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ORIGINAL ARTICLE

Osteolipoma in posterior maxilla: A case report

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Abstract:

Osteolipoma is a histologic variant of lipoma characterized by bone formation, which rarely occurs in the oral cavity. This condition usually is easily recognized by microscopic examination and it has a good prognosis. However, this lesion may occur in different sites of the oral cavity and may present different clinical aspects, being a diagnostic challenge at the time of clinical examination. The aims of this article are to report a case of osteolipoma located in the buccal aspect of the posterior maxilla and discuss the main clinical and histological findings of this rare oral lesion. A 46-year-old woman presented with a painless mass of about 2 cm in the vestibular portion of the posterior maxilla. The lesion had a hard consistency and color similar to adjacent mucosa. Imaging findings revealed a well-defined and circumscribed hypodense lesion with hyperdense areas. The histopathological examination showed mature adipose tissue among trabeculae of vital lamellar bone, which was consistent with the diagnosis of osteolipoma. No signs of recurrence were observed after 3 years of follow-up.

Keywords: Lipoma; Neoplasms, Adipose Tissue; Mouth; Maxilla.

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INTRODUCTION

Lipomas are common benign soft tissue neoplasms composed of mature adipose tissue, which may affect oral and maxillofacial region[1]. This neoplasm may present histological variants based on the presence of other lesional tissue, besides adipose one. Examples of these variants are: fibrolipoma, spindle cell lipoma, chondrolipoma, osteolipoma, sialolipoma, angiolipoma and myxoid lipoma[2].

Osteolipoma is a rare histologic variant of lipoma caracterized by bone formation[1]. Only 26 cases of oral cavity lipoma with osseous change were described in the English language literature[3-12] since 1961. The oral sites affected were tongue, floor of the mouth, hard palate, buccal mucosa, buccal sulcus, mandibular buccal mucosa, mandibular buccal alveolar mucosa, mandibular buccal vestibule, lower labial vestibule (3), mandibular lingual alveolar mucosa[8], retromolar trigone[4], lower lip[9,11] and mandible extending to labial sulcus[6,12]. Here, we report a case of osteolipoma located in maxillary buccal vestibule and discuss important features of this rare oral lesion.

CASE REPORT

A 46-year-old woman with slight facial asymmetry (Fig. 1A) presented with a painless mass located in the vestibular portion of the posterior right maxilla. The lesion exhibited hard consistency, and it was covered by undamaged mucosa of similar color to adjacent tissue (Fig. 1B). According to the patient, the lesion had appeared during childhood and it has slowly grown up to its present size, which was about 2 cm in diameter. The patient has a totally edentulous upper jaw and wears full dentures. After clinical examination, the diagnostic hypothesis was a benign fibro-osseous lesion.

Computed tomography scan was performed and a well-defined hypodense lesion with hyperdense areas was identified in the vestibular aspect of the posterior region of right maxilla (Fig. 1C). An incisional biopsy disclosed the presence of compact lamellar bone that surrounded mature adipose tissue, rendering a diagnosis of an intra-osseous lipoma. During the excisional biopsy, it was possible to observe that the lesion was attached to the maxillary bone by a wide base (Fig. 1D).

The microscopic examination revealed mature adipose tissue interspersed within mature trabeculae of vital lamellar bone with different sizes and shapes. Osteoblasts were observed around some trabeculae and the lesion was completely surrounded by a layer of compact lamellar bone. Congested blood vessels and areas of hemorrhage were also observed in the specimen (Fig. 2). Based on these findings, the diagnosis was osteolipoma. The patient has been followed up and no signs of recurrence were observed after 3 years (Fig. 3).

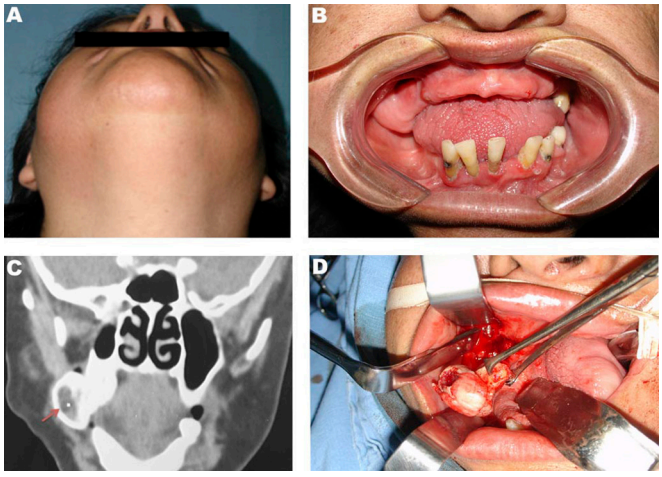

Figure 1. Photograph of the patient showing a slight facial asymmetry of the right side caused by the lesion (A), which was located in the vestibular portion of the posterior right maxilla (B). The axial CT image demonstrated a well-defined hypodense lesion with hyperdense areas (arrow) (C). The excisional biopsy showed that the lesion was attached to the maxillary bone by a wide base (D).

DISCUSSION

Lipoma is the most common benign neoplasm in adults. It is more common in obese individuals, although its pathogenesis is unknown[1]. In the oral cavity, the most affected place is the buccal mucosa[2,13]. Lipomas may appear within the subcutaneous/submucous tissues (superficial lipoma); within the deep soft tissues (deep lipoma), such as in the muscle (intramuscular lipoma)[1], associated with minor and major salivary glands (sialolipoma)[14]; on bone surfaces (parosteal lipoma)[1] and within the bone (intraosseous lipoma)[1].

Besides the location, lipomas may also present histological variants based on lesional tissue, beyond adipose one. The presence of significant fibrous, myxoid and cartilaginous tissues in the lipomas, and even the presence of spindle cells and many blood vessels have been reported as fibrolipoma, myxoid lipoma, chondrolipoma, spindle cell lipoma and angiolipoma, respectively[2].

To describe the osseous change variant, different names such as osteolipoma[15], lipoma with osseous metaplasia[16] or ossifying lipoma[17] have been used. Some theories have emerged in the literature to explain the osseous changes affecting lipoma. Some authors have suggested that osteolipoma is a kind of "mesenchymoma"

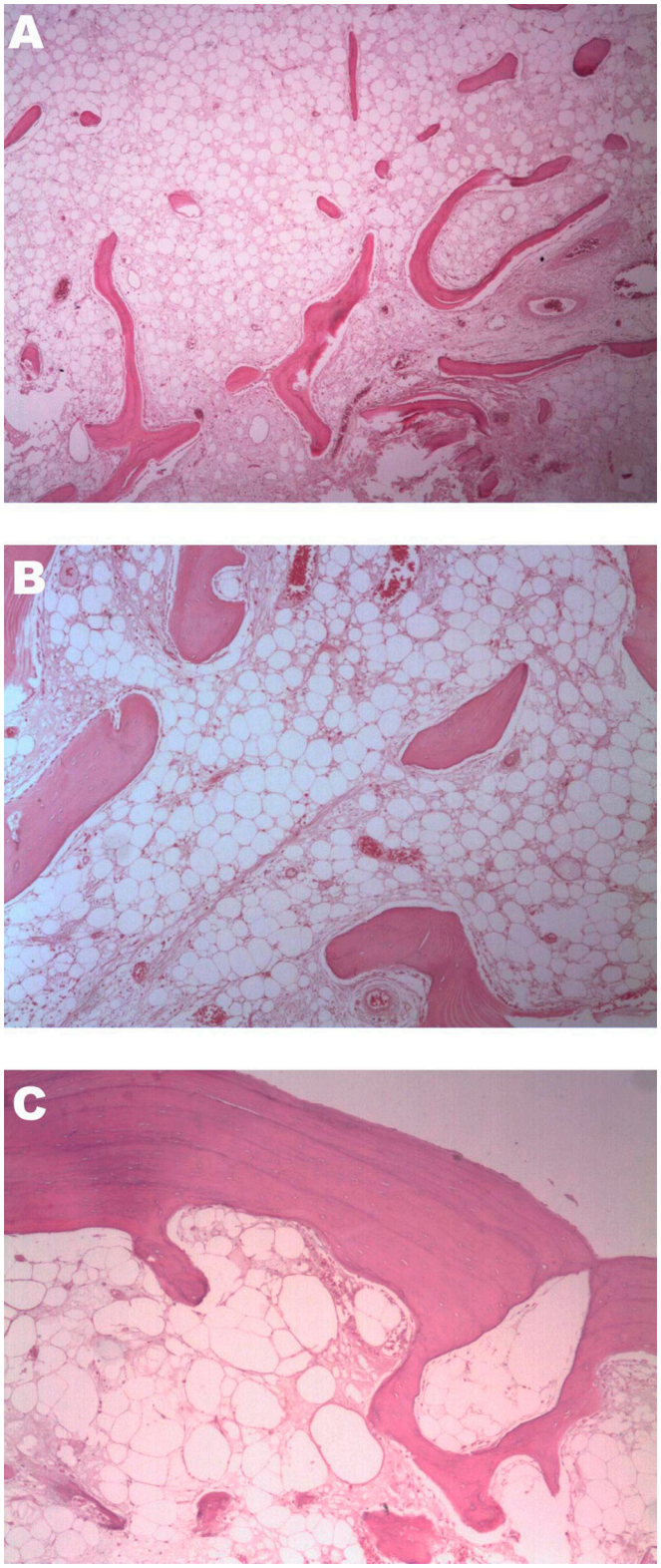

Figure 2. Photomicrograph of the lesion showing mature adipose tissue containing trabeculae of mature lamellar bone with different sizes and shapes (A and B). The lesion was delimited by bone and attached to it (C) (Hematoxylin and eosin, original magnification 200× (A) and 400× (B and C)).

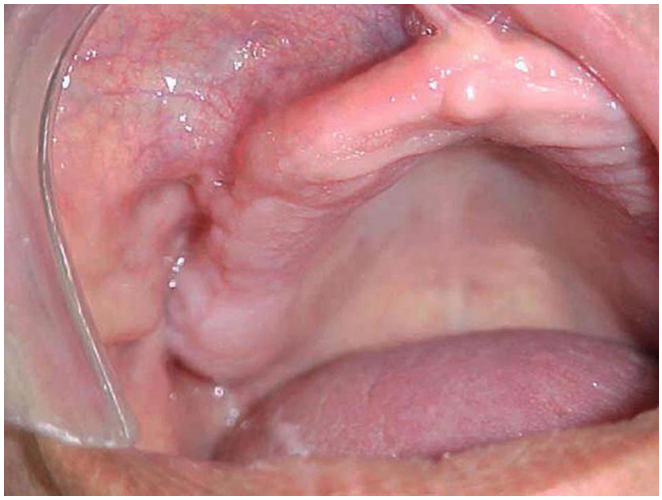

Figure 3. Postoperative photograph showing no signs of recurrence after 3 years of follow up.

and both adipose and osseous tissue of osteolipoma have originated from two types of undifferentiated mesenchymal cells[18]. Makiguchi et al. suggested that the bone components of osteolipoma could originate from multipotent adipose-derived stem cells, in response to growth signals[19]. On the other hand, the bone within lipoma could be originated from metaplastic transformations due to mechanical stress, the contact with periosteum and even still unknown reasons[20].

Few cases of osteolipoma have been described in head and neck, including cases in parotid region[21], submandibular area[22], parapharyngeal space[23], nasopharynx[24], mandible[17] and coronoid process[25]. Osteolipomas are rare in the oral cavity in which only 26 cases have been described in the English language literature from 1961 until June 2017[3-12]. When the osteolipoma appears close to the bone, it may be attached to it (parosteal), as in the present case, or a subtype non-attached to the bone, which is completely independent of it[26].

In the oral cavity, osteolipomas occur more frequently in adults between 31 and 70 years of age and there is no predilection of gender[4]. Only one case involving a child (congenital) was reported[27]. In our case, although the patient has been diagnosed with 46-year-old, she reported that the lesion had appeared in the childhood (more than 30 years of evolution). The most affected site is the buccal mucosa[4], the same of classic lipomas (2, 13). Some cases have been reported in the vestibule, as the present case, although they have occurred in the mandible[3,4].

Clinically, ostelipoma presents as a painless mass or nodule with hard or soft consistence. A normal or a

yellowish color may be observed in the mucosa covering the lesion. With concern to the size, osteolipomas ranging from 0.8 cm to 7 cm in their largest diameter have been reported. In some cases, there was facial asymmetry associated with the lesion[3]. Different imaging methods have been used as an adjunct to the clinical examination such as radiography, computed tomography and ultrasonography. In general, a radiopaque/hyperdense mass or a radiolucent/hypodense mass with areas of calcification have been observed[3,15].

The differential diagnoses depend on the location, clinical and radiographic features of each lesion. As mentioned before, different sizes of osteolipoma in the oral cavity have been described, besides the fact that this lesion may be hard or soft, probably due the amount of calcification. Osteoma cutis and osteocartilaginous choristoma were described as possible differential diagnoses[28]. Although the clinical hypothesis of fibro-osseous lesion, in the present case, the diagnosis possibilities also include osseous choristoma or osteoma due to hard consistency.

Osseous choristoma is a tumor-like growth of normal bone tissue occurring in the soft tissue (soft tissue osteoma). This kind of choristoma is more common in the tongue, however a case has been reported in mandibular buccal vestibule[29]. Osteolipoma also was a hypothesis, once this lesion is partially calcified, which could justify the hard aspect. Malignant neoplasms were not included in the differential diagnosis because the lesion was covered by undamaged mucosa and radiographically it was well defined, with no apparent infiltrative behavior. Moreover, the duration of the lesion, present since childhood, was more consistent with the slow growth of a benign tumor.

Although osteolipoma may present many clinical differential diagnoses, after microscopic examination the diagnosis is established without difficulty. It usually presents mature adipose tissue, with no atypia, separated by fibrous connective tissue septa and trabeculae of bone which may be immature, mature, or both mature and immature[15].

Some osseous choristomas present a histologic organization that resembles osteolipoma, once spongy bone trabeculae with abundant bone marrow spaces filled with adipose tissue may be observed. However, osseous choristomas also present hemopoietic marrow[30] which is not found in osteolipoma[15]. In the present case, the histogical findings was entirely compatible with osteolipoma, since no foci of hematopoietic cells were observed.

Osteolipomas must be treated by conservative surgical excision. The prognosis is good[15], as well as the prognosis observed in classic lipomas[13]. None recurrence of osteolipoma in the oral cavity has been reported yet[15]. Although the recurrence is not expected, the follow up is important especially if the lesion is attached to bone, as in the present case, due the difficulty in its excision, which may lead to its incomplete removal[26].

In summary, osteolipoma is a rare lesion that is easily diagnosed after histological evaluation. Clinically, this lesion may occur in different sites of the oral cavity as a hard or soft mass that affects mainly adults, with no gender predilection. Due to a wide range of clinical possibilities, osteolipoma should be included in the differential diagnosis of bone-containing benign masses affecting the oral cavity.

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