

## Previously unreported: The bone ring – uniformly thick, constantly active, always immature bony wall around an intraosseous hemangioma

### Abstract

**Background:** Hemangiomas within the mandible or maxilla are rare and typically present as radiolucencies with well-demarcated borders. It is rare to have focal or diffuse sclerosis at the border, but we present a case with an extremely unusual and completely unexplained sclerotic ring around such a vascular lesion.

**Case description:** A 24-year-old male presented with a firm, sessile, asymptomatic cortical expansion of his maxillary left premolar region. Radiographs showed a 1.9 mm diameter rounded radiolucency surrounded by a uniformly thickened (2.0 mm) “ring” of diffusely radiopaque tissue with a very thin outer and inner border of more intense sclerosis; a less sclerotic form of this ring was visible in a 12-year-old radiograph. Biopsy showed the central radiolucency to be a cavernous hemangioma, with the surrounding bone ring consisting entirely of immature or newly formed bone with moderate osteoblastic activity and occasional osteoclastic activity. The vascular tumor did not extend into the surrounding ring of bone.

**Conclusion:** We report the first example of a uniformly thick encapsulation of a benign lesion by an independent sphere of radiographically dense and microscopically immature, actively remodeling bone. We suggest that it is an innocuous ring but its pathoetiology and pathophysiology are completely unknown at this point.

**Keywords:** Hemangioma; Central hemangioma; Intraosseous hemangioma; Sclerotic rimming; Sclerotic border; Bony wall; Immature bone; Newly formed bone.

### Introduction

Jawbone radiolucencies with well-demarcated borders typically represent benign cystic or neoplastic processes. Not uncommonly the border will show regular or patchy radiopacities (sclerotic rim, cortical shell) as an apparent response to the expanding pressures from slow lesional enlargement. Such responses occur at the lesional border and are typically very thin or appear as small, somewhat globular radiopacities (Figure 1). This sclerotic rimming is generally seen as evidence of benign behavior of the associated central lesion, with slow, even expansion outwardly rather than invasion through the border [1].

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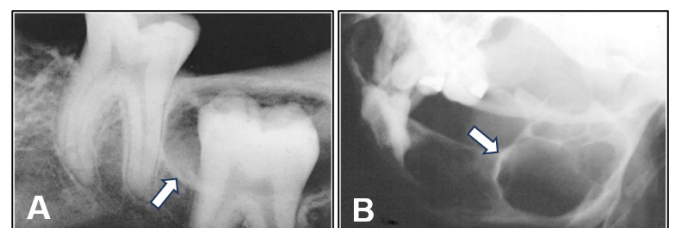
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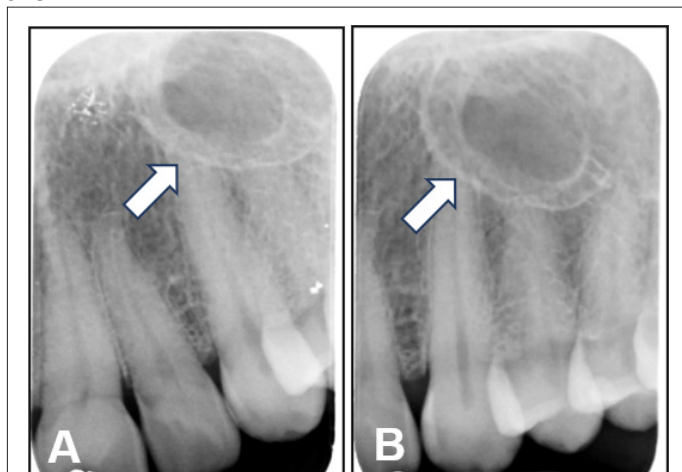


**Figure 1:** Examples of routine sclerotic rimming of radiolucent lesions. (A) Dentigerous cyst with uniformly thin radiopaque line at the border (arrow); (B) Odontogenic keratocyst with sclerotic rimming of only a portion of the border (arrow).

We have observed several examples of a different, previously unreported, type of border sclerosis: a uniformly thick (2.0-2.5 mm), uniformly sclerotic “ring” around benign neoplasms or certain cysts. The bone rings may have thin outer and inner layers of more intense radiopacity and the outer border of the ring, even without this thin layer, has always been, in our experience, as well-demarcated as the inter layer of the ring (the well-demarcated border of the central radiolucency). Of course, the ring is seen only with routine dental radiographs, we assume that it is part of an encasing sphere of bone which covers the lesion entirely, and the single example we have seen via CBCT confirms this. We offer herein the first description of this unusual variant of sclerotic rimming.

### Case description

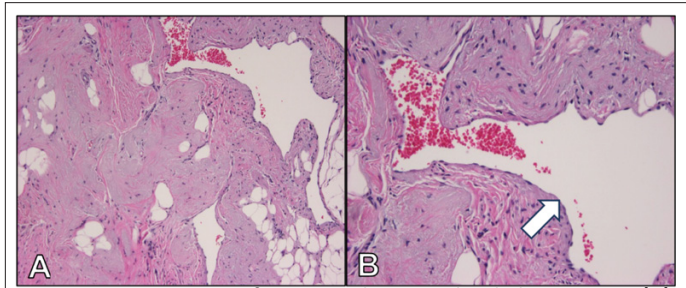
**Clinical/radiographic appearance.** A 24-year-old male presented for his annual dental examination with a firm, sessile, asymptomatic mass of the facial surface of his maxillary left premolar region. The overlying mucosa was normal in appearance and there were no symptoms in the area. Periapical radiographs showed a 1.9 mm diameter rounded radiolucency surrounded by a uniformly thickened (2.0 mm) radiopaque “ring” of diffusely radiopaque tissue (Figure 2). The ring showed very thin outer and inner borders of more intense sclerosis and the central radiolucency had a diffuse and slight gray “haze” to it, similar to the “ghost marrow” of bone marrow edema. The lesion was positioned near the apex of the first premolar but was not otherwise associated with a tooth; all teeth in the area were vital to electric pulp testing. A radiograph from 12 years earlier showed the ring very faintly, although it was the same size.



**Figure 2:** Radiographic appearance of the cortically expanded lesion. (A) A well-demarcated uniformly thick sclerotic ring (arrow) around a well-demarcated oval radiolucencies has very thin layers of more sclerotic tissue at the outer and inner margins (arrow); (B) Same lesion as A, from difference radiological angle.

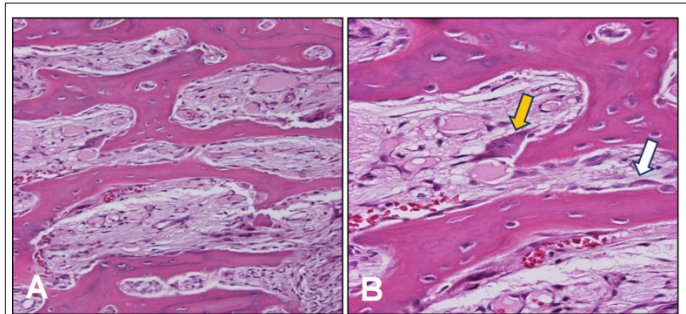
### Treatment and histopathology

During surgical exploration, there was no uncontrolled or extensive hemorrhage and there was no pulsing of the central mass once it was accessed (as would be felt if it was an arteriovenous malformation). The central radiolucency was curetted out of the ring, which was actually a complete sphere around the central lesion. It proved to consist of a background stroma of moderately cellular, somewhat immature fibrous tissue admixed throughout with often dilated veins and scattered adipocytes (Figure 3). Inflammatory cells were not present, nor were dysplastic cells, and bony trabeculae were not noted in the stroma.



**Figure 3:** Appearance of tissue in the central radiolucent area. (A) An immature fibrous stroma contains greatly dilated veins and scattered, normal adipocytes; (B) Higher power view of a dilated vein (arrow) surrounded by immature fibrous tissue.

The central vascular mass shelled out easily; it was not firmly attached to the surrounding bone. After its removal, the bony ring/sphere was surgically curetted out. Microscopically, the ring consisted of very active, immature bone with a somewhat immature fibrous stroma (Figure 4), similar to routine newly formed bone that one would find in an extraction socket 3-6 weeks after the extraction. The bone was formed in parallel layers, like plywood, similar to the layers forming the “onion skinning” radiographic pattern seen outside the bone in Garre’s osteomyelitis (proliferative periostitis).



**Figure 4:** Microscopic appearance of peripheral ring around the central lesion. (A) Immature or newly forming bone with moderate osteoblastic activity and with congestion (dilated vessels) of the background fibrous stroma; (B) Higher power of (A) shows dilated vessels, consistent with congestion, but osteocytes appear normal and several areas show a layer of active osteoblasts lining the immature bone (white arrow), with an active osteoclast also present (yellow arrow).

### Diagnosis

The microscopic appearance of the central mass in this case was that of a cavernous hemangioma, almost certainly a developmental anomaly rather than a true neoplasm. Its childhood onset and lack of radiographic enlargement over the previous dozen years would seem to confirm this, as does the relative immaturity of the background fibrous stroma. Additionally, it did not have the pulsing features of an arteriovenous malformation, nor did it have the excess surgical hemorrhage or the admixture of venous and arterial vessels of that entity. The surrounding bone ring consisted entirely of normal-appearing immature bone with active osteoblasts and occasional osteoclasts. It was not inflamed in any way.

### Discussion

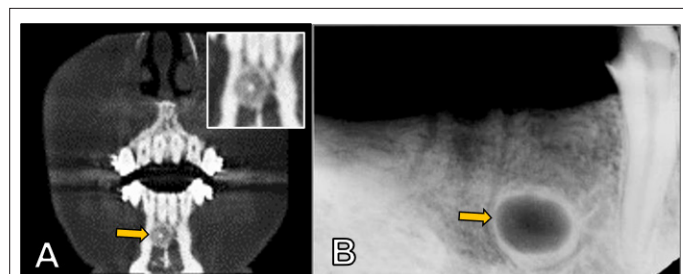
The hemangioma is primarily a childhood tumor, affecting 10-12% of infants, with the H&N region being the most common site of occurrence [2-4]. Hemangioma inside the mandible or maxilla, however, is quite a rare phenomenon, especially in the maxilla [1]. When found in the jaws it is usually in the posterior mandible, usually in females, usually asymptomatic, occasionally causes mild cortical expansion, is usually 2-3 cm

in diameter and it almost always reaches its maximum size by puberty or the early teen years [1,2].

Radiographically, an intraosseous hemangioma is almost always a well-demarcated radiolucency. It can be unilocular or multilocular in pattern, but it may present with one of two relatively unique patterns: 1) A honeycombed pattern, with radiolucent "bubbles" of similar size and shape clustered together and; 2) A central sunburst pattern with thin streaks of bone parallel to lesional vessels radiating outward from a central core (presumably the earliest part of the anomaly). This latter feature, stimulation of bone by the vessels may be pertinent to the present case: perhaps mediators released by the vessels are stimulating the bony ring around the tumor?

It would seem not to be so. There was no bone within the vascular tumor itself, and the bone ring had no bone which followed blood vessels of any type. Moreover, we have seen identical bone rings around a lymphangioma, an osteoblastoma, an odontogenic fibroma, an odontogenic hamartoma and a residual cyst (Figure 5), all presenting as newly forming or actively remodeling bone microscopically [5].

The bone ring, then, does not seem to be specific to a certain type of tissue mass. It is further unusual in being comprised of immature or newly forming bone throughout, with active and apparently continuous remodeling occurring for years. We have previously mentioned the subperiosteal bone of Garre's osteomyelitis, with its constantly immature bone creating the radiographic onion skin pattern. That pathologic bone is created by the periosteum itself, but there appears to be no such structure beneath or above a bone ring. Additionally, the bone in a bone ring is microscopically identical to the immature bone seen throughout the skeleton in osteogenesis imperfecta, but that has an obvious genetic component which does not seem to apply to a local response to expansile pressures.

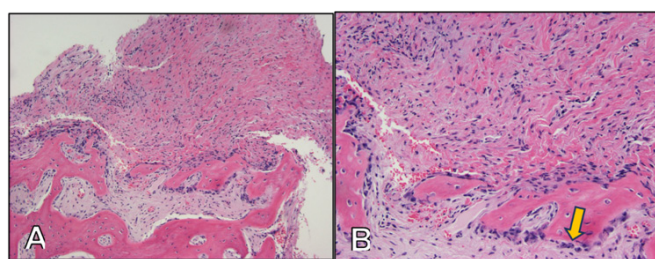


**Figure 5:** Other examples of bone rings. (A) Osteoblastoma of the anterior mandible, with a duration of more than 1.5 years; (B) Residual cyst of right mandible, with a duration of 8 years.

We know of only one entity which is routinely surrounded by immature or newly forming, constantly remodeling bone, although the bone is often not calcified enough to produce a strong, radiographically visible ring similar to a bone ring; 17.5% have at least some border areas of sclerosis [6]. That entity, periapical infection (periapical granuloma, periapical cyst) is one with numerous growth factors and other obvious mediators of tissue production and turnover, hence, has a logical explanation for the production of a surrounding bony barrier [6-8].

In an unpublished investigation (JEB) of more than 50 periapical infections, the surrounding bony encapsulation was immature in all cases, with active osteoblasts and minimal inflammation of the stroma (Figure 6). This bony barrier was similar for each case and seemed effective in keeping the apical infection from spreading, and to be able to do so for months and years.

Such bony encapsulation is almost never seen in inflammatory lesions of extragnathic bones. Acute osteomyelitis in other bones routinely expands by destroying surrounding marrow and bone, with numerous perforations of overlying cortical bone.



**Figure 6:** Representative bony wall surrounding a periapical granuloma of at least 7 months duration. (A) Fibrous periphery of granuloma (top of photo) directly contacts an irregular wall of newly formed bone with mature fibrous stroma; (B) Higher power shows numerous active osteoblasts (arrow) lining immature bone.

### Conclusion

We present a previously unreported and rather mysterious radiographic phenomenon, one for which we have no explanation as to its pathophysiology. This bone ring is comprised of continuously immature and remodeling bone completely surrounding a benign, relatively small soft tissue mass. It seems not to remodel into mature or lamellar bone, even after more than a decade. We suggest that the bone ring has an innocuous biological behavior and can be associated with a variety of benign developmental, neoplastic or cystic phenomena.

### Declarations

**IRB exemption:** The West Virginia University IRB rules exclude individual case reports from their purview.

**Conflict of interest:** None of the authors have conflicts of interest for this research.

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