

Osteolipoma: a rare tumor in the oral cavity



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Osteolipoma is a rarely reported histologic variant of lipoma that exhibits bone formation. To the best of our knowledge, only 13 well-documented case reports of osteolipoma in the oral cavity have been published in the English literature. This study presents the clinical, radiographic, and histologic features of an osteolipoma in the oral cavity and reviews the pertinent literature. The patient was a 29-year-old female, who presented with an 8-month history of a painless, progressively enlarging, well-defined, movable submucosal mass in the left posterior buccal mucosa. The lesion had a hard consistency. Imaging findings revealed a spherical radiopacity with an irregular trabecular pattern. The lesion was excised and the diagnosis of osteolipoma was established. No recurrence was observed after a 5-year follow-up. (Oral Surg Oral Med Oral Pathol Oral Radiol 2016;122:e8-e13)

Lipomas are benign mesenchymal neoplasms of soft tissue, which may affect the oral cavity. Previous studies have suggested that 20% of cases occur in the head and neck region, with only 1% to 4% occurring in the oral cavity.¹⁻³ Oral lipomas primarily affect the buccal mucosa, floor of the mouth, and lips.^{2,3} Histologic variants of lipoma are described on the basis of the predominant type of lesional tissue, including fibrolipoma, angioliipoma, myoliipoma, leiomyoliipoma, myxoliipoma, spindle cell lipoma, osteoliipoma, chondrolipoma, and sialoliipoma.^{2,3} Osteoliipoma, a lipoma exhibiting bone formation, is a rarer histologic variant among oral lipomas, accounting for less than 1% of cases described in the literature.⁴⁻⁶ Osteoliipomas are classified as *intraosseous* when located in bone and as *parosteal* or *periosteal* when located adjacent to bone. Lipomas that exhibit bone formation but are independent of bone tissue (nonattached osteoliipomas) have been reported in very few cases.⁵

To the best of our knowledge, only 13 well-documented case reports of osteoliipoma in the oral

cavity have been published in the English language literature. Due to its rarity, knowledge about this tumor is incomplete. We describe a case of osteoliipoma affecting the buccal mucosa.

CASE REPORT

A 29-year-old female patient presented to the Oral Pathology Clinic at Pontifícia Universidade Católica de Minas Gerais with a painless mass in the left buccal mucosa with an 8-month evolution. The patient had no history of trauma in that region. The medical history and systemic review were noncontributory. Physical intraoral examination showed a 1.5 × 1.5 cm, well-defined, movable submucosal mass in the left posterior buccal mucosa, near the retromolar triangle (Figure 1A). The lesion had a yellowish appearance and hard consistency. The overlying mucosa showed telangiectasia but no ulceration or erythema (Figure 1B).

Because the mass presented a hard consistency, a radiographic examination was performed. Imaging findings revealed a round area of radiopacity, with an irregular pattern of trabeculae, and no evidence of cortical abnormality or influence on the surrounding structures was found (Figure 2).

The clinical differential diagnosis included oral osseous choristoma (soft tissue osteoma), cartilaginous choristoma, osteoliipoma, chondrolipoma, pleomorphic adenoma with ossification, and other salivary gland or connective tissue tumors with dystrophic calcification. It was free from the periosteum and not attached to any adjacent structures, such as the mandibular bone. With the patient under local anesthesia, the lesion was completely excised. It was free from the periosteum and not attached to any adjacent structures, such as the mandibular bone. The surgical specimen was sent to the Oral Pathology Laboratory.

Grossly, the resected specimen consisted of a mass measuring 1.8 × 1.5 × 1.2 cm, which was well circumscribed and yellowish with soft and focal hard consistency (Figure 3A). On sectioning, the mass exhibited a yellow soft tissue with various intermixed thin lamellar bony structures and a fibrous capsule (Figures 3B and 3C).

Microscopically, the mass presented abundant mature adipose tissue, with no atypia, and was separated by thin fibrous

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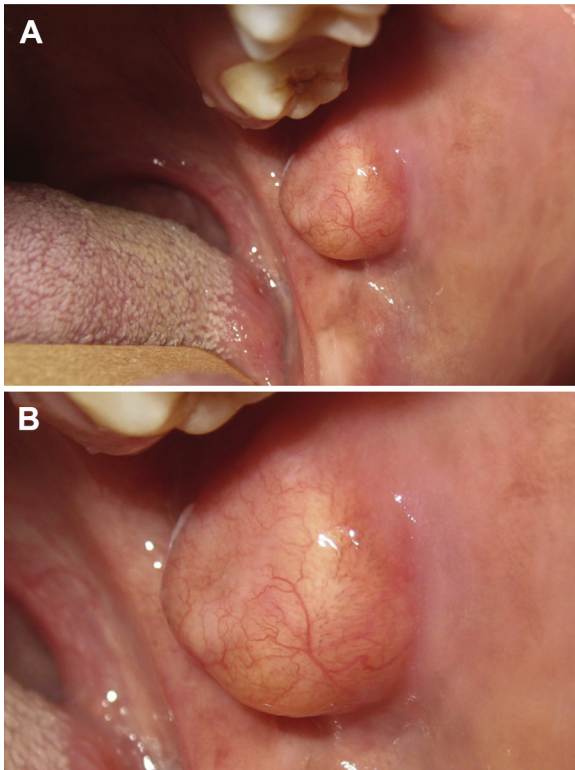


Fig. 1. Clinical aspects of oral osteolipoma. **A**, Well-defined, movable submucosal mass showing yellowish appearance, hard consistency and a size of 1.5 × 1.5 cm located in the left posterior buccal mucosa, near the retromolar triangle. **B**, The overlying mucosa showed telangiectasia without ulceration or erythema.

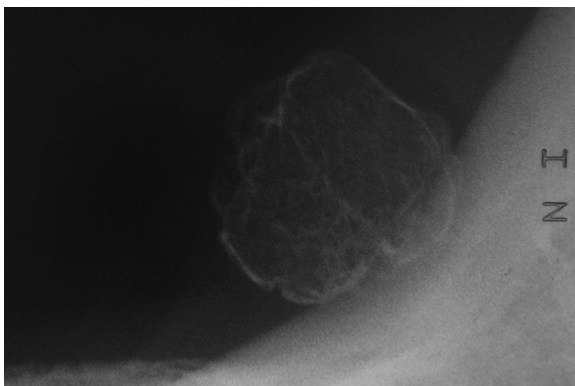


Fig. 2. Radiographic aspects of oral osteolipoma. Radiography revealed a spherical radiopacity with an irregular pattern of trabeculae, and no evidence of cortical abnormality or influence on the surrounding structures was found.

connective tissue septa. Randomly distributed irregular trabeculae of immature bone, with osteoblastic activity, were found throughout the tumor. No foci of hematopoietic cells were observed (Figures 4A-D). The lesion was surrounded by a thin osseous layer and a fibrous capsule (see Figure 4A). The surgical margins were tumor-free.

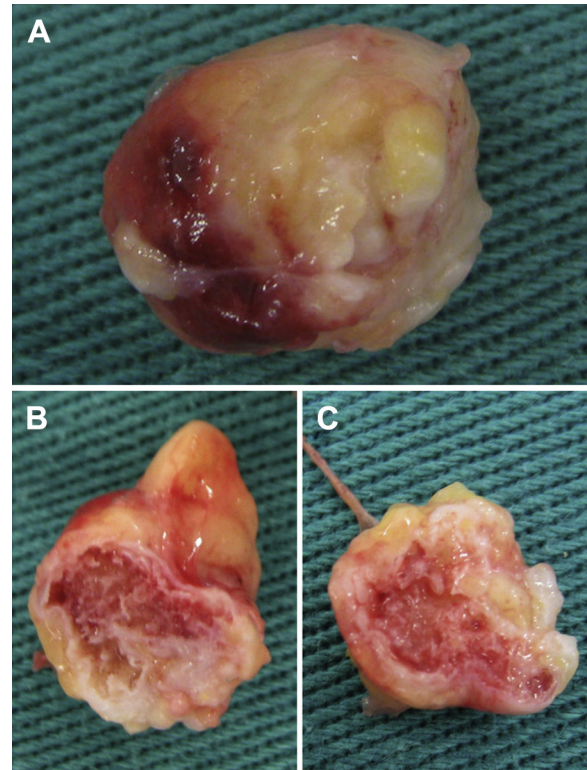


Fig. 3. Gross appearance of oral osteolipoma. **A**, The resected specimen consisted of a mass measuring 1.8 × 1.5 × 1.2 cm that was well circumscribed and yellowish, with soft and focal hard consistency. **B** and **C**, On sectioning, the mass revealed a yellow soft tissue with various mingled thin lamellar bony structures and a fibrous capsule.

A final diagnosis of osteolipoma was established. The recovery course was uneventful, and no recurrence was detected after a 5-year long follow-up.

DISCUSSION

Lipomas are common benign soft tissue tumors,^{7,8} appearing most commonly in areas of the body where adipose tissue is present.⁹ The subcutaneous or submucosal region is the usual site, but these can develop between skeletal muscle fibers, on bone surfaces, or within deep soft tissues.⁷ The tumor may undergo a variety of changes. An uncommon change is ossification.^{1,5,6,8-17} To our knowledge, only 13 cases of oral osteolipomas have been reported in the English language literature. Table I summarizes their features.

Although oral lipomas are believed to be more common in men,³ no clear gender preponderance has been observed in patients with osteolipomas of the oral cavity (see Table I), except for one case of a young boy with an osteolipoma in the palate, most patients affected by oral osteolipoma were adults. Most cases were reported in patients over 40 years of

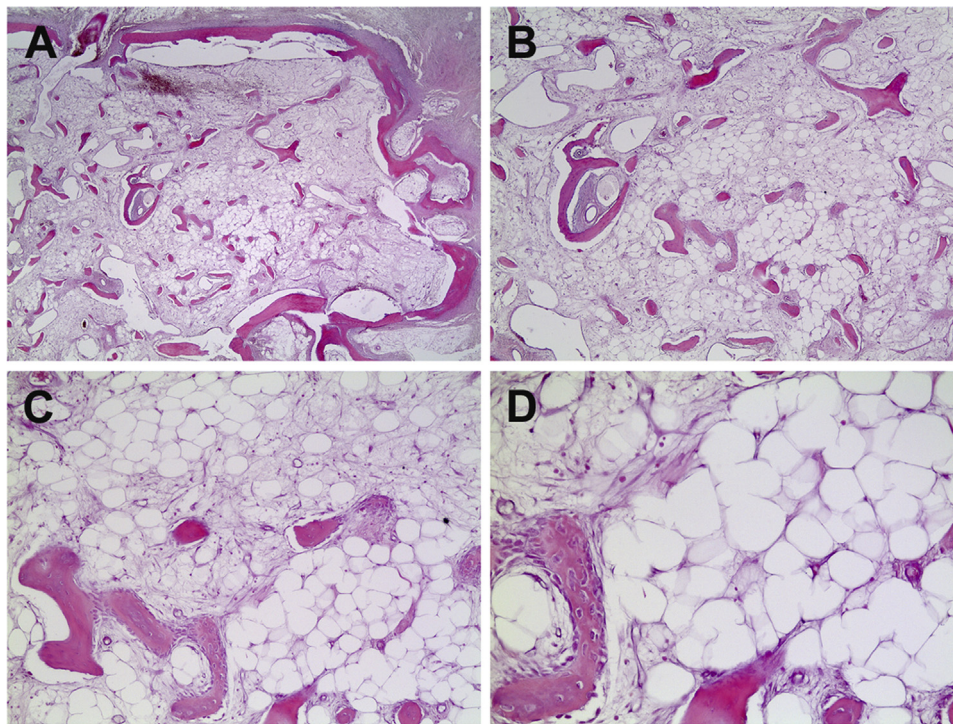


Fig. 4. Histopathologic features of oral osteolipoma. Abundant mature adipose tissue with no atypia and separated by thin fibrous connective septa was observed. Randomly distributed irregular trabeculae of immature bone exhibiting osteoblastic activity were found throughout the tumor. The tumor was surrounded by a thin osseous layer and fibrous capsule. No foci of hematopoietic cells were observed (A, H&E, $\times 20$; B, H&E, $\times 40$; C, H&E, $\times 100$; D, H&E, $\times 200$). A high-resolution version of this slide for use with the Virtual Microscope is available as eSlide: VM01035.

age. A long history of slow progression is commonly observed, although our patient had the lesion for only 8 months.

As evident in Table I, osteolipomas of the oral cavity can occur in various anatomic sites, regardless of the proximity to bone. Four cases occurred in the buccal mucosa, four in the mandibular buccal mucosa (vestibule or alveolar mucosa), three in the hard palate, two in the floor of the mouth, and one in the lateral tongue. In the cases of tongue osteolipoma, osteocartilaginous choristoma, osteosarcoma, liposarcoma with metaplasia, and post-traumatic chondrification had been considered in the differential diagnosis.⁹ In one case involving the alveolar mucosa, exostosis, peripheral giant cell granuloma, fibrous hyperplasia, and fibroma with calcifications had been considered⁵; and in a case of a palatal osteolipoma, the differential diagnosis had included osteoma, neurofibroma, and intraosseous palatal cyst.⁶ Additionally, with regard to the differential diagnosis, in osteolipomas exhibiting radiographic features, such as the present case, the clinician should consider oral osseous choristoma (soft tissue osteoma), cartilaginous choristoma, chondrolipoma, pleomorphic adenoma with ossification, and other salivary gland or connective tissue tumors with dystrophic calcification.

Suggestive clinical and radiographic features (hard or soft peripheral mass generally associated with a radiopaque area), as well as the histopathologic appearance of mature fatty tissue with diffuse bone trabeculae, usually confirm the diagnosis of osteolipoma (Table I). However, these features are also found in oral osseous choristoma. The term *oral osseous choristoma* was coined by Krolls et al.¹⁸ in 1971 to describe benign bone formations in oral soft tissue, previously referred to as *soft tissue osteomas*. Although oral osseous choristoma generally exhibit a mass of dense lamellar bone,¹⁹ which is easily differentiated from an osteolipoma, some of these lesions can display a cancellous pattern. These osseous choristomas, such as the case described by Hodder and MacDonald²⁰ in 1988, display spongy bone trabeculae containing abundant bone marrow spaces filled with fatty tissue, a histologic organization suggestive of that found in osteolipoma. Generally, foci of hemopoietic marrow are encountered in osseous choristomas²⁰ but not in osteolipomas.⁴ Hence, this characteristic may be important in the differential diagnosis. Both lesions are benign connective tissue lesions, which are treated by conservative surgical excision and exhibit similar prognoses.

Table 1. Osteolipomas of the oral cavity previously reported in the English language literature (current case included)

Author	Age (Years)	Gender	Site	Clinical presentation	Imaging findings	Duration	Histopathology
Godby et al. ¹¹	54	Male	Floor of the mouth	Painless and soft mass. Size: 7.0 × 6.0 × 3.0 cm (gross examination)	Radiopaque mass (occlusal radiography)	1 year	Mature adipose tissue, well-formed cancellous bone, fibrous connective tissue and striated muscle
Hughes CL ¹²	69	Male	Mandibular buccal vestibule	Painless and soft yellowish “walnut-size” mass	No evidence of alterations (radiography)	NA	Fat cells with foci of ossification surrounded by fibrous connective tissue
Allard et al. ¹³	81	Female	Mandibular buccal vestibule	Slight facial asymmetry Painless and hard “walnut-size” mass	Well defined radiopaque mass with a pattern of irregular trabeculae (occlusal radiography)	30-40 years	Homogeneous adipose tissue containing fibrous septa and irregular trabeculae of lamellar bone without hematopoietic tissue
Piattelli et al. ⁹	49	Female	Lateral margin of the tongue	Painless and hard mass Size: 0.8 cm in diameter	NA	8 years	Mature adipose tissue containing lamellar bone surrounded by a fibrous pseudocapsule
Castilho et al. ⁸	65	Female	Buccal mucosa	Painless and soft yellowish mass Size: 1.0 × 1.0 × 0.8 cm (gross examination)	NA	NA	Mature fat cells supported by fibrous septa. Focal areas of woven bone surrounded by fusiform-shaped mesenchymal cells
Saghafi et al. ⁵	68	Male	Mandibular buccal alveolar mucosa	Painless and hard mass. Size: 1.5 × 1.0 cm	No evidence of cortical abnormality or influence on the surrounding structures (radiography)	4 years	Adipose tissue with foci of lamellar bone surrounded by mesenchymal cells
Gokul et al. ¹⁴	6	Male	Hard palate (associated with cleft of hard and soft palate)	Painless and soft mass. Size: 3.0 × 2.0 cm	Well-defined hypodense lesion arising with mixed density, showing a well-defined radiodense body (computed tomography)	Congenital	Lobules of adipose tissue separated by fibrovascular connective tissue septa and showing the presence of mature bone
de Castro et al. ¹	47	Female	Buccal mucosa	Facial asymmetry Painless nodule. Size: 1.5 cm in diameter	An irregular and radiopaque structure (radiography of the surgical specimen)	1 year	Trabeculae of lamellar bone inside a mature adipose tissue surrounded by thin septa of conjunctive tissue
Adebisi et al. ⁶	37	Female	Hard palate	Painless and hard mass. Size: 3.0 × 4.0 cm	Patchy areas of radiopacity (occlusal radiography)	10 years	Mature adipose tissue with scattered trabeculae of lamellar bone
Hsu et al. ¹⁵	71	Male	Buccal mucosa	Painless and hard mass. Size: 4.0 × 2.5 cm	NA	4 years	Lobules of mature adipose tissue separated by fibrous septa Randomly distributed trabeculae of mature lamellar bone and foci of woven bone
Bajpai et al. ¹⁶	55	Male	Hard palate	Painless and hard yellowish mass Size: 1.5 × 1.4 cm	Patchy area of radiopacity (occlusal radiography)	4 years	Bone trabeculae surrounded by lobules of mature adipocytes separated by fibrous septa

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Table I. Continued

Author	Age (Years)	Gender	Site	Clinical presentation	Imaging findings	Duration	Histopathology
Amaral et al. ¹⁰	51	Male	Mandibular buccal mucosa	Slight facial asymmetry. Painless and hard mass. Size: 2.0 × 1.5 cm	Well-defined hyperchoogenic mass with areas of calcification (ultrasonography)	3 years	Proliferation of mature fat cells with central areas of lamellar bone trabeculae and fibrous septa
Raghunath and Manjunatha ¹⁷	20	Female	Floor of the mouth	Painless and hard yellowish mass. Size: 6.0 × 6.0 cm	Well-defined, hypodense lesion with irregular hyperdense areas (computed tomography)	3 years	Central areas of osseous trabeculae and lobules of matured adipose tissue
Omonte et al. (current case)	29	Female	Buccal mucosa	Painless and hard mass. Size: 1.5 × 1.5 cm	Spherical radiopacity with an irregular trabecular pattern (radiography)	8 months	Mature adipose tissue with fibrous septa and irregular trabeculae of immature bone Tumor surrounded by a thin osseous layer and a fibrous capsule

NA, not available.

The pathogenesis of osteolipomas has not yet been clarified. Some authors have suggested that osteolipomas may originate from two types of undifferentiated mesenchymal cells that separately form adipose and bone cells.²¹ An alternate theory proposes that multipotent adipose-derived stem cells within adipose tissue may be involved in osteolipoma development. Thus, bone tissue develops inside a lipoma in response to growth signals.²² This theory hinges on the assumption that adipose-derived stem cells have the potential to differentiate into fat, bone, cartilage, and vascular components.²³ Some other authors have suggested that repetitive trauma, metabolic changes, or ischemia may lead to metaplasia of pre-existing fibrous elements within lipomas, which then develop into osteoblasts.^{24,25} The conditions that induce the differentiation of mesenchymal stem cells into bone remain to be investigated.

CONCLUSIONS

Although osteolipoma is a very rare entity in the oral cavity, it should be considered when a patient presents with a peripheral lesion that has a hard consistency on palpation, areas of radiopacity in image examinations of peripheral lesions, and mature adipose tissue associated with bone trabeculae in histopathology. It has the same prognosis as a simple lipoma, and surgical excision is the recommended treatment. Recurrences have not been reported.

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