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Case Report

A rare case of osteoblastoma of the mandible

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ABSTRACT

Osteoblastoma is a rare bone-forming tumor, accounting for approximately 1% of all primary bone tumors. It commonly affects males within the age range of 10–30 years and typically arises in the vertebral column and long bones. Clinically, patients have pain and swelling. Osteoblastoma of the jawbones is rare, and thus, little is known about it. Here we report a rare case of osteoblastoma of the mandible in a 28-year-old pregnant female with a past history of tooth extraction. The patient experienced slight discomfort in the right mandible approximately 2 years after mandibular third molar extraction. Radiographic examination revealed a mixed radiolucent/radiopaque lesion in the mandible during pregnancy. The lesion, which measured approximately 2.5 cm in diameter, was not associated with the adjacent molar root. Although there was no significant change in the state of the lesion during pregnancy, severe jaw pain and swelling occurred shortly after parturition. Surgical excision was performed and a histopathological diagnosis of osteoblastoma was made. The severe jaw pain completely disappeared after surgery. The patient was followed up for a year and a half and was disease-free. In consideration of the drastic change in symptoms, we speculate that tooth extraction and parturition might have some effect on the progression of osteoblastoma.

1. Introduction

Osteoblastoma is a rare bone-forming tumor, accounting for approximately 1% of all primary bone tumors. It typically affects young males within the age range of 10–30 years and most commonly occurs in the vertebral column and long bones. Osteoblastoma of the jaws is rare, and till date, only around 100 cases have been reported in the literature [1–4].

Clinically, osteoblastoma mainly presents with pain and swelling [5]. Radiographic features are variable, usually showing a mixed radiolucent/radiopaque lesion with relatively defined borders [5]. Histologically, osteoblastoma is characterized by osteoid and bone formation with the presence of numerous osteoblasts [5].

The differential diagnosis of osteoblastoma mainly includes cementoblastoma, osteoid osteoma, and osteosarcoma. Typically, cementoblastoma is associated with the tooth root, and the size of osteoid osteomas is generally smaller than 1.5 cm [5,6]. However, osteoid osteoma and cementoblastoma are essentially histologically identical to osteoblastoma and some investigators regard these three entities as variants of a single entity [5–8]. Osteosarcoma shows atypical mitotic figures, cellular pleomorphism, neoplastic cartilage, and permeative growth into adjacent bone tissues [4,5,7].

In view of the purported benign nature of this tumor, surgical excision is the treatment of choice. The recurrence of osteoblastoma is rare (13.6%) and is usually attributable to incomplete excision [5,9].

Here we report a rare case of osteoblastoma of the mandible in a 28-year-old pregnant female with a past history of tooth extraction.

2. Case report

A 28-year-old Asian female was referred to our hospital for a bone lesion of the right mandible. The patient experienced slight

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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discomfort in the right mandible approximately 2 years after right mandibular third molar extraction by a general dentist, although there was no obvious abnormality both in the intraoperative findings and during the preoperative dental X-ray investigation of the wisdom tooth. Two years after the tooth extraction, another general dentist suspected a bone lesion on panoramic radiograph and referred her to our hospital.

The patient was 26 weeks pregnant and otherwise healthy. Physical examination revealed a low degree of diffuse swelling and tenderness in the right mandible. Panoramic radiography showed a mixed radiolucent/radiopaque lesion with a radiolucent rim in the right mandibular angle region, measuring approximately 2.5 cm in diameter. The lesion was not associated with the adjacent tooth root (Fig. 1) and was not identified on the previous panoramic radiograph taken 2 years before the tooth extraction (data not shown). A provisional diagnosis of benign bone tumor was made and surgical treatment after childbirth was planned.

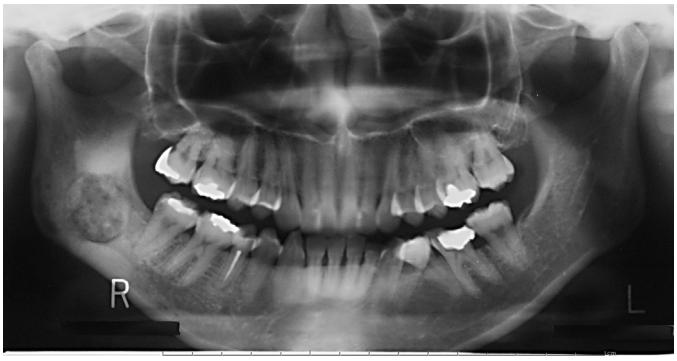

Fig. 1. Panoramic radiograph exhibiting a mixed radiolucent/radiopaque mass in the right mandibular angle region.

Shortly after parturition, the patient experienced increasing pain and swelling of the right mandible and had limited mouth opening, although her condition had been stable during pregnancy. Computed tomography revealed a predominantly radiopaque mass with a radiolucent border in the right mandibular angle region and revealed cortical thickening and masseter muscle swelling around the lesion (Fig. 2). Complete excision was performed through an incision in the retromolar area. Intraoperatively, the lesion was well circumscribed, without attachment to the adjacent tooth root (Fig. 3). Histological examination revealed dense bony trabeculae with osteoid formation in the fibrovascular connective tissue. Numerous plump osteoblasts surrounding the islands of new bone formation were observed. Mitotic figures were rare and no atypical figures were identified. Osteoclast-like giant cells and scattered foci of osteoclastic bone resorption were also observed; however, no cartilage formation was observed (Fig. 4). A diagnosis of osteoblastoma of the mandible was made. Excision of the lesion completely relieved the pain. The patient had an uneventful postoperative course, and there was no evidence of recurrence at the one and a half-year follow-up.

3. Discussion

Osteoblastoma is a rare bone tumor, accounting for approximately 1% of all primary bone tumors. It most commonly affects males during the second decade and often arises in the vertebral column and long bones. Osteoblastoma of the jaws is very rare, and thus, little is known about it [1–4].

As described above, skeletal osteoblastoma predominantly occurs in males [1]; however, there is no clear gender predilection for osteoblastoma of the jaws [2,3]. Osteoid osteoma and cementoblastoma are essentially histologically identical to osteoblastoma [5,7,8]. Jaw osteoid osteoma shows no gender predilection, unlike the male predominance observed in skeletal osteoid osteoma [10]. Similarly, cementoblastoma also does not show a definitive gender preference [11,12]. These data suggest that factors that differ between skeletal bones and jaw bones might influence the development of these entities, including osteoblastoma.

The etiology of osteoblastoma is not clear. Jaffe [13] and Lichtenstein [14] independently suggested that osteoblastoma was a true neoplasm of osteoblastic derivation, whereas other investigators have suggested that osteoblastoma occurs as a result of trauma or inflammation [15,16]. Temple et al. reported that one-third of 329 patients with osteoblastoma had an associated history of antecedent trauma [17]. Gordon et al. also reported that at least eight of 58 patients with osteoblastoma of the jaw had a history of antecedent trauma or tooth extraction [18]. In the present case, the patient had a history of tooth extraction and subsequent chronic mandibular discomfort. The lesion was approximately 2.5 cm in diameter at 2 years after tooth extraction. In reports of osteoid osteoma, which has a lower growth potential than osteoblastoma, the annual growth rate is approximately 5 mm in diameter [19]. Given the potential growth rate of osteoblastoma, this lesion might have occurred around the time of tooth extraction. However, it was unclear whether tooth extraction affected the osteoblastoma or was merely a trigger for the patient's subjective symptoms.

As previously mentioned, osteoblastoma commonly occurs in adolescent and young adult males [1–4], in whom sex steroid hormones play an important role in bone metabolism [20–23]. This epidemiological feature suggests that osteoblastoma could be a sex steroid hormone-dependent tumor [21]. In the present case, symptoms of pain, swelling, and trismus worsened considerably shortly after parturition, despite remaining stable during pregnancy. This

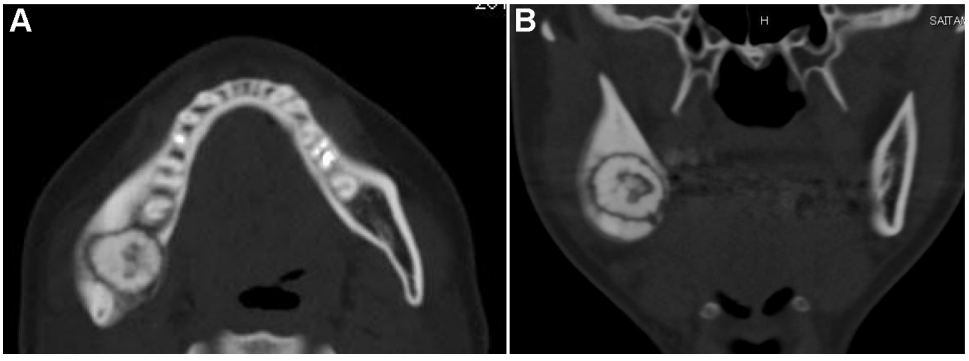

Fig. 2. Computed tomography (CT) images showing a predominantly radiopaque mass with a radiolucent border in the right mandibular angle region, as well as cortical thickening and masseter muscle swelling around the lesion. A: Axial section. B: Coronal section.