

Treatment of Mandibular Hemangioma-like Osteoblastoma via Resection and Immediate Replantation of the Segment: A Case Report

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Introduction: Vascular anomalies are extremely challenging lesions. Management of these lesions vary according to the nature of the lesion. **Case Report:** We represent an aggressive osteoblastoma that clinically bear a resemblance to vascular lesion. **Results:** Resection of the lesion and immediate reimplantation of the mandibular body was attained for the patient. 7-year follow up revealed complete healing of the lesion. **Conclusion:** Resection and Immediate Replantation of the Segment may be an appropriate option for the involved patients for an aggressive osteoblastoma with vascular component.

Keywords: Hemangioma-like Osteoblastoma, Immediate Replantation, Resection

Introduction

Benign osteoblastoma is a rare primary bone tumor that was first recognized as a distinct neoplasm by Lichtenstein (1) and Jaffe (2) in the same year, 1956. It is a vascular osteoid and bone-forming benign tumor characterized cytologically by the abundant presence of osteoblasts. It constitutes approximately 1% of all primary bone tumors (3). Its occurrence in the skull and jaw bones is relatively rare and represents only 15% of all osteoblastomas (4). Mandible is affected more frequently than the maxilla (4). The tumors range from 2 to 10 cm in diameter and radiologically appear as spherical calcified areas surrounded by a well-defined radiolucent zone (5). The tooth-bearing areas of the mandible are most affected. Painful tender swelling with expansion and erosion of the affected bone cortex may be seen depending on the site and size of the lesion (6). Benign osteoblastoma is a bone forming tumor with a histologically vascular rich scaffold, but (7). To extend of our knowledge, vascular anomalies in association with the aggressive osteoblastoma has not been reported yet. Here in we report a patient with clinically hemorrhagic lesion mimicking vascular malformation, but histologically resembles aggressive osteoblastoma. Resection of the manibular body and curettage of the lesion with immediate reimplantation of the resected segment was done for the patient. Seven-year follow up revealed complete healing of the lesion with functional bone formation in the region.

Report of a Case

A 7-year-old girl was referred to Taleghani hospital, department of oral and maxillofacial surgery, for the assessment of the left mandibular intraosseous lesion. Her dentist had noticed the extrusion and gradual mobility of deciduous tooth number 75, with continuous oozing of the blood from the gingival crevice. Mild facial asymmetry was observed in the left side during the facial examination. Extrusion and dental mobility on tooth 75 with a hemorrhagic, gingival granulomatous enlargement and a slight swelling of the left mandibular buccal cortex was demonstrated during the intra oral examination. No vascular bruit or thrill was detected but aspiration was positive in the clinical examination. The initial differential diagnosis suggested central giant cell granuloma, aneurismal bone cyst, and hemangioma or vascular malformation. Routine laboratory examination revealed that the hematologic factors were within the normal range. The initial panoramic investigation illustrated extensive multilocular radiolucency extending from tooth 73 to the inferior cortical border of left mandible, root resorption in tooth 36 and displacement of teeth bud (Figure 1). The mandibular CT scan confirmed the extent of the lesion, with erosion mainly at the expense of the buccal cortex and extension to buccal soft tissue. Severe hemorrhage after intra oral mucosal incision restricted the incisional biopsy. Hemostasis was achieved by packing and replacement of a buccal flap.

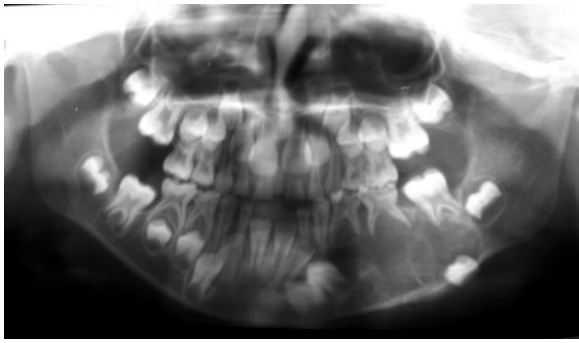


Figure 1. Multilocular radiolucent lesion in the left mandibular angle area

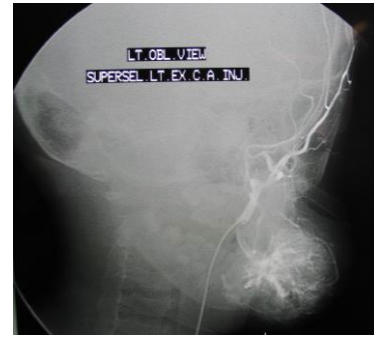


Figure 2. Subtractive digital angiography; note the engorgement of the internal maxillary artery branch to the lesion

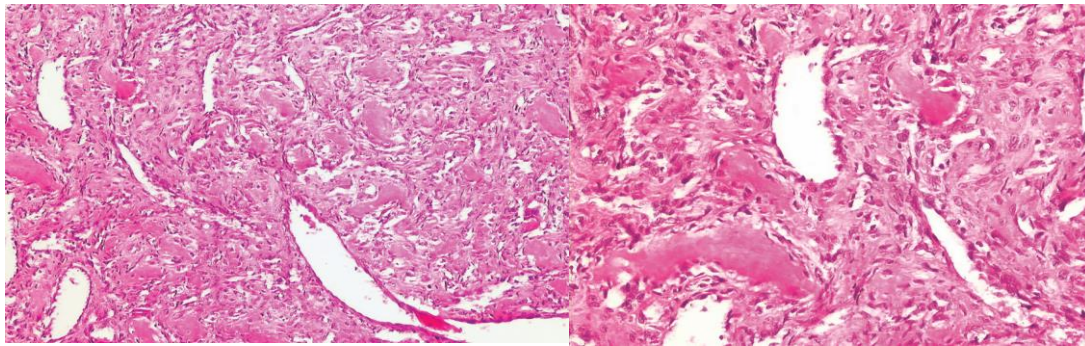


Figure 3. Massive osteoid tissue was seen in the vascular enrich scaffold with enormous active osteoblasts (H&E staining 100×), B. Active osteoblast and epithelioid cells was seen adjoining bony trabeculae (H&E staining 200×)

The patient was discharged once proper hemostasis was attained. Digital subtractive angiography bared the huge vascular proliferating tumor of the left mandibular body, which did not resemble vascular malformation or bony cavernous hemangioma. The radiologist suggested vascular proliferating tumor of the mandible that extended to the buccal soft tissue of the lesion (Figure 2). The patient was readmitted, transferred to operating room and the mandibular body with the safety margin of about 1 cm was resected due to the hemorrhagic nature and clinically vascular malformation like features of the lesion. Curettage of the lesion, extraction of involved teeth and immediate reimplantation of the resected mandible was accomplished based on the previous experience of the author (8, 9). Histologically decalcified sections of some areas of the resected material from patient showed multiple fragments of bony lesion characterized by presence of sheets and trabeculae of osteoid and calcified bone, which was lined by large and active epithelioid osteoblasts. Intermixed with bony trabeculae there are numerous vascular lumina with plump endothelial cells (Figure 3 A, B). Based on the histological examination the most probable diagnosis was an aggressive osteoblastoma. Postoperative assessment at 6 months confirmed excellent gingival healing, no signs of infection, and maintenance of facial symmetry and complete ossification of the re-implanted site. Up to 7 years follow

up revealed integration of re-implanted mandibular body and functional rehabilitation of the patient (Figure 4 A-C).

Discussion

Aggressive osteoblastoma is rare bone tumor and characterized by locally aggressive behavior with frequent recurrences, although metastatic spread is not seen. Aggressive osteoblastoma occupies a position between osteoblastoma and osteosarcoma in terms of biologic potential. The peak incidence is in the 3rd and 4th decades. 10 The clinical and radiographic features of aggressive osteoblastoma are also generally like osteoblastoma (10). The most consistently described microscopic feature of aggressive osteoblastoma is the presence of large epithelioid osteoblasts. The epithelioid osteoblasts line the surfaces of osteoid trabeculae and also form solid sheets within the intertrabecular (11). Other paraclinical and radiological data with unusual features of the hemangioma like scaffold advocated a hemangioma like lesion. The angiographic images of reported case did not match any type of vascular malformation or bony hemangioma; the histological examination showed excessive bone formation with epithelioid cells, resembling aggressive osteoblastoma.

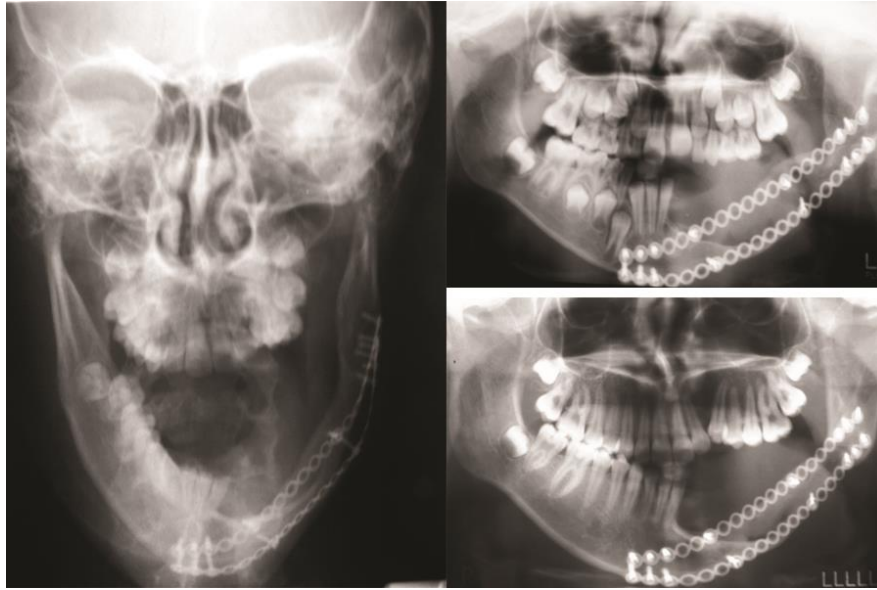


Figure 4. A-C) Immediate reconstruction of the mandibular segment via resection and immediate reimplantation. 6-month to 7-year follow up of the degenerative process of the healing of the lesion

Initial radiographic evaluation and clinical examination of this case showed similarity with vascular anomalies. Continuous gingival oozing and severe hemorrhage during the initial biopsy confirm the VM like nature of the lesion. Curettage of the resected fragment with immediate reimplantation does, however, reduce the morbidity associated with the procedure and the difficulty of reconstruction, seems to be the best approach (12).

Conclusion

Long term follow up showed, the selected approach for an aggressive osteblastoma with vascular component may be an appropriate option for the involved patients.

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Conflict of Interest: 'None declared'.

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