

CASE REPORT

Osteoblastoma of Anterior Maxilla: Route to Diagnosis

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ABSTRACT

Osteoblastoma, often referred to as “Benign Osteoblastoma” (BO) is a rare bone forming neoplasm which comprises only about 1% of all bone tumors. It occurs with a greater frequency in the long bones of the appendicular skeleton and the vertebrae. The occurrence of osteoblastoma in the bones of the skull and the jaw bones is rare. A predilection for occurrence in the mandible has been reported when it affects the jaw bones. It is usually seen in the second to fourth decade of life with a definite male predilection. Because of its clinical and histopathologic similarities with other bone-forming lesions of the jaw, BO can pose a diagnostic challenge. Here, we report a rare case of BO in the anterior maxilla of a 16-year-old female patient, and its route to diagnosis, along with a brief review of the literature.

Keywords: Jaw tumor, Maxilla, Osteoblastoma.

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INTRODUCTION

Benign osteoblastoma (BO) is a rare bone forming neoplasm, comprising of about 1% of all bone tumors and 3.5% of all benign bone tumors.¹ The sites of predilection for the tumor are vertebral column and the long bones of the appendicular skeleton. They are relatively rare in the skull and gnathic bones, constituting only 10–12% with a reported predilection for the mandible.^{1,2} It usually occurs in the second to fourth decades of life with a mean age of 17 years. A definitive male predilection has been reported.¹ The diagnosis of BO is made by correlating the clinical and radiographic features with its histopathology and needs to be differentiated from other similar bone pathologies like osteoid osteoma, ossifying fibroma, and even low-grade osteosarcoma. Here, we describe

the case of a 16-year-old female patient with BO of the anterior palate.

CASE REPORT

A 16-year-old female patient visited our hospital with a chief complaint of swelling on the anterior palate region of the maxilla. The swelling was first noticed 6 months back on the anterior part of the hard palate behind the upper left central and lateral incisors and subsequently exhibited a slow, gradual enlargement. A sharp pain which was intermittent and aggravated during mastication was also felt by the patient in relation to the swelling. The patient denied any history of trauma to the site. The patient gave a history of a visit to a local dental clinic on account of the painful swelling and was put on a course of antibiotics following root canal treatment presuming the swelling to be a periapical infection from the upper left central and lateral incisors. No relief in the symptoms was reported.

The patient has been taking periodic non-steroidal anti-inflammatory medication to get relief from the pain but with no effect. An intraoral examination revealed a well-defined solitary swelling on the anterior part of the hard palate behind the upper left central and lateral incisors (Fig. 1). On palpation, the swelling was firm to hard, non-fluctuant and tender. The related teeth showed grade I mobility and appeared tender on percussion. Both the teeth had temporary restorations placed on their crowns, following the root canal treatment that was performed earlier. Intraoral periapical radiograph



Fig. 1: Intraoral photograph showing a well defined swelling in the anterior maxilla, behind the left central and lateral incisors

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(IOPAR) and cone beam computed tomography (CBCT) revealed a lesion with well-defined margins, around 1.5 cm in diameter. It was radiolucent with multiple patchy areas of radiopacity within (Fig. 2). Displacement of the roots of both the involved teeth was observed. Based on the clinical and radiographic findings, a differential diagnosis of osteoid osteoma, osteoblastoma, and ossifying fibroma was considered. Since the root outlines of both the involved teeth were normal on IOPAR, benign cementoblastoma was not considered. The lesion was excised under local anesthesia. The excised specimen appeared to be a solid mass, hard in consistency, and measured 1.5 cm in diameter (Fig. 3). After decalcification and processing, sections were prepared and stained with hematoxylin and eosin stains. Histopathological examination revealed a lesion composed of numerous irregular trabeculae of lamellar and woven bone, of varying sizes in a highly vascular fibrous connective tissue stroma. The trabeculae were lined by plump osteoblasts which were round to ovoid in shape showing

hyperchromatic nuclei (Fig. 4). Numerous large osteocytes were seen within the trabeculae. Many multinucleated giant cells (osteoclasts) were seen in lacunar spaces on the resorptive side, both singly and in small clusters (Fig. 5).

The above histopathologic features were common for both osteoid osteoma and benign osteoblastoma. But taking into consideration, the larger size of the lesion (1.5 cm), pain in relation to the lesion that was not relieved on medication, and prominent giant cells with increased stromal vasculature in histopathology, a final diagnosis of benign osteoblastoma was made. The patient was symptom-free on evaluation after 3 months.

DISCUSSION

Benign osteoblastoma is a rare primary tumor of bone characterized by the proliferation of osteoblasts in a highly vascular connective tissue stroma.³ It was first described by Jaffe and Mayer in 1932 under the title, "An Osteoblastic Osteoid Tissue-Forming Tumor of a Meta-

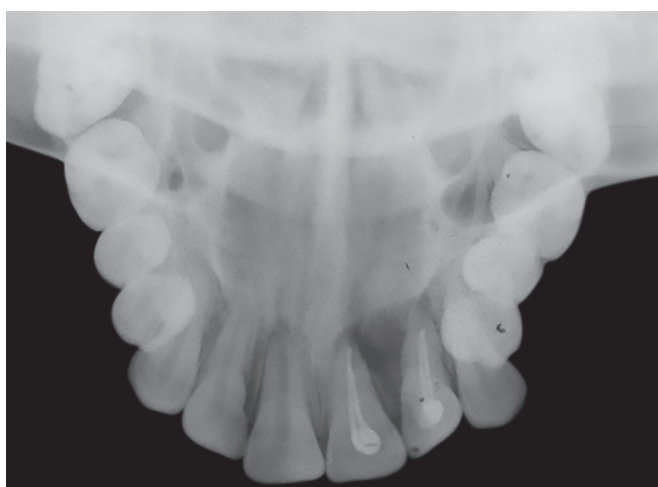


Fig. 2: Intraoral radiograph with an evident radiolucent lesion between the root-canal treated upper left central and lateral incisors

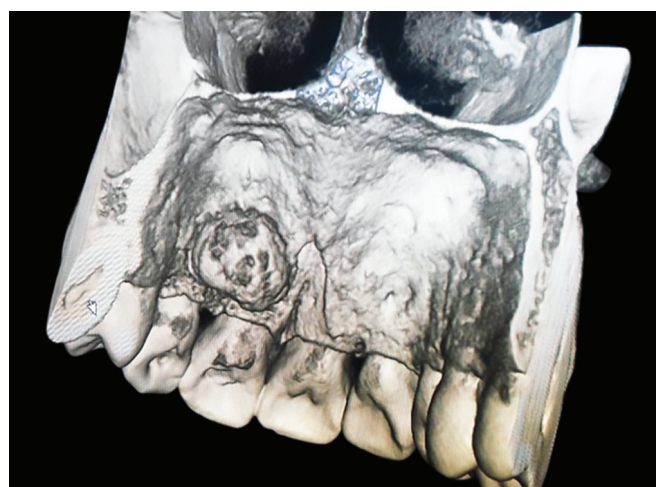


Fig. 3: Cone beam computed tomography image showing the lesion within the maxilla in relation to the upper left incisors



Fig. 4: Gross specimen of the excised lesion

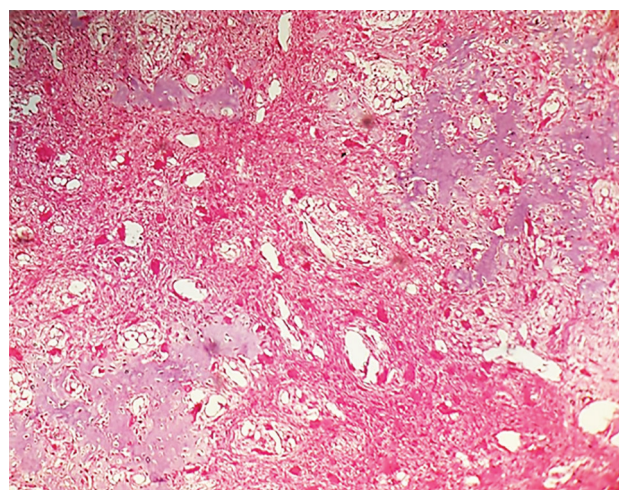


Fig. 5: Photomicrograph showing numerous multinucleated giant cells within the stroma (H and E; 10X)

carpal Bone".⁴ It was subsequently named "Osteogenic Fibroma" by Lichenstein in 1951 and "Giant Osteoid Osteoma" by Dahlin and Johnson in 1954.⁵⁻⁷ Jaffe and Lichenstein in 1956 proposed the terminology "Benign Osteoblastoma", which has since become the most widely accepted designation for this tumor.^{8,9} The first case of BO of the jaws was reported by Borello and Sedano in 1967.¹⁰ The vertebral column, long bones and small bones of the hands and feet are the sites of predilection of BO. Its occurrence in jaw bones is rare, making up only around 10–15% of all osteoblastomas. A search of related English literature shows that mandible is more frequently involved than maxilla among the reported cases of gnathic BO. Generally, BO affects young adults, with 90% of the cases arising in persons younger than 30 years of age with a mean age of 17 years. A definite male predilection has also been reported.^{1,2}

The true nature of this lesion though widely debated in the literature, still remains unclear. Jaffe and Lichenstein suggested that BO is a true neoplasm derived from osteoblast cells.^{8,9} Trauma, inflammation, localized alteration in bone physiology, and abnormal response to tissue injury are some of the other causes that have been cited in the literature as the etiologic factors of this tumor.¹¹

The clinical picture of BO comprises swelling localized to the tumor site and pain of varying intensity, the duration being just a few weeks to a year or more.¹² The pain in BO is relieved by salicylates, unlike in osteoid osteoma. The size of the BO is clinically and radiographically reported to range between 1.5 cm and 4 cm in diameter.¹³ A combination of radiolucent and radiopaque patterns is usually appreciated on the radiographs of BO. They appear as well-demarcated lesions showing a mixed appearance depending on the amount of bone produced.¹¹ Histologically, BO is a bone forming lesion characterized by numerous bony trabeculae showing varying degrees of calcification. These are paved by abundant plump proliferating osteoblasts. Moderate numbers of multinucleated giant cells are scattered in the tissue. The connective tissue is scanty and contains many dilated capillaries.^{12,13}

Due to its close resemblance with other bone lesions of the jaw, BO may often pose a diagnostic challenge. It needs to be differentiated from lesions like osteoid osteoma, ossifying fibroma, and even low-grade osteosarcoma.¹¹

Osteoid osteoma, unlike BO, rarely exceeds 1 cm in diameter and is characterized by severe pain out of all proportion to its small size, and is usually relieved by salicylates. At the microscopic level, osteoid osteoma lacks

giant cells and has less vascular stroma than BO.^{14,15} In ossifying fibroma, the proportion of fibrous connective tissue stroma will be much more than in BO, where the stroma is scanty and the major histologic hallmarks are numerous proliferating osteoblasts as well as more number of irregular trabeculae of new bone. Due to the presence of plump, actively proliferating osteoblasts and osteoid tissue, BO may be confused with low-grade osteosarcoma in histopathology. Careful microscopic examination to ascertain the absence of cellular atypia and lack of abnormally high as well as bizarre mitosis will aid in confirming the diagnosis of BO.¹³

Some reports state that BO treated by curettage or conservative surgical excision have a good prognosis with a recurrence rate of about 13.6%.¹¹

CONCLUSION

Benign osteoblastoma of the jaw is a rare-bone forming neoplasm which shows a close resemblance to other lesions like osteoid osteoma, ossifying fibroma, and low-grade osteosarcoma, thereby posing a diagnostic dilemma. A thorough history with the proper clinical, radiographic examination and histopathological examination can provide a clue to an accurate diagnosis. Surgical excision with adequate follow up is also essential.

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