Case Report

Osteolipoma in the retromolar trigone: A case report and review of literature

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ABSTRACT

Osteolipoma is the distinct histological variant of lipoma. Until now, a total of 17 cases have been documented in English literature. It is most commonly seen in buccal mucosa, floor of the mouth, tongue, palate, and parapharyngeal spaces of the oropharyngeal region. The aim of the present communication is to report an unusual presentation of osteolipoma in the retromolar trigone. Conventional radiographs have revealed "whorled pattern" of radiographic appearance in our case that might be due to uniform distribution of bony trabeculae within tumor; such type of finding has not been reported till now. Hence, here, we add these new features to existing literature.

Keywords: Osteolipoma, retromolar trigone, whorled pattern

INTRODUCTION

Osteolipoma is more prevalent in the appendicular skeleton. Based on the position of the tumor, these are subcategorized into endosteal, parosteal, and soft tissue variety. It is a benign mesenchymal tumor which accounts for <1%–4% of all lipomas of the oral cavity.[1] On the review of literature, a total of 17 cases have been reported[1,2] until now and overview of the literature is given in Table 1.

In the present communication, we add two additional findings; (a) site of occurrence and (b) "whorled pattern" radiographic appearance have not been reported earlier. Even though osteolipoma is a benign one, in the diagnostic point of view, it is important for clinician because it mimics the soft tissue sarcomas.

CASE REPORT

A 55-year-old female patient presented to the Department of Oral and Maxillofacial Surgery with the history of a painless, nodular swelling in the right retromolar region 6 years. Her medical history was not contributory. She had undergone extraction of the right third molar 8 years back. No abnormal extraoral findings were detected.

Intraoral examination revealed smooth, well-defined, sessile nodule, color similar to oral mucosa which was observed in the retromolar trigone and edentulous area of the third molar, measuring 3 cm × 2 cm, firm to hard in consistency, mobile, nontender with lobulated surface [Figure 1]. The fine needle aspiration cytology test has not yielded anything material.

Ultrasonography demonstrated diffused hyperechoic masses [Figure 2]. A panoramic radiography (orthopantomography [OPG])

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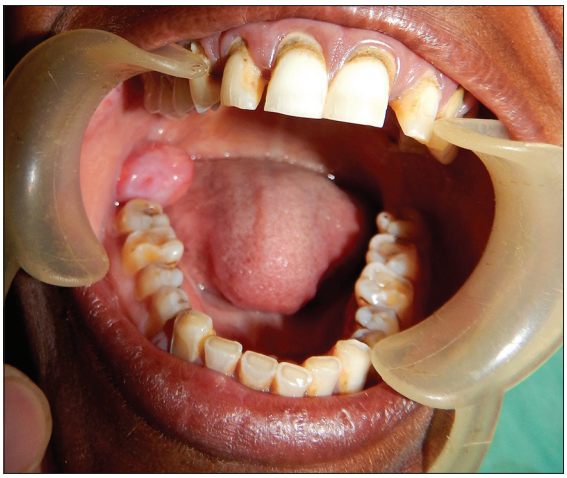

Figure 1: Intraoral photograph showing solitary nodule in retromolar trigone

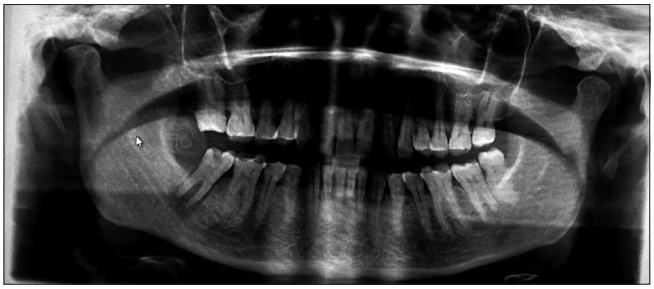

Figure 3: Orthopantomography showing multiple radiopaque masses which were arranged as concentric layers

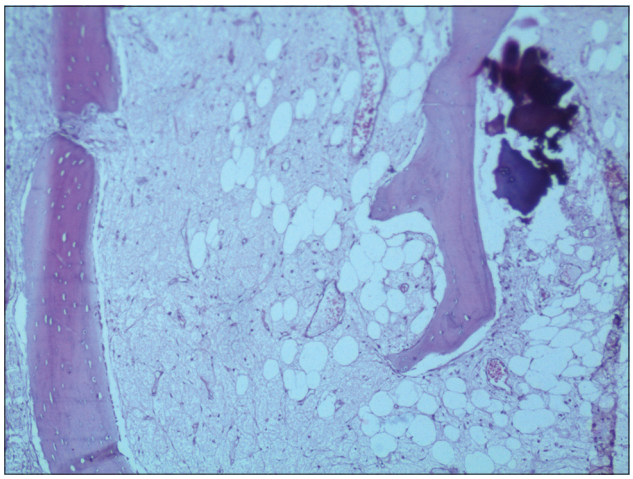

Figure 5: Histopathological sections showing mature adipocytes with intervening by woven bony trabeculae

[Figure 3] has shown multiple islands of bone, which were arranged in a whorled pattern. Based on clinical and radiological features, the provisional diagnosis of ossifying fibroma was given. An excisional biopsy was done under local anesthesia. A radiograph of the resected specimen revealed the presence of irregular and radiopaque structures which were arranged in a concentric manner like petals of flower [Figure 4]. Gross examination of the specimen showed that the tumor was well encapsulated, yellowish-white mass with lobulated surface that floated in formalin 10%.

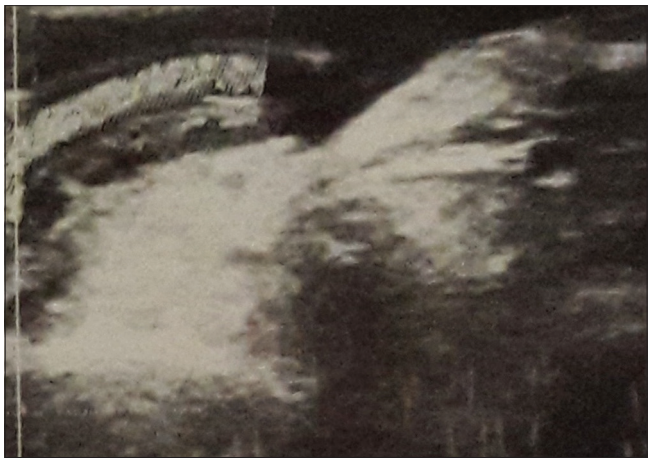

Figure 2: Ultrasonography revealed multiple hyperechoic masses

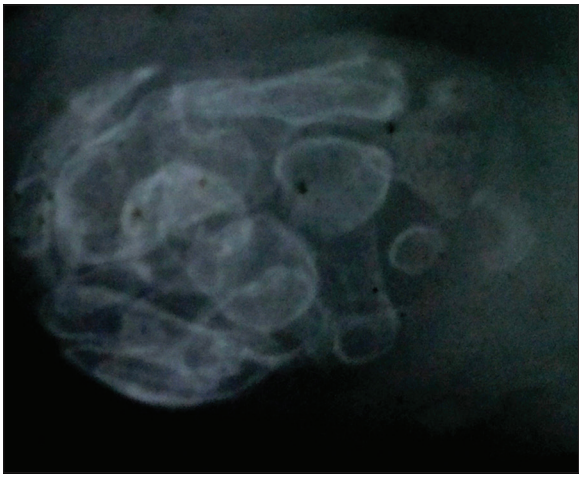

Figure 4: Radiographic features of excised specimen showing uniform distribution of radiopaque masses in a whorled pattern

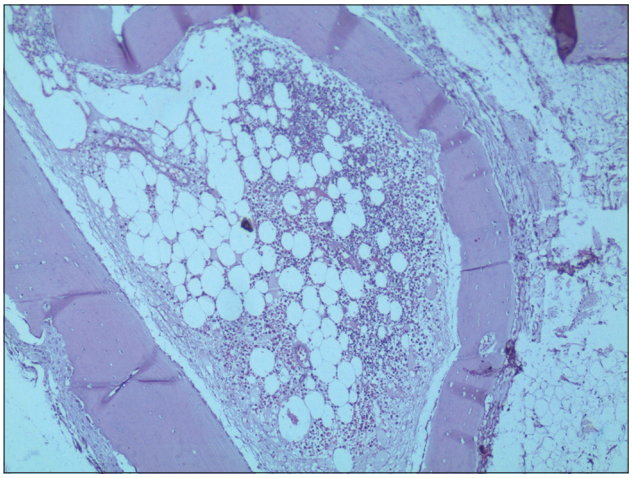

Figure 6: Histopathological picture showing infiltration of osteolipoma with mononuclear lymphocytes and small focal blood vessels

Microscopic examination showed thin trabeculae of bone with numerous adipocytes without any surface epithelium [Figure 5]. The arrangement of adipocytes into distinct lobules was observed. These lobules were separated with thin woven

Table 1: Overview of the results in percentage and ratios about review of literature of osteolipoma[2,3]

1. Demographic data	
Male to female ratio	1:1
Age range	
10-20 years	12.5%
21-30 years	6.25%
31-40 years	12.5%
41-50 years	12.5%
51-60 years	18.75%
61-70 years	18.75%
71-80 years	6.25%
81-90 years	6.25%
2. Location	
a. Buccal mucosa	35.29%
b. Floor of the mouth	11.76%
d. Palate	17.64%
e. Tongue	5.88%
f. Vestibule	23.52%
g. Alveolar mucosa of cleft	5.88%
3. Clinical appearance	
Painless swelling	70.58%
Fullness of the face	17.64%
Congenital cleft	5.88%
Not reported	5.88%
4. Radiographic appearance	
Articles not reported radiographic features	47.05%
Single Radio-dense body	11.76%
Patchy mineralization	35.29%
Homogeneous distribution	5.88%
5. Parosteal variants	17.64%
Soft tissue variants	82.35%

bone trabeculae. Focal area showed numerous blood vessels which were suppressed by mature adipocytes, and this tumor was infiltrated with mononuclear lymphocytes [Figure 6]. No pleomorphic features or mitotic figures were observed. By correlating all these histological features, the final diagnosis of osteolipoma was given. The patient has been kept under regular follow-up.

DISCUSSION

The etiological factors for lipoma are still unknown, but a total of three hypotheses have been mentioned in the literature; (1) trauma or infection or chronic irritation or hormonal alterations could be responsible for metaplasia of fibroblasts into osteoblasts,[3] (2) proliferations of lipoplastic embryonic cell nests,[3] and (3) osteoinducing factors released by blood-borne monocytes that enter the fatty tissue could be responsible for transformation of fibroblasts into osteoblasts.[4]

Here, we support the hypotheses 1 and 3 because in our case there was continuous trauma from opposing molars, and histologically, the entire tumor was infiltrated with numerous mononuclear cells which could bring the metaplasia of fibroblasts to osteoblasts [Figure 5].

Raviraj *et al*.[2] have given comprehensive review of literature from 1961 to 2016 about incidence, age/sex, clinical details, duration of the lesions, radiographic presentations of osteolipomas.

Clinically, indeed, osteolipoma is asymptomatic, but slow growth could be painful if the tumor is in contact or compress or displaces the neurological structures in pharyngeal region. The reporting time to the clinics varies from 2 months to 40 years since the first appearance of the tumor in their oral cavity.[2] In majority of the cases, it leads to facial asymmetry; however, in one case, it was associated with congenital cleft palate.[5]

The percentage of reported sites in the oral cavity was buccal mucosa (35.29%), mandibular buccal vestibule (23.52%), palate (17.64%), tongue (5.88%), and floor of the mouth (11.76%). Patient's age varies widely from birth to elderly without sex predilection. The mean age of the patients at the time of diagnosis was 51.77 years, with most cases occurred between fifth and sixth decades of life. The size of the tumor varies from 8 to 70 mm[1,2]

Our case was also in a 55-year-old female patient, with a painless nodular swelling, but the location of the tumor was retromolar trigone and edentulous area and had not been reported site earlier.

Conventional radiographs are adequate for the early detection of calcifications in the osteolipoma. OPG and periapical intraoral radiograph are the cost-effective and simple radiographs which can aid in the diagnosis. A total of two cases[2,3] have been documented in literature (5.88%) with "multiple dense homogeneous radiopaque structures" similar to our case [Figure 4].

However, the observed radiographic finding in our case was different from reported ones, with "homogeneous and equal distribution of ossifications in a whorled pattern or in concentric layers" which has not been reported until now [Figures 3 and 4]. These calcifications and ossifications are the secondary features of osteolipoma as well as several benign and malignant tumors. Ultrasonography reveals it as hyperechoic mass with areas of focal calcifications[1,2] which was also revealed by our case also.

Histologically, osteolipomas are characterized by the scattering lamellar bone trabeculae among proliferating mature adipocytes.[1,2] However, in our case, the trabeculae were distributed equally in a uniform manner throughout the tumor-like "concentric layers."

Lipomas are subclassified into 12 types based on the appearance of the stroma or matrix in histological sections; these variants are[6,7] (1) fibrolipoma, (2) chondrolipoma, (3) intramuscularlipoma, (4) angiolipoma, (5) infiltrating type angiolipoma, (6) noninfiltrating type angiolipoma, (7) myolipoma, (8) chondrolipoma, (9) chondrolipoma with ossification, (10) spindle cell lipoma, (11) pleomorphic lipoma, and (12) sialolipoma.

The differential diagnosis includes pyogenic granuloma, traumatic fibroma, peripheral giant cell granuloma, peripheral ossifying fibroma, peripheral osteoma, phleboliths, myositis ossificans, neurofibroma, pleomorphic adenoma, cysticercosis, chondrolipoma, chondroblastoma, chondro myxofibroma, chondrosarcoma, osteosarcoma, and liposarcoma.

Reactive lesions of the oral cavity such as pyogenic granuloma, fibrous hyperplasia or traumatic fibroma, and peripheral giant cell granuloma are similar to lipoma in clinical appearance as isolated nodules. However, in radiographs, resorption of the adjacent interseptal bone is commonly seen in both

traumatic fibroma and peripheral giant cell granuloma which is absent in pyogenic granuloma. Oral pyogenic granuloma shows histological findings of prominent capillary over growth in hyperplastic granulation tissue. Traumatic fibroma shows histological findings of stratified squamous epithelium and solid nodular mass of dense hyalinized fibrous connective tissue. Peripheral giant cell granuloma shows multinucleated giant cells in histopathological sections.

Peripheral ossifying fibroma is a nonneoplastic enlargement of the gingiva tissue; histologically, it shows dense aggregates of spindle-shaped fibroblasts, bundles of collagen fibers along with irregular bony trabeculae, and cementum-like material.

Peripheral osteoma is a benign often-asymptomatic neoplasm. Radiographically, the lesion appears as well-circumscribed radiopaque densities. Histologically, it consists of well-differentiated mature bone either compact or cancellous bone.

Phleboliths or vein stones are calcifications in the veins. Histologically it contains dilated vessels with calcified thrombus in a concentric manner. Myositis ossificans is progressive, heterotrophic ossification of muscles. Based on histological findings of muscle fiber bundles and classical zonal architecture, this can be excluded from the diagnosis of the present case report.

Neurofibroma is a benign tumor of nerve cell, circumscribed, and nonencapsulated tumor. Histologically, it appears as nodular tumor mass which consists of nerve tissue in fascicles.

Pleomorphic adenoma of the minor salivary gland clinically appears as a submucosal nodule; however, histopathologically, an epithelial tumor of complex morphology possessing epithelial and myoepithelial elements arranged in a variety of patterns, which embeds in mucopolysaccharide stroma.

Cysticercosis is also clinically appears as a solitary nodule and shows elongated densely calcified bodies in radiographs as "rice grain pattern." However, the histopathological examination confirms the diagnosis by the detection of cystic cavity containing cysticercus cellulosae.

Clinically, it is impossible to diagnose the particular type of lipoma because all these tumors appear as solitary nodules. Both chondrolipoma and osteolipoma appear same in consistency. However, on microscopic examination, chondrolipoma consists of chondroid matrix whereas osteolipoma consists of bony trabeculae in the adipose matrix. Kate and Jayabhaye[8] reported the presence of ossifications in chondroid lipoma in their case report.

Chondroblastoma and chondromyxoid fibroma are benign cartilaginous tumors of long bones in young patients. Radiographically, both mimic osteolipoma in the feature of "internal calcifications." However, histologically, both the lesions contain chondroid tissue, which is absent in osteolipoma. Radiography cannot differentiate osteolipoma from malignant tumors such as chondrosarcoma, osteosarcoma, and liposarcoma because all these pathologies also contain internal calcifications, so the definitive diagnosis should be done based on histological features of cellular atypia.

The syndromes which are commonly associated with multiple lipomas are Cowden's syndrome or multiple hamartomatous syndromes, neurofibromatosis, Gardner syndrome, encephalo cranio cutaneous lipomatosis, multiple familial lipomatosis, and proteus syndrome, so these syndromes also should be considered in differential diagnosis.

Complete surgical excision is the treatment of choice. Radiotherapy is indicated in infiltrating type of angiolipoma, but its role has not proven yet in osteolipoma.[9] The malignant transformation of osteolipoma has not been reported until now.

CONCLUSION

This is the first case report of occurrence of osteolipoma in retromolar trigone, which depicted the radiographic appearance of "whorled pattern." Hence, clinicians should be aware of this presentation in retromolar region and include in differential diagnosis. Even though radiographically it mimics the sarcomas with internal calcifications, surgeons should treat these lesions conservatively and should avoid the radical treatment.

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Conflicts of interest
There are no conflicts of interest.

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