors are uncommon lesions, presenting as slow-growing exophytic masses. Myxofibrolipoma (MFL) is a lipoma variant comprising mature fat cells admixed with variable quantities of mucinous and fibrous stroma.

Case Presentation: We present a case of a 15-year-old female presenting with swelling on the inner side of her left cheek, which she had noted for two weeks. With clinical differential diagnoses of mucocele, fibroma, and granuloma, the swelling was excised. Histopathological examination revealed it to be MFL with chondroid foci. Considering the site of the lesion and the absence of morphological signs of injury or irritation, labeling the chondroid foci as 'metaplastic' seemed doubtful in our case.

Conclusion: The presented case highlights the plausible capacity of mesenchymal tumor cells in lipoma variants to exhibit divergent differentiation and reaffirms the importance of having a standardized nomenclature.

Keywords: Oral, Neoplasia, Metaplasia

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Introduction

The oral cavity is known to be a site for a myriad of mesenchymal neoplasms with intriguing histogenesis and varied histological combinations of tissues. Lipomatous tumors are one such category of tumors that can occur as pure lipomas or in combination with other tissue elements. Many unconventional variants have been described in literature with an admixture of mesodermal elements along with mature adipose-tissue-like vascular components, cartilage, myxoid areas, and fibrous elements; they have been named angiolipoma, chondrolipoma, myxolipoma, and fibrolipoma respectively.[1,2] Myxoid fibrolipoma is one such entity composed of myxoid areas and dense fibrous tissue. Cartilage, when present, can be due to metaplasia attributed to inflammation/irritation or as an indigenous part of the lesion itself. We report a case of myxoid fibrolipoma with chondroid foci without histological or clinical signs of inflammation. This finding is rare and leaves the question of the origin of these chondroid foci unanswered.

Case Report

A 15-year-old female presented to the dental outpatient department with a complaint of swelling on the inner side of her left cheek for two weeks. There was no history of trauma or dental procedures. Dental hygiene was good. There was no increase in the size of the swelling and no pain associated with the swelling. On examination, a single firm globular swelling was noted in the inner cheek buccal mucosa (Figure 1a). With a clinical differential diagnosis of mucocele, fibroma, and granuloma, the swelling was excised and sent for histopathological examination. Gross examination revealed a firm grey-white nodule measuring 2.5 × 2 × 1 cm. The external surface was unremarkable. The cut section revealed a well-encapsulated mass, showing pearly white areas admixed with homogenous yellow foci (Figure 1b).

Microscopy showed an intact surface lining of stratified squamous epithelium. Beneath was an admixture of myxoid, lipomatous, and fibrous tissue in varying proportions with a few congested blood vessels. The chondroid tissue foci were intermingled with lipofibromatous areas. No atypia/increased mitotic activity or features of malignancy were noted. A final diagnosis of myxoid fibrolipoma of the left cheek mucosa with chondroid areas was made (Figure 2a and 2b).

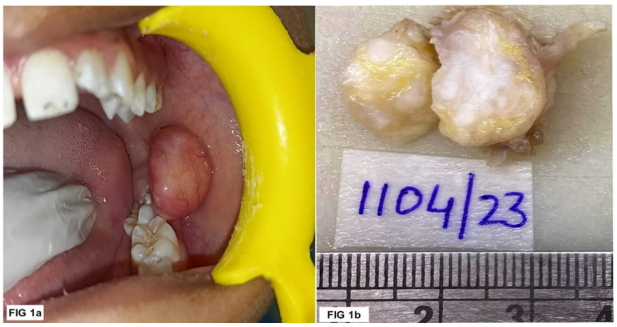

Figure 1. (a) Globular, smooth surfaced swelling in the inner cheek buccal mucosa. **(b)** Cut section of the tumor showing a well-encapsulated mass with pearly white areas admixed with homogenous yellow foci

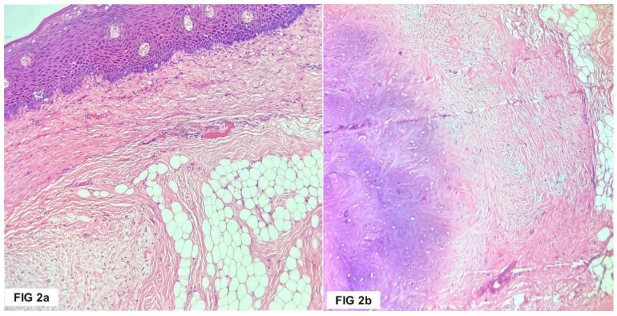

Figure 2. **(a)** Low-power view of the excised intraoral mass showing an intact surface lining of stratified squamous epithelium with a sub epithelium showing an admixture of myxoid, lipomatous, and fibrous tissue in varying proportions (H & E 10X). (**2b**) Chondroid areas admixed with myxoid, lipomatous, and fibrous tissue in the periphery (H & E 10X)

Discussion

The oral cavity is the site for various mesenchymal tumors with varying histogenetic and morphological patterns. As a result, their nomenclature is based on a mixture of tissue elements seen morphologically on microscopy. Myxoid fibrolipoma (MFL) is one such rare histological variant of lipoma. These lesions are predominantly non-pedunculated, solitary, and dome-shaped.[3] Though the most common site of lipoma and its variants is the trunk, shoulders, upper arms, and neck, cases in oral locations have been reported, constituting 1%–4% of such lesions.[4] Oral lipoma and its variants occur in all age groups, most frequently in subjects 40–60 years old.

Histologically, myxoid fibrolipomas comprise an admixture of mature adipose tissue with fibrous and myxoid components. The nuclei of fibroblasts show varied staining from negative to focal positive for CD34 and negative for S-100 and smooth-muscle actin. Spindle cell lipomas are distinct histological differentials that must be considered due to the presence of unique cytogenetic aberrations. They have been postulated to originate from adipose tissue composed of collagen-forming spindle cells with dendritic cytoplasmic processes and extensive myxoid changes. They are desmin-negative and express loss of chromosomes 12q and/or 16q, which may confirm the diagnosis when doubtful.[5] The absence of spindle cells is one of the differentiating features of myxoid fibrolipoma. Pedunculated lipofibroma and nevus lipomatosus cutaneous superficialis share similar microscopic features, like the presence of mature adipocytes between the collagen bundles of the dermis and the presence of myxoid material. However, clinically pedunculated lipofibromas are usually solitary pedunculated lesions with a predilection to the thigh and buttocks, in contrast to myxoid lipofibroma.[6,7] The term "myxofibrolipoma" is proposed for similar lesions in McKee's *Pathology of the Skin*, 5th edition. However, it can be semantically confused with other soft tissue lesions like fibromyxolipoma, myxofibrosarcoma, and superficial fibromyxoma. As there are no fixed criteria to define this lesion, very little conclusive literature could be retrieved about its clinical features, etiology, and pathological features.

In the present case, an additional rare finding was the presence of chondroid/cartilaginous foci. Very few cases of fibrolipoma variants with chondroid or osseous metaplasia have been reported in the literature. Osseous and chondroid metaplasia in the lipomatous lesions are rare, usually seen in long-standing cases, and are proposed to have been produced by the differentiation of undifferentiated mesenchymal cells of the stroma.[8] Literature search revealed two hypotheses are proposed for the origin of chondroblasts and osteoblasts in mesenchymal tumours. One is that the undifferentiated mesenchymal cells undergo neoplastic transformation that later differentiate into lipoblasts, chondroblasts, or osteoblasts and fibroblasts. The other theory is that only the adipose cells have a neoplastic transformation, and differentiation of undifferentiated mesenchymal cells of stroma produces the cartilage and bone. Though our case showed a mixture of foreign elements, the use of the term choristoma was avoided as typical oral cavity cartilaginous choristomas are seen in the midline and are predominated by cartilaginous elements, both of which were absent in our case.[9,10]

The current case adds to the sparse information about the occurrence of chondroid areas in myxoid fibrolipoma. The admixture of these chondroid areas can be labeled metaplastic when occurring secondary to trauma or irritation. Considering the site of the lesion and the absence of clinical and morphological signs of injury or irritation, labeling these foci as 'metaplastic' seems doubtful in our case. Further large-scale studies on similar lesions with similar combinations of mesenchymal elements may shed more light on such cases.

Conclusion

In our case, the oral mesenchymal tumor showed a varying combination of lipomatous, fibrous, and myxoid areas, leading to its categorization as myxoid fibrolipoma, an established entity in literature. In addition, a few

chondroid foci were seen without signs of inflammation or irritation, rendering the metaplastic nature of the cartilage doubtful. When a varying combination of elements is present in a tumor, we propose to use standardized nomenclature with mention of unclassified/ doubtful elements in the final impression, as in our case; future research may confirm the histogenesis of these intriguing changes.

Authors' Contribution

Conceptualization: Archana Shetty and Vijaya Mysorekar.
Data curation: Hima Shree Edupuganti and Aakanksha Koul.
Formal analysis: Sharmila G.S.
Investigation: Archana Shetty and Hima Shree Edupuganti.
Methodology: Sharmila G.S.
Project administration: Archana Shetty.
Resource: Sharmila G.S.
Software: Archana Shetty.
Supervision: Vijaya Mysorekar.
Validation: Vijaya Mysorekar.
Visualization: Archana Shetty.
Writing-original draft: Hima Shree Edupuganti and Aakanksha Koul.
Writing-review & editing: Archana Shetty.

Competing Interests

None declared.

Data Availability Statement

Confidentiality of the data has been maintained.

Ethical Approval

Not applicable; however, patient consent has been acquired.

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