

Osteolipoma of the buccal mucosa

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Abstract

Lipomas are benign mesenchymal neoplasms of soft tissue that can be found in any part of the human body. Conversely, their presence in the oral mucosa is rather uncommon, with approximately 4% of the cases occurring in the oral cavity. In such cases, they are likely to have originated from mature adipose tissue and to be among several described histological variants of lipomas, which are identified according to the predominant type of tissue. There is a rare lipoma, known as an osteolipoma or an ossifying lipoma; however, little has been written this type of lipoma characterized by a classical lipoma with areas of osseous metaplasia. Considering the few cases of oral osteolipomas previously described in the English-related literature and the consequent risk of misdiagnosis and overtreatment, this paper describes an extreme case of an osteolipoma affecting the buccal mucosa of an adult patient. This paper focuses particularly on the pathogenesis of this lesion and the discussion of a correct diagnosis.

Key words: Lipoma, lipomatous tumors, osteolipoma, metaplasia.

Introduction

Although not common in the mouth, lipoma is the most common benign mesenchymal neoplasia of soft tissues in adults. Around 20% of the cases involve the head and the neck region and only 1 to 4% occurs in the oral cavity. Oral lipomas mostly occur in the buccal mucosa, floor of the mouth, tongue and lips (1-7).

Many benign histological variants of lipoma are known and described based on the type of tissue present and predominant in the lesion: fibrolipoma, angioliipoma,

myoliipoma, myxoliipoma, spindle cell lipoma, osteoliipoma and chondrolipoma (1-4, 8). Lipomas with osseous or cartilaginous metaplasia are rare histological variants. Osteoliipomas are less common than chondrolipomas and normally are presented in large and long-term evolution lesions (1,4,5,7,9).

The occurrence of osteoliipomas is seen in many anatomic sites, including the scapula, vertebral spine, neck, skull, suprasellar region, and tuber cinereum. In the oral region there are few cases described and in a re-

cent review of the available English-language literature, Saghafi et al. (7) revealed only 6 cases of osteolipoma arising in the oral cavity and 4 pharyngeal cases also showing some oral manifestations. Considering the small number of cases of oral osteolipomas previously reported in the English-related literature and the consequent lack of information regarding this rare occurrence, this paper aims to describe an additional case of osteolipoma that affected the buccal mucosa. In addition, the authors have revised the existing pertinent literature concerning osteolipoma.

Case Report

A leukoderma female patient aged 47 years old, revealed a facial asymmetry with a mobile nodule on the left cheek region, with no color variations on the surface, during an external oral examination (Fig. 1A). During the intraoral examination, a mobile yellowish and well circumscribed submucous nodule of approximately 1.5 cm in diameter was seen in the left buccal mucosa; the nodule was painless upon palpation and had been present for one year (Fig. 1B). A differential diagnosis of

lipoma and benign minor salivary gland neoplasia was proposed. After the excisional biopsy, a soft red mass with yellowish areas was easily enucleated (Fig. 2A). A radiograph of the surgical specimen revealed the presence of an irregular and radiopaque structure (Fig. 2B). The microscopic analyses revealed thin septa of conjunctive tissue separating lobules of adipose tissue formed by mature and uniform adipose cells. Trabeculae of vital laminated bone were observed all over the lesion, especially along the fibrous septa. No nuclear atypia, cell pleomorphism or mitosis were noticed (Fig. 3). Follow-up visits to the dental clinic were performed and the patient did not present post-surgical complications or recurrent lesions.

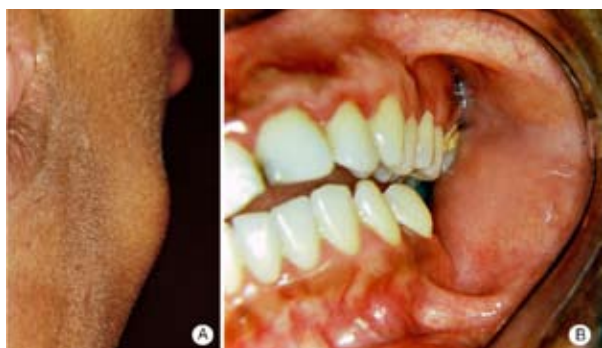


Fig. 1. A. Extrabuccal nodule in the left cheek region. B. Yellowish sub mucous nodule in the left buccal mucosa region.

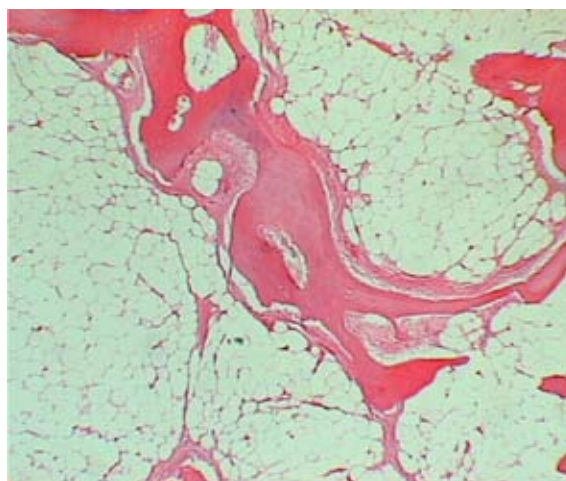


Fig. 3. Mature fat tissue and area of lamellar bone surrounded by fibrous capsule (H.E. 40X).

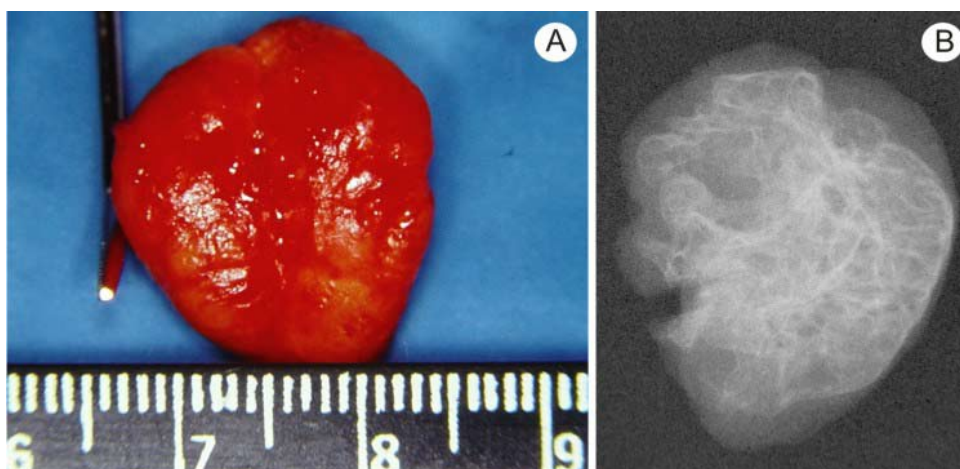


Fig. 2. A. Surgical specimen. B. Radiographic features of surgical specimen.

Discussion

Lipomas are tumors formed by mature adipose cells. Besides being mature adipocytes they can occasionally be composed of endothelial, muscular, osseous and chondroid tissues (1,3,4). Osteolipoma is a rare variant described for the first time in 1958 by Plaut and his partners.

Castilho et al. (4) and Obermann et al. (9) showed a greater incidence of lipomas in adult patients, with age ranging from 40 to 50 years old. In most cases mentioned the patients reported that there had been a long period of time between the appearance and the diagnosis of the tumor. In the present case, the patient was 47 years old and reported that the lesion had been present in the buccal mucosa for approximately one year, which is in accordance with the age group and the estimated evolution time found in the literature.

Clinically, lipomas tend to be presented as extensive well circumscribed masses with a wide implantation base or a pedicle. Depending on the depth of the lesions they can have a yellowish or pink color like the adjacent mucous membrane (5). Microscopically speaking, lipomas are formed by a mature adipose tissue without cellular atypias (1-4, 6). The histological variant osteolipoma presents osseous metaplasia and mature adipose tissue intermixed with conjunctive tissue. In the present case, the patient presented a yellowish well circumscribed submucous nodule in the buccal mucosa. The microscopic analyses revealed osseous trabeculae inside a mature adipose tissue surrounded by thin septa of conjunctive tissue. These characteristics are also similar to the ones described in the literature Piattelli et al. (1) and Castilho et al. (4).

Furlong et al. (3), when analyzing a group of 125 lipomas in the oral and maxillofacial region, did not find any histological variant that presented osseous metaplasia. The same was verified by Fregnani et al. (2), who studied 46 cases of oral lipoma. When revising the literature, we found only six cases affecting exclusively the oral cavity (1,4, 10-12) being this presented case the second one observed in the buccal mucosa.

The pathogenesis of the osteolipomas remains uncertain and some authors have suggested that many multipotent cells, or cells from different lineage, differentiate into lipoblasts, chondroblasts or osteoblasts, and fibroblasts, which could characterize a "mesenchymoma". This pathology is defined as a rare soft tissue lesion composed of fibrous tissue associated with two or more types of mesenchymal cells well differentiated, that would not normally be found in the same site. Another theory in-

volving the pathogenesis of this lesion would be that only the adipocytes would be able to transform into neoplastic tissue and the cartilage or bone would be the result of metaplasia of the fibroblasts in chondroblasts or osteoblasts (1,4,9,13,14). The microscopic characteristics in the present case, especially the presence of osseous trabeculae accompanying the fibrous septa through the adipose neoplasia reinforces the second hypothesis proposed.

The prognosis of lipomas with ossification is favorable, as well as the one expected in conventional lipomas. Usually the chosen appropriate treatment is surgical excision and recurrences have not been reported yet (2-5, 14).

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