

Psammomatoid juvenile aggressive ossifying fibroma of mandible

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ABSTRACT

A female patient, aged 14 years, came to dental OPD, with a large swelling of the right mandibular region, progressing since 4–5 years. The swelling initially started with a small size and later it became a large swelling extending from left lower premolar region to right mandibular angle region. Inferiorly, the swelling was extending below the inferior border in the symphysis region. No paresthesia was experienced by the patient. There was quite a marked facial disfigurement of the right mandibular body and symphysis region extending to left side. No other bones were involved and no other abnormalities were found on general physical examination. The medical history was noncontributory. There was no family history of skeletal disease. The case was surgically treated by segmental resection of the mandible and reconstruction was done using stainless steel reconstruction plate. With the follow-up period of 1 year, there was no recurrence.

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INTRODUCTION

Juvenile aggressive ossifying fibroma of the mandible is a very unusual benign fibro-osseous lesion that affects the craniofacial skeleton in young patients. This type of lesion is equally seen in both the sexes. Juvenile ossifying fibromas (JOFs) are non-odontogenic lesions by the signs and symptoms that they produce.

There are two distinct types of JOF: trabecular JOF and psammomatoid JOF. Psammomatoid is more commonly seen and affects patients from a wider age range (3rd month to 70 years), occurring more commonly in sino-nasal and orbital bone region.

The JOF has been also called as juvenile aggressive ossifying fibroma, the aggressive psammomatoid ossifying fibroma, and the juvenile active ossifying fibroma. The clinical diagnostic characteristics suggestive of JOF are patient's age, rapid increase in lesion size, and absence of pain, paresthesia, and bruit with normal arrangement and complement of teeth,



Figure 1: Preoperative photograph

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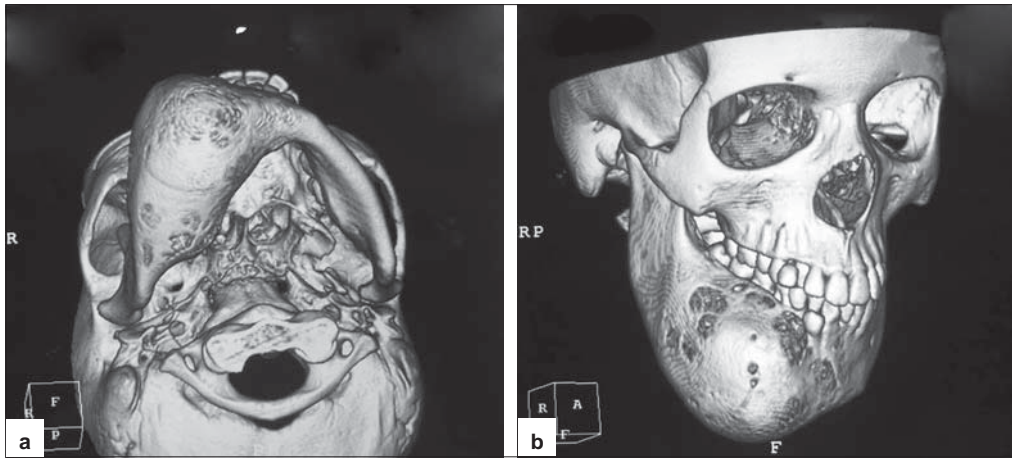


Figure 2a and b: Preoperative CT scan

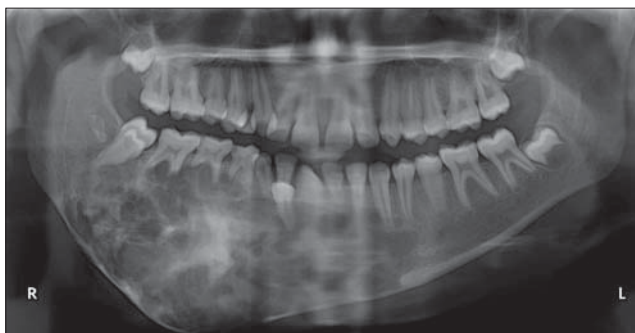


Figure 3: Preoperative OPG

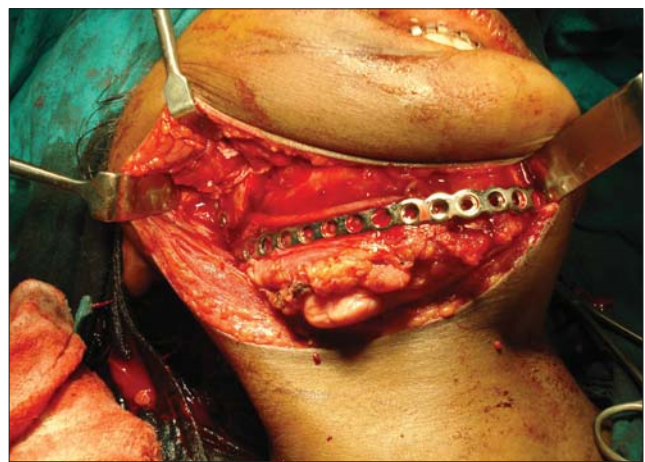


Figure 4: Intraoperative photograph



Figure 5: Postoperative photograph



Figure 6: Postoperative OPG

regular outline of the mass, and absence of the bone surrounding the lesion.^[1]

CASE REPORT

A 14-year-old girl was referred to the department of oral and maxillofacial surgery with a slowly enlarging right-sided mandibular swelling of 4–5 years duration.

Extraorally, there was right-sided facial swelling extending from left corner of the mouth to the right angle of the mandible anteroposteriorly, from the superior border of the external oblique ridge of the mandible to the lower border of the mandible superoinferiorly and caused significant facial asymmetry. There was no history of any kind of paresthesia along the whole distribution of the right inferior alveolar nerve. On intraoral examination, all molars and premolar

teeth showed grade II mobility and the mandibular swelling was non-tender on palpation, bony hard in consistency, covered by normal mucosa and extended from the right angle of the mandible to the mental region of opposite side and had caused expansion of the lingual and buccal cortical plates. No other bones were involved and no other abnormalities were found on general physical examination. The medical history was noncontributory. There was no family history of skeletal disease.

On radiological examination, orthopantomogram (OPG) and computerized tomography (CT) scan showed well-defined radiopacities surrounded by radiolucency with sclerotic border [Figure 1]. Routine hematological and urine investigations were normal.

Aspiration of the lesion was done which showed negative aspiration; so, incisional biopsy was performed under local anesthesia in 43 and 44 regions and sent for histopathological examinations which revealed psammomatoid JOF [Figures 2a-b and 3].

Later, the patient was taken under general anesthesia (GA) and the tumor mass was resected along with 5 mm of safety margin. Reconstruction was done using stainless steel reconstruction plate to give stability to the remaining part of the mandible and to re-establish the lost contour of the mandible [Figure 4].

Histological picture of the specimen was same as incisional biopsy report, and the report was interpreted to be juvenile psammomatoid ossifying fibroma. A cross-reference of the slide was also done and it coincided with the report as of juvenile psammomatoid ossifying fibroma.

DISCUSSION

Ossifying fibromas are more commonly seen in Blacks than Whites as stated by Fujimoto *et al.*^[2] Most of the fibro-osseous lesions of the jaw are asymptomatic and slowly progressive, but an unusual clinical presentation showing aggressive and destructive growth may be expected when the lesion is seen in younger patients aged below 15 years.

Juvenile aggressive ossifying fibroma (JAOF) is benign but aggressive neoplasm commonly seen in children below 15 years of age and has been thought to arise as a result of differentiation of mesenchymal cells of periodontal ligament, the multipotential precursor cells, into cementum, osteoid, or fibrous tissue.^[3]

Cementum and alveolar bone are distinguished by

anatomic, morphologic, and functional criteria, and presently, no biochemical marker exists that can distinguish cementum matrix or cells from those of bone. In spite of their common embryologic origin, the pathway of cellular differentiation, incompletely characterized extracellular protein matrices and the lack of cementum specific markers does not necessarily be the conclusion. Cementum and alveolar bone are identical tissues. Reflective of the vascular component and pathway of formation using a cartilaginous precursor, the extracellular matrix of bone is considerably more complex than that of cementum. Therefore, it is possible to biochemically distinguish bone from cementum. The development of cementum is dependent on epithelial cells and cell product because cellular phenotype is a direct consequence of a set of genes expressed by a given cell type. Final resolution of whether cementum is distinct from bone will probably be answered using molecular biologic approaches.^[1]

Recently, Pimenta reported the association of a new tumor suppressor gene (*HRPT2*) mutation with ossifying fibroma and suggested that these lesions could arise as a result of haploinsufficiency of the particular gene.^[4]

JAOF is a rare fibro-osseous lesion of the jaw occur at early age of onset, i.e., below 15 years of age, and a tendency to recur, and appears as a unilobulated/multilobulated lesion in its early stages of development followed by radiopaque appearance surrounded by radiolucent trimming at later stages.

The computed tomography has added value to the diagnosis and is used to confirm the extent of the destruction.^[5]

Recurrence rates ranging from 30 to 58% have been reported, which also include incomplete excision.^[6,7]

Aggressive enucleation and limited resection are acceptable procedures for JOF when the lesion is recognized early and is confined to a well-defined space within the bone.^[8,9] Attempt to enucleate this patient's lesion and preserve mandibular continuity would not have been successful and could have been improper treatment. The possibility of inadequate excision of this lesion which demonstrated extension beyond the bone and high probability of intraoperative or postoperative fracture mitigated against anything, but resection in this instance, and the remaining part of the resected mandible was reconstructed by using stainless steel reconstruction plate. During the follow-up period of 1 year, there was no recurrence [Figures 5 and 6].

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