



## Aggressive Osteoblastoma of the Mandible- A Journey of 2 Rare Cases

Deepthi Shetty<sup>1-a\*</sup>, Harshini B<sup>2-b</sup>, Anil Kumar Desai<sup>1-c</sup>, Niranjn Kumar<sup>1-d</sup>

<sup>1</sup> Department of oral and maxillofacial surgery, SDM Craniofacial surgery and research Centre, SDM college of dental sciences and hospital, A Constituent unit of Shri Dharmasthala Manjunatheshwara University, Sattur, Dharwad, Karnataka, India.

<sup>2</sup> Department of oral and maxillofacial surgery, Sibar Institute of Dental sciences, Takellapadu, Guntur, Andhra Pradesh, India.

\*Corresponding author

### Research Article

#### History

Received: 11/01/2023  
Accepted: 12/06/2023

#### License



This work is licensed under  
Creative Commons Attribution 4.0  
International License

### ABSTRACT

Bone tumours are a distinct entity among which osteoblastoma is a rare variant constituting less than 1% of all bone tumours. The bones most commonly involved include the backbone, sacrum, calvaria, long bones and the small bones of the hand and feet. Osteoblastomas have only 15 % occurrence in the head and neck region with a higher incidence in the mandible. Osteoblastomas are known to have a benign or conventional form and the aggressive form which exhibits locally aggressive behaviour and a higher proneness for recurrence. We have reported the journey of two rare cases of aggressive osteoblastoma in the mandible. The treatment was planned considering the aggressiveness of the lesion and the reconstruction was carried out with free fibula graft to maintain the aesthetics, function, and to achieve a good quality of life for the patients.

**Key words:** Fibula, mandibular prosthesis şplantation, osteoblastoma.

a [kdeepthishetty@gmail.com](mailto:kdeepthishetty@gmail.com)  
c [anildes2006@yahoo.co.in](mailto:anildes2006@yahoo.co.in)

<https://orcid.org/0000-0001-5999-2479>  
 <https://orcid.org/0000-0003-1513-2383>

b [harshinib27@gmail.com](mailto:harshinib27@gmail.com)  
d [dniranjankumar@sdmmmedicalcollege.org](mailto:dniranjankumar@sdmmmedicalcollege.org)

<https://orcid.org/0000-0002-7563-4476>  
 <https://orcid.org/0000-0002-4708-5801>

How to Cite: Shetty D, BH, Desai AK, Kumar N. (2023) Aggressive Osteoblastoma of the Mandible- A Journey of 2 Rare Cases, Cumhuriyet Dental Journal, 26(2): 216-219.

### Introduction

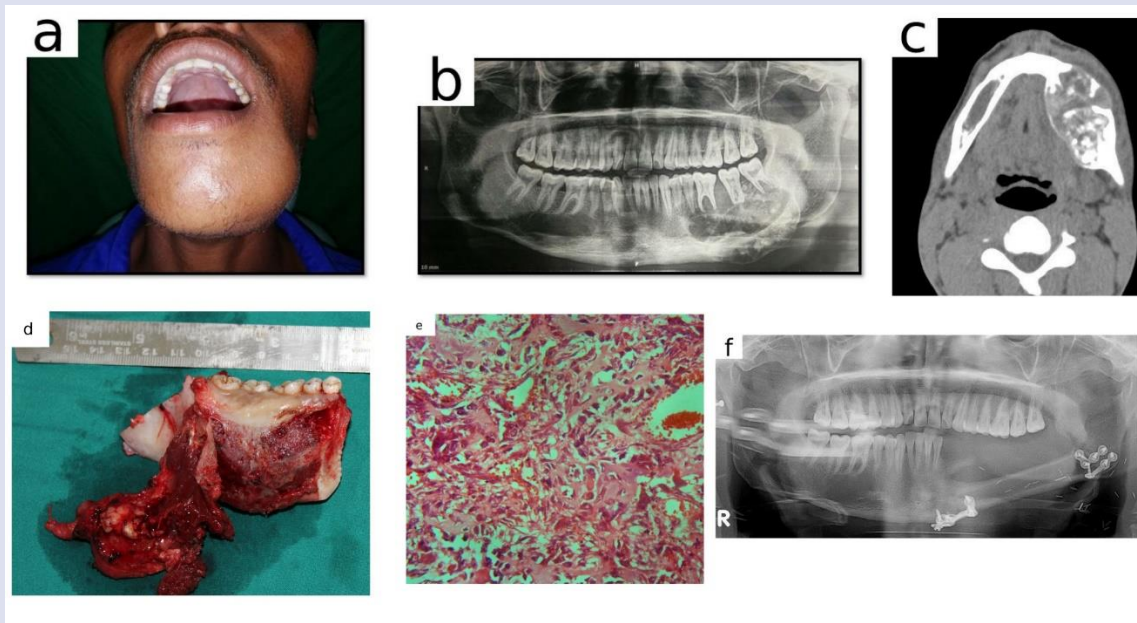
Lichtenstein and Jaffe first described osteoblastoma as a distinct neoplasm and uncommon primary bone tumour in 1956.<sup>1</sup> Osteoblastoma constitutes less than 1% of all the osseous tumours and most commonly involves the long bones, sacrum, calvaria, spinal column, and small bones of the hand and feet.<sup>2</sup> In the head and neck area it has an incidence of 10-15% and higher propensity to occur in the mandible.<sup>2</sup> Patients in their first three decades of life are usually affected, with male to female ratio of 2:1.<sup>3</sup> The two major clinicopathological forms of osteoblastoma are the conventional or benign form and the aggressive form which exhibits locally aggressive behaviour with a tendency to recur.<sup>4</sup> We have reported 2 rare cases of aggressive osteoblastoma of the mandible which was managed aggressively and was functionally and cosmetically rehabilitated.

### Case Report-1

23-year-old male reported to our department with the chief complaint of pain and swelling in the left lower 3rd of the face. On extraoral examination, a well-defined solitary swelling in the left lower third of the face was

noted which was bony hard, and tender on palpation (Figure 1a). Orthopantomogram (OPG) showed a well-defined osteolytic lesion with areas of radiopacities within the lesion extending from left premolar to 3rd molar region (Figure 1b). Contrast-enhanced Computerized tomography (CECT) showed a fairly well-defined large expansile lytic lesion, with thin internal septations and few areas of ill-defined cortex noted representing cortical break involving the mandible on the left side measuring approximately 5.5(AP) \*4.4(CC)\*3.5(ML) (Figure 1c).

Incisional biopsy of the lesion was done which was suggestive of aggressive osteoblastoma. The patient underwent wide excision of the lesion and segmental mandibulectomy and the defect was reconstructed with a free fibula graft (Figure 1d). The final histopathology report showed proliferative epithelioid osteoblasts arranged in sheets and overlying broad bony trabeculae rimmed by these osteoblasts with many osteoclastic giant cells which confirmed the diagnosis of aggressive osteoblastoma (Figure 1e). The patient had an uneventful postoperative course and is on regular follow-up with no evidence of recurrence till to date (Figure 1f).

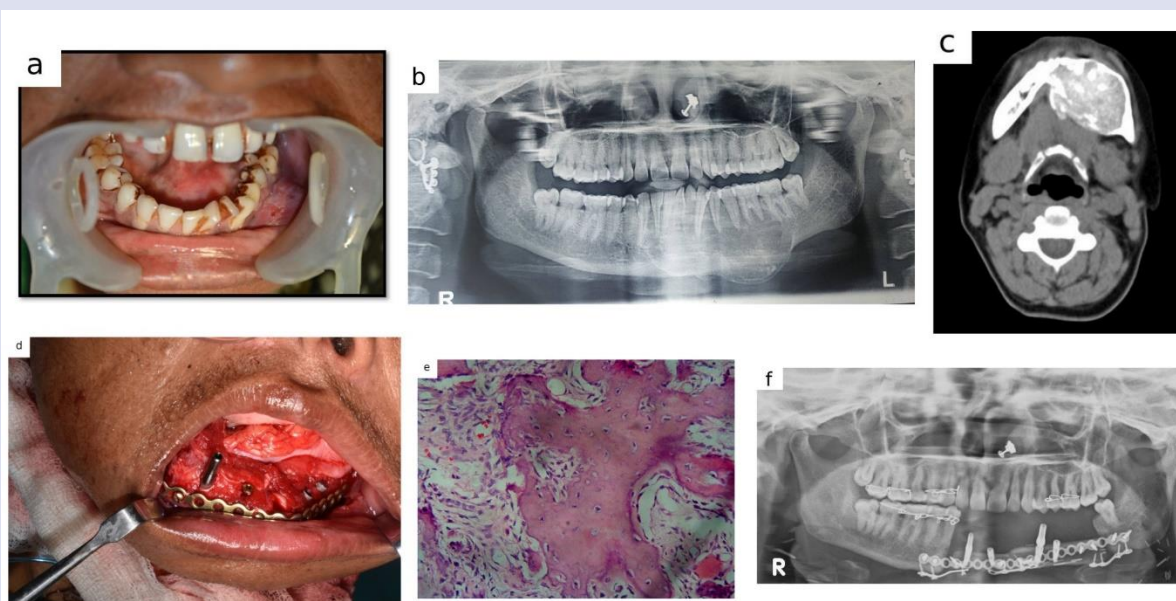


**Figure 1.** a) Extraoral clinical photograph shows facial asymmetry  
 b) Orthopantomogram (OPG) showing ill-defined lesion with various radiopaque focal areas with disruption of inferior border of mandible  
 c) Contrast enhanced Computed tomography (CECT) axial section shows expansile intramedullary lesion  
 d) Resected portion of mandible showing the tumour  
 e) Haematoxylin & Eosin-stained specimen shows highly cellular and vascular proliferative connective tissue consisting of epithelioid cells with abundant multinucleated osteoclast like giant cells and large areas of haemorrhage is evident throughout the lesion  
 f) Post-operative OPG.

### Case Report-2

35-year-old female reported to our department with swelling in the left side of the face for 4 months. On examination, there was restricted mouth opening and the swelling was firm to hard in consistency with bi cortical expansion extending from left mandibular canine to 2nd molar (Figure 2a). OPG revealed an ill-defined radiolucent lesion in the left body of the mandible with focal radiopaque areas with disruption of the inferior border of the mandible (Figure 2b). CECT revealed a large expansile intramedullary lesion with ground glass matrix, calcification noted involving the symphysis and body of the mandible on the left side associated with cortical expansion and significant thinning measuring 3.5cm x 5.0cm x 3.2cm (AP x ML x CC) (Figure 2c).

The patient underwent an incisional biopsy of the lesion which was suggestive of aggressive osteoblastoma. She underwent wide excision of the lesion with segmental mandibulectomy followed by reconstruction with free fibula graft and rehabilitation was done by immediate placement of implants (Figure 2d). The final histopathology report revealed the interconnecting bony trabeculae of coarsely woven bone rimmed by relatively plump osteoblasts to form an anastomosing net-like pattern. Many multinucleated osteoclast-like giant cells are also present along the bony trabeculae confirming the diagnosis of aggressive osteoblastoma (Figure 2e). The patient had an uneventful postoperative course and is on regular follow-up with no evidence of recurrence till to date (Figure 2f).



**Figure 2.** a) Intraoral examination shows swelling with bi-cortical expansion  
 b) OPG showing radiolucent lesion extending from canine to molar with disruption of inferior border of mandible  
 c) CECT scan (axial section) shows large intramedullary expansile lesion with calcification noted involving the symphysis and body of the mandible  
 d) Reconstruction was done using free fibula flap followed by immediate implant placement  
 e) Haematoxylin & eosin-stained final specimen shows interconnecting bony trabeculae of coarse woven bone rimmed by plump osteoblasts. Many multinucleated osteoclast-like giant cells present along the bony trabeculae. Connective tissue has multiple thin-walled blood capillaries  
 f) Post-operative OPG.

## Discussion

Jaffe and Lichtenstein in 1956 described osteoblastoma as a rare vascular osteoid and bone-forming benign tumour which is characterised by abundant osteoblasts.<sup>5</sup> Even though osteoblastoma comprises less than 1% of all the osseous tumours, it has an incidence of 10-15% in head and neck region with increased chances of occurring in the mandible.<sup>2,5,6</sup> Both our patients were in their first three decades of life and had lesions occurring in the posterior mandible.

The pathologic entity of 'aggressive' osteoblastoma was first proposed by DORFMAN in 1973.<sup>7</sup> Osteoblastoma is classified into two major clinicopathological forms which includes the Conventional osteoblastomas (CO) and the Aggressive osteoblastoma (AO). CO grow slowly not exceeding 4 cm in diameter and have restricted growth potential.<sup>2,7</sup> They have a well-defined sclerotic margin and are completely excised. Aggressive osteoblastoma (AO) usually exhibits locally aggressive behaviour and has a diameter of more than 4cm. Clinically they present mainly with pain, swelling, and expansion of the bony cortex.<sup>1</sup> Both our patients exhibited similar symptoms. Mobility of the teeth and pain are the early manifestations of these tumours, thus may be misdiagnosed as odontogenic infections or osteomyelitis.<sup>7</sup> Radiographically AO present a variety of patterns. It may be characterised by an osteolytic lesion with focal radiopacities and cortical expansion, or as sclerotic masses with well or ill-defined

borders.<sup>7,8</sup> Our patients also exhibited expansile osteolytic lesion with areas of radiopacities. AO may also be seen as a radiopaque nidus, within a radiolucent lesion, similar to osteoid osteoma.<sup>3</sup> The histopathological pathognomonic feature of AO is plump epithelioid osteoblasts larger than CO, with prominent nuclei and nucleoli, abundant and slightly eosinophilic cytoplasm. They have increased multinucleated giant cells of osteoclast type with increased mitotic activity and non-trabecular sheets or lace-like areas of osteoid production.<sup>5,9</sup>

AO has to be differentiated from conventional osteoblastoma and osteosarcoma to plan accurate treatment for the patient and to prevent recurrence.<sup>2,3</sup> AO has locally aggressive behaviour and a propensity to recur, hence aggressive treatment is recommended including surgical resection with safety margins.<sup>2</sup> Such aggressive treatment could result in complex facial defects which requires reconstruction to achieve functional and aesthetic rehabilitation.<sup>2</sup> Hence, we have managed our patients too aggressively with segmental mandibular resection and functionally and cosmetically rehabilitated using free fibula graft.

Gordon et al emphasizes the importance of long-term follow-up as the recurrence rate for conventional osteoblastoma is 13.6%, whereas for aggressive osteoblastoma is 50%.<sup>3,10</sup>

## Conclusions

To summarize, we have described the journey of 2 rare cases of aggressive osteoblastoma of the mandible which was diagnosed accurately and managed aggressively. Both the patients were successfully rehabilitated functionally and cosmetically thereby improving their quality of life.

## Conflict of Interest

The authors have no relevant financial or non-financial interests to disclose.

## Acknowledgments

None

## Funds

The authors declare that no funds, grants, or other support were received during the preparation of this manuscript.

## References

1. Capodiferro S, Maiorano MSE, Giardina C, Lacaita MG, Muzio LL, Favia G et al. Osteoblastoma of the mandible: clinicopathologic study of four cases and literature review. *Head & Neck* 2005; 27:616-621. PubMed ID: 15880408
2. Castro PHS, Molinari DL, Stateri HQ, Borges AH, Volpato LER. Aggressive osteoblastoma in a seven-year-old girl's mandible: Treatment and six-year monitoring. *International journal of surgery case reports* 2016; 27:5-9.
3. Pontual MLA, Pontual AA, Gempel RG, Campos LRM, Costa ALL, Godoy GP. Aggressive multilocular osteoblastoma in the mandible: A rare and difficult case to diagnose. *Brazilian dental journal* 2014; 25:451-456. PubMed ID: 25517784
4. Mardaleishvili K, Kakabadze Z, Machavariani A, Grzelidze T, Kakabadze A, Sukhitashvili N, et al. Benign osteoblastoma of the mandible in a 12 year old female: A case report. *Oncology letters* 2014; 8:2691-2694.
5. Pamuk AE, Yesilirmak A, Kuscu O, Kosemehmetoglu K. A rare case of aggressive osteoblastoma in maxilla. *Oral Surgery* 2017; 10:152-156.
6. Harrington C, Accurso BT, Kalmar JR, Iwenofu OH, Agarwal A, Allen CM. et al. Aggressive osteoblastoma of the maxilla: A case report and review of the literature. *Head and neck pathology* 2011; 5:165-170.
7. Vigneswaran N, Fernandes R, Rodu B, Baughman RA, Siegal GP. Aggressive osteoblastoma of the mandible closely simulating calcifying epithelial odontogenic tumor: Report of two cases with unusual histopathologic findings. *Pathology-Research and Practice* 2001; 197:569-576.
8. Capellozza ALA, Dezotti MSG, Alvares LC, Fleury RN, Santana E. Osteoblastoma of the mandible: systematic review of the literature and report of a case. *Dentomaxillofacial Radiology* 2005; 34:1-8.
9. Lypka MA, Goos RR, Yamashita DDR, Melrose R. Aggressive osteoblastoma of the mandible. *International journal of oral and maxillofacial surgery* 2008; 37:675-678.
10. Gordon SC, MacIntosh RB, Wesley RKO. A review of osteoblastoma and case report of metachronous osteoblastoma and unicystic ameloblastoma. *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology* 2001;91:570-575.