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Osteoid osteoma of mandibular bone: Case report and review of the literature

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ABSTRACT

Osteoid osteoma is a benign bone-forming tumor and characterized by its limited growth potential, not exceeding 2 cm. The radiological hallmark of this tumor is a nidus, which is a small round area of relative radiolucency. Osteoid osteoma can involve any bone but is most commonly found in long bones and is extremely rare in the head and neck region. This disease characteristically presents with dull pain, worse at night, and sometimes relieved with NSAIDs. A 24-year-old Japanese woman presented with spontaneous pain and tenderness on the lingual side of her mandibular second molar on the right side. The patient reported that her pain had gradually increased, becoming more continuous and severe and no longer responding to NSAIDs. An initial panoramic radiograph revealed an oval, internally non-uniform, somewhat obscure boundaries in the right mandible. Computed tomography (CT) scan revealed a sclerotic lesion with a delineated central calcified nidus surrounded by a radiolucent band. The interior of the nidus was a non-uniform, irregularly shaped area of high absorption. The nidus was removed with intralesional curettage under general anesthesia. The histopathology of the specimen consisted of actively proliferating osteoblasts mixed with an interlacing network of immature bone and osteoid trabeculae. Immunohistochemistry revealed that hardly detected osteoblasts or fibrous stromal cells with intense nuclear immunoreactivity for p16 and/or murine double minute 2 (mdm2). We thus distinguished the tumor from Low-grade central osteosarcoma (LGCO) with immunohistochemical findings. The histopathological diagnosis was thus osteoid osteoma.

1. Introduction

Osteoid osteoma is a benign bone-forming tumor, first recognized as an entity by Jaffe in 1935. These tumors typically present in young individuals and at a male-to-female ratio of 3:1. Osteoid osteoma is characterized by its limited growth potential, not exceeding 2 cm. Clinical and radiological findings strongly suggest the diagnosis, which is confirmed with histopathological examination [1]. The radiological hallmark of this tumor is a nidus, which is a small round area of relative radiolucency. Osteoid osteoma can involve any bone but is most commonly found in long bones, especially the femur or tibia. This disease characteristically presents with dull pain, worse at night, and sometimes relieved with NSAIDs [2]. Previous studies have reported high levels of prostaglandin E2 and prostacyclin inside the nidus, providing an argument for the use of NSAIDs. However, the pain is not completely relieved without surgical removal of the nidus. Surgical treatment is thus the recommended therapeutic option. En block removal or cortical shaving and curettage of the nidus in long bones is recommended and can provide immediate relief of symptoms. To determine which surgical treatment is appropriate, localization of the nidus is important.

Osteoid osteomas of the head and neck have rarely been reported, with only a few reported cases in mandibular bone. We herein report a rare case of osteoid osteoma of the mandibular bone successfully treated with surgical removal of the nidus with

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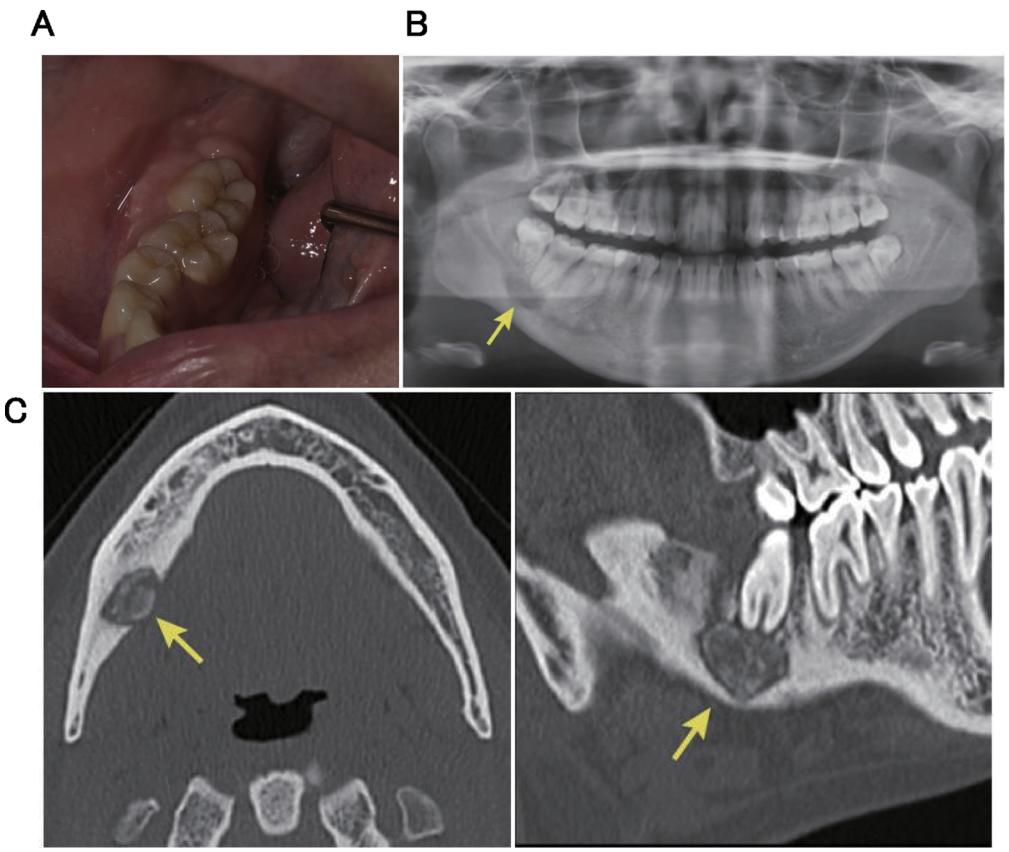

Fig. 1. (A) The lingual side of her mandibular second molar on the right side. (B) Panoramic radiograph shows a diffuse radiolucent area in the right mandibular molar region (yellow arrow). (C) (a) Axial CT view and (b) Sagittal CT view of the patient's mandible. The nidus (yellow arrows) appears as an opacity surrounded by a radiolucent band(For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

intralesional curettage.

2. Case report

A 24-year-old Japanese woman was referred to the Department of Oral and Maxillofacial Surgery at our institution in September 2017 with a 3-year history of spontaneous dull pain in the right mandible. The patient had no other remarkable medical history at that time. The values of routine blood examination were within normal limits. She had no abnormal findings on extraoral examination. The overlying skin and mucosa were normal, with no flare or increased temperature. The regional lymph nodes were non-palpable. Intraoral examination revealed a caries-free dentition and well-maintained oral hygiene. However, the patient had mild spontaneous pain and tenderness on the lingual side of her mandibular second molar on the right side (Fig. 1A). Her third mandibular molar on the right side was impacted. The right mandibular bone bulged slightly on the lingual side. The mandibular second molar was vital on electric pulpal examination. The patient reported that her pain had gradually increased, becoming more continuous and severe and no longer responding to NSAIDs.

An initial panoramic radiograph revealed an oval, internally non-uniform with somewhat obscure boundaries in the right mandible, above the inferior alveolar nerve (Fig. 1B). The mandibular bone around the lesion had diffusely increased X-ray opacity. On the basis of radiographic findings, osteoid osteoma, cementoblastoma, complex odontoma, ossifying fibroma, osteoblastoma, and idiopathic osteosclerosis were considered in the differential diagnosis. Computed tomography (CT) scan revealed a sclerotic lesion, with a delineated central calcified nidus (14 × 8 mm) surrounded by a radiolucent band. The interior of the nidus was a non-uniform, irregularly shaped area of high absorption. These findings were consistent with those of osteoid osteoma (Fig. 1C).

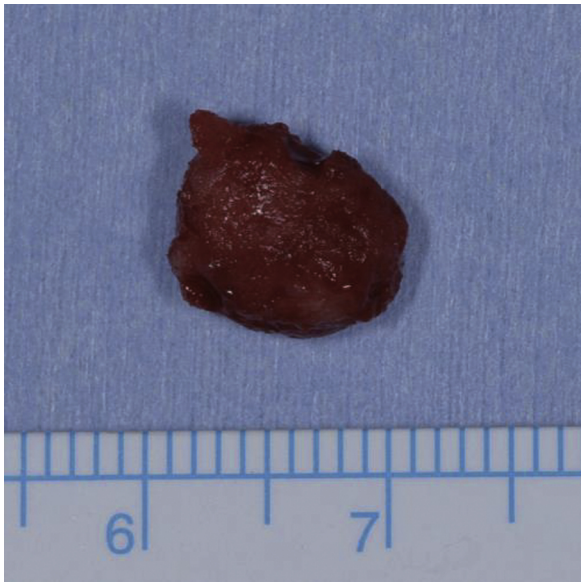

Fig. 2. The lesion was covered with a layer of soft tissue.

Mandibular wisdom tooth extraction was performed under local anesthesia in November 2017; however, biopsy from the tooth extraction did not confirm the diagnosis. Surgical excision of the nidus was performed under general anesthesia in June 2018 (Fig. 2). The nidus was removed with intralesional curettage.

Histologically, the inner lesion contains immature trabecular bone and osteoid bordered by many osteoblasts, which lack severe cellular

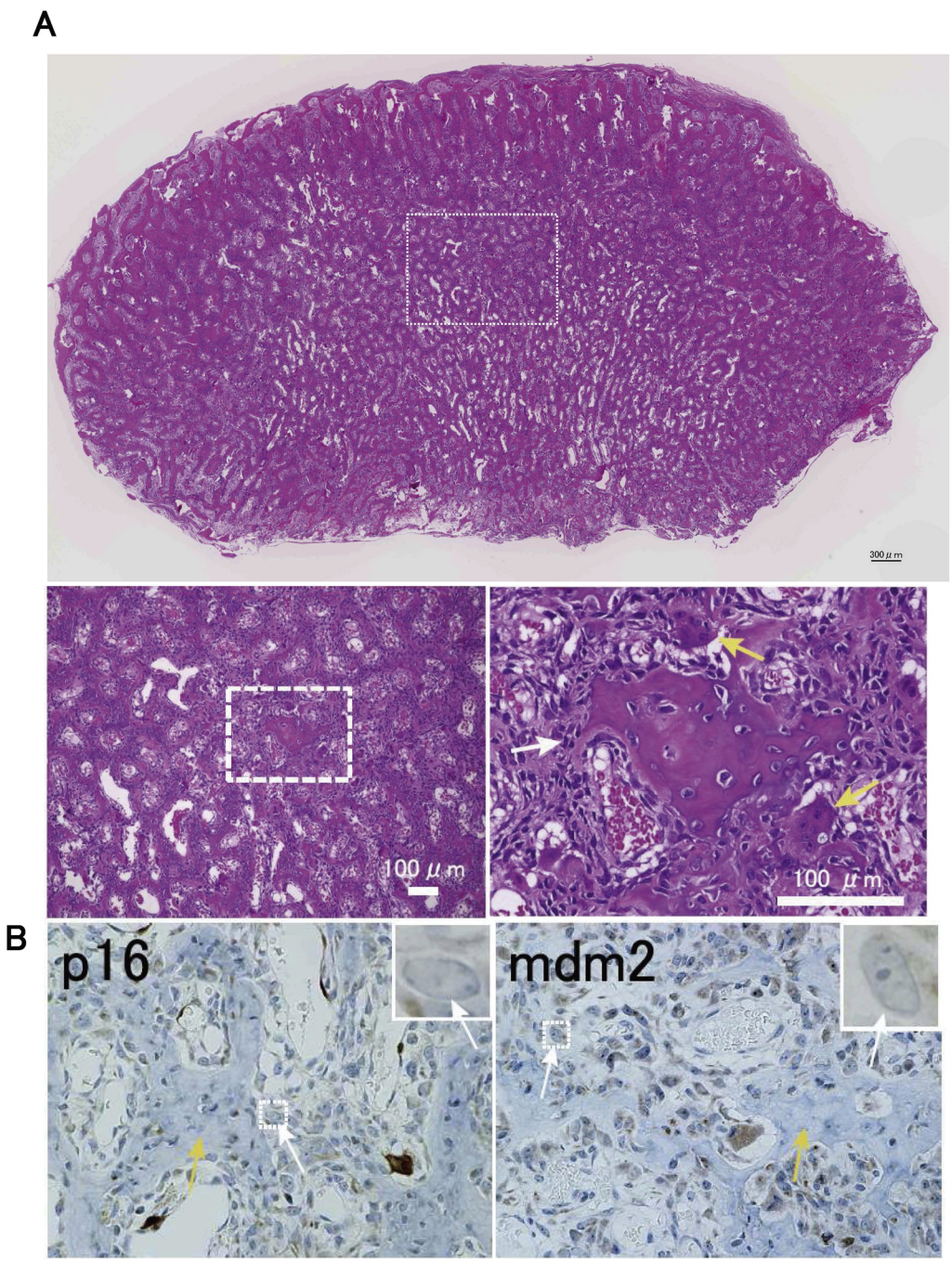

Fig. 3. (A) Tissue samples were fixed in formalin, embedded in paraffin, and sectioned. Histological findings show immature bone tissue and fibrous tissue with many dilated capillaries (H&E). White arrow indicates osteoblast and yellow arrow indicates multinucleated osteoclast. (B) Immunohistochemistry revealed that hardly detected osteoblasts or fibrous stromal cells with intense nuclear immunoreactivity for p16 (brown) and/or mdm2 (brown). Formation of immature trabecular bone and osteoid is seen (yellow arrow). Most osteoblasts in the immature bone are p16- and mdm2-negative (white arrow) (For interpretation of the references to colour in this figure legend, the reader is referred to the web version of this article).

pleomorphism. Amid the trabecular bone in the present case, highly vascularized and fibrous connective tissue with a small number of fibroblasts was found (Fig. 3A). Immunohistochemistry revealed that hardly detected osteoblasts or fibrous stromal cells with intense nuclear immunoreactivity for p16 and/or murine double minute 2 (mdm2), which negatively regulates p53 activity, around the immature trabecular bone and osteoid (Fig. 3B).

3. Discussion

Osteoid osteoma is a benign tumor characterized by an intracortical nidus, with a variable amount of calcification, sclerosis, and bone marrow edema. Growth of these tumors is limited, with lesion size generally < 2 cm according to WHO classification criteria [3]. Osteoid osteoma is extremely rare in the head and neck region. About 80% of osteoid osteomas occur in the long bones, whereas fewer than 1% occur in the jaws [4]. As shown in Table 1, only 22 cases of osteoid osteoma in the craniomaxillofacial region have been reported to date. In these reports, females were more often affected than males, at a ratio of 1.2:1 (12 women, 10 men; mean age 28.0 ± 16.1 years). Most of these tumors occurred in the mandible rather than the maxilla. Most patients (19/22, 87%) with osteoid osteoma had spontaneous pain and most (17/22, 77.3%) were treated with excision.

One challenge in diagnosing the current case was differentiating the tumor from osteoblastoma. Both osteoblastoma and osteoid osteoma are well-known benign osteoblastic lesions of bone. They have similar

Table 1
Overview of the literature: osteoid osteoma in the maxilla and mandible.

Author	Publication	Sex	Age	Location	Signs and symptoms
Rushton et al. [10]	1951	M	27	Mandible	Pain
Foss et al. [11]	1955	F	26	Mandible	Pain
Nelson et al. [12]	1955	M	17	Maxilla	Pain
Stoopack et al. [13]	1958	M	25	Mandible	Swelling, pain
Lind et al. [14]	1964	M	48	Mandible	Joint ankylosis, pain
Hillman et al. [15]	1965	F	4	Maxilla	–
Greene et al. [16]	1968	F	45	Maxilla	Dull pain
Brynolf, et al. [17]	1969	M	77	Maxilla	–
Zulian et al. [18]	1987	F	17	Mandible	Pain
Tochihara et al. [2]	2001	F	21	Mandible	Pain, trismus
Liu et al. [19]	2002	M	18	Mandible	Swelling, pain
Ida et al. [4]	2002	F	26	Mandible	Swelling, pain
Vasconcelos et al. [20]	2007	F	25	TMJ	Pain, limited mouth open
		F	23	TMJ	Pain, limited mouth open
Badauy et al. [21]	2007	M	26	Mandible	Swelling, pain
Manjunatha et al. [22]	2009	F	18	Mandible	Swelling, pain
Rahsepar et al. [23]	2009	M	21	Mandible	Swelling, dull pain
Karandilar et al. [24]	2011	M	14	Mandible	Swelling, deformity
Mohammed et al. [25]	2013	M	20	Mandible	Tenderness, swelling
Deferm et al. [26]	2017	F	56	TMJ	Swelling, dull pain
		F	39	TMJ	Pain

Temporomandibular joint; TMJ.

histological features and probably represent different expressions of one pathologic process.

Osteoid osteoma is small, self-limiting, and benign, whereas osteoblastoma is often larger (> 2 cm), more aggressive, and can become malignant. Osteoblastoma may be locally aggressive and may recur after removal. Although it is usually treated successfully with curettage, wide excision should be considered along with careful long-term follow-up because of the possibility of recurrence or malignant transformation. A central nidus is usually not present in osteoblastoma [5]. We distinguished osteoid osteoma from osteoblastoma in the current case on the basis of clinical features (intermittent pain relieved by NSAIDs), radiographic characteristics (nidus on CT measuring less than 2 cm), and histological findings.

Low-grade central osteosarcoma (LGCO) is a rare subtype of osteosarcoma that is less aggressive than conventional osteosarcoma. LGCO in the jaw can occasionally mimic benign lesions such as osteoid osteoma or osteoblastoma and may consequently be misdiagnosed in many patients. Immunohistochemical expression of mdm2, CDK4, and p16 is specific and provides sensitive markers for the diagnosis of LGCO, helping to differentiate it from benign osteoid tumors [6,7]. We also distinguished the tumor from LGCO with immunohistochemical findings (small numbers of nuclear immunoreactivity for p16- and/or mdm2 in osteoblasts).

Initial treatment of osteoid osteoma is generally aspirin or other NSAIDs, not surgery. A previous study reported that many patients can achieve lasting pain relief with NSAIDs. Cyclo-oxygenase is present in the nidus of osteoid osteomas and mediates increased production of prostaglandins in the tumor [8,9]. Surgical intervention is generally indicated for patients whose pain is unresponsive to medical therapy. The lesion can be removed with intralesional curettage or wide resection, depending on the clinical situation, location within the bone, and suspicion of malignancy. Successful treatment depends on complete marginal resection of the nidus.

Osteoid osteoma is a benign bone lesion that occurs very rarely in the jaw bones. We herein reported a case of osteoid osteoma that occurred in the mandible of a young adult. Clinical, radiographic, and pathological features were evaluated to arrive at the correct diagnosis. Surgical removal of the nidus with intralesional curettage completely relieved symptoms.

Ethical approval

This study design was approaved by the Ethics Committee of Kyushu University, Japan, and written informed consent was obtained from the patient.

Conflict of interest

No conflict of interest exists.

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