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A RARE CASE OF OSTEOID OSTEOMA IN MANDIBLE MIMICKING A PERIAPICAL INFLAMATORY LESION- case report

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Abstract

Osteoid osteoma is an uncommon non odontogenic benign bone neoplasm that rarely occurs in the jaw bones.

This article presents a rare case of osteoid osteoma in mandible mimicking a periapical inflammatory lesion. The patient reported without symptoms and in good general health. Intraorally, there was a swelling in the periapical region of the left mandibular first bicuspid. Vitality test was negative.

Orthopantomography depicted a well defined unilocular periapical radiolucency incasing the apical third of the mandibular first bicuspid, with osteosclerotic rim on its distal prospect. Such clinical findings were appropriate for chronic periapical inflammatory lesion, so periapical surgery was scheduled.

During surgery, apicoectomy was performed, the lesion removed and sent for histopathology. The result showed irregularly proliferated trabeculae with disrupted architecture and visible osteoid accumulations typical for osteoid osteoma. Periodic follow ups were scheduled. No recurrence was noted.

Although rare, osteoid osteoma in mandible can mimic a periapical inflammatory lesion. Surgical removal and histology contributed to proper diagnostics. Complete surgical removal prevented recurrence.

***Keywords**:* osteoid osteoma, mandible, periapical inflammatory lesion, surgery, nonvital bicuspid, histology

Introduction

Osteoid osteoma is an uncommon non odontogenic benign bone neoplasm marked by production of unmineralized bone termed osteoid. First described in 1930 by Bergstrand and later classified by Jaffe in 1935 [1], it was characterized as an offbeat clinical entity [2].

Lichtenstein defined osteoid osteoma as “a small, oval, or roundish tumor-like nidus composed of osteoid and trabeculae of newly formed bone deposited within a substratum of highly vascularized osteogenic connective tissue” [3,4]. The appearance in flat or skull bones is very unusual, around 1% of cases. [5,6,7,8]. It rarely occurs within the jaws, with the mandible more commonly affected than the maxilla [9,10]. Multiple osteomas of the jawbones are seen in Gardner syndrome [11].

There is one documented case of osteoid osteoma associated with teeth. Mohammed et al.[12] reported an unusual case of osteoid osteoma associated with the apex of a mandibular second premolar and first molar. According to their report, there was a positive response of the associated teeth to the vitality tests. These lesions are usually associated with severe pain that worsens at night, at which stage relief may be obtained by non-steroidal anti-inflammatory drugs [8].

If it is encountered in the mandible it is radiographically observed as a lesion developed in the body of the mandible. It has appearance of a mixed lesion with sclerotic borders. The true nature of this lesion is unknown. [13].

Because osteoid osteoma rarely occurs in the jaw bones, it is easily misdiagnosed [14,15].

This article reports a rare case of osteoid osteoma in mandible mimicking a periapical inflammatory lesion.

Case presentation

A 38year old female was referred to the University department of oral surgery by reason of a suspected apical cyst. The patient was symptomless and in good general health. Intraorally, in the periapical region of the left mandibular first bicuspid a swelling with normal color was visible, hard and painless on palpation. The tooth was carious, unsensitive on vertical and horizontal percussion. Vitality test was negative.

Orthopantomography depicted a well defined unilocular periapical radiolucency incasing the apical third of the mandibular first bicuspid, with osteosclerotic rim on its distal prospect (figure 1).

The clinical and radiographic findings were appropriate for chronic periapical inflammatory lesion, so the patient was scheduled for periapical surgery.

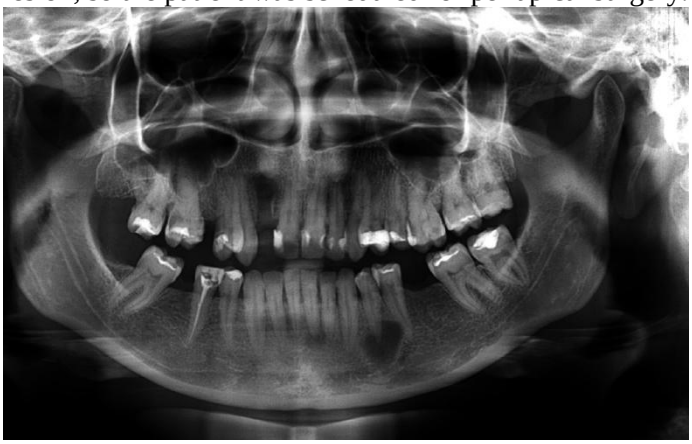

Figure 1. Unilocular radiolucency with osteosclerotic rim around the root of 34.

The surgery was performed under local block anesthesia. Triangular flap was raised. The exposed vestibular lamina was intact, but extended. After periapical osteotomy, the periapical lesion was exposed and completely removed in multiple fragments.

Apicectomy with orthograde canal filling was performed. The removed fragments were sent for histopathological verification. The result showed structure of irregularly proliferated trabeculae with disrupted architecture and visible osteoid accumulations that were built from single row of osteoblasts and proliferated fibroblasts and fibrocytes that were placed in scanty collagen matrix with preserved vascularization. Also, rare osteoclasts with irregular distribution in the lesion were seen.

Mitotic activity was absent. Such histomorphology was compatible with osteoid osteoma (figure 2). Periodic follow ups were scheduled. No recurrence was noted.

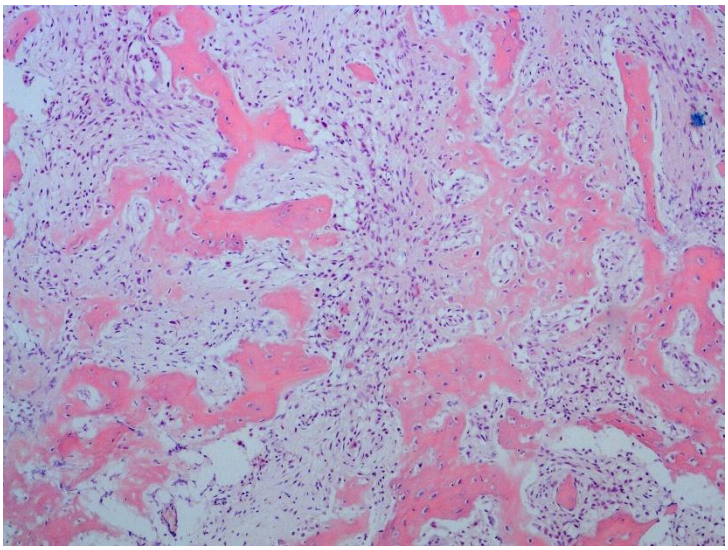

Figure 2. A photomicrograph of anastomosing bony irregular trabeculae with variable mineralization and loose, fibrovascular stroma.

Discussion

Pain is the first symptom in patients with osteoid osteoma. According to some authors, mild pain is the principal symptom of osteoid osteoma [16,17,18,19].

Others report an intense nocturnal pain in the bone tissue, and this pain is relieved with use of anti-inflammatory drugs.

There may be edema and tenderness at the surrounding soft tissue. These typical symptoms are reported in almost 80% of patients [6]

Various theories were brought forward to elucidate the reason for the pain. Arteriolar blood supply was observed to the lesion whereas others reported pressure exertion upon the surrounding bone, suggesting that the pain was produced by the vascular tumor lying within the confines of the sclerosed bone trabeculae. [20].

There is a hypothesis that the intensity of the pain is due to the rich vascularity of osteoid osteoma, which leads to innervation of the free nerve endings into the lesion, and the production of prostaglandins; this is why non-steroidal anti-inflammatory drugs affecting prostaglandins could bring relief to osteoid osteoma-associated pain [21].

Sometimes, as pointed out in our case, pain may not be the main symptom of osteoid osteoma when located in jaw bones. Representative number - around 30% of patients with osteoid osteoma affecting the jaws seem to be pain free by the time of discovering [22].

According to Boscainos PJ at al. the absence of pain can be attributed to the tumor's slow growth.

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 manifestations [5].

The patient reported to our clinic without any symptoms and the additional clinical findings correlated with a diagnosis of chronic periapical inflammatory lesion.

Dental granuloma, radicular cyst and periapical abscess represent periapical changes of frequent occurrence.[20].

Apical periodontitis lesions generally have an etiology that is associated with necrosis and infection of the root canal system that manifests itself as the host defense response to microbial challenge [23,24]; they are usually identified as radiolucency located in the apex of the teeth on radiographic examinations.

These lesions could be chronic (eg, radicular cysts, granulomas, and chronic abscesses) or acute (eg, periradicular abscess or cellulitis) and represent approximately 90% of all periapical lesions. [25]

The progressive stages of inflammation and periapical bone structure resulting in resorption are identified as exhibiting radiolucency on radiographic exams [26].

However, there are lesions of neoplastic sources, cystic lesions of non endodontic origin, and anatomic variations such as a Stafne bone cavity (SBC) that when located in the periapical area of the teeth might radiographically and clinically mimic lesions of endodontic origin, especially when associated with teeth with pulp necrosis or that were previously treated endodontically, leading to misdiagnosis and an ineffective therapeutic protocol [27,28].

When in the mandible, osteoid osteoma has been described as unifocal radiolucent lesion with prominent borders containing radio-opaque foci inside. Sclerotic borders and sclerotic bone reaction are the typical features of osteoid osteoma. (29].

The radiographic aspect consists of a lytic, small and round area in the subjacent cortical bone surrounded by sclerotic bone [21].

Endodontic treatment is a treatment of choice for periapical inflammatory lesions. Taking into account the cystic nature of the lesion, it's size and the need for histopathology we proceeded with periapical surgery. The histopathology report determined the diagnosis of osteoid osteoma in absence of any symptoms or image findings archetypal for osteoid osteoma.

Surgical excision is the treatment of choice and could include the affected teeth. In general, the prognosis of this condition is favorable, and recurrence is rare after surgical treatment [12]. Osteoid osteomas do not cause resorption or displacement of teeth [30].

It can cause cortical perforation at advanced stage[9]. Recurrence depends upon the complete removal. Although malignant transformation is very rare, one case of an osteoid osteoma has been reported in literature, which transformed into an aggressive (low grade) osteoblastoma[31].

Conclusion

The presented rare case demonstrated that osteoid osteoma in mandible can mimic a periapical inflammatory lesion. Surgical removal and histology contributed to proper diagnostics. Complete surgical removal prevented recurrence.

References

1. H. Jaffe, "A benign osteoblastic tumor composed of osteoid and atypical bone," Archives of Surgery, vol. 31, pp. 701–728, 1935.
2. S. Karandikar, G. Thakur, M. K. S. Tijare, K. Agrawal, "Osteoid osteoma of mandible," BMJ Case Reports, vol. 2011, no. dec 05, 2011. bcr.10.2011.4886.indd (nih.gov)
3. C. Walia, P. Devi, V. B. Thimmarasa, and S. Jayadev, "Osteoid osteoma of the mandible: a rare entity," Journal of Indian Academy of Oral Medicine and Radiology, vol. 22, no. 3, pp. 162–164, 2010.
4. Manjunatha BS, Nagarajappa D. Osteoid Osteoma. Indian J Dent Res. 2009;20(4):514–5. Osteoid osteoma Manjunatha B S, Nagarajappa D - Indian J Dent Res (ijdr.in)
5. Boscainos PJ, Cousins GR, Kulshreshtha R, Oliver TB, Papagelopoulos PJ. Osteoid osteoma. Orthopedics. 2013;36:792-800. http://m2.wyanokecdn.com/512102fb8ed227a778fd0e03c13cd979.pdf
6. Dorfman HD, Czerniak B, editors. Bone Tumours. St. Louis, MO: Mosby; 1998. Benign osteoblastic tumours; pp. 85–127.
7. Atesok KI, Alman BA, Schemitsch EH, Peyser A, Mankin H: Osteoid osteoma and osteoblastoma. J Am Acad Orthop Surg 19(11): 678-689, 2011. https://pubmed.ncbi.nlm.nih.gov/22052644/
8. 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(5):639-50.
9. B. Rahsepar, A. Nikgoo, and S. A. Fatemitabar, "Osteoid osteoma of subcondylar region: case report and review of the literature," Journal of Oral and Maxillofacial Surgery, vol. 67, no. 4, pp. 888–893, 2009.
10. E. O. Kirwan, P. A. Hutton, J. L. Pozo, and A. O. Ransford, "Osteoid osteoma and benign osteoblastoma of the spine. Clinical presentation and treatment," Journal of Bone and Joint Surgery, vol. 66, pp. 21–26, 1984.
11. E. J. Gardner and R. C. Richards, "Multiple cutaneous and subcutaneous lesions occurring simultaneously with hereditary polyposis and osteomatosis," American Journal of Human Genetics, vol. 5, p. 139, 1953.
12. Mohammed I, Jannan NA, Elrmali A. Osteoid osteoma associated with the teeth: unusual presentation. Int J Oral Maxillofac Surg. 2013;42(2):298-302.
13. Shafer WG, Hine MK, Levy BM. A textbook of Oral Pathology, 4th Edition, WB Saunders Company, Philadelphia: Pennsylvania; 1993. p. 164-5.
14. Zulian MA,Vincent SK, Hiatt WB. Osteoid osteoma of the mandibular ramus. J Oral Maxillofac Surg. 1987; 45: 712.
15. Shatz A, Calederon S, Mintz S. Benign osteoblastoma of the mandible. Oral Surg Oral Med Oral Pathol. 1986; 61: 189.
16. Mirra JM, Gold RH, Picci P. Osteoid Osteoma. In: Mirra JM, ed. Bone Tumors. Philadelphia, PA: Lea Febiger; 1989:226–48.
17. Schajowicz F. Bone-forming tumours 1.1.2 Osteoid osteoma and osteoblastoma. In: Schajowicz F, ed. Histological Typing of Bone Tumours. Second Edition Berlin: Springer-Verlag; 1993:7–9.
18. Resnick DL, Kyriakos M, Greenway G. Tumors and tumor- like lesions of bone: imaging and pathology of specific lesions. In: Resnick DL, ed. Diagnosis of Bone and Joint Disorders. Third Edition Philadelphia, PA: Saunders; 1995:3629–47.
19. Ayala AG, Ro JY, Raymond AK. Bone tumors. In: Damjanov I, Linder J, eds. Anderson's Pathology. Tenth Edition St Louis: Mosby; 1996:2531–73.
20. Neville BW, Damm DD, Allen CM, Chi AC. Oral and maxillofacial pathology. 4th ed. St. Louis: Elsevier; 2016.
21. Liu CJ, Chang KW, Chang KM, Cheng CY. A variant of osteoid osteoma of the mandible: report of a case. J Oral Maxillofac Surg. 2002;60(2):219-21. v1a19.pdf (gn1.link)
22. 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.
23. Nair P.N. Pathogenesis of apical periodontitis and the causes of endodontic failures. Rev Oral Biol Med. 2004; 15: 348-381. Pathogenesis of Apical Periodontitis and the Causes of Endodontic Failures - P.N.R. Nair, 2004 (sagepub.com)
24. Oliveira L.D.Carvalho C.A.Carvalho A.S.et al. Efficacy of endodontic treatment for endotoxin reduction in primarily infected root canals and evaluation of cytotoxic effects. J Endod. 2012; 38: 1053-1057.
25. Faitaroni L.A.Bueno M.R. De Carvalhosa A.A. et al. Ameloblastoma suggesting large apical periodontitis. J Endod. 2008; 34: 216-219.
26. Nair PNR. Biology and pathology of apical periodontitis. In: Estrela C. Endodontic science. 2nd ed. São Paulo: Artes Médicas; 2009. p. 285-347.
27. de Moraes Ramos-Perez F.M. Soares U.N. Silva-Sousa Y.T. et al. Ossifying fibroma misdiagnosed as chronic apical periodontitis. J Endod. 2010; 36: 546-548.
28. Rodrigues C.D.Estrela C. Traumatic bone cyst suggestive of large apical periodontitis. J Endod. 2008; 34: 484-489.
29. White SC, Pharoah MJ. Oral radiology principles and interpretation. 6th ed., St. Louis, Mosby; 2009.

30. A Singh and MC Solomon. Osteoid osteoma of the mandible: A case report with review of the literature. J Dent Sci. 2017 Jun; 12(2): 185–189. Osteoid osteoma of the mandible: A case report with review of the literature (nih.gov)
31. Pieterse A.S., Vernon-Roberts B., Paterson D.C., Cornish B.L., Lewis P.R. Osteoid osteoma transforming to aggressive (low grade malignant) osteoblastoma: a case report and literature review. Histopathology. 2007;7:789–800.