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Case Report

Osteoid Osteoma of the Maxilla: Literature Review and Case Report

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Abstract

Osteoid osteoma (OO) is a benign osteogenic tumor, usually less than 2cm in size. It is associated with nocturnal pain, and in most cases, is alleviated with non-steroidal anti-inflammatory drugs (NSAIDs). It usually presents in the first three decades of life, and is predominant in men. Osteoid osteoma usually affects long bones, and rarely occurs in the jaws. When it does, it is mostly localized in the mandible. Differential diagnosis of OO includes fibro-osseous lesions, tumors of osseous origin, and odontogenic tumors. Because of overlapping and superimposition of anatomical structures on conventional radiographs, it is difficult to visualize the architecture of this type of lesion using radiography. Cone beam computerized tomography allows observing the different planes of the lesion, without superimposed structures, and should therefore be the method of choice to analyze jaw lesions suspicious for OO. We herein present a case of OO in the maxilla, which brings the total number of reported cases to six.

Keywords: Osteoid osteoma; Jaws; Pain

Introduction

Osteoid osteoma (OO) is a benign osteogenic tumor, which is usually less than 2cm in size. It is associated with nocturnal pain, and in most cases, is alleviated with non-steroidal anti-inflammatory drugs (NSAIDs). It usually presents in the first three decades of life, and is predominant in men [1,2]. Osteoid osteoma accounts for 12% of benign skeletal bone tumors [3], and usually affects lower limb long bones, like the femur (40%), tibia and fibula. Location in the humerus and vertebrae is also frequent [3,4]. Less than 1% of OOs affect the jaws. In these cases, the lingual aspect and lower border of the mandible are usually affected [5]. Osteoid osteoma can present a diagnostic challenge since it has the same clinical, radiographic, and histologic features as other lesions, such as osteoblastoma, cementoblastoma, osteosarcoma, and fibro-osseous lesions [6,7].

Imaging studies are critical to the diagnosis of this pathology. Radiographically, the tumor presents a rounded or oval radiolucent area, the "nidus", surrounded by a radiopaque rim corresponding to a peripheral sclerotic reaction zone. The "nidus" may also be surrounded by a radiolucent halo [3,8,9].

The first description of osteoid osteoma was made by Berg strand in 1930. The author reported two cases: one in the metatarsal and the other case in the phalanx of a finger. Clinically, both were thought to be osteogenic sarcoma; the metatarsal was resected and the finger was amputated. The detailed clinical features and photomicrograph left no doubt about the identity of the lesions, which were interpreted as rare benign osteoblastic lesions, and were attributed to embryonic rests [10].

Although osteoid osteoma had been reported as "chronic bone abscess", "non suppurative sklerosing osteomyelitis", and "osteomyelitis with annular sequestrum", Jaffe is due the credit for establishing it as a distinct entity in 1935. In his original report, Jaffe studied five cases of OO and evaluated all their clinical and histopathological features. The author identified certain features that were common to all the cases of OO he included in his study: 1) all patients were adolescents or young adults; 2) the presenting complaint was pain, which increased during the night; 3) radiographically, all lesions were rounded and well delimited; 4) the lesions were small and similar in size; 5) surgical excision was performed in all cases on the assumption that the lesion was inflammatory – osteomyelitis or abscesses – although there was no evidence of infection in any of the cases; 6) excision of the lesion resulted in disappearance of all symptoms without recurrence of the local condition [11].

In 1945, Jaffe reported 62 proven and treated cases, and confirmed most of the features of the tumor he had reported in his first publication on 5 cases in 1935. In 1966, Eideken identified three types of osteoid osteoma, the cortical, medullary, and subperiosteal type, depending on the radiographic location of the nidus, and found the cortical type to be the most common [13].

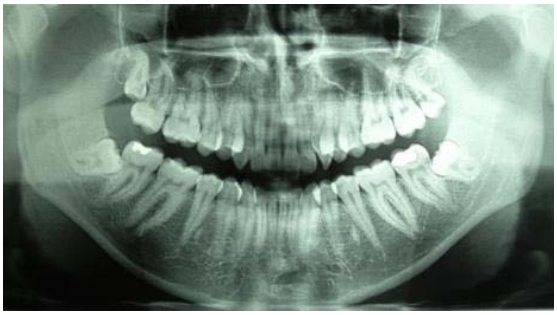

Figure 1: Panoramic radiograph. The image does not allow clear visualization of the lesion.

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Table 1: Review of the literature of case reports of osteoid osteoma in the maxillofacial region.

Case	Author/Year	Age	Sex	Location	Size	Symptoms
1	Rushton (1951) [14]	27	M	Mandible	6mm	Pain
2	Foss (1955) [29]	26	F	Mandible	5mm	Pain
3	Nelson (1955) [18]	17	M	Mandible	25mm	Pain
4	Stoopack (1958) [19]	25	M	Mandible	-	No
5	Lind (1964)	48	M	Mandibular condyle	3mm	Pain
6	Hillman (1965) [20]	4	F	Maxilla	-	No
7	Borello (1967) [30]	21	M	Maxilla	30x20 mm	No
8	Greene (1968) [31]	45	F	Maxilla	10mm	Pain
9	Brynolf (1969) [32]	77	M	Maxilla	4mm	No
10	Gupta (1985) [33]	18	F	Mandible	30mm	Pain
11	Zulian (1987) [34]	17	F	Mandible	10mm	Pain
12	Tochihara (2001) [35]	21	F	Mandibular condyle	8mm	Pain
13	Liu (2002) [36]	18	M	Mandible	12mm	Pain
14	Ida (2002) [37]	26	F	Mandible	5x10 mm	Pain
15	Badauy (2007) [38]	26	M	Mandible	8mm	Pain
16	do Egito Vasconcelos (2007) [39]	23	F	Mandibular condyle	8x11 mm	Pain
17	Rahsepar (2009) [40]	21	M	Mandible	6x8 mm	Pain
18	Manjunatha (2009) [41]	18	F	Mandible	10mm	Pain
19	Walia (2010) [42]	18	M	Mandible	10x20 mm	Pain
20	Karandikar (2011) [21]	14	M	Mandible	35mm	No
21	Singh (2012) [8]	20	M	Mandible	35mm	Pain
22	Mohammed (2013) [7]	20	-	Mandible	30mm	Pain
23	An (2013) [15]	10	M	Mandible	7x20 mm	No
24	Adouly (2015) [16]	11	F	Bilateral mandible	6mm - 25mm	Pain
25	Khaitan (2016) [5]	40	M	Maxilla // Upper jaw	10x20 mm	Pain
26	Maccotta (2016) [9]	20	M	Mandible	10mm	Pain
27	Martin (2016) [6]	14	M	Mandible	17mm	No
28	Raja Devathambi (2016) [43]	13	F	Mandible	10mm	Pain
29	Infante-Cossio (2017) [44]	44	F	Mandible	10mm	Pain
30	Our case (2017)	21	M	Maxilla	4mm	Pain

Osteoid osteoma of the maxillofacial region is very rare. It was first described by Rushton et al. in 1951; the authors reported a 27-year-old male patient presenting a 6mm lesion on the mandibular alveolar ridge associated with the lower first molar [14]. Since that first report, only 29 cases of OO in the jaws (Table 1) have been reported, 83% of which were located in the mandible and 17% in the maxilla. Unlike OO in the remaining skeletal areas, no sex predilection was observed in the reported cases of OO in the jaws. Two of 29 cases were multifocal [15,16].

When OO is located in the jaws, it is necessary to establish differential diagnosis from fibro-osseous lesions, bone tumors, and odontogenic tumors. The main differential diagnosis is from osteoblastoma. Although they are very similar lesions, osteoblastoma is usually larger9. Treatment of choice for OO in the jaws is enucleation, since it eliminates the pain. Recurrence is infrequent, and is usually due to incomplete excision of the tumor [1,9].

Case Presentation

A twenty-one year old male patient presented at the Oral Maxillofacial Surgery and Traumatology Department of the School of Dentistry, University of Buenos Aires, with a 6mm diameter swelling and intense pain in the buccal aspect of the alveolar ridge at the level of the right upper second premolar. The mucosa covering the lesion appeared normal. Panoramic radiograph revealed a radiopaque mass. Cone beam computerized tomography showed a radiopaque lesion measuring 5mm in diameter, with a radiolucent core and peripheral reactive sclerosis (Figure 2).

Under local anesthesia, the lesion was enucleated and the cavity was burred. The patient reported total remission of pain after surgery.

The histopathological study of the surgical specimen showed formation of immature bone trabeculae with marked cellularity and lined by plump epithelioid-like osteoblasts separated by highly

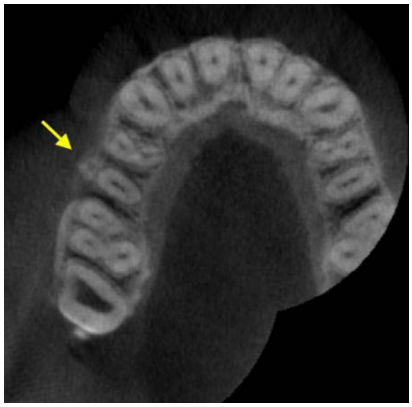

Figure 2: CBCT axial section. Note the well demarcated lesion on the buccal plate of the maxilla at the level of the second upper right premolar.

vascular, edematous and congested fibrous stroma; osteoclast-like giant multinucleated cells were also found. Thinning of the cortical bone was observed in some areas of the periphery (Figure 3). The histopathological study of the lesion together with the clinical and radiographic data confirmed diagnosis of osteoid osteoma. The patient was followed up post-operatively, and had no signs or symptoms of recurrence.

Discussion

The etiopathogenesis of osteoid osteoma is controversial. Some authors consider it to be a neoplasm, while others consider it as an inflammatory reaction. Although the traumatic etiology of osteoid osteoma has not been demonstrated to date, there are reports of the lesion occurring following trauma [18-21]. Genetic changes in OO have been scantily studied, and few cases have been reported in the literature [22].

The lesion is usually associated with severe pain that increases during the night and is relieved by NSAIDs. The tumor has been shown to have high levels of prostaglandins, especially PGE2 and PGI2, and abundant nerve fibers within the nidus, which would explain the presence of pain [23,24].

Several authors have suggested that osteoid osteoma and osteoblastoma have the same origin and are of the same nature [25]. Both are typically seen in the second decade of life, with a marked male predilection. They also have similar histological features, showing increased osteoid tissue formation surrounded by vascular fibrous stroma and perilesional sclerosis. Nevertheless, they also have distinct characteristics. The main difference is that osteoblastomas are usually larger than OO, exceeding 20mm in diameter [26,27]. Osteoid osteoma usually occurs in long bones, like the femur and the tibia, presents with pain that increases at night, and is relieved with NSAIDs. Osteoblastoma, on the other hand, is more frequently located in the axial skeleton; the pain is not worse at night and is less likely to be relieved with NSAIDSs. Osteoblastoma can be locally aggressive, whereas OO has limited growth potential [26]. There are some case reports of osteoid osteoma that recurred as an osteoblastoma after surgery [25,28].

Osteoid osteoma of the jaws is very rare. Twenty-four (83%) of the 29 reported cases were located in the mandible, and only 5 (17%) were located in the maxilla. The case reported here brings the world series of OO of the maxilla to 6 cases [29]. Because of overlapping and superimposition of anatomical structures on conventional radiographs, it is difficult to visualize the architecture of this type of lesion using radiography. Cone beam computerized tomography (BCCT) allows different spatial views of the lesion, with no superimposition of structures, and should therefore be the method of choice to observe lesions suspicious for OO of the jaws.

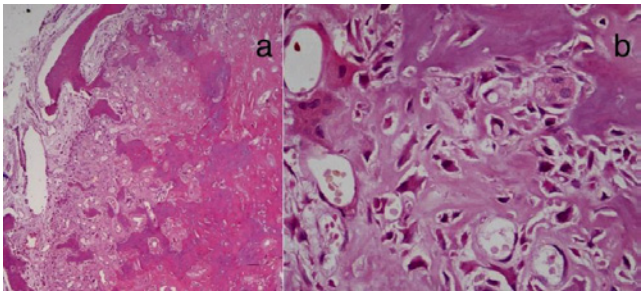

Figure 3: Histological section of the lesion. a) Bone trabeculae and osteoid with a cellular vascular fibrous stroma. Peripherally, cortical with reduced thickness and fenestration. H-E Orig. Mag. x10. b) Osteoblasts with immature bone and isolated osteoclasts. H-E Orig. Mag. x40.

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