

Labial osteolipoma, clinical and surgical management of an infrequent disease

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Abstract:

Lipomas are relatively infrequent benign neoplasms of connective tissue that can involve oral mucosa. Different histological subtypes of oral lipomas were previously described in current literature. The aim of this work is to report a clinical case of an 83-year-old male, who presented with a 20-years-old labial mass. Surgical removal was performed, and the histopathological diagnosis reported an oral Osteolipoma (OL). Discussion: There are previous reports of oral lipomas that have histologically presented a combination with other cell lines such as bone and cartilaginous tissue within the tumor. The rarity of its presentation makes this entity an infrequent diagnosis for clinicians and specialists in Oral Medicine. The provisional diagnosis could be aided with X-ray when the mass exam revealed indurated areas and calcifications. There are few cases described of OL in this oral subsite. Because of its unusual presentation, OL of the oral mucosa is a true challenge in oral diagnosis.

Keywords: Lipoma. Soft Tissue Neoplasms. Surgical Procedures, Operative. Diagnosis, Differential

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INTRODUCTION

Lipomas are considered benign mesenchymatic neoplasms of soft tissues with an incidence of 1-4,4% when oral mucosa is involved¹. In such cases, the buccal mucosa, the tongue and the floor of the mouth are the most frequent sites². Lipomas usually present as an asymptomatic tumor growing in size during months even years until its diagnosis³.

The yellowish colour is a distinctive feature and the thin epithelial surface lets superficial blood vessels become transparent. The differential diagnosis could involve other benign tumours such as neurofibromas, irritative or traumatic fibromas, adenomas, etc². Histologically, lobules of mature lipocytes separated by collagen fibres are the most important findings for diagnosis⁴. Although in these cases the histopathological diagnosis is simple, different kinds of histological subtypes have been described: fibrolipoma, angioliipoma, pleomorphic lipoma, fusiform cell lipoma, condrolipoma, etc³. Osteoliipoma (OL) is considered a rare subtype due to the presence of bone tissue within the tumor mass⁵.

The aim of this study is to report a clinical case of a 20-years-old-labial mass with a histopathological diagnosis of OL.

CASE REPORT

An 83-years-old male patient presented denoting an increase of the labial volume. During anamnesis hypertension and four cardiac by-pass surgeries were recorded. The patient also reported a basal cell carcinoma surgery located on the surface of the auricular pavilion.

During the oral exam, a notorious mass was observed on the lower labial mucosa. The patient revealed that the lump had 20 years old of evolution and emphasised a very slow growth during that period of time. However, he noted an increasing aesthetic discomfort. The yellowish mass surface showed a lobed and smooth surface, painless and a cartilaginous consistency (Figure 1 A-B). Previous lab exams, surgical removal was performed. The clinical differential diagnosis included pleomorphic adenoma with ossification and lipoma, and other salivary gland or connective tissue tumours with dystrophic calcification... The lesion was easily enucleated, showing an easy divultion in depth from the muscular planes (Figure 2 A-B-C). The specimen (Figure 2 D) was fixed in Formalin 10% for its histopathological examination.

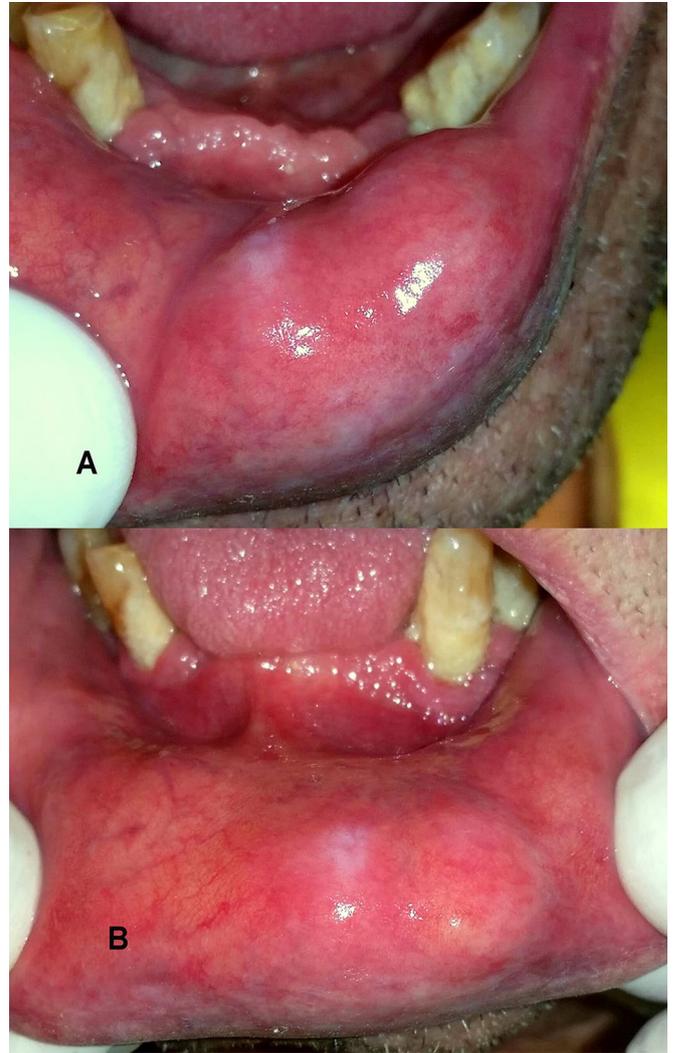


Figure 1. A-B Clinical presentation of the tumoral labial lesion of 3.5 cm x 1.6 cm. The overlying mucosa showed normal appearance. An irregular zone could be observed on the distal region of the lesion. A hardness consistency was associated with calcifications.

On sectioning, the mass exhibited a yellow soft tissue with various bony structures and a well-limited capsule.

Histopathological examination with H&E staining revealed a mass composed by mature adipose tissue with absence of cell atypia and vascularized connective tissue with acidophilic areas of bony tissue (Figure 3 A). Adipocytes did not show any morphological anomaly and they were in contact with thin bands of collagenous fibres and well-formed bone trabeculae which harboured osteocytes without atypia (Figure 3 B). A final diagnosis of OL was considered.

The patient had a satisfactory response and no signs of remission were showed two weeks after the surgical removal.

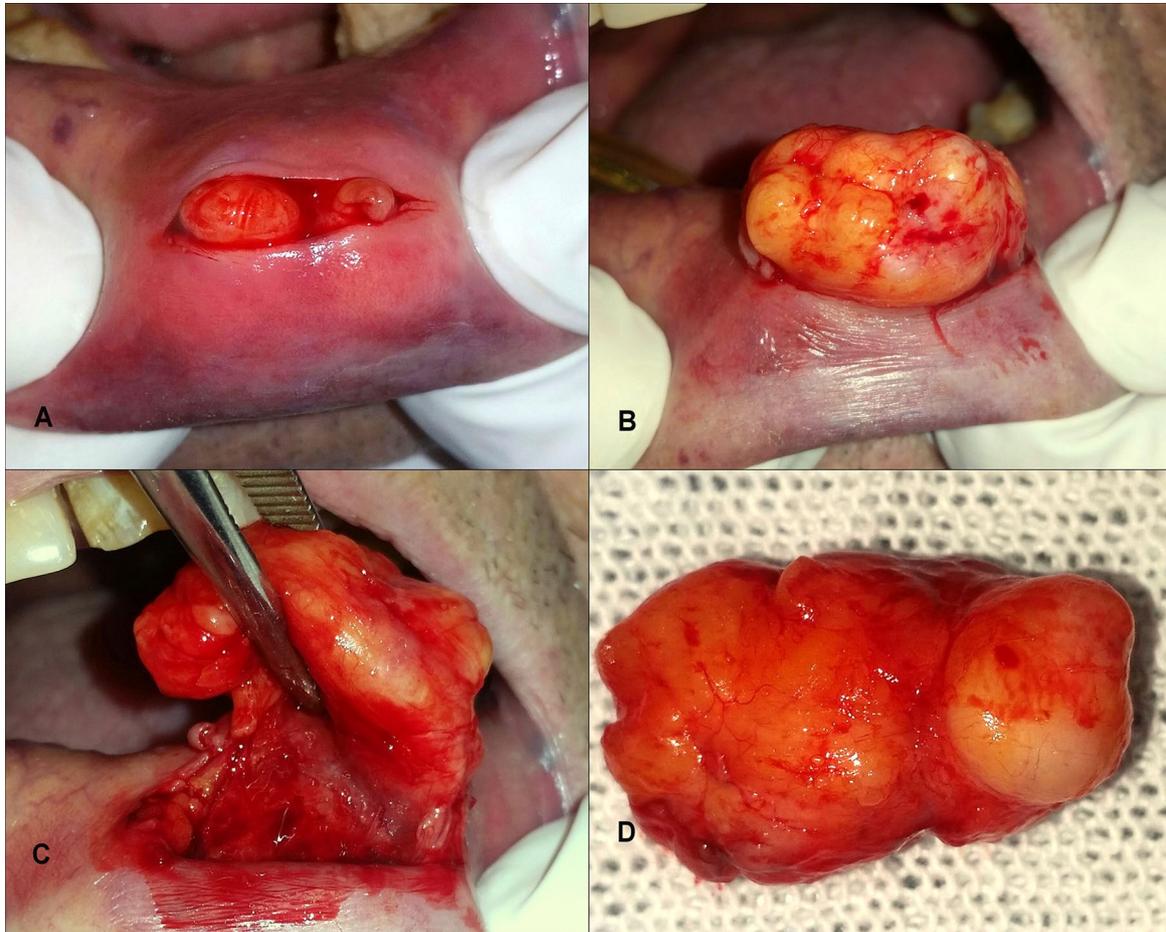


Figure 2. Surgical removal. A) Linear incision on the labial mucosa. It could be observed a yellowish mass. B) An irregular mass with multiple yellowish lobes and blood vessels could be observed over the mucosal surface. On the right side, a whitish bony area is visible, probably associated with calcified areas. C) Complete surgical excision. Muscular plane could be easily separated from the tumor mass. D) Obtained surgical specimen.

DISCUSSION

Lipomas are soft tissue tumours of scarce incidence in oral cavity, being 1% of all benign neoplasms⁶. While pathogenesis of OL was not clearly understood, some mechanisms such as chronic irritation, trauma, muscular metaplasia and fatty degeneration have been previously proposed by different authors^{5,7-10}. Gokul et al, described an extremely rare case of OL in a 6-years-old boy which was located in palate¹¹. In this case, authors proposed that the presence of centers of calcified tissue in the adipose mass could be due to a common mesenchymal origin of bone, cartilage and adipocyte cells, which would be influenced by a common factor giving rise to a tumor called "Mesenchymoma". The presence of a necrotic process due to ischemia, followed by an exuberant granulation tissue, would lead to a dystrophic calcification. The proximity of lipomas like lesions to periosteal tissues, tendons or joint capsules would increase the possibility of these metaplastic changes^{11,12}.

In our case, labial involvement is an anatomical region free of periosteal, tendinous or articular structures. The hypothesis of OL pathogenesis described above could not be applied in our case, however, during the interrogation the patient highlighted the chronic mechanical irritation generated by anterior tooth on the tumor surface. This repeated and persistent mechanical irritation could have contributed as an external stimulus to the progression of the mass and the formation of calcifying foci. Omonte et al described that OLs can be found intraosseous or adjacent to bone. Those lipomas that exhibit bone formation but whose location is independent of bone tissue (called OL not adhered) such as the one presented here, have been reported very rarely¹³.

Raviraj et al, presented in their study 17 cases of OL published between 1961 and 2015^{7,13}. 10 of the 17 cases presented were OL not adhered. However, only one case involved lower labial mucosa. The last case description coincides with some aspects of our case,

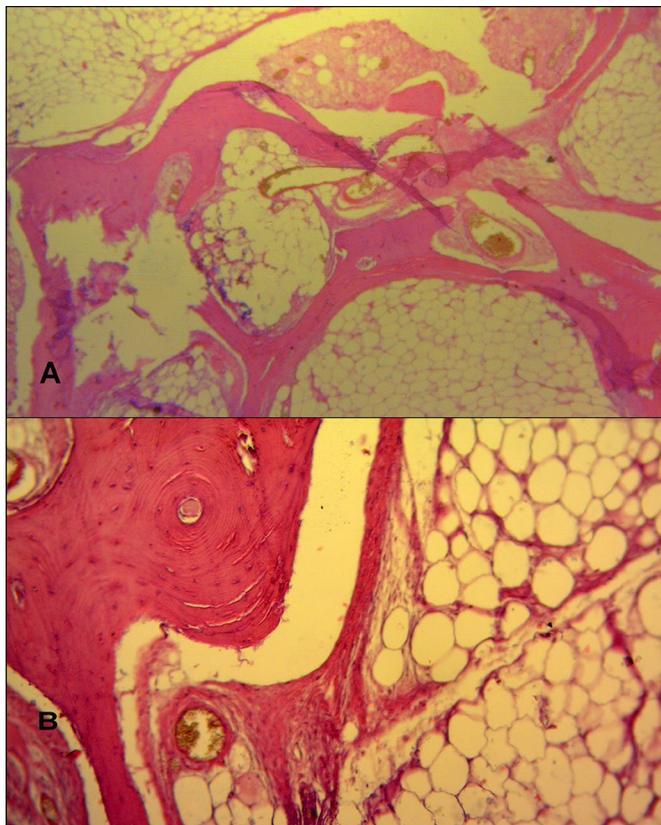


Figure 3. Histopathology A) Mature adipose tissue is observed in the connective, with vascularized areas and an acidophilic region corresponding to bone tissue (4X). B) Presence of grouped adipocytes without particularities, bands of collagen tissue and trabecular areas of mature bone with osteocytes without atypia (10X).

but with an ostensibly shorter evolution time¹⁴. Most of OLs reported were of long evolution, which would indicate the presence of an asymptomatic growth process. The literature review of oral OL cases reported so far, highlights this condition as an uncommon finding. The long evolution of OL reported in the literature as well as in the present work, would be a data of clinical relevance to suspect possible metaplastic and / or calcifying phenomena.

In order to reach a provisional diagnosis or ruling out other kinds of oral tumors with calcified areas, radiographic images should be taken. In some cases of OL, radiopaque images with an irregular pattern of trabeculae were described, without evidence of abnormality. The radiographic examination of these lesions also helps to differentiate them from entities that could present a similar radiograph pattern such as soft tissue osteoma, cartilaginous coristoma, condrolipoma or pleomorphic adenoma with ossification. The presence of bone marrow foci are signs of soft tissue osteomas. This finding would not support the diagnostic hypothesis of OL¹³. In

our case, although the hardness of the labial mass was evident, the presence of calcified tissue was never suspected since the labial mucosa is a region devoid of bone, periosteum and/or cartilaginous tissue. The presence of this bony areas could be associated with a metaplastic evolution of the lesion or a dystrophic calcification. The differential diagnosis that was considered before surgical removal was Pleomorphic Adenoma because its frequent presentation on a region where physiologically, minor salivary glands are present¹⁵. In our case, no radiographic images were obtained. However, attempts were made to obtain X-rays of the paraffined-block, but it could not be reached any clear images.

CONCLUSION

Clinical presentation of osseous or cartilaginous masses within oral lipomas are not common nor easy diagnosis in Oral Medicine. Although OL is a rare entity, it should be considered as an alternative diagnosis in cases of firm tumoral lesions of large evolution. The prognosis is favourable, with a satisfactory postoperative response without tendency to recur.

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