Case Report

Painless osteoid osteoma in the maxilla of an elderly female patient

Iván Alejandro Díaz-Rengifo[1], Antonio José Díaz-Caballero[2], Katherine Redondo-De Oro[3], Yuritza Yoleinnis Hernández-Arenas[4]

[1]Physician and Surgeon, Rosario University, Young Researcher Colciencias in University of Cartagena, GITOUC Research Group, [2]Dentist, University of Cartagena, Periodontist, Javeriana University, Master in Education, Universidad Del Norte, Phd in Biomedical Sciences, University of Cartagena, Faculty of Dentistry Professor in University of Cartagena, GITOUC Research Group, [3]Physician Specialist in Pathology, University of Cartagena, Faculty of Medicine Professor in University of Cartagena, [4]Dentist, University of Cartagena, Research Assistant in GITOUC Research Group, Bolívar, Colombia

Abstract

Osteoid osteoma is an osteoblastic benign bone tumor more frequent in long bones of young male patients. It is the third most commonly diagnosed benign bone tumor and has distinctive symptomatology, nocturnal pain that relieves with nonsteroidal anti-inflammatory drugs. Nowadays, total resection is the preferred management. In the present paper, an unusual variant in the maxilla of a female elderly patient without previous symptomatology is exposed; it was surgically removed without signs of reappearance in 12 months of follow-up. Although it is not a common site of appearance, the manifestation of this tumor in the skull bones seems to be associated with a different pattern in contrast to the skeletal type as can be seen in the present case and others previously reported. The authors consider due to the clinical similarity between this and other tumors that it should be taken into consideration for future diagnosis dilemma.

Keywords: Bone neoplasms, jaw, maxilla, osteoid osteoma

Address for correspondence: Dr. Iván Alejandro Díaz-Rengifo, Faculty of Dentistry, University of Cartagena, Health Campus, Zaragocilla, Cartagena, Bolívar, Colombia, South America.
E-mail: ivandr93@hotmail.com
Received: 28.11.2018, **Accepted:** 03.04.2019

INTRODUCTION

Osteoid osteoma (OO) consists of a small tumor lesion usually limited with a distinctive pattern and classic symptomatology.[1] It is an osteoblastic tumor which produces a disproportional pain in contrast to the size of the actual lesion and that characterizes itself for a nocturnal time of presentation and response to nonsteroidal anti-inflammatory drugs (NSAIDs).[2] OO represents 2%–3% of resected bone tumors; however, it is the third most common diagnosed bone tumor in long bones around 10%–14% between benign bone neoplasias.[3] This is a self-limited entity whose natural tendency is regression, within 6–15 years without treatment and around 2–3 years when acetic salicylic acid (ASA) or NSAIDs are added.[2] Its characteristic features include a dull intermittent pain, which increases in duration, frequency and intensity in time, that typically worsen at night and response well to NSAIDs.[2,4] As a slow-growth tumor, its presentation could be delayed; therefore, the finding during the initial stages may be incidental due to the lack of clinical

Access this article online

Quick Response Code:

Website: www.jomfp.in

DOI: 10.4103/jomfp.JOMFP_295_18

How to cite this article: Díaz-Rengifo IA, Díaz-Caballero AJ, Oro KR, Hernández-Arenas YY. Painless osteoid osteoma in the maxilla of an elderly female patient. J Oral Maxillofac Pathol 2019;23:280-3.

manifestations.[2] These typical symptoms are reported in almost 80% of patients.[5] However as previously described in other reviews, a representative number around 30% of patients with OO affecting the jaws seem to be painless by the time of discovering.[6] OO is more frequent in adolescents and young adults, with a peak of incidence between 10 and 20 years. Typically, a more common male to female ratio is seen in relation to this entity but referencing the skeletal variety mainly in long bones such as tibia and femur or affecting the spine in around 6% of cases.[7] The appearance in flat or skull bones is very unusual around 1% of cases, and the manifestation in the jaw does not show gender predilection.[2,5,8,9] The most common site of presentation for OO within the bone itself is the cortical area, displaying typical radiographic findings in the X-rays; usually, a radiolucent nidus generally <1 cm in size surrounded by a sclerotic bone area or cortical thickening. Although descriptions from different sites of presentations are present in the literature, with variations in the imaging findings, there are reports of subcortical, subperiosteal, intraperiostial and intramedullary OO. Occurrence in bones with subcutaneous locations may present with local signs of inflammation.[2] In this paper, a clinical case of OO is presented as an incidental finding during the clinical examination for dental rehabilitation implants based in the left upper maxilla, which is an unusual site of presentation and apparently occurred without any previous manifestation.

CASE REPORT

A 69-year-old female patient attended for examination in need of dental prosthesis in the upper left maxilla. The patient refers that a few years ago she had an implant-supported dental prosthesis in the mandible without complications. As interesting aspects of her medical history, she only refers osteopenia and the use of bisphosphonates, discontinued in the last 6 months. During the physical examination, the upper right and left premolars appeared fractured and infected with a bilateral abscess formation. Simple X-rays shows a well-delimited radiopaque mass of 4 mm × 8 mm in the left maxilla [Figure 1].

After additional interrogation, the patient refers the finding of an anomaly in a previous X-ray of 10 years ago, which was reviewed, finding a small radiopaque mass of 4 mm × 7 mm similar to the one recently found. Nevertheless, she denies any previous symptoms or history of trauma at the site of injury. Further investigation with computed tomography identified a bone mass of 5 mm × 8 mm with clear edges and neither invasion of the cortical nor the tissue surround, there was no ganglionar reactivity [Figure 2].

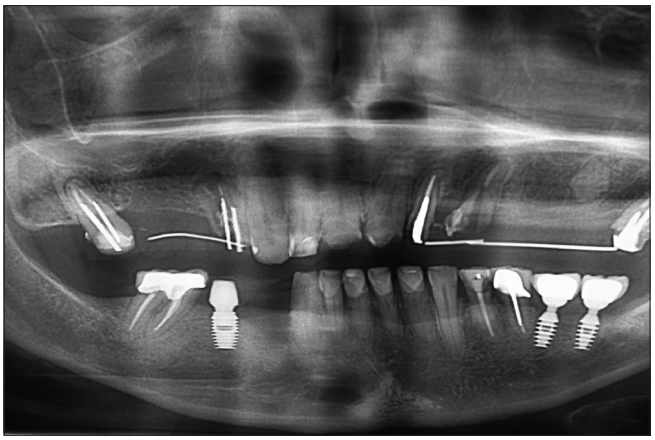

Figure 1: Presurgical X-rays. Simple X-rays from mandible and maxilla, it shows a radiopaque small well-delimited lesion of 4 mm × 8 mm, in the left side of the maxilla

Due to the lack of growth, small size, delimited borders and absence of locoregional compromise (no ganglionar reactivity in physical examination or imaging), surgical removal of the lesion was performed considering a benign bone growth and dental implants were implanted during the same surgical time. A full-thickness flap was developed with a light osteotomy for easy liberation of the lesion using surgical burs; complete liberation was performed in a conservative way. The complete specimen was sent to pathology in 10% neutral-buffered formalin, the report described a benign differentiated bone lesion close to the cortical, inside presents thick trabeculae of mature bone with laminar and sclerotic formations such as Haversian system, there is a dense bone mass with medullar activity without osteoblastic or osteoclastic activity; suggesting an OO [Figure 3].

After 12 months of follow-up, the patient has proper healing without clinical or radiographic evidence of recurrence.

DISCUSSION

Around 15 cases of OO in the jaw can be found in the literature of the past 10 years, their main characteristic is the accurate definition of the lesion like our case; however the most frequently referred symptom is the dulled pain sometimes accompanied by swelling of the area,[6] not uncommonly followed by surgical trauma, none of these seen in our patient. Despite the age predilections, this osteoblastic tumor can manifest at any age like seen in this report. As mentioned before, the painless variety in flat bones seems to be greater, even up to 30%.[6] These could

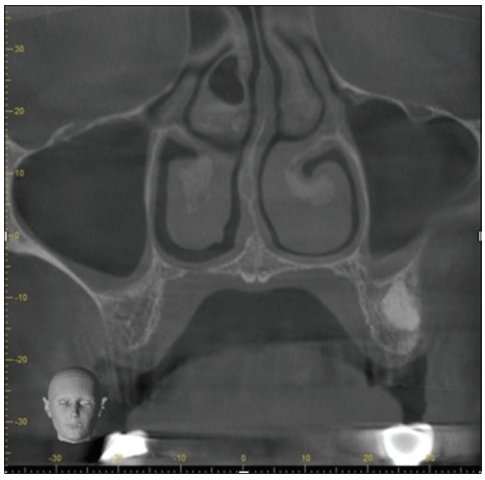

Figure 2: Presurgical cone beam computed tomography. Cone beam computed tomography, transversal view, a small radiodense lesion in contact with lower maxillary sinus of 5 mm × 8 mm consistent of an intracortical nidus with a variable amount of mineralization and cortical enlargement

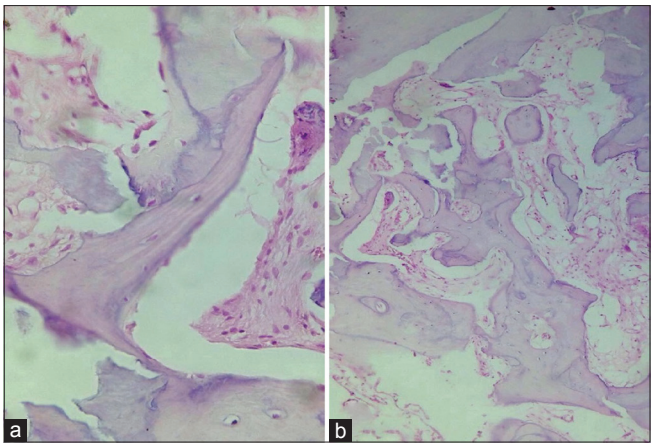

Figure 3: Biopsy results. (a) A ×10 amplified sample where Havers-like formations can be seen with active bone marrow within. (b) A ×40 amplified image where the osteoid tissue with small cellularity can be appreciated

be in relation to the small size and slow-growing rate of the lesion in a nonproblematical area like the jaw, which is not under axial pressure as other parts of the body, so it could easily be associated to other symptomatology, which is considered what happened in the present report. Initial approximation to OO includes a simple X-ray as the study of choice, whose typical findings have been mentioned above. These findings, however, may become difficult to detect according to the affected area and surrounding structures.[2] In long time cases, secondary osteopenia and changes in bone morphology may become more evident, which was not this case despite the apparent 10 years of evolution. An *et al.*, 2013 found that approximately 38% of OO may have dense radiographic appearance rather than radiolucent, like the presented case where no radiolucent nidus was evident at the moment of evaluation; they also reported 38% of mixed radiographic appearance in their review.[6] Despite the multiple radiographic features, the best way to characterize an OO is with computer tomography, even better than resonance images.[10] A tomography is recommended to analyze for more lesions and a better description of the current one in the preoperative scenario.[2] As done confirming the lack of clinical and radiographic signs of malignancy. The main differential diagnosis for an OO is an osteoblastoma, with which it shares many imaging and even histological features.[11,12] However, some particularities may be noticed: in regard to radiographic size, OO is usually smaller than 1 cm and a mass of 1.5 cm or over is suggestive of an osteoblastoma.[2,3,5] Multiple simultaneously nidi are rare, notwithstanding more common in OO than osteoblastoma.[13] Symptoms in osteoblastoma may be absent or can manifest as pain and tenderness similar to OO, however, does not have the characteristic night pain that improves with ASA and NSAID.[5] Ultimate diagnosis with this or any other entity will be directed by the pathological findings as it was with this patient who did not present any typical symptomatology or additional imaging finding rather than the small bone mass. In histology can be seen well defined limited structures of irregular bone trabeculae with a varying mineralization degree, surrounded by a reactive bone formation structure with loose of the fibrovascular stroma.[2] The tissues around may show augmented vascularity with vessels that provides nutrition to the nidus as they tend to become smaller and get into it.[14] Chronic inflammatory infiltrates may be found in chronic lesions,[2] which did not happen in this report of a 10-year lesion. The cellular component that can be usually found consist of osteoblast around the trabeculae and multinucleated osteoclast-like cells have also been described.[5] Nowadays, a complete excision of the lesion is the recommended management, and it associates with full remission of symptomatology. Recurrent lesions could suggest incomplete removal or the presence of multiple not previously detected nidi.[15]

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

REFERENCES

1. Jaffe H. "Osteoid osteoma": A benign osteoblastic tumor composed of osteoid and atypical bone. Arch Surg 1935;31:709-8.
2. Boscainos PJ, Cousins GR, Kulshreshtha R, Oliver TB, Papagelopoulos PJ. Osteoid osteoma. Orthopedics 2013;36:792-800.
3. Unni KK. Dahlin's bone tumors. General Aspects and Data on 11,087 Cases. 5th ed. Philadelphia, PA: Lippincott-Raven; 1996. p. 121-42.
4. Lee EH, Shafi M, Hui JH. Osteoid osteoma: A current review. J Pediatr Orthop 2006;26:695-700.
5. Dorfman HD, Czerniak B, editors. Benign osteoblastic tumours. In: Bone Tumours. St. Louis, MO: Mosby; 1998. p. 85-127.
6. An SY, Shin HI, Choi KS, Park JW, Kim YG, Benavides E, *et al.* Unusual osteoid osteoma of the mandible: Report of case and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol 2013;116:e134-40.
7. Saifuddin A, White J, Sherazi Z, Shaikh MI, Natali C, Ransford AO. Osteoid osteoma and osteoblastoma of the spine. Factors associated with the presence of scoliosis. Spine (Phila Pa 1976) 1998;23:47-53.
8. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H. Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg 2011;19:678-89.
9. 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.
10. Davies M, Cassar-Pullicino VN, Davies AM, McCall IW, Tyrrell PN. The diagnostic accuracy of MR imaging in osteoid osteoma. Skeletal Radiol 2002;31:559-69.
11. Alvares Capelozza AL, Gião Dezotti MS, Casati Alvares L, Negrão Fleury R, Sant'Ana E. Osteoblastoma of the mandible: Systematic review of the literature and report of a case. Dentomaxillofac Radiol 2005;34:1-8.
12. 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.
13. Kyriakos M, El-Khoury GY, McDonald DJ, Buckwalter JA, Sundaram M, DeYoung B, *et al.* Osteoblastomatosis of bone. A benign, multifocal osteoblastic lesion, distinct from osteoid osteoma and osteoblastoma, radiologically simulating a vascular tumor. Skeletal Radiol 2007;36:237-47.
14. de Chadarévian JP, Katsetos CD, Pascasio JM, Geller E, Herman MJ. Histological study of osteoid osteoma's blood supply. Pediatr Dev Pathol 2007;10:358-68.
15. Streitparth F, Teichgräber U, Walter T, Schaser KD, Gebauer B. Recurrent osteoid osteoma: Interstitial laser ablation under magnetic resonance imaging guidance. Skeletal Radiol 2010;39:1131-7.