SUBSCRIPTION INFORMATION

Journal of Oral and Maxillo Facial Pathology, the official publication of Indian Association of Oral and Maxillofacial Pathologists is published half yearly. All the members of the association are entitled to receive a free copy of the journal. The subscription rates for others are as follows:

Non IAMOFP members (within India)	Rs. 300
Institutions and libraries (Within India)	Rs. 500
Non IAMOFP members (Outside India)	US$ 30
Institutions and libraries (Outside India)	US$ 50

All subscriptions are to be made in the form of demand draft in favour of "Journal of Oral and Maxillo Facial Pathology" drawn on any nationalized bank, payable at Chennai. This is not applicable to foreign subscriptions.

EDITORIAL OFFICE
Department of Oral and Maxillo Facial Pathology,
Meenakshi Ammal Dental College and Hospital,
Alapakkam main road, Maduravoyal,
Chennai - 600 095, Tamilnadu, India
Tel: 91-44-2378 2552 (Direct), 91-44-2378 2566, ext. 125 (College)
Fax: 91-44-2378 1631
E-mail: jomfp@yahoo.co.in, drsivapatham@yahoo.co.in

Website: www.jomfp.in

JOMFP

JOURNAL OF ORAL AND MAXILLO FACIAL PATHOLOGY

Vol 11 **Issue 2** **July - December 2007**

PUBLICATION OFFICE
Medknow Publications, A-108/109 Kanara Business Centre, Ghatkopar (E), Mumbai - 400075, India.
Phone: 91-22-6649 1818 / 1816, E-mail: publishing@medknow.com, Web: www.medknow.com

CASE REPORT

Osteoid osteoma of mandible

Mayur Chaudhary, Meena Kulkarni
Department of Oral and Maxillofacial Pathology, Dr. D.Y. Patil Dental College and Hospital, Pimpri, Pune - 411 018, Maharashtra, India

Correspondence: *Dr. Mayur Chaudhary, Department of Oral and Maxillofacial Pathology, Dr. D.Y. Patil Dental College and Hospital, Pimpri, Pune - 411 018, Maharashtra, India.*
E-mail: drmayur111@yahoo.co.in

ABSTRACT
Osteoma are benign osteogenic lesions characterized by proliferation of either cancellous or compact bone and can be central, peripheral or extraskeletal. They may arise in medullary (endosteal) bone or on the bone surface as a polypoid or sessile mass (periosteal). The most common site is in the skull. When affecting the facial bones, they are frequently found in the mandible, the most common locations being the posterior lingual surface and the mandible angle area. Here, a case of osteoid osteoma of the mandible in a 43-year-old female patient is presented with a literature review.
Key words: Osteoid osteoma, mandible

INTRODUCTION

Osteoid osteoma is a distinct benign entity. It has a nidus less than 2 cm in diameter composed either of immature osteoid, woven bone or a mixture of both. The nidus causes considerable pain and can often provoke reactive sclerosis in contagious tissue of the host bone. Pain is very characteristic of this lesion and is accompanied by vasomotor disturbances, which occur long before characteristic radiographic and histopathology findings become evident. The lesion occurs predominantly in children, adolescents and young adults between 10 and 25 years of age. It is distinctly rare in patients aged more than 30 years.[1] We report a rare case of osteoid osteoma in a 43-year-old female in mandibular first molar area.

CASE REPORT

A 43-year-old female reported with pain in the lower left posterior region of the jaw since three days. Pain was dull and continuous in nature and aggravated on applying pressure on the lower left posterior teeth. The patient also gave a history of swelling in that area since six months. She also felt a loss of taste sensation from the anterior part of tongue. There was no relevant medical history. Extraoral examination did not reveal any swelling, but left submandibular lymph nodes were tender on palpation. Intraoral examination revealed tenderness on vertical percussion in 35, 36 and 37. There was no clinical mobility of the involved teeth nor were there any periodontal problems, wasting diseases, fracture of the involved teeth or any evidence of caries. Intraoral periapical radiograph of 35, 36, 37 region and orthopantomogram revealed a well-defined small oval-to-round radiolucency surrounded by well-defined corticated border in association with 36. Irregular radiopacity was seen within the radiolucency in the center of the lesion, and it appeared that it was attached to the apical one-third of the Distal root of 36 [Figures 1 and 2].

Based on clinical and radiographic findings, benign cementoblastoma, cystic odontome, cementifying /ossifying fibroma, sclerosing osteitis, periapical cemental dysplasia were considered in the differential diagnosis. Surgical enucleation of the lesion was done as a part of the treatment [Figure 3]. Histopathology of the lesional tissue revealed islands of mature bone containing osteocytes within the lacunae. Osteoblastic rimming was also seen. Scanty connective tissue stroma showed numerous fibroblasts, collagen fibers and vascular spaces [Figure 4]. The postoperative healing was uneventful.

DISCUSSION

Osteoid osteoma was described as a specific entity by Jaffe in 1935, and since then hundreds of cases have been published which bear out his original criteria: (1) the lesion is a benign neoplasm; (2) it formed large amounts of osteoid which became calcified; (3) there was little evidence to suggest that the lesion was an inflammatory process; (4) there were characteristic X ray changes, such as focal rarefaction and reactive bone, which appeared some distance from the lesion; (5) the lesion occurred most frequently in young adults; (6) pain is an outstanding feature and (7) complete removal is the treatment of choice.[2]

Jaffe described a type of nidus which appeared as a hard osseous core composed of densely set trabeculae of newly formed bone which was atypical.[3] In review by Greene GW *et al.*,[4] cases involving the jaws revealed a type of nidus, more brittle in nature, in which the osteoid tissue predominated. Lind and Hillerstrom classified these two types of nidi and observed that

Figure 1: Intraoral periapical radiograph of 35, 36, 37 region showing an irregular radiopacity with a radiolucent rim

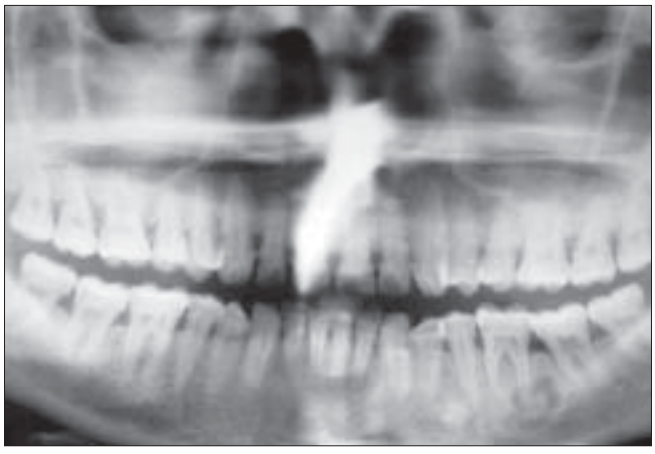

Figure 2: Orthopantomograph region showing an irregular radiopacity attached to the distal root of 36 and surrounded by a radiolucent rim

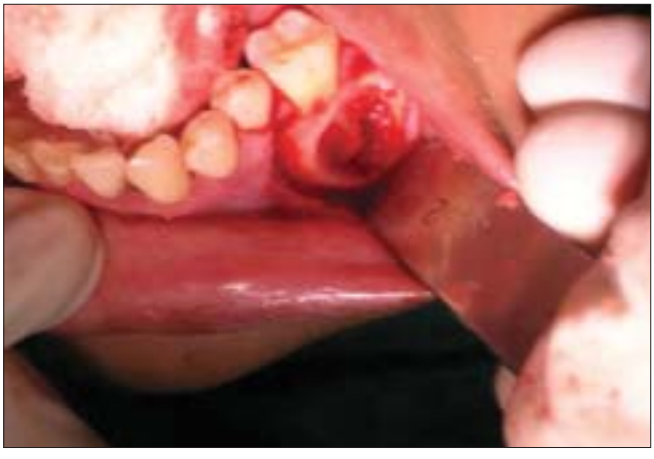

Figure 3: Surgical site showing the mass attached to the distal root of 36

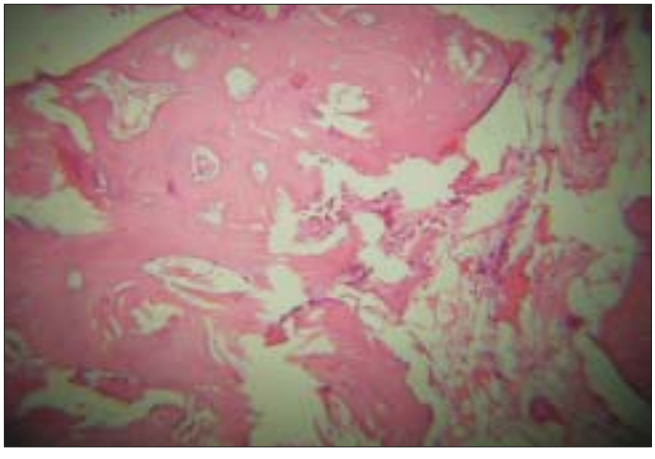

Figure 4: Microphotograph showing islands of mature bone with osteoblastic rimming

the latter type exhibited active and productive osteogenesis.[5] Lichtenstein related that microscopically, a broken nidus may be mistaken for granulation tissue and that the older lesion showed atypical bone modeled from sheets of osteoid trabeculae.[6]

Jaffe perceived the initial notable changes in this lesion as an increased vascularization and destruction with replacement by new atypical bone following resorption of the destroyed tissue. The stroma consisted of osteogenic connective tissue containing numerous blood channels. A cortical lesion which produced this bony replacement stimulated the overlying periosteum to lay down new bone of fairly normal architecture.[3]

Some have suggested that the lesion represented developmental aberrations of embryologic anlage. Others have observed that the histologic features of inflammation were associated with lesions that they studied.[7,8] Pines and associates observed inflammatory changes in a percentage of their cases.[9] The solitary nature of the lesion, the lack of cardinal signs of inflammation and the absence of systemic manifestations were seen by Sherman. In her study, negative cultures were obtained in 19 of the 22 cases.[10] Flaherty, Pugh and Dockerty obtained similar results in their culture studies of this lesion.[11]

The most common symptom of osteoid osteoma was pain; and descriptions of its quality, duration and frequency were consistent throughout several large studies.[12-15] Golding believed that the marked vascular elements were responsible for both the pain and the osseous reaction common to the lesion.[16] Jaffe regarded the curious pain seen in osteoid osteoma as being attributable to the arteriolar blood supply to the lesion.[3] Sherman and McFarland found that the blood vessels in the reactive fibrous zone surrounding the nidus were accompanied by non-myelinated nerve fibers and postulated that these might be the cause of pathognomonic pain occurring in cases of lesions of this type.[17] Byers described several such lesions where axonal fibers occurred singly or in pairs and coursed irregularly through the stroma.[18] Schulman and Dorfman postulated further that the pain was generated and transmitted by autonomic nerves sensitive to vascular pressure.[19] Localized swelling and

tenderness were common clinical findings of this lesion. The pain was often dull and boring and frequently became worse at night. The pain was also described as being referred to a nearby joint; and in one study, this occurred in 27 of the 80 cases.[13]

Majority of the patients found were in the first two decades of life. Freiberger and associates found that 60 of the 80 patients were between the ages of 5 and 20 years, the youngest being 17 months and the oldest, 56 years.[13] Rushton and co-workers found that in 62 patients, 43 lesions occurred before the age of 21; the youngest patient was 21 months and the oldest, 58 years of age. Males were affected between two and three times as often as females.[15]

Jaffe emphasized that the roentgenographic features of the osteoid osteoma were most important in the definitive diagnosis of the lesion. He stated that the nidus was more radiolucent than radiopaque and that it was surrounded by a reactive radiopacity that extended a variable distance from the nidus. The less mature lesion was more likely to have radiopaque nidus, whereas the fully mature osteoid osteoma had a radiolucent nidus.[3] Prichard and Mckay reported that calcification of osteoid in the later developmental stages, deposited on calcified trabeculae, resulted in a central opaque body which varied in density as the calcification progressed.[14] Stafne described the roentgenographic features as a radiopaque nidus surrounded by an area of dense bone.[20] Foss, Dockerty and Good also stated that calcification of the radiolucent nidus occurred.[21]

Huvos distinguishes between three distinct evolutionary stages of modification. The initial stage is characterized by the presence of actively proliferating, densely packed prominent osteoblasts in a highly vascularized stroma. In the intermediate phase, the osteoid is deposited between the osteoblasts. In the mature stage of the lesion, the osteoid is transformed into well-calcified, compact trabeculae of atypical bone, which are histologically peculiar since they are neither typically woven nor typically lamellar.[1] Ultrastructural investigation of five cases of osteoid osteoma by Steiner revealed the morphology of the osteoblasts similar to that of normal osteoblasts, although atypical mitochondria can be seen. For comparison, the osteoblasts of benign osteoblastoma were studied and were found to be identical with those of osteoid osteoma. He concluded that both these lesions are closely related.[22]

Differential diagnosis of osteoid osteoma includes endostosis, intracortical bone abscess, sclerosing forms of osteomyelitis, condensing osteitis, benign osteoblastoma and early stages of Ewing's tumor.[4]

There is general agreement in the literature that the treatment of choice for osteoid osteoma is the complete removal of the nidus. Several instances of regression of the untreated lesion have been recorded in literature. The symptoms regressed following episodes of pain ranging in duration from two to nine years.

Coley related a case in which osteogenic sarcoma developed nine years after X-ray therapy.[23] Gold described a case of osteoid osteoma which recurred twice and subsequently resulted in mandibular resection.[24]

CONCLUSION

The lack of knowledge concerning the genesis of osteoid osteoma and its confusion with similar lesions in the bone make accurate compilation of data concerning the lesion a difficult problem. It is obvious that the small number of reported cases of osteoid osteoma in maxilla and mandible prohibits any lasting deductions concerning this lesion's behavior in the jaws. It is not unreasonable to assume that its occurrence in these areas is more common than the literature would indicate, and it is hoped that the dentists' awareness of it will result in additional cases being reported in the literature.

REFERENCES

1. Makek M. Osteoid osteoma. *In*: Krager S, editor. Clinical Pathology of Fibro-Osteo-Cemental lesions in the Cranio-Facial and Jaw bones: A new approach to differential diagnosis. Basel: Switzerland; 1983. p. 78-83.
2. Jaffe HL. Osteoid osteoma. Arch Surg 1935;31:709-28.
3. Jaffe HL. Tumors and Tumorous conditions of the bones and joints. Lea and Febiger: Philadelphia; 1958. p. 92-105.
4. Greene GW Jr, Natiella JR, Spring PN Jr. Osteoid osteoma of jaws. Oral Surg Oral Med Oral Pathol 1968;26:342-51.
5. Lind PO, Hillerstrom K. Osteoid osteoma in the mandibular condyle: Case report and survey of the literature. Acta Otolaryngol 1964;57:467-74.
6. Lichtenstein L. Bone tumor. 2nd ed. The C.V. Mosby Company: St. Louis; 1959. p. 84-108.
7. Brailsford JF. Chronic Subperiosteal abscess. Br J Radiol 1942;15:313-7.
8. Brown RC, Ghormley RK. Solitary cortical abscess in bone. Surgery 1943;14:541-53.
9. Pines B, Lavine L, Grayzel DM. Osteoid osteoma. J Int Coll Surg 1950;13:249-77.
10. Sherman MS. Osteoid osteoma. J Bone Joint Surg 1947; 29:918.
11. Dockerty MB, Flaherty RA, Pugh DG. Osteoid osteoma. Am J Roentgenol Radium Ther Nucl Med 1956;76:1041-51.
12. Dahlin DC. Bone tumors. 2nd ed. Thomas CC, editor. Springfield, Ill; 1967.
13. Freiberger RH, Loitman BS, Helpern M, Thompson TC. Osteoid osteoma: A report on 80 cases. Am J Roentgenol Radium Ther Nucl Med 1959;82:194-205.
14. Prichard JE, McKay JW. Osteoid osteoma. Canad MAJ 1948;58:567.
15. Rushton JG, Mulder DW, Lipscomb PR. Neurologic symptoms with Osteoid osteoma. Neurology 1955;5:794-7.
16. Golding JS. The natural history of Osteoid osteoma: With a report of twenty cases. J Bone Joint Surg Br 1954;36:218-29.
17. Sherman MS, McFarland G Jr. Mechanism of pain in Osteoid osteomas. South Med J 1965;58:163-6.
18. Byers PD. Solitary benign osteoblastic lesions of bone: Osteoid osteoma and benign osteoblastoma. Cancer 1968;22:43-57.
19. Schulman L, Dorfman HD. Nerve fibres in Osteoid osteoma.

J Bone Joint Surg Am 1970;52:1351-6.
20. Stafne EC. Oral roentgenographic diagnosis. W.B. Saunders Company: Philadelphia; 1959. p. 192.
21. Foss EL, Dockerty MB, Good CA. Osteoid osteoma of the mandible: Report of a case. Cancer 1955;8:592-4.
22. Steiner GC. Ultrastructure of osteoblastoma. Cancer 1977;39: 2127-36.
23. Coley BL. Neoplasms of bone and related conditions. *In*: Hoeber PB, editor. 2nd ed. New York, Inc; 1960. p. 102.
24. Gold L. Classification and pathogenesis of fibrous dysplasia of jaws. Oral Surg Oral Med Oral Pathol 1955;8:628-38,725-37, 856-61.

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