Osteoblastoma (Aggressive Form) of the Mandible – Report of a Case

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Osteoblastoma is a rare osteoblastic bony neoplasm characterized microscopically by proliferation of osteoblasts with trabeculae production within a vascularized fibrous connective tissue stroma [1]. Although Jaffe and Lichtenstein regarded this disease entity to be a true neoplasm of osteoblastic origin [2, 3], the exact pathogenesis of this lesion remains varied and its true origin is still controversial. Reviewing English-language literatures, potential etiologies including trauma, inflammation, abnormal local response of the tissue injury and local alteration in bone physiology are reported to be associated to this tumor formation.

To date, two subtypes of osteoblastoma are classified: the benign form and the aggressive form. The former is featured with a well-vascularized slow grow lesion containing a well-defined corticated margin [4]. The latter form represents a locally aggressive lesion with a tendency to relapse and manifests atypical microscopic features, requiring differential diagnosis with low-grade osteosarcoma [5]. Furthermore, this disease entity can be categorized as cortical, medullary, and periosteal types with reference to which component of the bone is affected [6]. Here, we present a rare case of mandibular osteoblastoma (aggressive form) in a 47-year-old female patient.

CASE PRESENTATION

A 47-year-old female patient visited her family dentist with a chief complaint of pain and swelling over her right mandibular body for an uncertain duration. The family dentist then referred the patient to our institution for further clinical examination and treatment.
Figure 1 (A) Clinical presentation of the right mandibular lesion. (B) Cropped panoramic radiograph showed a multilocular radiolucency at the right mandibular body. (C) Histological examination of the specimen revealing proliferation of osteoblasts with trabeculae production within a vascularized fibrous connective tissue stroma (Hematoxylin-eosin stain ×100). (D) The supporting stroma contained scattered vascular channels demonstrating positivity on CD34 immunohistochemical staining (×40).

The patient was denied to have any systemic diseases. In addition, she had no oral risk factors of betel-quid chewing, cigarette smoking as well as alcohol drinking. On extraoral examination, there was a diffuse tender swelling over the right facial area. Intra-oral examination revealed a buccal-lingual expansion covered with normal appearing oral mucosa extending from distal aspect of lower second premolar to the retromolar area (Figure 1A). The swelling was hard in consistency and measured about 2.5 × 1.5 cm in maximum diameter. Missing of tooth 46 and 47 as well as a malposed 48 was observed in the swelling region. Neither paresthesia nor cervical lymphadenopathy was noted. Panoramic examination revealed a well-defined multilocular radiolucency with a sclerotic margin over right posterior mandibular body, extending from distal aspect of tooth 45 to anterior aspect of right mandibular ramus and from right mandibular alveolar ridge down to the inferior border of right mandibular body, downward displacing and destroying the superior border of right inferior alveolar canal, measured approximately 3.5 cm in maximum diameter. This lesion causes external root resorption of tooth 45, crown & root resorption and displacement of tooth 48 (Figure 1B).

Incisional biopsy was performed under local anesthesia. The specimen was subsequently sent for histopathological examination. The decalcified hard tissue specimen was
characterized by trabeculae of immature woven bone surrounded by numerous osteoblasts demonstrating ample cytoplasm and hyperchromatic nuclei. Large osteoblasts with increased mitotic activity and osteoid production were also found (Figure 1C). The supporting stroma contained scattered dilated vascular channels showing positivity on CD34 staining (Figure 1D). Hence, based upon these microscopic findings, osteoblastoma (aggressive form) was rendered. A treatment plan of complete excision of the entire tumor under general anesthesia with post-surgical radiation therapy and/or chemotherapy was suggested to the patient; however, the patient would like to delay the treatment and to seek further evaluation from other sources.

COMMENTS
Due to rarity of osteoblastoma in the jawbones and its close similarities to other jawbone tumors, this bony tumor may present a diagnostic challenge during the initial clinical presentation. Differential diagnosis should comprise fibro-osseous lesions as well as non-odontogenic and odontogenic bony tumors. Significantly, osteosarcoma must be considered for differential diagnosis. Distinction of osteoblastoma from osteosarcoma is principally based on the histopathological findings of the lack of abnormal mitotic figures, cellular pleomorphism, neoplastic cartilage and permeative growth into adjacent bone tissue [7]. In the present case, the bone-forming tumor characterized by osteoid and woven bone deposition and abundant osteoblasts with atypical histological findings but lack of abnormal mitoses were observed.

Osteoblastoma, typically occurs in young adults with an average age of 20 years old, is clinically presents mainly with pain, swelling and expansion of bone cortex with a limited growth potential and usually do not more than 4 cm in diameter [8]. Radiographic findings of osteoblastoma is non-consistent and may vary case by case depending on the duration [7]. A mixed radiopaque and radiolucent patterns, depending on the degree of calcification without perilesional sclerotic border is a general radiographic features for osteoblastomas. By contrast, our case was found in a middle-aged adult showing a well-defined multilocular radiolucency with a sclerotic margin.

The mostly accepted treatment for osteoblastomas is surgical excision. A more conservative approach of surgical curettage has been suggested in the literature [6]. However, considering a recurrence rate could be as high as approximately 14% for osteoblastomas [8, 9], surgical excision of the whole tumor would be the preferred treatment of choice as curettage would have a high propensity to recurrence. On the other hand, if aggressive osteoblastoma is suspected, a complete resection with enough safe margin and post-surgical radiotherapy and/or chemotherapy is recommended as the treatment of choice [4]. Unfortunately, for the current case, the patient delayed our proposed treatment planning. Then, the patient was then loss of clinical follow-up.

CONCLUSION
We presented a rare case of aggressive osteoblastoma affecting mandible in a middle-aged female patient demonstrating multilocular radiolucency radiographically.

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Aggressive osteoblastoma

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