Osteoblastoma of the jaws: report of a case and review of literature

Bhari Sharanesha Manjunatha,[1] Patel Sunit,[1] Mahajan Amit,[2] Shah Sanjiv[1]

[1]Department of Oral and Maxillofacial Pathology; [2]Department of Oral and Maxillofacial Surgery, KM Shah Dental College and Hospital, SumandeepVidyapeeth, Piparia, India

Abstract

Benign osteoblastoma (OB) is a rare tumor of bone representing less than 1% of all tumors of the maxillofacial region. Vertebral column, sacrum, long bones, and calvarium are more frequent sites of this tumor. Clinically, patients present with pain and swelling. Histologically, contain a well vascularized, osteoblastic connective tissue stroma and occasionally, osteoclasts along with osteoid and varying degrees of calcification, as well as immature bone may also be noted. The main aim of this report is present a case of OB occurring in the mandible and a brief review of English Medical Literature of this tumor. Many bone-producing lesions possess some overlapping clinical, radiographic or histopathological findings similar to osteoblastoma. Understanding and correlating all features helps in correct diagnosis and adequate management of this rare entity. Hence, osteoblastoma has to be differentiated from other bone lesions for correct diagnosis.

Introduction

Osteoblastoma (OB) is a benign tumor of bone characterized by osteoid and bone formation with the presence of numerous osteoblasts.[1] This tumor was first described by Jaffe and Mayer in 1932.[2] The current term *Benign Osteoblastoma* was proposed by Jaffe and Lichtenstein in two separate reports in 1956.[3,4] Osteoblastoma constitutes about 1% of all primary bone tumors. In approximately 15% of cases it is found in the maxillofacial skeleton, with a greater frequency occurring in the mandible.[5,6] Borello and Sedano reported the first case of osteoblastoma involving the jaws.[7] Very few cases of osteoblastomas occurring in the jaws have been reported in the literature. To the best of our knowledge, since the original description by Jaffe and Mayer in 1932, about 108 well documented examples of osteoblastoma arising both in the maxilla or mandible have been reported in the English literature till date.[8-10] Here we report one such rare case of OB occurring in posterior mandible and a brief review of English Literature.

Case Report

A 25-year-old man presented with chief complaint of a painless swelling in mandibular lower left back region since 3 years. The lesion was small and increased in the past 3 years to the present size. The clinical examination revealed a firm palpable hard mass, which was fixed to the underlying bone. The swelling is noted on the left posterior part of the ramus of the mandible with no facial symmetry. Intraoral examination showed a mass of about 5 × 4 cm in diameter and was well circumscribed. Overlying mucosa was intact and normal in color. Based on the patient history and clinical findings, a provisional diagnosis of cementoblastoma, osteoblastoma, ossifying fibroma was given. Orthopantomogram showed a round to oval radio opaque mass with a radiolucent border at the apex of left mandibular 1st molar (Figure 1). The lesion was excised under general anaesthesia and the surgical specimen comprised of both tooth and a hard tissue mass attached to it (Figure 2). Histologically, H and E stained sections revealed both hard and soft tissue mass mainly comprising of bone arranged both in trabacular as well as lamellar pattern with new bone formation and osteoid matrix in a highly vascular connective tissue stroma (Figure 3A and 3B). The lamellar pattern mainly consists of calcified cellular inclusive mainly osteocytes. Based on histopathological findings, the diagnosis of Osteoblastoma was confirmed.

Discussion

Osteoblastoma is a rare benign neoplasm of bone, recognized as separate entity and distinguished from osteoid osteoma. Though Osteoblastoma is considered as benign, the true nature of this lesion is unknown.[2] Jaffe and Lichtenstein suggested that this lesion is a true neoplasm of osteoblastic derivation.[3,4] Other investigators suggested that it occurs as a result of trauma or inflammation.[11,12] Smith *et al.*, have regarded this entity as an abnormal local response of the tissues to injury, or even possibly as a localized alteration in bone physiology, rather than as a true neoplasm.[13]

Osteoblastoma seldom involves the jaws and very few cases in the literature have been reported to occur in the jaws.[14] Osteoblastoma may be classified into cortical, medullary, and periosteal types. Osteoblastomas of jaws are either medullary or periosteal and not cortical which are common in extragnathic sites.[15] The mandible is affected more often than the maxilla with most mandibular lesions occurring in the body.[6]

Gordon et al have showed that 59% of osteoblastomas occur in men and 41% in women.[16] Mandible is affected more often than maxilla and most of the mandibular lesions tend to occur in the body region.[6] Age range varies between 5 years to 60 years. However, the most affected age group remains between 5 and 30 years.[10]

The systematic review of the English-language medical literature of OB of jaws showed only 108 cases published as various case reports as well as reviews.[8,9,10,17-22] PubMed and Medline search was done using the keywords *osteoblastoma*, *benign osteoblasoma*, *bone tumors* and *osteoblastoma of jaws* for the review of Literature.

The summary of clinical details such as age, gender, site and symptoms and radiographic findings of the previous cases of OB of jaws in the literature review are enlisted in Table 1.[8,9,17-22] Previous literature reviews had left out few cases in their study and the clinical data of the same had variations.[10] In the present review, an additional 13 cases have been included after the recent review of Literature.[17-22] Out of 108 cases excluding the present cases, the age range of patient was from 3 years to 78 years with the mean age of 22-23 years. The male to female ratio is almost equal (47% to 53%) with slight increase in females. Majority of the previously reported cases had swelling with pain or tenderness in 64%. Just swelling without any symptoms was noted in 21% of cases. About 10% of cases are asymptomatic and 5% cases have been report-

Correspondence: Bhari Sharanesha Manjunatha, Department of Oral and Maxillofacial Pathology, KM Shah Dental College & Hospital, SumandeepVidyapeeth, Pipariya-391760, Vadodara, India.
Tel. +91.02668.245266 - Fax: +91.02668.245292.
E-mail: drmanju26@hotmail.com

Key words: benign osteoblstoma, bone tumors, cementoblastoma, radiopaque mass.

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ed without any details about the nature disease. Eighty-one cases occurred in the mandible (74%) and twenty-nine cases are associated with the maxilla (26%). One case was seen both in the maxilla as well as posterior mandible bilaterally.[23] Of 110 cases, only 54 cases (49%) occurred in the posterior region and 9 cases occurred in the anterior region of both the jaws. However, 47 cases (43%) have not stated the location of occurrence the lesion in the jaws. The radiographic details of review of previous cases showed varied findings. Forty-three cases (39%) had mixed radiolucency and opacities with poorly and well defined borders. Radiolucent lesions were noted in 28 cases (25%) and 22 cases (20%) had radiopaque lesions. The details of radiographic findings in rest of the 12 cases (11%) were not available. All the above discussed data is summarized in Table 2.[8,9,10,17-22]

DeSouza and Frost[24] reviewed 24 cases of osteoblastoma, dividing their cases into three categories: i) Cortical osteoblastoma-synonymous with osteoid osteoma, showing a circum-

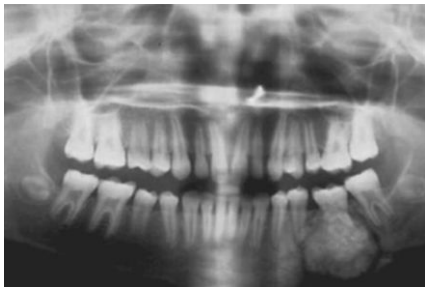

Figure 1. Orthopantomogram (OPG) showing a round to oval radio opaque mass with a radiolucent border at the apex of left mandibular 1st molar.

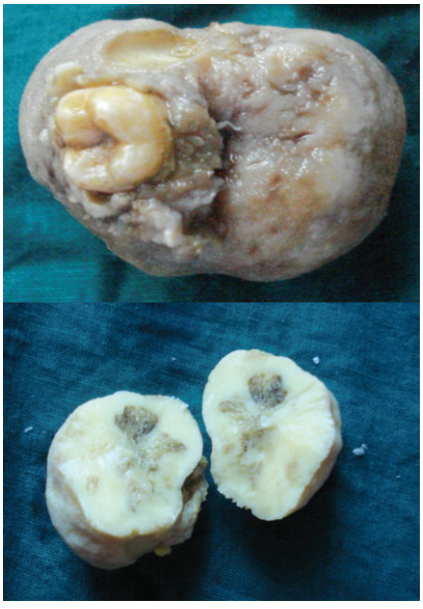

Figure 2. The surgical specimen comprising both tooth and an attached hard tissue mass.

Table 1. Details of English literature review of benign osteoblastoma cases.

Case	Authors	Year	Age*	Gender	Location	Symptom	Radiographic
1	Capodiferro, *et al.*[21]	2005	16	F	Mandible	Painful swelling	Mixed Poorly defined
2		2005	10	M	Mandible	Painful swelling	Mixed Poorly defined
3		2005	20	F	Mandible	Painful swelling	Mixed Poorly defined
4		2005	21	M	Mandible	Painful swelling	Mixed Poorly defined
5	Rawal, *et al.*[8]	2006	30	F	Parasymphsis body of Mandible	Painful swelling	Radiolucent Well-defined
6		2006	31	F	Body of Mandible	Painful swelling	Radiolucent Well-defined
7		2006	16	M	Maxilla, canine premolar	Painful swelling	Radiolucent Well-defined
8		2006	29	F	Body of Mandible	Painful swelling	Mixed Poorly defined
9		2006	18	F	Body of Mandible	Painful swelling	Non-description
10		2006	15	F	Body of Mandible	Painless swelling	Radiolucent Well-defined
11		2006	78	M	Body of Mandible	Painful swelling	Radiopaque
12	Angiero, et al.[22]	2006	8	M	Right posterior Maxilla	Painless swelling	Mixed Poorly defined
13		2006	24	M	Left posterior Mandible	Painless swelling	Mixed Poorly defined

Modified and Updated from Alvares Capelozza, *et al.*[10] and Jones, *et al.*[9] reports. F, Female; M, Male. *Age in years.

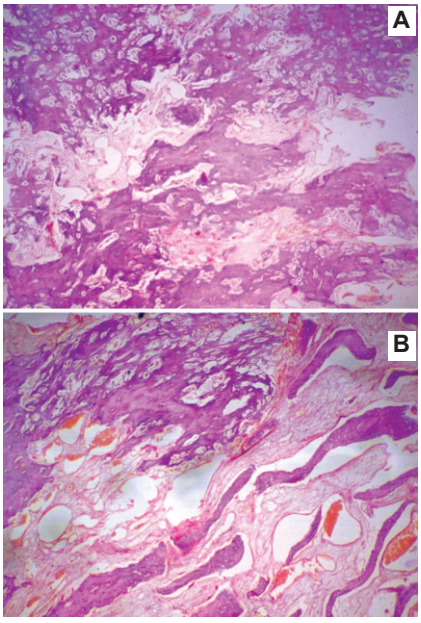

Figure 3. A) Photomicrograph showing Photomicrograph showing osteoid deposition with osteoblastic rimming and rich vascularity; B) Irregular woven bone in lace like pattern within a loose fibrocellular and vascular connective tissue stroma.

Table 2. Summary of 108 cases of osteoblastoma of the jaws reported in English literature till date.

Age	
Age range	3 to 78
Mean age	22.23
Gender	
Male	n=51(47.22%)
Age range	5 to 78
Mean age	22.04
Female	n=57(52.78%)
Age range	3 to 53
Mean age	22.42
Location	
Mandible	81(73.64%)
Maxilla	29(26.36%)
Posterior region	54(49.09%)
Anterior region	09(8.18%)
Not stated	47(42.73%)
Left side	36(32.73%)
Right side	18(16.36%)
Not stated	56(50.91%)
Symptom	
Asymptomatic	11(10%)
Swelling	23(20.91%)
Pain/tender	70(63.64%)
Not stated	6(5.45%)
Radiographic description	
Radiolucent	28(25.45%)
Radiopaque	22(20%)
Ground Glass	5(4.55%)
Mixed	43(39.09%)
Not stated	11(10%)
No finding	1(0.90%)

scribed appearance and striking perifocal bone reaction; ii) Spongious osteoblastoma-synonymous with little bone reaction; iii) Periosteal osteoblastoma-synonymous with poorly circumscribed, little bone reaction.

Smith *et al.*, have compared the osteoblastoma in gnathic and extragnathic sites, concluding that the lesion has similar clinical, radiologic and histologic features in both instances.[13] Osteoblastoma of the jaws when is associated with the roots of teeth, it is difficult to distinguish from cementoblastoma. Abrams *et al.* reported that the histologic similarities of cementoblastoma, osteoid osteoma, and benign osteoblastoma suggesting a close relationship among these three conditions.[25]

The attachment of the tumor to the roots of teeth should not be used as a hallmark for the diagnosis of cementoblastoma as osteoblastoma in the tooth-bearing area may envelop the roots of the involved teeth. However, the broad trabacular regions with limited cellularity seem to be a prominent feature, possibly unique to cemetoblastoma.[25] Furthermore, reversal lines that may impart a mosaic pattern to the calcified portion are a feature of cementoblastoma.

Osteoblastomas of the jaws are treated by curettage or local excision. En bloc resection is not commonly employed. Radiation should be considered only if surgical removal is not possible and there is evidence of continued aggressive behavior or multiple recurrences.[13] Most tumors are curetted and bone grafts are inserted in some cases.

Overall the prognosis of osteoblastoma is good. OB has to be differentiated from other bone producing lesions such as osteoid osteoma, osteosarcoma, cementoblastoma, ossifying fibroma etc.[26]

Yang C and Qiu WL have reported 24 new cases of osteoblastoma in the maxilla and mandible. Their results suggested that osteoblastomas are more common in females than previously reported. So, the overall percentage of osteoblastoma in females increased from 47.2% to 58.4%.[27]

Conclusions

Osteoblastoma has to be differentiated from other similar bone producing lesions like osteoid osteoma, osteosarcoma, cementoblastoma, ossifying fibroma etc for correct diagnosis. Complete surgical excision is only treatment available for osteoblastoma with an overall good prognosis.

A correct diagnosis helps in proper treatment planning. Many bone-producing lesions possess some clinical, radiographic or histopathological similarity to osteoblastoma. Understanding and correlating all these features is of utmost importance. This helps in correctly diagnosing and helps in adequate management of this rare entity, giving a good prognosis.

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