CASE REPORT

Parosteal osteochondrolıpoma of the mandıble

Sibel Elif Gültekin, Sevil Kahraman[1], Kıvanç Karadayı[1]
Departments of Oral Pathology and [1]Oral and Maxillofacial Surgery, Gazi University Faculty of Dentistry, Ankara, Turkey

Address for correspondence:
Dr. Sibel Elif Gültekin,
Department of Oral Pathology, Gazi University,
Faculty of Dentistry, Biskek caddesi 82. Sokak
Emek-Ankara, Turkey - 06510.
E-mail: sibel@gazi.edu.tr

ABSTRACT
Osteochondrolipoma is a rare benign soft tissue neoplasm. It is occasionally considered to be a variant of adipose tissue neoplasm 'lipoma' showing multiple differentiation pathways of pluripotent stem cells. As with the lipomas they can be seen at any location and show cartilagenous and osteoid differentiation when located parosteally. We present a case of osteochondrolipoma located at the symphysis of the mandible. To our knowledge, this is the first reported case of an oral osteochondrolipoma associated with parosteal localization.
Key words: Mandible, osseous and cartilaginous metaplasia, osteochondrolipoma

INTRODUCTION

Lipoma is the most common soft tissue mesenchymal neoplasm in adults, which is located in any part of the body. The majority occurs in the upper-half of the body, particularly the trunk, head, and neck region, and it is rarely described in the oral cavity. Conventional lipoma has been divided into three forms according to the origin of the localization: Superficial lipoma (arise within subcutaneous tissue), deep lipoma (arise within deep soft tissue) and parosteal lipoma (arise within the surfaces of bone).[1] The histological characteristics and types vary, which include angiolipoma, angiomyolipoma, myolipoma, osteolipoma and chondrolipoma.[1] Both osteolipomas and chondrolipomas are rare entities.[2-4] However, occurrence of osteolipomas or in other term "lipomas with oseous and cartilagenous metaplasia" are less common than other subtypes of lipomas.[4]

The term of osteochondrolipoma has been used for the lipomas containing both cartilage and bone which, are very rarely seen and generally associated with a paraosteal localization.[4] To our knowledge, oral ostochondrolipoma with parosteal localization has not been reported. We present the first case of osteochondrolipoma parosteally located at the mandible.

CASE REPORT

A 64 year old male patient with painless mass on his mandibulary symphysis region, was referred to the Department of Oral and Maxillofacial Surgery, Faculty of Dentistry on September, 2007. The lesion had been first noticed 2 months prior of the presentation to the clinic with no significant enlargement. There was no relevant medical and family history apart from diabetus mellitus. Clinical examination of the oral cavity revealed a well demarcated, firm, round and exophitic lesion of approximately 2 cm in diameter at the symphysis region of the left mandible. The lesion was firmly attached to the bone in the clinical examination and the overlying mucosa was intact and of normal color.

Radiographically, at the site of the lesion some degree of radioopacity was observed [Figure 1]. Under local anesthesia, the patient was taken to the operating room and the lesion was completely excised with an intraoral approach. During the surgical intervention, the encapsulated lesion was not adherent to the underlying periosteum and overlying mucosa, thus the lesion was dissected and removed easily [Figure 2]. The surgical specimen was histopathologically evaluated for final diagnosis at Department of Oral Pathology, Faculty of Dentistry, Gazi Universitys. The 6-month postoperative follow-up showed healing of the oral mucosa without any recurrency.

Macroscopically, the gross specimen was a 2.7 × 1.8 × 1.4 cm encapsulated mass mostly comprising of adipose tissue with a few number of encapsulated nodular structures. On section, the cut surface was yellowish-white with an amount of bony streaks.

Histological evaluation revealed a partially encapsulated lobular benign adipose tissue neoplasm. The large areas of mature fat cells supported with fibrous connective tissue septa. Focal areas of hyaline chondroid structures and lamellar bone with fatty marrow were seen throughout the lesion. Both islands of bone and cartilage were surrounded with well defined fibrous tissue [Figure 3]. There was no histological evidence of malignancy,

Access this article online
Quick Response Code:
Website: www.jomfp.in
DOI: 10.4103/0973-029X.99090

such as mitosis, cellular atypia, necrosis, and hemorrhage. Histochemistry with Alcian Blue Periodic acid-Schiff stain showed acid mucopolysaccaride presence in chondroid tissue; hence, this was hyaline chondroid. On immunohistochemistry, adipocytes were positively stained with vimentin antibody, while both adipocytes and chondrocytes were positive for S-100 protein [Figure 4]. The diagnosis was osteochondrolipoma.

DISCUSSION

Lipomas are the most common mesenchymal neoplasm of the soft tissue. However, they are relatively uncommon in the oral cavity with incidence of 1–4%.[5,6] Variants of lipoma especially osteolipoma and chondrolipoma are very rare lesions which are located in the oral cavity.[2,3,5,7] Furthermore, intaoral lipoma with both osteoid and cartilaginous metaplasia has been reported just in two cases.[7] We presented an osteochondrolipoma case that differed from the typical arrangement by two rare alterations.

First, the lesion showed well-defined cartilaginous and osseous areas within lipoma that is also considered as osseous and cartilaginous metaplasia. In the a study of Fregnani *et al*.,[5] fibrolipoma was reported as the dominant subtype in 46 lipoma cases, in which neither osteolipoma nor chondrolipoma was found. Two lipomas with cartilaginous or osseous metaplasia was reported out of 125 lipoma cases in another study, whereas none of the cases represented osteochondrolipoma.[6] Although, any case in the name of "oral osteochondrolipoma" has not been reported, lipoma with osteoid and cartilaginous metaplasia has been represented by two cases in the oral region.[7] The osteochondrolipoma is the specific term of the lipoma with mature cartilaginous and osseous differentiation;[4] thus, both terms represents the same entity.

The nomenclature of ostechondrolipomatous lesions is controversial. Some authors consider such type of lipoma as a benign mesenchymoma due to consisting of two or more mesenchymal elements such as lipocytes, chondrocytes, osteocytes, and fibroblasts.[8] Jones *et al*.[9] stated that the term of benign mesenchymoma should be used strictly to describe an unencapsulated neoplasm composed of two or more mature mesenchymal tissues not ordinarily associated with each other, excluding fibrous connective tissue. In the present case, as the tumor is well demarcated and partially encapsulated and moreover the prominent component is mature adipose tissue, the tumor is considered as osteochondrolipoma.

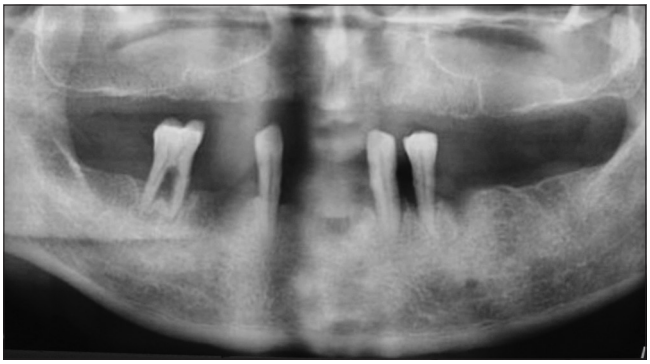

Figure 1: Preoperative panoromic radigoraph exhibiting the dense radioopacity (arrow) in the symphysis region of the mandible

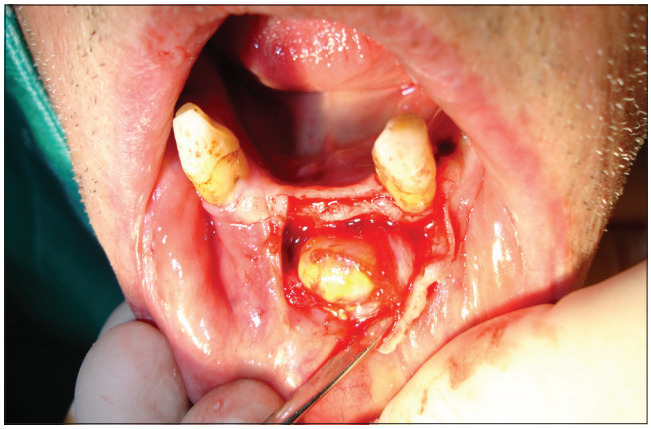

Figure 2: Intraoperative view of the lesion with encapsulation

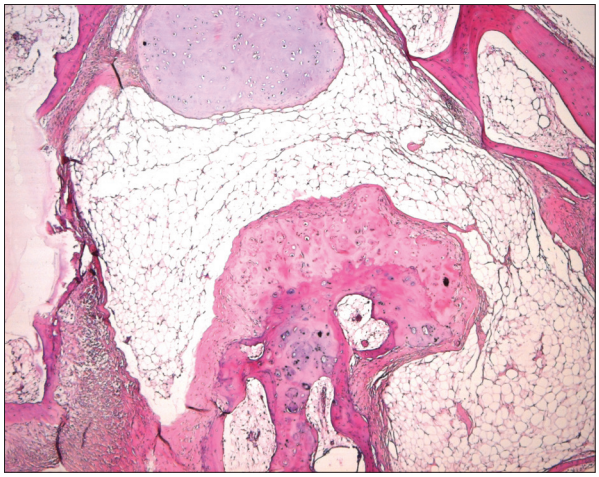

Figure 3: Histopathological features of osteochondrolipoma showing mature adipose tissue containing cartilage islands and lamellar bone trabecules (H and E, ×40)

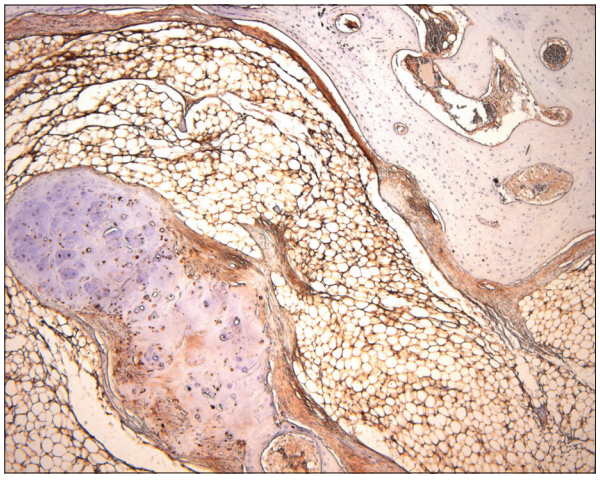

Figure 4: Chondrocytes and adipocytes demonstrating positivity for S-100 protein (Immunoperoxidase, ×40)

Second alteration that took part in this case, was the parosteal localization of the lesion. The parosteal lipoma which is defined as lipoma exhibits a conguous relationship with the periosteum, usually demonstrating some form of attachment to periosteum with an underlying osseous reaction, is rarely seen in the oral cavity.[10] Our case did not exhibit any adherence to the underlying bone and periosteum. Similar features was reported in an intraoral parosteal lipoma case.[10] Radiographically, at the site of the lesion some degree of radioopacity was observed. Various types of osseous cortical responses from a reactive overproduction of bone to cortical erosion can be seen underlying bone.[10] The radiopacity in our case may reflect either the reactive bone production of underlying bone or the density of cartilagenous and ossous component of the tumor.

The pathogenesis of osteochondrolipoma is very speculative. Multipotent cells of mesenchyme, different cell lines and metaplastic differentiation of the adipose tissue are three hypothesis regarding the origin of the tumor.[4] There were no reports of an osteochondrolipoma for the jaws in the literature. However, considering its parosteal location, histological findings of well-formed cartilage and bone tissue within the adipose tissue neoplasm, it is decided to be a true neoplasm of mesenchymal tissue stem cells showing characteristics of three different tissues originating from the mesenchyme.

REFERENCES

1. Feltcher DM, Unni KK, Mertens F. World Health Organization Classification of Tumors. Pathology and genetics of tumors of soft tissue and bone. Lyon: IARC Press; 2002. p. 19-20.
2. Castilho RM, Squarize CH, Nunes FD, Pinto Junior DS. Osteolipoma: A rare lesion in the oral cavity. Br J Oral Maxillofac Surg 2004;42:363-4.
3. Piattelli A, Fioroni M, Lezzi G, Rubini C. Osteolipoma of the tongue. Oral Oncol 2001;37:468-70.
4. Rau T, Soeder S, Olk A, Aigner T. Parosteal lipoma of the thigh with cartilaginous and osseous differentiation: An osteochondrolipoma. Ann Diagn Pathol 2006;10:279-82.
5. Fregnani ER, Pires FR, Falzoni R, Lopes MA, Vargas PA. Lipomas of the oral cavity: Clinical findings, histological classification and proliferative activity of 46 cases. Int J Oral Maxillofac Surg 2003;32:49-53.
6. Furlong MA, Fanburg-Smith JC, Childers EL. Lipoma of the oral and maxillofacial region: Site and subclassification of 125 cases. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2004;98:441-50.
7. Allard RH, Blok P, van der Kwast WA, van der Waal I. Oral lipomas with osseous and chondrous metaplasia; report of two cases. J Oral Pathol 1982;11:18-25.
8. Enzinger MF, Weiss SW. Soft Tissue Tumors. 2nd ed. Missouri: Mosby; 1988. p. 305-6.
9. Jones AC, Trochesset D, Freedman PD. Intraoral benign mesenchymoma: A report of 10 cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2003;95:67-76.
10. Steiner M, Gould AR, Rasmussen J, LaBriola D. Parosteal lipoma of the mandible. Oral Surg Oral Med Oral Pathol 1981;52:61-5.

How to cite this article: Gültekin SE, Kahraman S, Karadayi K. Parosteal osteochondrolıpoma of the mandıble. J Oral Maxillofac Pathol 2012;16:280-2.

Source of Support: Nil. **Conflict of Interest:** None declared.