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Case Report

A rare case of maxillary osteoblastoma in a 10-month-old infant

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ABSTRACT

Osteoblastoma (OSB) is an uncommon (3% of benign and 1% of all) primary bone neoplasm with extremely rare occurrence in head and neck, especially the maxillary bone region. OSB of the jaw mainly involves the mandible bone. We report a 10-month-old male who was admitted to the hospital because of a mass in the right maxillary jaw region. Clinicoradiopathologic features of the patient have been described. The histological examinations revealed OSB of the maxillary bone. Total surgical resection of the tumor was performed with 10 months of follow-up and there is no evidence of recurrence. To the best of our knowledge, it is the first report of maxillary OSB in an infant.

Key Words: Bone neoplasm, maxilla, osteoblastoma

INTRODUCTION

Osteoblastoma (OSB) is a benign neoplasm that histopathologically is described as a proliferation of osteoblasts forming osteoid tissue and primitive bone trabeculae set in a vascularized fibrous connective tissue stroma. OSB usually involves long bones and spine and rarely involves the head and neck region.[1]

OSB compromises for <1% of all tumors in the maxillofacial region. Young adults with ages between 20 and 30 years are the most affected range, with a higher prevalence in males with a 2:1 ratio. Among children under the age of 10, benign OSB involving the maxilla is a rare tumor.[2]

Many review articles discussed the epidemiological pattern of the OSB. Jones *et al*.[3] reported 43 cases of OSB. In total series of cases, there were only six case reports relating to children, and in all of them, the tumor was located in the mandible, although several recent articles reported OSB in the hard palate and maxillary region.[4,5]

The youngest case of OSB in the literature was an infant having OSB involving the temporal bone. The tumor site was not entirely limited to oral cavity and jaws. Also as mentioned, the tumor was a benign neoplasm.[6]

In this case report, we described a very rare case of OSB in the maxilla for the first time in an infant patient. The tumor involved the maxilla and hard palate region in a 10-month-old male patient. We reported clinicopathologic features and a review of the literature about OSB in children.

CASE REPORT

A 10-month-old infant presenting a swelling on the palate noticed by his parents was brought to

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the Oral and Maxillofacial Surgery Department of Children's Medical Center, Tehran University of Medical Sciences.

The time at which the lesion started to emerge was unknown because of the patient's age and his mother reported that there was not a noticeable swelling at the birth time. The neurological findings such as otorrhea, dizziness, tenderness, pain, or other symptoms could not also be reported because of the patient's age. Physical examination revealed a swollen mass in the right posterior part of the upper jaw which extended from tuberosity to the midline of the hard palate.

Multi-detector computed tomography (CT) of the hard palate, nasal cavity, and maxilla was performed. The internal structure of the lesion appeared as a mixed radiolucent–radiopaque. The radiographical findings showed a well-defined lesion expanded from the maxillary crest to the maxillary sinus perforating the sinus walls. Expansion of maxillary sinus walls and displacement of the lateral wall of the nasal cavity were visible as its effect on the surrounding structures [Figure 1]. According to radiographic findings, a suitable diagnosis was a cemento-ossifying fibroma.

The surgical procedure was carried out under general anesthesia using nasal intubation. All of the incisions were intraoral, starting from the palatal region of the left side maxilla. The whole lesion was extracted by enucleation, and curettage was done by peripheral ostectomy. There were neither signs of any teeth nor tooth buds in the extracted lesion.

Macroscopic view of the excised lesion demonstrated several fragmented pieces of tan to gray elastic tissue [Figure 2].

Microscopic examination of the lesion showed a benign neoplasm consisting of proliferation of plump epithelioid osteoblasts with extensive areas of thick trabecular bone formation, most of them surrounded by a plump osteoblastic rim. The tumoral cells were arranged in vascular myxoid to the hemorrhagic stroma. No atypia was seen among the tumoral cells [Figure 3].

There are lesions that can be considered as a histopathologic differential diagnosis for OSB, such as osteoid osteoma, cementoblastoma, ossifying fibroma, and osteosarcoma. In this case, due to clinical, histopathological, and radiological findings, the finial diagnosis was OSB.

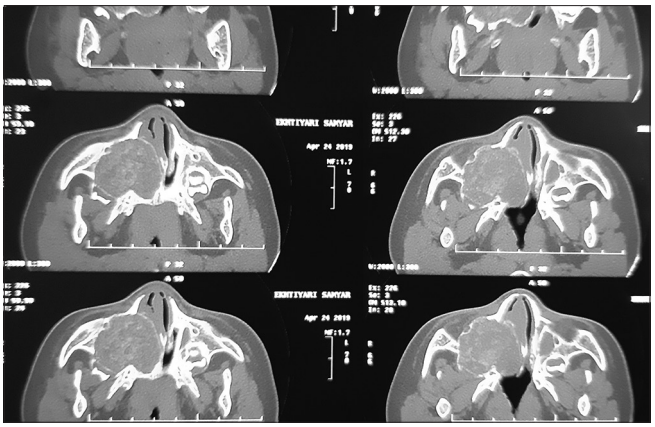

Figure 1: Axial sections of a multi-detector computed tomography demonstrate a large well-defined lesion involving maxillary jaw with some central areas of opacifications.

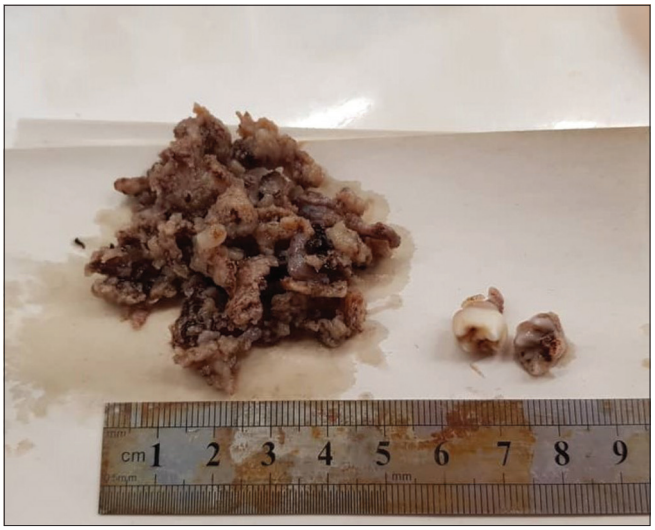

Figure 2: The gross appearance of the lesion after excision of the tumor mass. Note two deciduous molar teeth germs that are excised with the lesion.

After 10 months, the patient is in a good physical state with no evidence of recurrence.

DISCUSSION

OSB is considered a rare bone tumor, having a prevalence of 0.8%–1% among the population. It is commonly seen in young adults.[6] OSB has a rare prevalence in children. It seems that OSB in children includes mostly head and neck bones, with more predilection for mandible. The youngest reported case in the literature is a 7-month-old female infant with an OSB of temporal bone.[6] In the present case, the patient was a 10-month-old infant with maxillary OSB. Only 10% of cases appear in maxillofacial area.[7] González *et al*.[2] also reported a 7-year-old

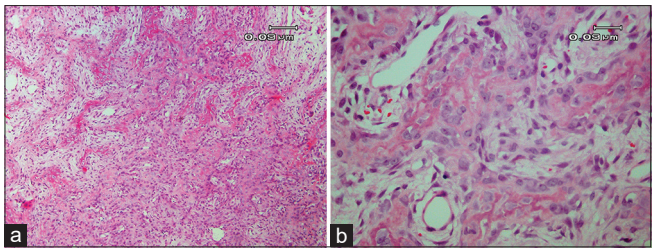

Figure 3: Histopathologic features of the lesion. (a) A bone-forming lesion composed of plump epithelioid cells admixed with woven bone trabeculae (H and E, ×100). (b) Sheets of large epithelioid cells which lined bone trabeculae with any atypical changes (H and E, ×400).

boy presenting a painful swelling on the left hard palate. Salmen *et al*.[4] reported an aggressive OSB in a 7-year-old patient. The lesion was large and fully involved the left maxilla, including maxillary sinus and nasal cavity. Recurrence was detected 2 months after enucleation and en-block resection of the maxillary segment.

The tumor has a predilection to male, but Jones *et al*. reported 83.3% of the cases occurring in female patients. They reviewed 24 patients with OSB of the maxilla and mandible and mostly involving the posterior areas of the jaws, mostly mandible.

OSB can have various clinical features including swelling, mild and long-standing pain and tenderness. Most of the patients present a localized swelling, tenderness, and warmth at the site of the tumor. In our reported case, the patient had a swelling in the posterior area of jaw which is a usual clinical finding in Osteoblastoma (OSB) patients, especially in younger ones. The tumor was 5 cm in the maximum dimension.

The radiological findings are various, including radiolucent, radiopaque, and mixed lesions, so it is not a reliable diagnostic tool but can be helpful in ruling out the other lesions.

The differential diagnosis of OSB includes osteoid osteoma, cementoblastoma, ossifying fibroma, and in aggressive tumors, osteosarcoma. Using clinical, histopathologic, and radiographic findings altogether, we can distinguish OSB from osteoid osteoma and cementoblastoma, although there is a difficulty to distinct an OSB from an OSB-like osteosarcoma. The most effective and helpful landmark is the radiographic and histopathologic margins. OSB is a circumscribed tumor with distinct and sharp margins making it seems separate from the peripheral tissues, but an OSB-like osteosarcoma shows infiltration of tumoral cells into the peripheral tissues. This invasive behavior of an osteoblasoma-like osteosarcoma is prominent, especially in radiological features as blending and ill-defined border at margins.

Shah *et al*.[8] reported a 7-year-old male patient with swelling and pain on the left side of the maxilla. Due to the homogeneous radio-opaque diagram, the primary diagnosis was a fibrous dysplasia. Thereby, no other examinations like CT or bone biopsy were performed even though the patient had pain in the region. Recurrence happened 6 months after the initial surgery. Histopathological examinations confirmed a benign OSB. Jones *et al*.[3] also reported a grounded-glass opacification which was difficult to differentiate from fibro-osseous lesions. It has been reported that the histopathological features alone are not reliable predictors of an aggressive behavior. Hence, it is important to consider the radiological and clinical findings altogether. The recurrence rate of conventional OSB is 13.6%; while in aggressive OSB, it is 50%.

Lin *et al*.[9] also presented two cases, that one of them was a 10-year-old boy with swelling of the left side of the face in the preceding 18 months. Histologically, the tumor was composed of small, irregular bony trabeculae and osteoids that were separated by a vascular stroma. In this case, an osteoblastic lesion can be considered as a differential diagnosis.

OSB presents a benign tumor composed of well-vascularized trabeculae of osteoid tissue surrounded by plump osteoblasts. In some lesions, it is difficult to differentiate it from a low-grade osteosarcoma. Mild cellular atypia can be seen in OSB. The larger tumors (more than 4 cm) more likely happen to be an aggressive type of osteoblstoma.

Although OSB is a benign tumor, the treatment is complete surgical removal of the tumor because of its high tendency to show recurrence. Enucleation followed by curettage and resection is mostly indicated. For a conventional OSB, enucleation is a safe treatment. Although the aggressive OSB is still a benign tumor, because of the aggressive nature of this type of OSB, resection of the lesion is recommended as soon as possible.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient's parents have given their consent for images

and other clinical information to be reported in the journal. The patient's parents understand that names and initials will not be published and due efforts will be made to conceal identity, but anonymity cannot be guaranteed.

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Conflicts of interest
The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

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