Case Report

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Osteoid Osteoma of Mandible as an Uncommon Site: A Case Report

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ABSTRACT

Osteoid osteoma is a benign bone-forming tumor. Besides, it is small and painful, consisting of osteoid and woven bone, and surrounded by a thick layer of reactive bone formation. The tumor is self-limited or needs to be removed by surgery. This report describes a 16-year-old male with congenital deafness who presented with a right mandibular swelling initiated 2 years ago. The patient underwent excisional surgery. Histological examinations revealed a benign bone-forming tumor, compatible with osteoid osteoma. Radiologic and histologic correlation confirmed the diagnosis. Base on the diagnosis, the patient received no more treatment. After 4 years of close follow-up examinations, no recurrence was observed in the patient. Osteoid osteoma is often confused with numerous other entities in uncommon sites. Thus, imaging and histologic correlations are required. Proper diagnosis is necessary for further patient management in this respect.

Introduction

Osteoid osteoma is a benign bone-forming tumor; it is small and painful, containing osteoid and woven bone, and delimited by a rim of reactive bone formation [1]. It is single and very infrequently has a multifocal or multicentric presentation [2]. No soft-tissue component was found in any osteoid osteomas [3]. Its etiology is not fully recognized and the role of trauma remains vague [4]. The lesion is most commonly located in the cortex of long bones where it is accompanying by dense, fusiform, and reactive sclerosis [5]. Osteoid osteoma is a benign painful bone tumor that usually originated in the lower extremities of children and young adults [6]. Its occurrence is extraordinary before 5 and after 30 years of age [7]. The lower extremity is much more frequently affected than the upper extremity. In the lower limb, the femur and tibia are involved in approximately 50% to 60% of the cases; however, it is very unusual in the skull

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[8]. When affecting the facial bones, they are regularly set up in the mandible; the most common situations being the posterior lingual surface and the mandible angle region [9].

Clinically, osteoid osteoma causes night pain, i.e. released with Nonsteroidal Anti-Inflammatory Drugs (NSAIDs) [10]. The physical findings of osteoid osteoma differ with the site of the tumor [11]. The clinical exhibition of osteoid osteoma in the jaw differs from that of osteoid osteoma in the more common sites; however, the radiographic features are parallel [12]. The diagnosis is commonly straightforward when imaging reflects a radiolucent nidus surrounded by variable degrees of reactive sclerosis [13]. The establishing of a preoperative diagnosis allowed the surgeon to eliminate the lesion without a biopsy. The histologic evaluation established the preoperative diagnosis in all cases [14]. Histologically, it is considered by the formation of the central nidus with surrounding sclerotic bone [15]. Computed Tomography (CT) scan vestiges the best imaging modality for the diagnosis of osteoid osteoma [16]. Osteoid osteoma, a benign bone tumor, is conventionally treated with operative excision [17].

It is extensively supposed to run a course concluding in spontaneous regression. The tumor can usually be removed by excision or ablation, although it may relapse locally [18]. The reported cases of osteoid osteoma in the mandible and maxilla are scarce. We reported a case of an osteoid osteoma of the right mandible, i.e. successfully treated with surgical excision without any recurrence.

Case Presentation

A 16-year-old male with congenital deafness was referred to the department of surgery in Sina Hospital affiliated to Tehran University of Medical Sciences (TUMS) with a two-year history of swelling in the right mandible. The patient's symptoms were no pain, fever, or other associated signs/symptoms. At the first outpatient examination, a diffuse palpable, non-motile swelling of the right mandible was noted. The patient's family and medical history were unremarkable. No neurological sign was found in the case. The patient underwent plain radiography, which suggested a calcified nodule on the inferolateral aspect of the right mandibular condyle. The radiological diagnosis was benign bone-forming tumors, including osteoma and osteoid osteoma (Figure 1). Thus, the patient was treated by excisional surgery. He well-tolerated the procedure without complications. Received specimen for pathology examination consisted of multiple pieces of brown tissue, measuring 3x2.5x1 cm.

Histological examinations revealed the portions of thick sclerotic bone trabeculae surrounded by an area containing anastomosing trabeculae of woven bone rimmed by osteoblasts in a fibrovascular stroma. Osteoid osteoma histologic examination revealed distinct demarcation between the nidus and surrounding reactive bone. Nidus contains uniform osteoid material resembling astomosing immature osteoid trabeculae (woven bone) with a sharp border of osteoblastic rimming. Uniform plump osteoblasts have regularly-shaped nuclei with abundant cytoplasm. The reactive zone was composed of a region surrounding the sclerotic border (Figure 2). Therefore,

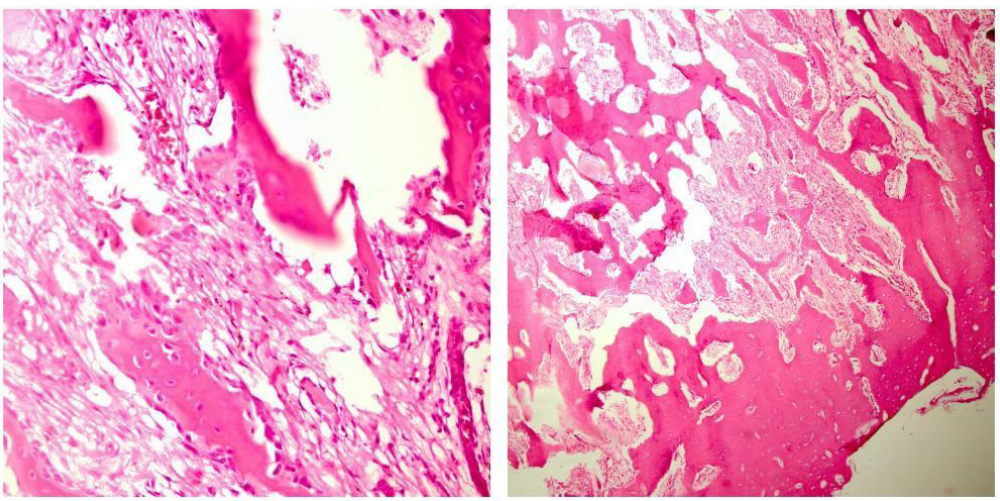

Figure 1. The section shows the anastomosing trabeculae of woven bone rimmed by osteoblasts in a fibrovascular stroma surrounded by sclerotic areas (x400 and 40)

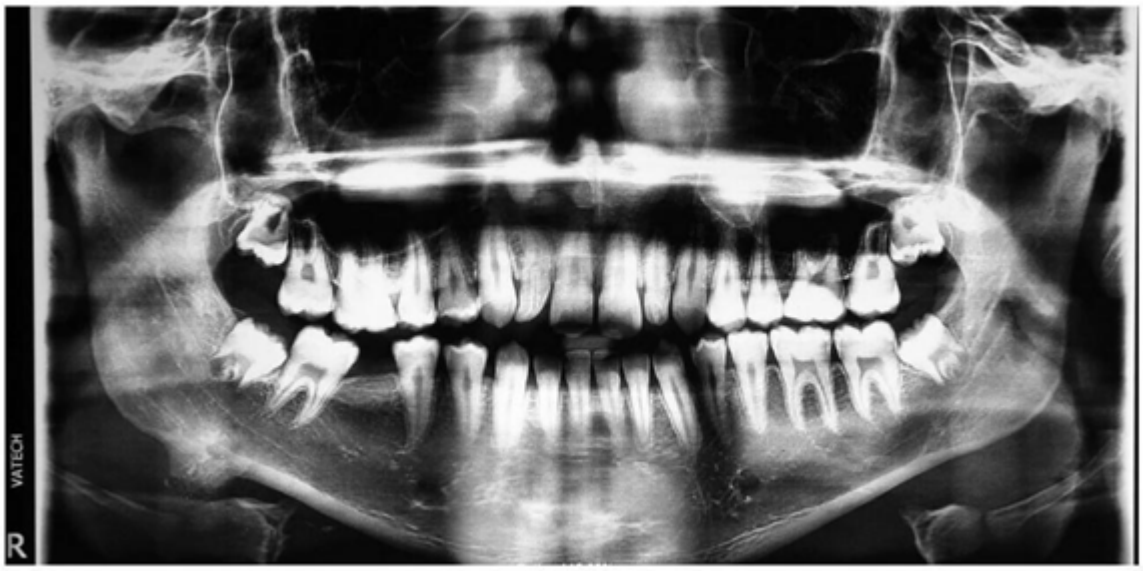

Figure 2. Radiologic examination shows a calcified nodule on the inferolateral aspect of the right mandibular condyle

our case was compatible with osteoid osteoma, and the diagnosis was confirmed. Based on the diagnosis, the patient received no additional treatment. After 4 years of close follow-up examinations, no recurrence was observed and the patient remains asymptomatic.

Discussion

Osteoid osteoma is a painful highly-vascularized benign tumor usually involving the long bone diaphysis cortex of young patients; it mostly appears as a small radiolucent nidus with or without central calcification and surrounding bone sclerosis on radiographs, and as a hot spot on scintigraphy [19]. Osteoid osteoma is often confused with numerous other entities. Although the pain was the most common clinical expression, it was absent in 12% of cases [20]. This is similar to our case which reported no pain. The proximal femur and spine are regular locations of involvement; however, almost any bone can be involved in this regard [21].

Osteoid osteomas are benign, slow-growing osteogenic tumors, infrequently occurring in the craniofacial bones [22]. Numerous similarities occurred among the osteoid osteomas of the jaws and those originated in much more common sites, such as the femur, tibia, and so on. It is significant to distinguish this uncommon entity to avoid the morbidity related to a prolonged delay in diagnosis [23]. Osteoid osteomas in the skull occurred (23.8%) in the maxilla and (76.2%) were in the mandible [24].

In 1935, Jaffe first described 5 cases of osteoid osteoma. Since Jaffe's report, a large series of cases of osteoid osteoma of the long bones have been reported. However, only a few instances presented the involvement of the mandible or maxilla [25]. According to other similar studies, osteoid osteoma is a rare finding in maxillofacial bones [22]. Osteoid osteoma and osteoblastoma histological manifestations are incredibly comparable [26]. CT and Magnetic Resonance (MR) imaging are essential in the diagnosis of benign bone tumors and the tumor-like lesions of bone [27]. When an Osteoid osteoma is clinically suspected in an extraordinary setting, a CT scan should be performed in the entire case [28]. Osteoid osteoma infrequently progresses to osteoblastoma. The decline of a patient's ability to relieve pain is a warning sign.

Approaching into such cases underlines the significance of close long-term radiological follow-up examinations in patients with conservatively-treated osteoid osteomas [29]. Additionally, a careful histologic study is essential to differentiate osteoblastoma from more aggressive tumors [30]. The expected history of osteoid osteoma is self-limited. Therefore, patients should be offered non-operative treatment, reserving ablative interventions for those who are unable or unwilling to take NSAIDs until their symptoms resolution [31]. If the nidus of the osteoid osteoma is removed, the patient will be free of pain [32]. Thus, a careful investigation of history, such as night-time pain and positive salicylate test, in addition to extensive imaging procedures, lead to the exact judgment previous to surgery [33].

Conclusion

When the typical clinical and radiological features are present, making the diagnosis is not difficult. Problems in the differential diagnosis may occur in association with a strange setting. They present frequent unchar-

acteristic clinical signs and radiological images that ultimately lead to insufficient treatment. The awareness of the possibility for spontaneous resolution may assist in the management of these tumors and tumor-like lesions of bone. Early correlation among juvenile joint pain and imaging abnormalities as well as the use of further imaging could decrease the time to identify the lesion and prevent avoidable morbidity.

Ethical Considerations

Compliance with ethical guidelines

All ethical principles are considered in this article. The participants were informed about the purpose of the research and its implementation stages.

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Conflict of interest

The authors declared no conflicts of interest.

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