

# FOCAL OSTEOPOROTIC BONE MARROW DEFECT: A MYSTIFYING ENTITY WITH SUBTLE LITERATURE

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## ABSTRACT:

Focal osteoporotic bone marrow defect of the jaws has been reported as a poorly demarcated radiolucent area affecting the posterior mandible. It rarely affects the jaws, usually asymptomatic and often confused with other radiolucent lesions of the jaws. This review provides an overview of the literature about focal osteoporotic bone marrow defect which will help the oral physician and clinician for the early and rightful diagnosis of this perplexing entity.

**Key-words:** Bone marrow, bone marrow disease, osteoporosis, mandible, osteoporotic bone marrow defect, ill-defined radiolucencies of jaws



## INTRODUCTION:

Focal osteoporotic bone marrow defect has been described in literature for more than 70 years.<sup>[1]</sup> It is a rare condition of the jaws discovered during routine radiographs. The condition has been reported since 1954, by Cahn, who described variations of the normal location of bone marrow within the jaws present as focal defects.<sup>[2]</sup>

In adulthood, foci of hematopoietic marrow that persist in certain region of the jaws, is usually restricted to the condylar process, angle of the mandible, and the maxillary tuberosity.<sup>[2,3]</sup> Focal osteoporotic bone marrow defect is a radiolucent area corresponding to the presence of hematopoietic tissue within the jaws.<sup>[4]</sup>

The condition is seldom included in the differential diagnosis for focal, ill-defined radiolucent areas of the jaws. Clinicians should be aware of the existence of this abnormality and understand that it

appears radiographically similar to other lesions, some of which are malignant; therefore a biopsy and histopathological analysis is necessary to establish a prompt diagnosis. The knowledge of the accurate clinical features with characteristic radiographic and histopathological findings of the condition, in association with an proper examination protocol are mandatory to distinguish it from other most common intrabony lesions such as odontogenic tumors or cysts, pseudocysts or primary or metastatic malignancies.

This paper is an attempt to critically review the literature in order to aware and update the oral physician and clinicians for this rare condition, so that the misdiagnosis and unnecessary treatments will be avoided.

**METHOD:** A web-based search for all types of articles published was initiated using PubMed, with the keywords such

as focal osteoporotic bone marrow defect, ill-defined radiolucent areas of the jaws. The search was subsequently refined to focal osteoporotic bone marrow defect. The sites of specialized scientific journals in the areas of oral and maxillofacial surgery, oral medicine, and oral pathology were also used.

## ETIOPATHOGENESIS

The etiology of the focal osteoporotic bone marrow defect is still unknown; but three major theories have been proposed regarding the etiopathogenesis of osteoporotic bone marrow defect of the jaws.

First, it was believed that the persistence of red embryonic marrow remnants in adult life within specific areas of jaws, that had no conversion to fatty marrow might lead to focal osteoporotic bone marrow defect.<sup>[5,6,7]</sup> In newborns, the marrow cavities in all the bones contain active hematopoietic tissue, known as red bone marrow. From the early postnatal period onwards, the hematopoietic tissue, mainly in the bones of the extremities, is gradually replaced by non-hematopoietic mesenchymal cells that accumulate lipid drops, known as yellow bone marrow.<sup>[8]</sup> The persistence of the red bone marrow in some particular bones might lead to a well-known “focal osteoporotic bone marrow defect”.<sup>[7]</sup>

Second, it was proposed that the compensatory bone marrow hyperplasia that occur secondary to increased functional demand for erythrocytes, for

e.g. in systemic diseases like sickle cell anemia may leads to bone resorption resulting in focal osteoporotic bone marrow defect.<sup>[5,6,7,9]</sup>

And third, alteration in bone repair phenomenon in areas of previous trauma such as a tooth extraction,<sup>[5]</sup> results in transient ischemia which induces the osteoporotic bone marrow defects.<sup>[10]</sup> Shankland and Bouquot<sup>[10]</sup> proposed that impaired blood flow due to the several factors, including trauma, is responsible for the most cases of the focal osteoporotic bone marrow defect and classified this condition as an ischemic jawbone lesion.

## CLINICAL FEATURES

Osteoporotic defects are predominantly diagnosed in middle-aged females;<sup>[10,11,12]</sup> most often between the fourth and sixth decades of life.<sup>[7]</sup> The most common location is the mandibular posterior region<sup>[3,13]</sup> and approximately 5% of the cases are diagnosed in maxilla.<sup>[2]</sup> From 72% to 91 % occur in the premolar and molar region of the mandible.<sup>[3,13]</sup> The condition frequently occurs in an edentulous region where tooth extraction was previously performed.<sup>[2,10]</sup>

It is rarely related to supernumerary teeth.<sup>[1,5,10]</sup> Chiang C-P et al<sup>[14]</sup> reported a case of focal osteoporotic bone marrow defect associated with two supernumerary teeth in the right posterior maxilla of a young woman.

In the literature, few cases of the focal osteoporotic bone marrow defect associated with dental implants has been described.<sup>[4,15]</sup> Sencimen et al<sup>[15]</sup> reported a clinical case in which the focal osteoporotic bone marrow defect occurring secondary to dental implant placement in posterior region of mandible, two years postoperatively apical to a dental implant and the diagnosis was established based on the combination of clinical, radiographic, and microscopic features. Lee et al<sup>[4]</sup> reported three clinical cases of displacement of dental implants into the focal osteoporotic bone marrow defect but the final diagnoses of this condition were based only on clinical and radiographic findings.

This condition is generally asymptomatic and discovered incidentally during routine radiographic examination of the jaws.<sup>[6,16,17]</sup> However, in rare instances, this condition may be accompanied by pain or swelling.<sup>[18]</sup> Some authors indicates the presence of pain and swelling in approximately 62–65% of the patients.<sup>[10,13]</sup> It rarely show cortical plate expansion.<sup>[2,16]</sup>

Clinically, the focal osteoporotic bone marrow defect occurs as isolated or multifocal radiolucency with several millimeters to centimeters in diameter and ill-defined borders.<sup>[6]</sup> Bilateral mandibular involvement of the osteoporotic bone marrow is not frequently found in the literature.<sup>[2,9,19,20]</sup> Of the 20 cases of osteoporotic bone marrow defect described by Makek and

Lello<sup>[2]</sup> only one was bilaterally located in the molar areas of the mandible. According to study conducted by Bouquot and collaborators<sup>[19]</sup> based in the literature review and report of 596 new cases, the bilateral occurrence of osteoporotic bone marrow defect within the jaws affected 3 % of patients.

## **RADIOGRAPHIC AND HISTOPATHOLOGIC FEATURES**

Radiographic appearance of focal osteoporotic bone marrow defect is generally described as a sharply defined radiolucency with distinct sclerotic borders to extremely ill-defined areas. Trabeculations were occasionally visible within the radiolucent areas.<sup>[3,5]</sup> Many are cast shadows over or near the apices of teeth, but lamina duras are intact, and teeth test vital.

Microscopically, the presence of hematopoietic marrow composed of monocytic, erythroid, granulocytic, and lymphocytic series as well as megakaryocytes associated with fatty marrow is required for diagnosis of focal osteoporotic bone marrow defect.<sup>[4,6]</sup>

## **DIFFERENTIAL DIAGNOSIS**

The differential diagnosis of the focal osteoporotic bone marrow defect includes various intraosseous lesions such as odontogenic tumors or cysts, pseudocysts like aneurysmal bone cyst or traumatic bone cyst and primary or metastatic malignancies.<sup>[21]</sup>

The bilateral radiolucent areas with indistinct margins suggested the presence of the mandibular benign cysts or tumors. Based on age, site, clinical and radiographic findings, the osteoporotic bone marrow defect was considered as a differential diagnosis. The final diagnosis of osteoporotic bone marrow defect should be established based on microscopic features rather than clinical parameters.<sup>[5,10]</sup>

Traumatic bone cyst of the jaws is usually an asymptomatic cystic lesions detected incidentally on routine radiographs or during surgery. The lesion is most frequently diagnosed in the second decade of life, causing expansion of the cortical plate (usually buccally) of the jaw bone. A characteristic of traumatic bone cyst which differ it from focal osteoporotic bone marrow defect is the pronounced scalloping of the endosteal surface of either the buccal or lingual plates (scalloping effect), when extending between the roots of the vital teeth.<sup>[22]</sup> Aneurysmal bone cysts of the jaws, although rare, occur usually below 20 years of age, causing swelling of the overlying soft tissues due to bony expansion<sup>[23]</sup>, and has been known to displace teeth with progressive malocclusion but tooth vitality is not violated (teeth remain vital). Propensity for extreme expansion of the outer cortical plates is the dramatic characteristic in these cysts than in most other lesions.<sup>[24]</sup> Another characteristic of aneurysmal bone cysts is gross findings at the time of operation. Upon entering the lesion, excessive bleeding is

encountered, the blood welling up from the tissue. The tissue has been described as resembling a blood-soaked sponge with large pores representing the cavernous space of the lesion.<sup>[24]</sup> Radiographically, it appears as well-defined radiolucent lesion, usually circular or "hydraulic" in shape, with internal aspect has a multilocular appearance having wispy, ill-defined septa. The bone is expanded in acentrically fashioned.<sup>[24]</sup> There may be destruction or perforation of the cortex and a periosteal reaction may be evident.<sup>[25]</sup> Aneurysmal bone cysts can be classified into three types. Conventional or vascular type (95%), solid type (5%) and mixed variant type. The solid type may present as a small asymptomatic lesion noticed as radiolucency on a routine radiograph or as a small swelling.<sup>[26,27]</sup> In contrast to traumatic bone cyst and aneurysmal bone cysts, the focal osteoporotic bone marrow defect rarely show cortical plate expansion with teeth remains vital having intact lamina dura. Trabeculations are occasionally seen within the radiolucent areas.

The primary or metastatic malignancies usually erode rather than expand the adjacent cortical plates. Consequently detectable expansion is not bony hard but firm on palpation. Also, pain is not a frequent complaint in malignancies, but occasionally it is present in later stage, when the tumor encroaches on sensory nerves within bone. However, the pain is usually of short duration because the tumor rapidly destroys the affected

nerve.<sup>[28]</sup> Metastatic lesions are generally radiolucent having ill-defined invasive margin. Typical of malignancy, the lesion effaces the lamina dura and can cause an irregular widening of the periodontal ligament space. Teeth may seem to be floating in a soft tissue mass and may be in an altered position because of loss of bony support.<sup>[29]</sup> All these features argued against focal osteoporotic bone marrow defect.

### TREATMENT

According to Barker, Jensen, and Howell,<sup>[3]</sup> a radiolucent shadow with distinct or ill-defined margins in the posterior aspect of the body of the mandible of a middle-aged woman suggests an osteoporotic bone marrow defect. If conflicting evidence is not present in the form of worrisome signs or symptoms such as pain or swelling, patient should be kept under

observation and on regular follow ups. Such area should be radiographed at regular intervals to ensure that any change is not occurring within the jaw bones with time. If any change is suspected, surgical exploration and biopsy are carried out.

### CONCLUSION:

In summary, the focal osteoporotic bone marrow defect is a rare condition of the jaws which is usually asymptomatic and discovered during routine radiographic examination. Although rare, it should be included in the differential diagnosis of radiolucent lesions of the jaws. Biopsy and histopathological analysis is particularly necessary for the correct diagnosis of the lesion, avoiding unnecessary treatments. Once the diagnosis has been established after histopathological analysis, no further specific treatment is required.

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