CASE REPORT

A Rare Case of Osteolipoma Presenting as a Mobile Swelling on the Buccal Sulcus

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Abstract:

Osteolipoma is an extremely rare histologic variant of lipoma that contains mature lamellar bone within the tumor and is independent of bone tissue. It is most commonly seen in buccal mucosa, floor of the mouth, tongue, palate, and parapharyngeal spaces of the oropharyngeal region. We present a case of 72 yr old female patient with a complaint of intraoral swelling in left lower buccal sulcus. The swelling was mobile and firm to hard in consistency. Excision specimen showed circumscribed mass of mature adipose tissue with metaplastic lamellated bone formation. The lesion was diagnosed as osteolipoma. The pathogenesis and the differential diagnosis are discussed with this case presentation.

Keywords: Lipoma, Osteolipoma, Buccal sulcus.

Introduction

Osteolipoma is a very rare histological variant of lipoma that exhibits bone formation (less than 1%)[1]. They are lipomas which exhibit bone formation but are independent of bone tissue. Osteolipoma are classified as intraosseous when located in bone and parosteal or periosteal when located adjacent to bone[2]. Due its rarity, knowledge about this tumor is incomplete. We describe a case of osteolipoma affecting the buccal mucosa.

Case report

A 72 year old female patient presented with a painless freely mobile mass in the left buccal sulcus of duration 6

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month (figure 1). The patient had no history of trauma in that region. No enlarged lymph nodes were detected in the clinical examination. The medical history and systemic examination were non contributory. The intraoral examination showed a 1.5 x 2 cm, well defined, movable mass in lower left buccal mucosa. The swelling was firm to hard in consistency with no rise in local temperature.

Fig1 - Black arrow showing intraoral swelling in the left buccal mucosa

Fig 2 - Surgical excision of the lesion

Fig 3 - Gross specimen showing an ovoid mass of yellow soft tissue

Fig 4 - Bisected gross specimen showing peripheral fatty tissue and central areas of dark brown osseous material

Under local anesthesia, the mass was excised from the buccal sulcus (figure 2) and sent for histopathological examination. There was no attachment of the lesion to the adjacent bone. The laboratory received a hard specimen covered with soft tissue of yellow colour measuring 1.5 × 2 cm. The gross specimen showed peripheral yellow soft tissue with central osseous tissue (figure 3 & 4). Microscopically, the mass presented with abundant mature adipose tissue, with no cellular atypia. The section showed spicules of mature lamellated bone within the lesion (figure 5 & 6). No evidence of hematopoietic bone marrow was seen in the section. Based on these histopathological findings the lesion was diagnosed as osteolipoma.

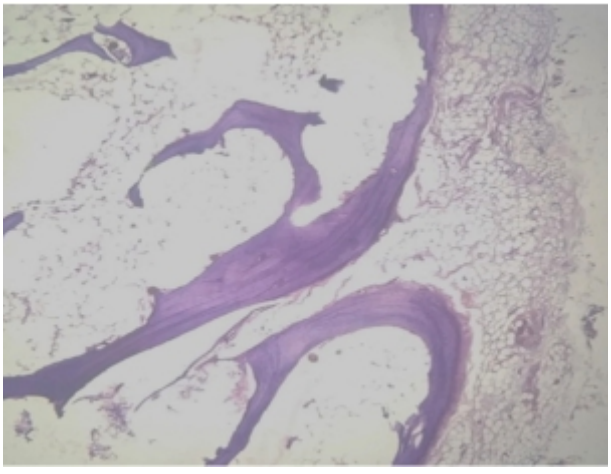

Fig 5. Scanner view (4X) round to oval varying size of mature adipocytes with spicules of mature lamellated bone

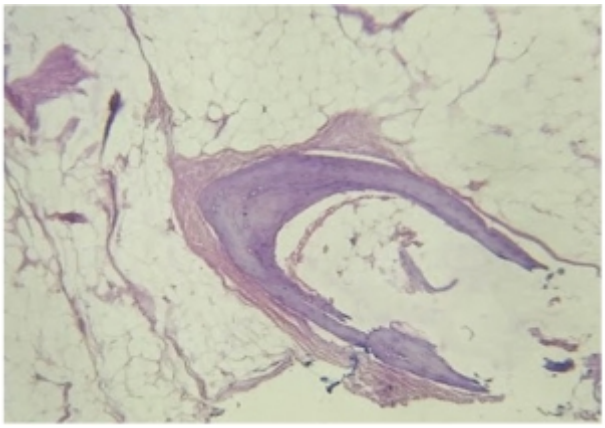

Fig 6. Low power (10X) showing mature adipocytes with spicules of mature lamellated bone covered with fibrous periosteum

Discussion

Lipomas, affecting any part of the human body, are the most common benign mesenchymal tumors of soft tissue[3]. According to previous studies, the head and neck region has been involved in 20% of the cases, but only 1-4% of the cases have been observed in the oral cavity[2]. The common sites affected are buccal mucosa, floor of the mouth, and lips[2]. According to literature till now 20 cases of osteolipomas have been reported[4].

Lipomas which have mature osseous elements are called osteolipoma. Other terms like ossifying lipoma, osseous lipoma and lipoma with osseous metaplasia are also used for osteolipoma[5].

Although patients with a wide range of ages (6-81 years) are affected by osteolipoma in the oral cavity, it usually presents in middle-aged or elderly patients with a history of slow progression[4]. Osteolipoma in the oral cavity affects both sexes, with a slight predilection in females. Osteolipomas clinically present as painless, mobile, and hard masses with yellowish colour[4]. In our case also the lesion had similar presentation.

Histopathologically, three differential diagnosis can be considered for this case which include

(1) osteocartilaginous choristomas

(2) chondrolipomas and

(3) Pleomorphic adenoma with ossification[4].

Osteocartilaginous choristomas (soft tissue osteoma) which are the most common choristomas of the oral cavity[4]. Osteocartilaginous choristomas histologically show a mass of dense lamellar bone that has the haversian canal system and they demonstrate hematopoietic marrow[4]. But in osteolipoma the osseous tissue does not show hematopoietic marrow as in our case.

Another histopathological differential diagnosis of osteolipoma is chondrolipomas. Chondrolipomas are rare benign variants of lipoma characterized by slow growth. The microscopic examination shows proliferation of mature adipocytes associated with variable amounts of mature cartilaginous tissue[4]. But in our case there were no chondroid tissue formation.

Third histopathological differential diagnosis is pleomorphic adenoma with osseous metaplasia. Pleomorphic adenoma is a salivary gland tumor of complex morphology possessing epithelial ductal and myoepithelial elements with osseous and chondroid metaplasia[4]. But in our case only mature adipocytes tissue was seen with no ductal elements, so this entity was also ruled out.

Pathogenesis of osteolipoma is still not clear, three main theories have been suggested in the literature[5]:

(1) Response to trauma, metabolic changes, or ischemia, neoplastic change which occur in the fat, later metaplasia of preexisting fibrous elements in the lipomas to osteoblasts that results in a secondary ossification[6].

(2) Develop due to the proliferation and differentiation of mesenchymal stem cells[7].

(3) Develop due to transformation of fibroblasts into osteoblasts owing to osteoinductive factors released by blood-borne monocytes that enter into the fatty tissue[8].

Treatment of osteolipoma is complete surgical excision. Osteolipoma has been reported to have similar prognosis as other lipomas with low recurrence[9]. In our case after the surgical excision of the lesion there was no recurrence after six months of follow-up.

Osteolipoma is a rare benign lesion in the oral cavity and till now only 20 cases have been reported in literature[1]. According to the report this lesion can present as a hard swelling in the oral cavity with a long duration[1]. So clinician and pathologist should be aware of this entity and consider them in the differential diagnosis for intraoral swellings.

Conflict of interest: None

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