

Osteolipoma of the palate: Report of a case and review of the literature

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Abstract

Oral lipomas, though rare, predominantly affect the buccal mucosa, tongue, and lips. The occurrence of lipomas in the palate is extremely rare. Osteolipoma is a very rare histological variant of lipoma accounting for less than 1% of all cases. Although a few cases involving the soft palate have been reported, there is only one reported case of osteolipoma of the hard palate in the English-language literature and it was a congenital osteolipoma associated with a cleft palate in a 6-year-old male child. This paper aims to describe an additional case uniquely located in the hard palate of an adult female.

Key words: Lipoma, osteolipoma, oral cavity, palate

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Introduction

Lipomas are benign mesenchymal neoplasms of soft tissue that can be found in any part of the human body. Around 20% of the cases involve the head and neck region and only 1 to 4% occurs in the oral cavity.^[1] Benign histological variants of lipoma have been named according to the type of tissue present and they include fibrolipoma, angioliipoma, myoliipoma, leiomyoliipoma, myxoliipoma, spindle cell lipoma, osteoliipoma, chondrolipoma, and sialoliipoma.^[2,3] Osteoliipoma which is a lipoma with osseous metaplasia is a very rare histological variant accounting for less than 1%^[4] and is seen in many anatomic sites, including the scapula, vertebral spine, neck, skull, suprasellar region, and tuber cinereum.^[1] A review of the available English-language literature by Saghafi *et al.*^[5] revealed only eight cases of osteoliipoma arising in the oral cavity and two pharyngeal cases also showing some oral manifestations.

Oral lipomas with osseous/chondroid differentiation are mostly situated in the tongue and lower lip.^[6] Considering the rarity of cases of oral osteoliipomas, this paper aims to describe an additional case uniquely located in the hard palate of an adult female. Although a few cases involving the soft palate

have been reported,^[7,8] this is the second reported case of a solitary osteoliipoma involving the hard palate. In addition, the authors have revised the existing pertinent literature.

Case Report

A 37-year-old female Nigerian presented with a complaint of a slow-growing and painless palatal swelling of 10-year duration. Both the medical history and systemic review were noncontributory. Extraoral examination was not significant, while intraorally there was an oval, firm to hard palatal swelling measuring about 3 × 4 cm in relation to the upper left second premolar and first and second molars [Figure 1]. The swelling was not tender and the overlying palatal mucosa was normal with no evidence of ulceration or erythema. Occlusal radiograph showed patchy areas of radiopacity [Figure 2]. Based on clinical and radiographic findings, a provisional diagnosis of cementifying fibroma was made. The differentials included osteoma, neurofibroma, and intraosseous palatal cyst. The lesion was surgically excised under local anesthesia after raising a palatal

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Figure 1: Clinical photograph showing the osteolipoma mass on the left half of the hard palate

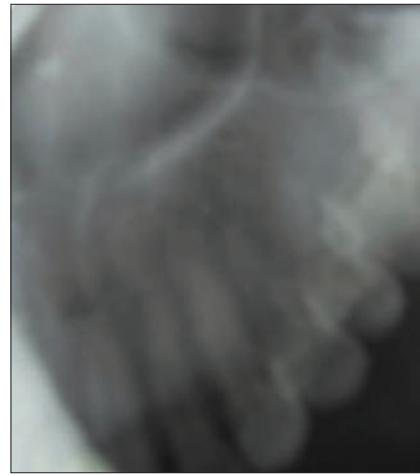


Figure 2: Radiograph of the lesion showing patchy areas of radiopacity



Figure 3: A palatal mucoperiosteal flap exhibiting the lesion

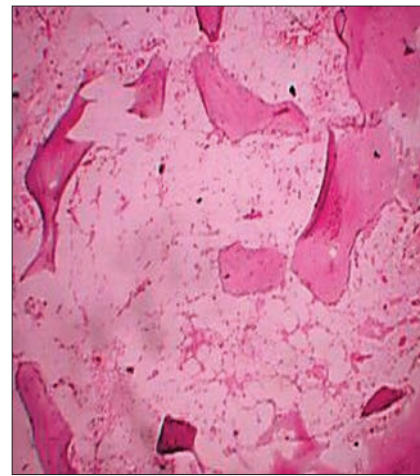


Figure 4: Photomicrograph of the lesion showing trabeculae of vital lamellated bone scattered among benign looking adipocytes (H and E, × 40)

mucoperiosteal flap [Figure 3]. The flap was repositioned and closed primarily. Analgesic and prophylactic antibiotics were prescribed and postoperative healing was uneventful.

The gross specimen measuring $3.2 \times 3.0 \times 1.6$ cm was yellow-gray in color. The cut surface was also yellowish-gray in color and had a gritty consistency. The specimen was decalcified and processed whole.

Microscopic examination revealed mature adipose tissue composed of adipocytes which were uniform in size and shape. Scattered among them were trabeculae of vital lamellated bone of varying sizes and shapes. No nuclear atypia, cell pleomorphism, mitosis, or necrosis was noticed [Figure 4]. Based on the foregoing, a histological diagnosis of osteolipoma was made.

Discussion

Lipoma is a benign tumor of adipose tissue commonly found

in adults and mainly affecting the trunk, upper extremities, back of the neck, and rarely the oral cavity. They are the most common soft tissue mesenchymal neoplasms with 15 to 20% of cases involving the head and neck region and 1 to 4% affecting the oral cavity. Oral lipomas predominantly affect the buccal mucosa, tongue, and lips.^[9] Occurrence of lipomas in the palate is extremely rare. Various pathogenic mechanisms that have been proposed for the development of lipoma include origin from lipoblastic embryonic cell nests, metaplasia of muscle cells, and fatty degeneration.^[10]

An additional histological feature in our case was the presence of osseous metaplasia. Kuyamo *et al.*, reviewing 16 cases of rare lipoma/fibrolipoma with osseous/chondroid differentiation, made an additional report of two cases, one of which was a fibrolipoma with consecutive osseous and chondroid differentiation. This is the first to be reported in the literature.^[6] There are sparse cases of osseous metaplasia in lipoma of the oral cavity reported, while those recorded were seen mainly in intraoral sites other than the palate.

In a recent review of the literature, de Castro *et al.*^[1] found only six cases of osteolipoma affecting exclusively the oral cavity. Although, ours will be the second case of a solitary osteolipoma of the hard palate to be reported, to the best of our knowledge, it is the first in an adult patient.

Clinically, depending on the depth of the lesion, lipomas can have a yellow or pink color like the adjacent mucous membrane. Microscopically, lipomas are composed of mature adipose tissue without cellular atypias.^[1,2,5] The histological variant osteolipoma presents osseous metaplasia and mature adipose tissue intermixed with conjunctive tissue. In the present case, the patient presented with a well-circumscribed submucous nodule overlaid by normal mucosa in the hard palate. The microscopic analyses revealed osseous trabeculae inside a mature adipose tissue.

The pathogenesis of osteolipoma is still not clear. Two main theories exist for the pathogenesis of osteolipomas.^[1,11] First, these tumors may originate directly from multipotent cells, or cells from different lineage which differentiate into lipoblasts, chondroblasts, or osteoblasts, and fibroblasts, characterizing a “mesenchymoma.”^[1,6] 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.^[1] Alternatively, it has been suggested to arise after repetitive trauma, metabolic changes, or possibly ischemia, leading to metaplasia of pre-existing fibrous elements within the lipoma and development into osteoblasts. Furthermore, huge lipomas showing fast enlargement may have cystic degeneration and necrosis. Consequently, necrotic tissue may ossify mimicking osteolipoma.^[11] Our histological findings and especially the presence of osseous trabeculae scattered among and intermixed with the adipocytes support the first hypothesis.

Reports in the literature show a greater incidence of lipomas in adult patients^[3] and there is usually a long interval between occurrence of the lesion and when the patient presents to the clinician. This is consistent with the present case, which occurred in a 37-year-old female who reported that the lesion had been present in the palate for a period of 10 years.

Results of MRI evaluation of 126 consecutive fatty masses by Gaskin and Helms^[12] showed that osteolipoma may mimic

well-differentiated liposarcomas, from which they are often hard to differentiate on imaging alone. Definitive diagnosis of the lesion can easily be done with histopathologic examination and treatment is by surgical excision. In addition, the identification of histological subtypes in already known variants of lipoma, such as low-fat and fat-free spindle cell lipomas, highlight the importance for careful microscopic evaluation of these tumors.^[3] Osteolipoma is considered to be benign, but close monitoring and long-term follow-up is advocated because of paucity of pertinent clinical information.

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