Osteoblastoma in the maxilla: report of a case

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Abstract: A case of maxillary osteoblastoma in a 29-year-old man is presented. The patient developed a painful swelling on the right hard palate. Panoramic radiographs revealed a mixed radiolucent/radio-opaque mass surrounded by a narrow radiolucent zone in the apical area of the right maxillary first molar. Under the clinical diagnosis of apical periodontitis, root canal treatment for the molar was carried out. Bleeding from the root canal was observed during the treatment, and the painful swelling disappeared immediately. Shortly afterward, when the bleeding from the root canal stopped, the painful swelling recurred. CT showed a subcortical nodule in the right maxilla. Histopathological examination revealed the lesion was composed of immature bone with broad osteoid matrix at the periphery. The osteoid matrix of the immature bone was fringed with numerous plump osteoblasts without nuclear atypia. Some small foci composed of the neoplastic osteoblasts were associated with the osteoid. Histopathological diagnosis of the biopsy was benign osteoblastoma, and excision of the tumor completely relieved the pain. There has been no recurrence for 7 years. Reduction in vessel pressure due to root canal therapy was considered to have temporarily relieved the pain of osteoblastoma. [Oral Med Pathol 2009; 14: 37-40 doi: 10.3353/omp.14.37]

Key words: bone tumor, cyclooxygenase, maxilla, osteoblastoma, root canal treatment

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Introduction

The term “osteoblastoma” was first used by Jaffe (1) and Lichtenstein (2). Osteoblastoma is a vascular, osteoid- and bone forming benign tumor of bone characterized cytologically by the abundant presence of osteoblasts (3). Osteoblastoma accounts for about 1% of all bone tumors, and involves the spine and sacrum. The tumor is more common in males and affects patients in the age range of 10-30 years, with extremes of 5-70 years old (4-9). Osteoblastoma arising in the maxilla is very rare, and jaw lesions produce tooth pain and/or swelling (9). Diagnosis of osteoblastoma of the jaw is necessary to distinguish osteoid osteoma, cementoblastoma, fibro-osseous lesions and osteomyelitis (9). We present here a very rare case of osteoblastoma arising in the maxilla, and discuss the differential diagnosis and temporary relief of the pain.

Case report

A 29-year-old man was referred to Nagasaki University Hospital for evaluation of a painful swelling on the right hard palate. He stated that the pain had been present for approximately two months and had worsened since its onset. Physical examination revealed a 2 cm in diameter, tender,
bony swelling of the right palatal mucosa of the maxillary first molar (Fig. 1). The overlying mucosa was intact and no pus discharge was noted. Electric pulp test of the right maxillary first molar indicated that it was non-vital. Panoramic radiography revealed a mixed radiolucent/radio-opaque round lesion surrounded by a narrow radiolucent zone above the right maxillary first molar (Fig. 2). Under the clinical diagnosis of apical periodontitis, we initially attempted root canal treatment of the tooth without local anesthesia, and as blood exited via the root canal, the pain and swelling of the right palatal mucosa subsided. Although bleeding from the root canal had stopped by the next consultation, the painful and tender bony swelling of the right palatal mucosa reappeared. CT showed a partial subcortical nodule measuring approximately 2 cm in diameter in the same area (Fig. 3). Clinical diagnosis was changed to osteomyelitis of the maxilla.

The biopsy was performed under local anesthesia. The biopsy specimen included immature bone fragments with broad osteoid matrix at the periphery and chips of normal cortical bone. The osteoid matrix of the immature bone was fringed with numerous plump osteoblasts without nuclear atypia. Some small foci composed of the neoplastic osteoblasts with large and plump cytoplasm were associated with the osteoid. Complicated irregular basophilic reversal lines were seen in the immature bone. The fibrous connective tissue around the immature bone contained many blood capillaries, and no inflammatory cell infiltration was found (Figs. 4a & 4b). There was no continuity between the normal cortical bone and neoplastic bone. Histological diagnosis of the biopsy was osteoblastoma, and the tumor was removed under general anesthesia. The surgical specimen showed peripheral radiating columns of immature bone (Fig. 4c), and scattered osteoclasts within the Howship’s lacunae (Fig. 4d), as well as the histological features of the biopsy. The postoperative course was uneventful, and the patient was discharged 10 days postoperatively. His pain disappeared after the operation. He was last seen 7 years after the operation and showed no sign of recurrence on either clinical or radiological examination.

**Discussion**

Osteoblastoma, first reported by Jaffe (1) and Lichtenstein (2) in 1956, is a rare benign bone forming neoplasm which produces bone spicules bordered by prominent osteoblasts. Osteoblastoma accounts for about 1% of all bone tumors and is more common in males (4, 9). The nature of osteoblastoma is still unknown. Jaffe and Lichtenstein suggested that osteoblastoma is a true neoplasm of osteoblastic derivation, but others (10-11) suggested that osteoblastoma occurs as a result of trauma or inflammation. Smith reported that the entity of osteoblastoma may be regarded as a peculiar local response of the tissues to injury, or even possibly as a localized alteration in bone physiology, rather than as a true neoplasm (11). In our case, there were no past histories of trauma or inflammation in the maxilla.

Because the clinical and radiographic features of osteoblastoma are nonspecific, a number of lesions must be considered in its differential diagnosis. Osteoid osteoma is the most closely related lesion from a histological aspect. Both entities are often histologically and radiographically similar. It is well recognized that the histological features of osteoid osteoma and osteoblastoma overlap (12). However, osteoid osteoma originates from the cortical bone, while osteoblastoma arises from the medullary bone (13). One of the important clinical criteria for discrimination between them is size. Dahlin et al. described that osteoid osteoma is usually less than 1 cm in diameter, while osteoblastoma is usually larger than 2 cm (14). With respect to jaw lesions, the diameter of the nidus of osteoid osteoma does not exceed 2.0 cm (15). Osteoid osteoma generally is more painful than osteoblastoma and possesses a radiolucent-opaque pattern surrounded by thin radiolucency, and a perilesional halo of sclerotic bone (16). However, it is known that osteoblastoma arising in the jaws also produces root pain and/or swelling (9). In the present case, the lesion was located approximately 2 cm beneath the cortical bone of the maxilla. Thus the
tumor could be diagnosed as osteoblastoma rather than osteoid osteoma.

Osteoblastoma of the jaws should also be distinguished from cementoblastoma. According to Abrams et al. the histological similarities of cementoblastoma, osteoid osteoma, and osteoblastoma suggested a close relationship among these three conditions (17). When osteoblastoma occurs in the jaws, it is difficult to distinguish it histologically from cementoblastoma. When making a differential diagnosis between them, it is very important to confirm whether or not the tumor is connecting with tooth root (18-19). However, the attachment of the tumor to the root of the teeth should not be used as a basis for the diagnosis of cementoblastoma (20). Our case is not considered as cementoblastoma because no continuity to the root of the tooth was found.

Osteoblastoma should be differentiated from benign fibro-osseous lesions, practically ossifying fibroma, which usually presents as a painless swelling that produces facial deformity (20). The radiographic appearance of early ossifying fibroma appears as a relatively well-demarcated radiolucency, but later the lesion becomes more mineralized and relatively less localized (21). Ossifying fibroma may show marked osteoblastic activity, but the inter trabecular spaces do not contain the large, plump osteoblasts observed in osteoblastoma (20).

The specimen of the biopsy and surgical material included neither inflammatory cell infiltration nor necrotic bone. Granulation tissue and fibrosis with regenerative bone formation were not found. Thus osteomyelitis was histologically denied.

Osteoblastoma may be misdiagnosed as osteosarcoma because of prominent large plump osteoblasts rimming the broad osteoid matrix. Malignant transformation of osteoblastoma is also known (22-23). Clinically, rapid growth and cortical destruction suggest such conditions. Histologically, high cellularity, cellular and nuclear atypism, a high mitotic rate, and bizarre mitoses should lead to the diagnosis of osteosarcoma (20). The present case showed neither malignant clinical nor histological features, except for the severe pain.

A very characteristic symptom of osteoid osteoma and osteoblastoma is spontaneous pain. In peculiar to osteoid osteoma, the pain at first is mild and intermittent, but later becomes severe, constant, and more pronounced during the night (24). This symptom is generally attributed to changes in the vessel pressure being registered by the abundant
innervations. Other explanations of the pain are direct irritation of nerve fibers in or near the calcified focus (24). Mungo et al. examined immunoperoxidase of cyclooxygenase-1 (COX-1) and cyclooxygenase-2 (COX-2) in osteoid osteoma and osteoblastoma tissues (25). As COX is the rate limiting enzyme in prostaglandin biosynthesis, they considered that COX expression was implicated in induction of the pain (25). However, Hasegawa et al. suggested that the increased density of nerve fibers around and within the nidus might play a more important role in the mediation of pain in osteoid osteoma than the effects of osteoblast produced prostaglandin E2 on the nerves and proliferated blood vessels (26). In the present case, the symptoms of pain and swelling were immediately and temporarily cured by root canal treatment. Bleeding from the root canal was thought to have temporarily reduced the vessel pressure of the lesion, thereby relieving the symptoms.

The prognosis of osteoblastoma is good if the lesion has been removed completely. Osteoblastoma is thought to exhibit benign behavior. However, long-term follow-up is recommended to detect any recurrence and malignant transformation in the early stages, because sarcomatous transformation of osteoblastoma has been reported (22-23).

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References

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