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RECURRENT AGGRESSIVE OSTEOBLASTOMA OF THE MAXILLA- A DIAGNOSTIC AND THERAPEUTIC CHALLENGE

Dental Science

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ABSTRACT

Aggressive osteoblastoma (AO) is a rare benign neoplasm of the bone commonly occurring in the vertebrae and long bones and rarely in the jaw bones[1]. It is locally invasive with high chances of recurrence compared to conventional osteoblastoma. Histologically AO is characterised by the presence of epitheloid osteoblasts. We present a case of a 48year old female with AO of the maxilla with four recurrences. The tumour could only be rightly diagnosed in the second recurrence. With other bone lesions having similar clinical, radiological and histopathological features, this lesion poses to be a diagnostic challenge to both the clinician and the pathologist.

KEYWORDS

aggressive osteoblastoma, maxilla, recurrence, differential diagnosis

INTRODUCTION

Osteoblastoma (OB) is a rare benign bone neoplasm characterized by highly proliferating osteoblasts[1]. Although benign, the aggressive variant raises concerns due to its potential for massive local destruction and tendency to relapse[2]. The incidence and distribution of aggressive osteoblastoma (AO) is currently unknown however occurrence in the jaws is extremely rare.

We present the case of a patient who underwent enucleation of a palatal bony mass followed by multiple recurrences over a period of 1 year. A definite histological diagnosis of AO could be obtained only after the second recurrence. Following a fourth recurrence, total resection of the tumour was performed along with irrigation using 5 Fluoro Uracil (5-FU). The patient has remained asymptomatic till date.

Case report

A 48 year old female patient reported with swelling in the left posterior region of the palate since 3 months. The swelling had gradually increased to the present size and was not associated with pain. The patient was a known diabetic & hypertensive and was under medication for the same.

A well defined palatal mass measuring 3x2 cm was noted adjacent to 24, 25 and 26 extending up to the midline with no surface changes (Fig 1). The growth was bony hard in consistency and non tender. 24, 27 and 28 were grade II mobile. Radiographically, the lesion presented as a well defined mixed radio opaque-radiolucent mass with no root resorption or displacement. A provisional diagnosis of ossifying fibroma was made. The lesion was enucleated in toto under local anesthesia (LA) as the patient was medically unfit for general anesthesia (GA). Histopathology was suggestive of chronic nonspecific osteomyelitis.

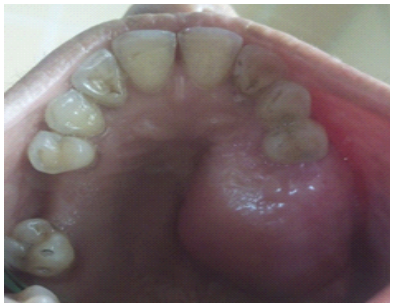

Fig 1: A well defined palatal mass measuring 3x2 cm was noted adjacent to 24, 25 and 26 extending up to the midline with no surface changes Four months later the patient reported with a recurrence in the same region measuring 1x1cm which was enucleated under LA. Histopathology features were suggestive of aggressive ossifying fibroma.

Three months later the lesion had recurred in the same region measuring 4x3cm. CBCT revealed mixed radiodense lesion extending from 24 to the maxillary tuberosity and from buccal cortex till the midline. The periphery was well demarcated from the underlying bone showing few areas of perforation/erosion of the palate and cortical plates The lesion was excised under GA with extraction of 23 and 24. Histopathology revealed plump large epitheloid like osteoblasts suggestive of AO (Fig 2).

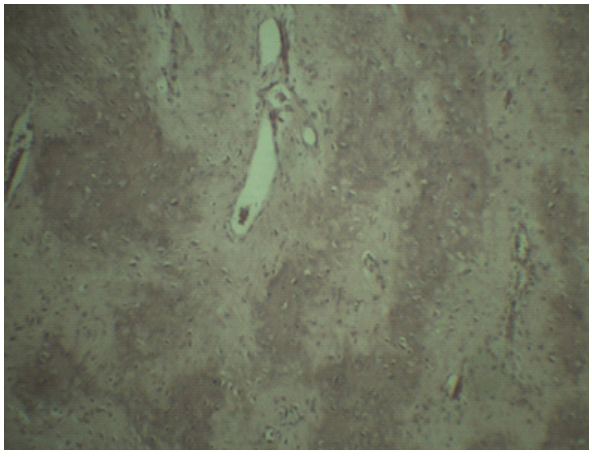

Fig 2: Large plump proliferating cells and irregular deposits of osteoid and trabeculae of bone lined by plump large epitheloid like osteoblasts with eccentrically placed nuclei and focal areas of cartilaginous tissue (haematoxylin and eosin, magnification×10).

Four months later the patient reported with a 3x2cm recurrence posterior to 22. Under GA a partial maxillectomy was performed with 0.5 cm of safe margin. 5FU was irrigated onto the resection site and washed away after 15 minutes. Buccal fat pad was advanced into the defect and the palatal & buccal mucosae were advanced to achieve primary closure. An acrylic feeding plate was secured in position using a stainless steel screw. The post-excision specimen was histopathologically reported as AO. The patient has been under close observation and has remained disease free since 6 months (Fig 3).

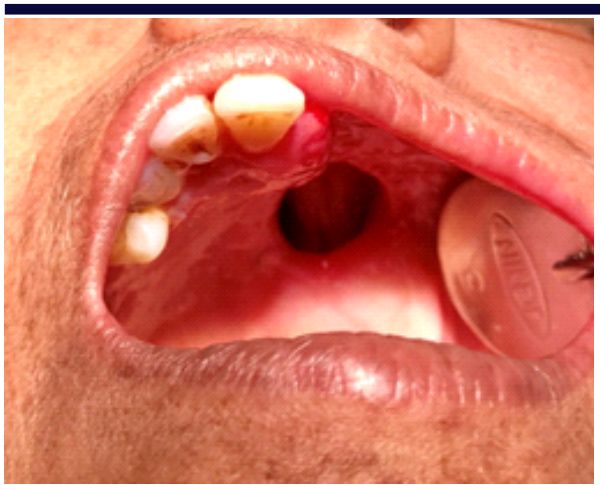

Fig 3: 6 month follow-up showing no signs of recurrence

DISCUSSION

OB was described by Jaffe and Lichtenstein as a true neoplasm derived from osteoblasts. Etiology include trauma, inflammation, abnormal tissue response to injury and local alteration in bone physiology[3].
OB commonly affects males in the 2nddecade[4], 90% occurring before age 30[1] whereas AO commonly presents in the third and fourth decades which was consistent with our case. Pain may or may not be present[1].

AO commonly presents as a unilocular or multilocular expansile mixed radiodense lesion with internal calcifications and ill or well defined borders[1]. The potential aggressiveness of the lesion may result in expansion, perforation, or cortical destruction.

Histopathologically the hallmark of AO is the presence of plump epitheloid osteoblasts[5]. Moderate mitotic activity and invasion into surrounding bone and soft tissue may be seen. Some authors believe that lesions described as AO are, in fact, well-differentiated osteosarcomas resembling osteoblastomas[6].

AO tends to be clinically and radiographically larger (>4 cm) than OB (<4 cm). The clinical aggressiveness appears more dependent on the precise location and size of the tumour than on its microscopic features. Hence, though epithelioid osteoblasts are critical to the diagnosis of AO, tumour size and location, which impact the surgeon's ability to completely remove the lesional tissue, are more important considerations with regard to treatment and prognosis[5]

Immunohistochemical analysis have suggested that MIB-1, apoptosis and p53 expression are probably involved at the onset or development of OB, but roles of these markers are not yet clear[7].

Management

Complete resection is the current treatment of choice due to the high chances of recurrence. The antimetabolite drug, 5-fluorouracil (5-FU) which we used following the resection, has been shown to induce apoptosis. 5-FU has been used topically to treat superficial BCCs. A study by Nicholas et al concluded that 5-FU is a novel, effective, targeted treatment for KOTs with lower recurrence rates and less morbidity compared to modified Carnoy's solution[8].

Role of Radiotherapy and Chemotherapy

There is no role for adjuvant radiotherapy or chemotherapy, except in recurrent or surgically unresectable cases[1], incomplete excisions and histologically aggressive tumours. The concerns with radiotherapy include possibility of radiation induced osteonecrosis and malignant malformations in the residual tumour. Methotrexate is an effective chemotherapeutic agent and has been used in combination with doxorubicin and cisplatin. Progression free survival up to 33 months has been reported with combination chemotherapy[4].

Prognosis

Mark et al. have reported two cases of AO with diverse biological behaviour. One patient died despite curettage, extensive chemotherapy and radiotherapy, whereas the other patient survived for the next 14 years on radiotherapy alone. Two cases of malignant transformations of maxillary OB into osteosarcoma have been reported. Khan et al reported three recurrences of AO in the ilium with distant pulmonary metastasis[9]

Despite multiple treatment modalities, AO remains a difficult proposition with recurrences, malignant conversion and even death. Gordon et al reported a recurrence rate of 13.6% for OB and 50 % for AO. Molecular analysis have revealed a splice mutation at the exon 5 donor site of the p53 gene, indicating a malignant potential of the tumor[10]. This underscores the need for an active follow-up of these patients and further research for other treatment options[4].

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