

Case Report

# Aggressive Osteoblastoma of the Palate: A Diagnostic Dilemma for Clinician and Pathologist

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## ABSTRACT

Osteoblastoma is a rare benign neoplasm of the bone commonly affecting long bones, vertebrae. Oral cavity is a rare site for its occurrence. It is known for its extremely aggressive behaviour with high potential for local recurrence and invasion, in which case it is termed as aggressive osteoblastoma. It is diagnostic dilemma for the clinician and pathologist owing to its difficulty in differentiating from the benign and malignant bone tumours which shows similar characteristics in its clinical, histological and radiological appearance. Here we report a rare case of an aggressive osteoblastoma on the hard palate with a brief review.

**Key words:** Osteoblastoma, low grade osteosarcoma, aggressive osteoblastoma, osteoid osteoma.

## INTRODUCTION

The term osteoblastoma was first described by Jaffe and Mayer (1932).<sup>[1]</sup> Osteoblastoma was independently termed by Jaffe and Lichtenstein (1956) under the name “benign osteoblastoma” to identify osteoblastic osteoid tissue forming tumour similar to osteoid osteoma but exhibiting greater potential growth.<sup>[2]</sup> This was the name that had been adopted by the World Health Organization Classification of Bone Tumours<sup>[3]</sup> and the Armed Forces Institute of Pathology.<sup>[4]</sup> Other names, such as giant osteogenic fibroma<sup>[5]</sup> and giant osteoid osteoma<sup>[6]</sup> have also been proposed. Conventional osteoblastoma (CO) are very rare benign tumour which corresponds to 3% of all benign bone neoplasias<sup>[7]</sup> and less than 1% of all primary bone tumours.<sup>[8-10]</sup> Predominantly seen in males during 2<sup>nd</sup> decade of life. Most commonly involved sites are vertebral column and long bones.<sup>[8,11]</sup> Face, extremities and skull cap bones are less frequently involved sites.<sup>[7]</sup> In the

head and neck region 10-12% of CO involves maxillofacial skeleton, and mandible is the most common site to be reported.<sup>[8,12]</sup> especially the posterior segments.<sup>[13]</sup> Herein, we report a rare case of aggressive osteoblastoma affecting the hard palate in a 47 year old patient which rendered challenge for us in diagnosis due to close differentials.

## CASE REPORT

A 47 year-old female patient reported to our Dental College Hospital with the chief complaint of swelling on the upper left back region of the oral cavity since three months. Initially the swelling was small, which had gradually increased in size, attaining the present dimensions. Patient never experienced any pain but had problems in chewing and swallowing. No history of trauma was elicited. Dental history revealed that patient had undergone multiple uneventful extractions. Medical history revealed patient was known diabetic

and hypertensive. Family history was non-contributory. Intraoral examination revealed the presence of a solitary, well defined, non tender, bony hard growth on the palatal aspect in relation to teeth 24, 25, 26 extending medially to point 2 to 3cm lateral to the midpalatine suture and laterally to the buccal aspect up to the mucogingival junction measuring 3x2cm with mobility in relation to teeth 25, 26, 27 (Figure 1) and a sessile growth over the occlusal aspect in relation to tooth 25 measuring over 0.5 x0.5cm (Figure 2) since 1 month. The intra oral periapical radiograph (Figure 3) revealed a single, well defined bony lesion on the palatal aspect in relation to teeth 24, 25, 26. Radiographic differential diagnosis of Central ossifying fibroma, Periapical/focal cement osseous dysplasia was made.



Figure 1: Intraoral clinical photograph showing presence of a solitary well defined bony hard growth on the palatal aspect.



Figure 2 : Intra oral clinical photograph showing sessile growth over the occlusal aspect.



Figure 3: The intra oral periapical radiograph showing a single, well defined bony lesion on the palatal aspect.

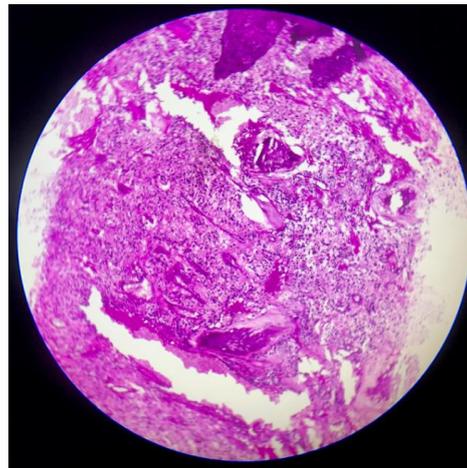


Figure 4: Histopathological examination showing necrotic bone

The lesion was surgically excised under local anesthesia along with the extraction of teeth 25, 26 and rehabilitated with palatal plate later. The excised mass was sent for histopathological examination. Histopathological examination of the resected specimen revealed a loosely arranged connective tissue stroma with sequestrum, involucrin, and wide areas of necrosis interspersed with chronic inflammatory cells (Figure 4). Adjacent areas showing myxoid changes, chondroid areas were seen. Based on the clinical, radiographical and histopathological findings, the final diagnosis of chronic nonspecific osteomyelitis was rendered and patient was kept under follow up.

Six months later patient reported to Department of Oral and Maxillofacial surgery with a recurrence in the same region which was again surgically excised under local anesthesia and the mass was sent for histopathological examination (Figure 5).

Histopathological examination revealed highly cellular and fibrous connective tissue stroma with numerous plump proliferating cells. Numerous bony trabeculae with osteocytes are found to be distributed in connective tissue stroma. Focal areas of cartilaginous tissue and overlying stratified squamous epithelium were also evident. Biopsy was suggestive of aggressive ossifying fibroma and patient was kept under follow up. (Figure 6)



Figure 5: gross specimen of the curetted lesion.

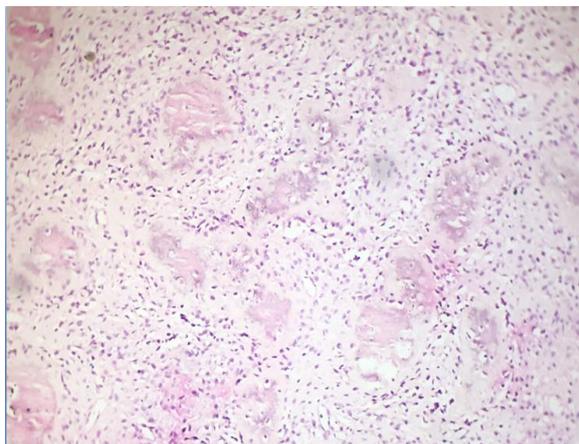


Figure 6: Photomicrograph of histopathological section reveals highly cellular and fibrous connective tissue stroma with numerous plump proliferating cells and bony trabeculae with osteocytes.

One year later patient again reported back to Department of Oral and Maxillofacial surgery with a recurrence in the same region. Investigations like CBCT were carried out. CBCT image revealed a lesion of mixed radio-density on upper left maxillae extending anterioposteriorly from 24 region to the region of maxillary tuberosity and mediolaterally from buccal cortex till midpalatine region. Predominant part of mixed lesion was seen extending

palatally (Figure 7A). The radio dense lesion appears to be well demarcated from underlying bone (Figure 7B). The palatal extension of the lesion showed multiple radiolucent areas (7A). The buccal cortical plate appeared to be lost (Figure 7A, 7B, 7C). Diagnosis of fibro osseous lesion on upper left maxillae was made based on CBCT findings. The radiological differential diagnosis included osteoblastoma, osteoid osteoma, ossifying fibroma, and low grade osteosarcoma. The lesion was excised completely in multiple fragments (Figure 8) including the overlying mucosa along with the extraction of premolar tooth and the mass was sent for histopathological examination. On gross examination the excised tissue was irregular in shape, greyish white in colour measuring about 5.5 cm in its greatest diameter. The tumour had a gritty consistency with softer parts and evidently firmer areas of bone tissue.

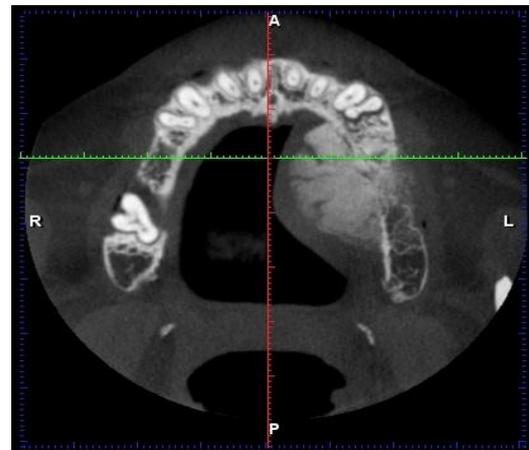


Figure 7A: Axial CT image showing mixed radio-density mass along the posterior part of upper left maxillae.

Histopathological examination revealed highly cellular connective tissue stroma with large plump proliferating cells. Irregular deposits of osteoid and trabeculae of bone is seen lined by plump large epithelioid like osteoblasts with eccentrically placed nuclei (Figure 9). Striking feature was the sheets and groups of round to polygonal cells showing hyperchromatic nuclei and prominent nucleoli (Figure 10). Focal areas of cartilaginous tissue were also evident

(Figure 11). The histopathological examination of the excised mass confirmed the final diagnosis of aggressive osteoblastoma. Patient is kept under strict follow up due to aggressive nature and to evaluate any sign of the lesion at the site of excision.

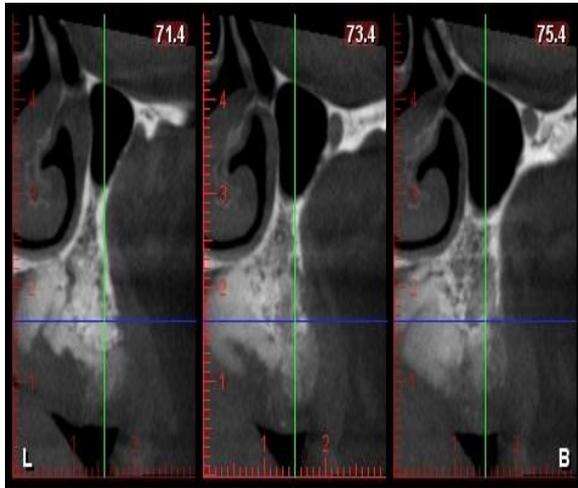


Figure 7B: Coronal view of the lesion.



Figure 7C: 3D CBCT image showing the lesion in the left posterior maxillary region.



Figure 8: gross specimen of the resected lesion.

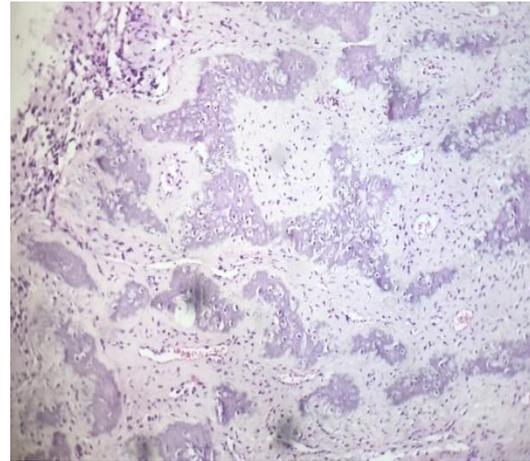


Figure 9: Photomicrograph of histopathologic section showing highly cellular connective tissue stroma with large plump proliferating cells. Irregular deposits of osteoid and trabeculae of bone is seen lined by plump large epithelioid like osteoblasts with eccentrically placed nuclei.

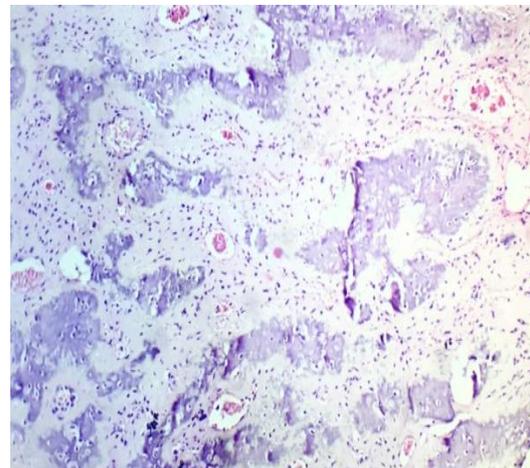


Figure 10: Photomicrograph of histopathologic section showing sheets and groups of round to polygonal cells showing hyperchromatic nuclei and prominent nucleoli.

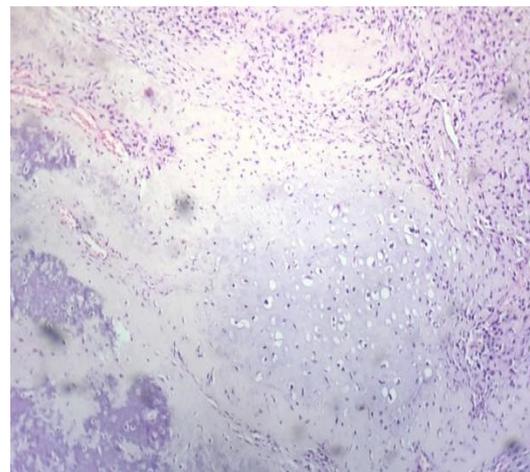


Figure 11: Photomicrograph of histopathologic section showing focal areas of cartilaginous tissue.

**Table 1: Comparison of conventional osteoblastoma (CO), aggressive osteoblastoma (AO) and osteoblastoma - like osteosarcoma (OB-like OS).**

Type of lesion	Tumor borders	Osteoid/woven bone	Intertrabecular	Nuclear Features	Mitosis	Cytologic features
conventional osteoblastoma	Well- demarcated	Long anastomosing trabeculae Solid or lace like Osteoblastic rimming	High vascularity	Round- oval nucleus	Rare to absent	Polygonal Moderate amount cytoplasm
aggressive osteoblastoma	Peripheral maturation without sharp demarcation	Irregular and wider trabeculae	Less vascular	Round to oval eccentric placed	Occasional typical mitosis	Epithelioid Eosinophilic cytoplasm
osteoblastoma-like osteosarcoma	Host bone Infiltration	Irregular trabeculae Anaplastic cells within osteoid	Highly cellular stroma	Round to oval, spindle Hyperchromatic nucleus	Occasional atypical Mitoses	Deeply stained cytoplasm

## DISCUSSION

Osteoblastoma of the jaw bones was first well documented by Borello and Sedano (1967).<sup>[14]</sup> In the review conducted by Dorfman (1972), out of 23 osteoblastoma cases, 4 were examined which exhibited aggressive behaviour with recurrence and locally destructive growth pattern.<sup>[15]</sup> In 1984 histologically it was named as “aggressive osteoblastoma” (AO) as an entity of borderline osteoblastic tumors and suggested that the presence of epithelioid osteoblasts was a characteristic histological feature in these variants.<sup>[16]</sup> The term 'aggressive' osteoblastoma was put forward by Dorfman as a rare tumour that represents a borderline lesion between benign osteoblastoma and osteosarcoma which have the tendency to recur, but do not metastasize, and are characterized microscopically by the presence of epithelioid osteoblasts.<sup>[17]</sup> The incidence and distribution of AO are unknown due to rare occurrence.<sup>[18]</sup> Gnathic AO reports are relatively rare.<sup>[19-21]</sup> Osteoblastoma exhibiting atypical histopathological features, locally aggressive behaviour and recurrence potential always puts the clinician and pathologists in diagnostic dilemma owing to its difficulty in differentiating from low grade osteosarcoma.<sup>[22]</sup>

The tumour of interest which is always demanding for a pathologist to give a definitive diagnosis are AO, CO and osteoblastoma - like osteosarcoma (OB-like OS) due to close overlap among these bone lesions.<sup>[8,23,24]</sup> (Table 1)

AO usually exceeds >4 cm whereas conventional osteoblastomas normally do not exceed 4 cm in diameter.<sup>[25]</sup> Our case had involvement of upper jaw with swelling of 5.5 cm in diameter. The aggressive osteoblastoma affects the advanced age group, whereas benign osteoblastoma most commonly seen during the third or fourth decade.<sup>[8]</sup> Our patient was 47 year old at the time of presentation. In 1985 Bertoni et al<sup>[11]</sup> reviewed 17 patients with age ranging from 11 to 58 yr old patients with osteosarcomas that histologically resembled osteoblastomas.<sup>[23]</sup> In 1993 another group of 11 patients aged between 11 to 47 years were reported from the Rizzoli Institute.<sup>[24]</sup> OB like OS is low grade osteosarcoma which radiographically exhibits lytic to sclerotic with well defined borders typical for osteoblastoma or ill defined borders with cortical destruction suggesting malignant process.<sup>[23,24]</sup> Imaging of our case revealed mixed radio-density lesion on upper left maxillae extending anteroposteriorly from upper left 1<sup>st</sup> premolar to the region of maxillary tuberosity and mediolaterally from buccal cortex till midpalatine region. Predominant part of mixed lesion was seen extending palatally. The radiodense lesion appears to be well demarcated from underlying bone. The palatal extension of the lesion showed multiple radiolucent areas. The buccal cortical plate appeared to be lost suggesting the aggressiveness of the lesion.

Sometimes we often get confused mainly because of the presence of atypical histopathological features like plump or

bizarre appearing osteoblast-like cells in aggressive osteoblastoma, imitating the atypical cells of osteosarcoma. In such scenario, the diagnosis is greatly facilitated by careful evaluation of the histopathological, clinical behaviour and radiological features. [25] Treatment of choice for AO is complete surgical resection with reconstruction and prosthetic replacement as needed. [19] Other treatment strategies recommended are post-surgical radiotherapy and/or chemotherapy. [26] According to Gordon et al, the recurrence rate potential for aggressive osteoblastoma is 50% and for conventional osteoblastoma is 13.6%. [9] Long term follow-up is mandatory due to the possibility of high chance of recurrence and to rule out the tumour undergoing malignant transformation.

In conclusion, we have presented a case of aggressive osteoblastoma of the palate where it was presented initially with loosening of teeth and swelling mimicking osteomyelitis or dental infection. Even though the cause was removed, multiple recurrences were observed in the above case, finally diagnosing it as aggressive osteoblastoma due to clinico- radiological and histopathological correlation. In a scenario such as in our case the patient should be under periodic observation to look for whether there is any recurrence. Appropriate diagnosis is very important step in order to plan treatment. Wrong diagnosis or misdiagnosis of these bone lesions can lead to recurrence and further complications.

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