Osteoid Osteoma of the Mandible: A Rare Entity

Cherry Walia, Parvathi Devi, Thimmarasa VB, Jayadev S

Postgraduate Student, Department of Oral Medicine and Radiology, Rama Dental College, Hospital and Research Center, Kanpur, Uttar Pradesh, India
Professor and Head, Department of Oral Medicine and Radiology, Rama Dental College, Hospital and Research Center, Kanpur, Uttar Pradesh, India
Professor, Department of Oral Medicine and Radiology, Rama Dental College, Hospital and Research Center, Kanpur, Uttar Pradesh, India
Reader, Department of Oral Medicine and Radiology, Rama Dental College, Hospital and Research Center, Kanpur, Uttar Pradesh, India

Correspondence: Cherry Walia, Department of Oral Medicine and Radiology, Rama Dental College, Hospital and Research Center, A 1/8, Lakhanpur, Kanpur-208024, Uttar Pradesh, India, e-mail: drcherry2004@yahoo.co.in

ABSTRACT

Osteoid osteoma is a benign tumor of the bone which has seldom been described in the jaws. It was first described as a distinct clinical entity by Jaffe in 1935. Lichtenstein defined osteoid osteoma as a "small, oval or roundish tumor like nidus which is composed of osteoid and trabeculae of newly formed bone deposited within a substratum of highly vascularized osteogenic connective tissue." The most interesting clinical feature of osteoid osteoma is the exquisite pain produced by a very small lesion, never greater than one centimeter in diameter. It accounts for 3% of all primary bone tumors, and about 10% of benign bone tumors. About 80% of cases of osteoid osteoma occur in long bones, while less than 1% occur in jaws.

We report a case of an 18-year-old male patient with osteoid osteoma of right side of the mandible.

Keywords: Nidus, Osteoid osteoma, Pain, Radiopacity.

INTRODUCTION

Osteoid osteoma was first described as a distinct clinical entity by Jaffe in 1935. It is a benign tumor of bone which has seldom been described in jaws. Jaffe and Lichtenstein have suggested that the lesion is a true neoplasm of osteoblastic derivation, but other workers have reported that lesion occurs as a result of trauma or inflammation. Jaffe refers to the osteoid osteoma as “sui genris”, denoting the lesion’s small, self-limiting nature. The exquisite pain produced by a very small lesion, never greater than one centimeter in diameter, is the most interesting clinical feature of osteoid osteoma. This pain is usually dull, but severe enough to interfere with the patient’s sleep causing considerable discomfort. Green et al reviewed the literature and reported the total number of cases of osteoid osteoma of the jaws to be seven, of these four have occurred in the mandible and three in the maxilla (one in the antrum). We report a rare case of osteoid osteoma in the mandible of an 18-year-old male patient.

CASE REPORT

An 18-year-old male patient reported to the Department of Oral Medicine and Radiology with a chief complaint of severe pain on the right side of the lower jaw since two and a half months. Pain was sharp, continuous and was relieved on taking analgesics and recurred after sometime. Patient had also noticed swelling in the same region since one month which had gradually increased in size and later became stable. There was no relevant medical and family history. On general physical examination the patient was moderately built and nourished and all the vital signs were in the normal limit. Extraoral examination revealed a diffused swelling on the right side of the lower jaw extending from the corner of the mouth to 4 cm anterior to the angle of the mandible anteroposteriorly, from the corner of the mouth to 1 cm above the lower border of the mandible superoinferiorly measuring approximately 2 × 1 cm in size. The swelling was firm to hard in consistency and tender on palpation. On intraoral examination, an oval shaped diffused swelling was seen in relation to 44, 45 region with obliteration of the buccal vestibule (Fig. 1). On palpation it was firm to hard in consistency and tender. Considering the history and clinical examination, a provisional diagnosis of benign tumor of the jaw was given with a differential diagnosis of central giant cell granuloma, osteoma, cementoma and cemento ossifying fibroma.

Routine blood investigations were carried out and all the values were in normal limit. 45 was non vital on electric pulp test. IOPA of 44, 45 region (Fig. 2) showed an oval shaped radiopacity extending from distal aspect of 44 to the distal aspect of 45 with deviation of the root of 44 mesially. Radiopacity was surrounded by a radiolucent rim. Mandibular lateral occlusal projection (Fig. 3) showed periosteal new bone formation in relation to 44, 45 region. Panoramic radiograph revealed an oval shaped radiopacity which was surrounded by a thin radiolucent rim.

Surgical resection of the lesion was done along with extraction of 45. Histopathological section (Fig. 4) showed proliferation of lamellar bone. The overlying peristeam exhibited new bone formation and newly formed osteoid tissue was rimmed by osteoblasts, in between fibrovascular tissue was seen. Few multinucleated giant cells were also present. The overall picture was suggestive of osteoid osteoma of the mandible.
The postoperative course was uneventful and the patient is still being followed up. He was last seen eight months after the excision with no relevant clinical and radiographic finding.

**DISCUSSION**

The osteoid osteoma is characterized clinically as a small but painful bony lesion which has seldom been described in the jaws. Lichtenstein defined osteoid osteoma as “a small, oval, or roundish tumor like nidus which is composed of osteoid and trabaculae of newly formed bone deposited within a substratum of highly vascularized osteogenic connective tissue.” The tumor itself, often referred to as a nidus, is extraordinarily small considering the mischief it causes. It usually occurs in young persons, seldom developing after the age of 30 years. Young children under the age of five years are also frequently affected. Males are affected twice as often as females. It is generally located in the tibia or the femur (50 percent) but it may involve all bones. Other common sites include the fibula, humerus and vertebral arch. Even the skull may be involved. Jaffe (1935) first described the lesion as a distinct clinical entity and set the criteria for osteoid osteoma. According to him (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 radiographic changes, such as focal rarefaction and reactive bone formation, 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.

The true nature of the lesion is unknown but some investigators think it as a variant of osteoblastoma. Various theories have been given relating to the nature and genesis of the osteoid osteoma. Jaffe as already mentioned considered it as a benign tumor. He observed that particularly in its latest stages of development the osteoid osteoma manifests histological patterns of a pronounced neoplastic lesion. Bergstrand (1930) thought that the lesion was of embryonal nature and considered it as a hamartoma. Pines et al in 1950 and Lofgren in 1953 described it as an inflammatory lesion as pain which is its most constant feature is more usual in inflammation than on neoplasia.
The clinical hallmark of this lesion is local pain which is dull, throbbing and intermittent, but subsequently it usually increases in severity. Nocturnal pain is common. Point tenderness over the lesion is present along with slight local swelling. Dramatic relief of pain is obtained in most cases with aspirin.\(^5\)

Jaffe regarded the curious pain seen in osteoid osteoma as being attributable to the arteriolar blood supply to the lesion.\(^5\) Golding reported that osteoid osteoma exerts pressure upon the surrounding bone, presumably because of its vascular nature. This evidence of increased pressure suggests that the pain is produced by the highly vascular tumor lying within the confines of the sclerosed bony trabeculae.\(^9\) Sherman et al demonstrated nerve fibers in the fibrous zone around the nidus and implicated them as mediators of pain. Schulman and Dorfman demonstrated nerve fibers within matrix of the nidus in sixteen of eighteen lesions studied. These fibers were associated with blood vessels and were found in greatest abundance adjacent to arterioles. They postulated that neural elements demonstrated are sensitive to changes in vascular pressure.\(^5\) Kolody (1929) and Rowbotham (1939) believed that the vascular lesion puts the bone into a state of metabolic activity, particularly around the venous sinuses draining the tumor, and commented on the extremely slow growth of the lesions. Kolody also stressed the fact that the most massive reactions occur in the young, stating that it is the dilating vascular channels that result in new bone formation.\(^9\)

Osteoid osteoma is classified as cortical, cancellous, or subperiosteal osteoid osteoma. Cortical tumors are the most common. The radiolucent nidus is within the cortical bone, where it is surrounded by a fusiform sclerotic cortical thickening or solid or laminated periosteal new bone formation. Cancellous osteoid osteoma also referred to as medullary osteoid osteoma is intermediate in frequency. In this type osteosclerosis is usually mild to moderate and may be distant from the lesion. Subperiosteal osteoid osteoma is a rare form of the disease that usually presents as a rounded soft-tissue mass adjacent to a bony cortex, which it excavates. Surrounding reactive changes are usually absent in this type.\(^10\)

The classical roentgen appearance of osteoid osteoma is a small, radiolucent intracortical nidus, less than one centimeter in diameter, surrounded by a large, dense sclerotic zone of cortical thickening. Evidence of a laminated periosteal reaction is often present.\(^4\) Jaffe emphasized that the roentgenographic features of 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.\(^8\) Prichard and Mckay reported that a central opaque body which varied in density and that extended a variable distance from the nidus.\(^8\) Prichard and Mckay reported that a central opaque body which varied in density and that extended a variable distance from the nidus.\(^8\) Foss et al also reported that there may be calcification or ossification of a portion or all of the radiolucent nidus.\(^9\)

CT has been suggested to be one of the most useful modalities to diagnose bone tumors in the complex anatomical site of the jaws. The characteristic features of osteoid osteoma such as a radioluency with central calcification, located in the cortical bone with surrounding sclerotic bone are best elucidated by CT. Bone scanning is considered a useful adjunctive test in some cases because the rich bone-producing osteoid osteoma is associated with a “hot” radionuclide bone scan. Radionuclide scanning is a sensitive technique, and findings may be positive before radiographic changes are apparent.\(^11\)

Lindborn et al demonstrated pronounced hypervascularization of the nidus in angiography which is a valuable finding as it helps in differentiating osteoid osteoma from osteomyelitis.\(^6\)

The tumor consists of an ovoid or spherical nidus of osteoid-rich tissue and interconnected bone trabeculae superimposed on a background of highly vascularized connective tissue containing large dilated vascular channels. The amount of osseous and osteoid tissue varies within the nidus and is reflected in its radiopacity. The histological appearance varies with the age of the lesion and its site. There are some irregular calcified trabeculae of new bone lying in the osteoid tissue and in older lesions these are frequently pronounced. In general the younger lesions are more highly vascular.\(^7\) Osteoclasts and foci of bone resorption are also usually evident. Ultrastructural investigations of five cases by Steiner revealed the morphology of osteoblasts to be similar to that of normal osteoblasts although atypical mitochondria could be seen.\(^8\)

Complete excision is considered as the treatment of choice. The surgeon must try to remove the nidus intact; if the entire osteoid osteoma lesion is not removed or destroyed, the clinical complaints will either remain, or disappear and recur at a later date.\(^7\) The tumor has no malignant potential. There is fairly good circumstantial evidence that spontaneous regression may occur in at least some untreated cases.\(^7\)

To conclude osteoid osteoma is a painful benign tumor usually affecting the long bones but can occur in jaws also. The diagnosis of this entity is usually obtained by radiographs and is confirmed by histopathological examination. Surgical excision of the lesion usually renders the patient asymptomatic, provided the nidus is removed thoroughly. Therefore it is recommended that follow-up should be carried out at regular intervals to prevent recurrence.

REFERENCES