J. Oral Diag. 2017; 02:e20170039

ORIGINAL ARTICLE

An unusual presentation of osteoblastoma

Victor Perez Teixeira [1,2]
José Narciso Rosa Assunção Júnior [1]
Letícia Almeida Oliveira [1]
Ana Maria Hoyos Cadavid [3]
Celso Augusto Lemos Júnior [4]
João Alberto Lopes Sousa Júnior [5]

Abstract:

Osteoblastoma is a rare benign bone tumor that accounts for less than 1% of all primary bone tumors. Due to a low occurrence and varied histopathological aspects, this pathology becomes a diagnostic challenge. Therefore, our objective of reporting a case of osteoblastoma in the maxilla is to discuss another presentation of this tumor. Male patient, suffering from leucoderma, 50 years old, presented an expansion in right superior vestibular groove with pain during palpation and an extensive osteolytic lesion in the right maxilla, of solid content, observed in computed tomography. With an anatomopathological diagnosis of osteoblastoma by an incisional biopsy, conservative surgery was performed to remove the lesion. The definitive analysis of the excised tumor confirmed the preoperative diagnosis. The patient has been followed up for a period of 5 years without relapses of the neoplasia. Therefore, due to the rarity and the difficult diagnosis, describing cases like this one and their presentation characteristics is extremely important to enhance knowledge of this pathology among professionals in the area of diagnosis and maxillofacial surgery.

Keywords: Osteoblastoma; Diagnosis, Oral; Maxilla.

[1] UNIMES, School of Dentistry - Santos - São Paulo - Brasil
[2] UNIMES, School of Medicine - Santos - São Paulo - Brasil
[3] FOUSP, Department of Oral Pathology, School of Dentistry - São Paulo - São Paulo - Brasil
[4] FOUSP, Department of Stomatology, School of Dentistry - São Paulo - São Paulo - Brasil
[5] Hospital Dr. Luis Camargo da Fonseca e Silva, Serviço de Cirurgia de Cabeça e Pescoço - Cubatão - São Paulo - Brasil

Correspondence to:
Victor Perez Teixeira.
E-mail 1: teixeira.victorp@gmail.com
E-mail 2: victorpteixeira@hotmail.com

Article received on July 18, 2017.
Article accepted on October 5, 2017.

DOI: 10.5935/2525-5711.20170039

INTRODUCTION

Osteoblastoma is a rare benign bone tumor that accounts for less than 1% of all primary bone tumors. It most commonly affects young adults, with a mean age of 20 years, usually occurring in long bones and vertebral column. Only 10 to 20% of osteoblastoma occur in the maxillary region, representing approximately 0.7% of the bone neoplasms of this region[1-3].

According to the clinical history and histological aspects, there are two well-defined types of osteoblastoma, named a conventional or benign osteoblastoma that presents with slow growth and evolution time of years, well defined sclerotic margin, sparse vascularization, and medial inflammatory infiltration. The other form is known as aggressive osteoblastoma, which is fast-growing, locally invasive, with a propensity to recur, and has atypical histopathological features, often hindering differentiation from low-grade osteosarcoma[4,5].

The most common presentation of the benign osteoblastoma involves a swelling with expansion of the bone cortex and slight pain, its size typically do not exceed 4 cm[6,7].

The radiographic aspect of the osteoblastoma is not very consistent and varies depending the duration. Generally, it consists in an image with radiopaque and radiolucent patterns, depending on the degree of calcification. In most cases, it´s not possible to identify a perilesional sclerotic border[7].

The differential diagnosis of this lesion must include osteoid osteoma, cementoblastoma e fibroma ossificante. Sometimes, the difficult in the diagnostic persists even in the histopathologic analysis, which can resemble other bone-producing lesions, fibrous bone lesions and also osteosarcoma in some cases[8,9].

A conservative approach with surgical excision and curettage have been suggested in literature. The recurrence is rare and have been attributable to an incomplete excision[9].

Due to the low occurrence and varied histopathological aspects, this pathology becomes a diagnostic challenge. Therefore, our objective of reporting a case of osteoblastoma in the maxilla is to discuss another presentation of this tumor.

CASE REPORT

A 50-year-old male patient, who suffered from leucoderma, presented with the complaint of lack of adaptation of his superior total prosthesis to the ridge, for approximately 2 years, in addition to a complaint of pain if trying to use it. At the extraoral physical examination, the patient presented with facial asymmetry owing to the increase of volume in right zygomatic region with nasolabial groove erasure. In oroscopy, presented with a large expansion in right superior vestibular groove, measuring approximately 4.5 cm on its largest axis, consistent and sore to palpation. The mucosa of the region showed normal and preserved staining.

In computed tomography, we observed an extensive osteolytic lesion in the right maxilla, of solid content, extending laterally up to the zygomatic bone, medially to the nasal fossa, and posteriorly to the tuber region (Figure 1).

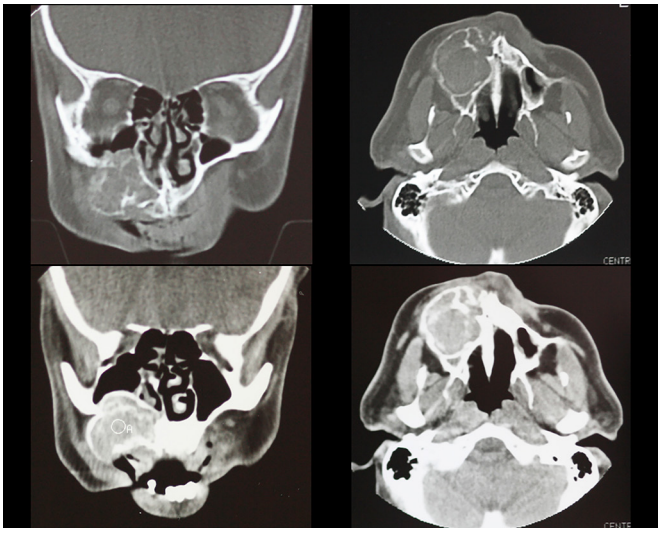

Figure 1. Computed tomography, coronal and axial scan, window for soft and hard tissues, showing extensive osteolytic lesion in the right maxilla, with solid content, extending laterally up to the zygomatic bone, medially to the nasal fossa, and posteriorly to the tuber region.

Due to the clinical picture and the tomographic aspect, an incisional biopsy was performed having as diagnostic hypotheses to be clarified an odontogenic tumor and a central ossifying fibroma. In this first procedure, we obtained as the histopathological result a fibrous dysplasia. However, as the images were not compatible with this pathology, we submitted the piece for a new evaluation, this time in the Department of Oral Pathology of FOUSP. This new exam showed an anatomopathological diagnosis of osteoblastoma.

In view of this diagnosis, conservative surgical removal and curettage of the tumor were performed (Figure 2). The excised tumor was submitted to a definitive anatomopathological diagnosis. The histological

sections showed fragments of benign bone neoplasia, characterized by the presence of osteoid tissue and bony trabeculae immersed in a richly cellularized and vascularized dense connective tissue stroma, showing spindle fibroblasts. The numerous bony trabeculae that composed the lesion are surrounded by broad osteoblasts which present oval and basaloid nuclei besides abundant eosinophilic cytoplasm. Different degrees of mineralization are verified in the trabeculae and osteoid formations. Within the bony trabeculae, cells with large nuclei are observed and were diagnosed as benign osteoblastoma (Figures 3 and 4).

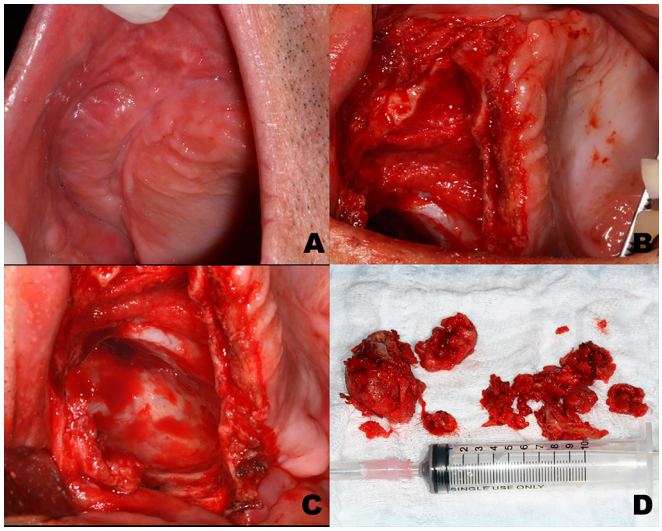

Figure 2. (A) Expansive increase of volume in right maxilla. (B) Intraoperative aspect of the lesion. (C) Surgical cavity after complete tumor removal. (D) Excised tumor.

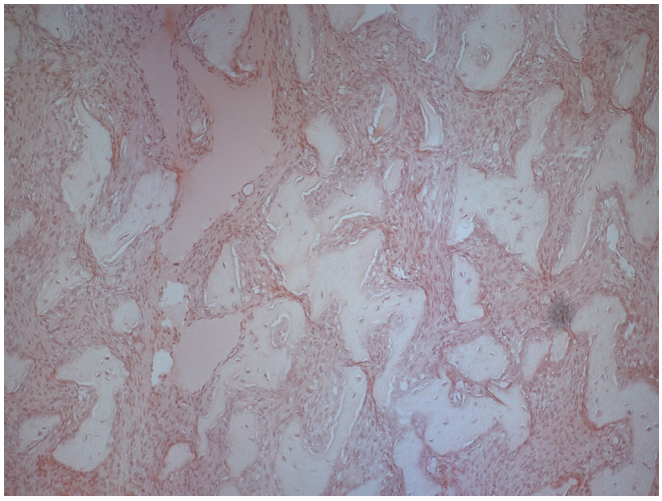

Figure 3. Photomicrograph 10X (H&E): osteoid tissue and bony trabeculae with different degrees of mineralization immersed in a richly cellularized and vascularized dense connective tissue stroma, showing spindle fibroblasts. Into the bony trabeculae, cells with large nuclei are observed.

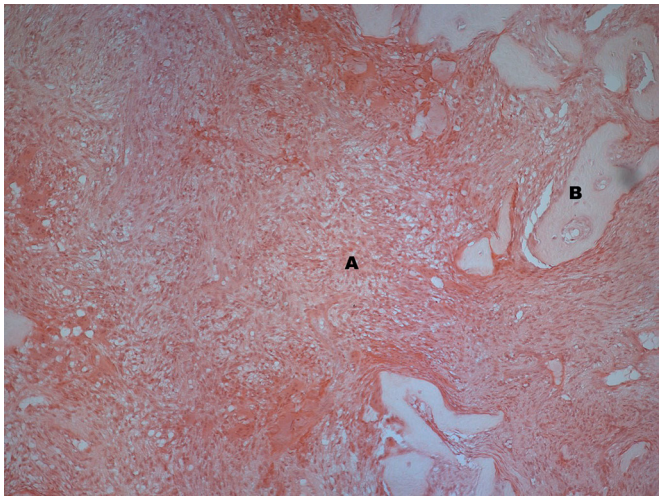

Figure 4. Photomicrography 10X (H&E): (A) Dense connective tissue, richly cellularized. (B) Bony trabeculae, showing cells inside.

The patient has been in a clinical radiographic control for five years without signs of relapse.

DISCUSSION

Although osteoblastoma could affect any bone in the human skeleton, it usually develops in long bones followed by the cervical spine (47-69%). Only about 10% of cases occur in maxillo-mandibular complex, with an apparent predilection for jaw[1]. In 2006, Jones *et al.*[10] reviewed 77 cases of osteoblastoma and observed that only 28.6% of cases occur in maxilla. It usually occurs in young adults, with the mean age of 20 years[6], so our case is unusual because of its location of development and age of diagnostic.

Clinical presentation of this pathology is quite varied. Pain, often mild and longstanding, is the most present symptom[11]. In the case reported here, the patient complained of pain only when using his total prosthesis. Regarding the clinical alterations, it is generally presented as an expansion of slightly painful cortical bone to palpation, and limited growth does not exceed four centimeters in diameter, in most cases[6,11]. This report shows a lesion with slow but continuous growth, assuming dimensions that resulted in asymmetry and facial deformity.

Radiographically, its presentation is quite varied, and there is no pattern. Normally, a mixed pattern of radiopacity/radiolucency can be found, depending on the calcification degrees and duration of the frame, in addition to the absence of perilesional sclerotic border[12]. Therefore, fibro- osseous lesions and odontogenic tumors can be included in a differential diagnosis. Normally, fibrous dysplasia has a ground glass appearance

with poorly discernible borders, diverging from the aspects generally observed in osteoblastoma. In contrast, ossifying fibromas manifest as central radiopaque masses surrounded by a radiolucent rim akin to osteoblastoma, although they have a predilection for jaw and are generally well delimited[13,14].

On CT scan, we could observe the absence of well-defined lesional limits, as well as a mixed image aspect, which led us to consider whether it is an odontogenic tumor or central ossifying fibroma. Although we can observe odontogenic tumors with a similar radiographic presentation, such as cementoblastoma, odontoma, and desmoplastic ameloblastoma, anatomopathological features are quite different[13].

According to the histopathological features, osteoblastoma has been subdivided into conventional (benign) and aggressive. The term benign osteoblastoma was first proposed by two authors in 1956, Jaffe[15] and Lichtenstein[16]. At that time, they used this term to identify an osteoid matrix forming osteoblastic lesion similar to osteoid osteoma, but with higher growth potential.

Later, in 1972, Dorfman[17] included osteoblastoma in a review of malignant transformation of benign bone lesions. In a review of his osteoblastoma cases, he noted that some of them exhibited recurrent behavior and non-specific histologic features that would deserve the name of aggressive osteoblastoma. Since then, the term has been used for clinical patterns of greater aggressiveness and speed, having a higher rate of recurrence after conservative therapies.

Initially, osteoblastoma can be a diagnostic challenge. Due to the low incidence in the population, its clinical and anatomopathological standards are often confused with fibro-osseous lesions or odontogenic tumors[7]. This difficulty was found in our case, in which, at first, we received the provisional microscopic diagnosis of fibrous dysplasia, and only after a slide review, at a reference center in oral pathology, we obtained the description for osteoblastoma.

Osteoblastoma can be included in the microscopic differential diagnosis of fibro-osseous lesions, such as fibrous dysplasia, by the possibility of observing certain similar patterns in these pathologies[18] and also of osteoid osteoma, which can be distinguished only by adding the clinical characteristics[19]. The literature shows that the central feature to distinguish osteoblastoma from other fibro-osseous lesions is that the stroma does not consist of cellular spindle cells but rather is a loose vascular stroma with numerous prominent epithelioid-type osteoblasts[18].

In the case presented by us, the lesion was treated conservatively, by means of excision and curettage. In the literature, the therapeutic modality employed ranged from more conservative[20] to more aggressive excision in the block, such as maxillectomies or mandibulectomies[6,11,13]. The most aggressive treatment, with a safety margin, has been indicated in order to reduce recurrence. However, in 2001, Gordon *et al.*[21] described the likelihood of recurrence for conventional lesions around 13.6%.

However, recurrences of the lesions, when described in the literature, were related to the difficulty of access, facilitating incomplete approaches[22,23] or even inadequate initial treatment due to incorrect preoperative diagnosis[11]. Therefore, the possibility of recurrence would be more related to these factors than to the pathological behavior of osteoblastoma.

Therefore, due to the rarity and the difficult diagnosis, describing cases like this one and their presentation characteristics is extremely important to enhance knowledge of this pathology among professionals in the area of diagnosis and maxillofacial surgery. In our case, the correct diagnosis was only possible when a maxillofacial pathologist was consulted, highlighting the importance of such professional.

REFERENCES

1. Lucas DR, Unni KK, McLeod RA, O'Connor MI, Sim FH. Osteoblastoma: a clinicopathologic study of 306 cases. Hum Pathol. 1994;25:117-34.
2. Rawal YB, Angiero F, Allen CM, Kalmar JR, Sedghizadeh PP, Steinhilber AM. Gnathic osteoblastoma: clinicopathologic review of seven cases with long-term follow-up. Oral Oncol. 2006;42:123-30.
3. Matsuzaka K, Shimono M, Uchiyama T, Noma H, Inoue T. Lesions related to the formation of bone, cartilage or cementum arising in the oral area: a statistical study and review of the literature. Bull Tokyo Dent Coll. 2002;47:173-80.
4. Woźniak AW, Nowaczyk MT, Osmola K, Golusinski W. Malignant transformation of an osteoblastoma of the mandible: case report and review of the literature. Eur Arch Otorhinolaryngol. 2010;267:845-9.
5. Kulkarni MM, Shah AS, Ahire S. Aggressive osteoblastoma of the mandible: a case report. Int J Contemp Dent. 2011;2:135-8.
6. Angiero F, Mellone P, Baldi A, Stefani M. Osteoblastoma of the jaw: report of two cases and review of the literature. In Vivo. 2006;20:665-70.
7. Bokhari K, Hameed MS, Ajmal M, Togoo RA. Benign osteoblastoma involving maxilla: a case report and review of the literature. Case Rep Dent. 2012;2012:351241.
8. Peters TE, Oliver DR, McDonald JS. Benign osteoblastoma of the mandible: report of a case. J Oral Maxillofac Surg. 1995;53:13479.

9. Mardaleishvili K, Kakabadze Z, Machavariani A, Grdzelidze T, Kakabadze A, Sukhitashvili N, *et al.* Benign osteoblastoma of the mandible in a 12-year-old female: A case report. Oncol Lett. 2014;8:2691-4.
10. Jones AC, Prihoda TJ, Kacher JE, Odingo NA, Freedman PD. Osteoblastoma of the maxilla and mandible: a report of 24 cases, review of the literature, and discussion of its relationship to osteoid osteoma of the jaws. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2006;102:639-50.
11. Shah S, Kim JE, Huh KH, Yi WJ, Heo MS, Lee SS. Recurrent osteoblastoma of the maxilla. Dentomaxillofac Radiol. 2013;42:20100263.
12. Saikhedkar R, Wanjari S, Nirwan A, Prasad H. Osteoblastoma involving maxilla: an unusual report with review of literature. J Orofac Sci. 2010;2:53-7.
13. Pandiar D, P M S, Gulati S, Banyal VS, Kumar A. Benign osteoblastoma of maxilla: a case report emphasizing the histopathological differential diagnosis. J Clin Diagn Res. 2014;8:ZD21-3.
14. Alawi F. Benign fibro-osseous diseases of the maxillofacial bones. A review and differential diagnosis. Am J Clin Pathol. 2002;118Suppl:S50-70.
15. Jaffe H. Benign osteoblastoma. Bull Hosp Joint Dis. 1956;17:141-51.
16. Lichtenstein L. Benign osteoblastoma; a category of osteoid- and bone-forming tumors other than classical osteoid osteoma, which may be mistaken for giant-cell tumor or osteogenic sarcoma. Cancer. 1956;9:1044-52.
17. Dorfman HD. Proceedings: Malignant transformation of benign bone lesions. Proc Natl Cancer Conf. 1972;7:901-13.
18. McCarthy EF. Fibro-osseous lesions of the maxillofacial bones. Head Neck Pathol. 2013;7(1):5-10.
19. Cury SEV, Silva BSF, Rocha AC, Lemos-Junior CA, Pinto Junior DS. Osteoid osteoma of the mandible: case report. J Oral Diag 2016;01:e20160019.
20. Manjunatha BS, Sunit P, Amit M, Sanjiv S. Osteoblastoma of the jaws: report of a case and review of literature. Clin Pract. 20111;1:e118.
21. Gordon SC, Macintosh RB, Wesley RK. A review of osteoblastoma and case report of metachronous osteoblastoma and unicystic ameloblastoma. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2001;91:570-5.
22. Ohkubbo T, Hemandez JC, Ooya K, Krutchkoff DJ. "Aggressive" osteoblastoma of the maxilla. Oral Surg Oral Med Oral Pathol. 1989;68:69-73.
23. Ataoglu O, Oygur T, Yamalik K, Yucel E. Recurrent osteoblastoma of the mandible: a case report. J Oral Maxillofac Surg. 1994;52:86-90.