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Case Report

The Aggressive Masquerade-Juvenile Ossifying Fibroma of Maxilla: An Interesting Case Report

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Abstract

Introduction: Juvenile ossifying fibroma is a rare benign fibro-osseous lesion. It is characterized by the early age of onset, the localization of the tumor, the radiological pattern and a tendency to recurrence. Juvenile ossifying fibroma (JOF) appears at an early age and in 79% of the patients are diagnosed before the age of 15. Males and females are equally affected. JOF originates from periodontal ligament and ranges 2% of oral tumours in children. The JOF is located mainly (85%) in facial bones, in some cases (12%) in calvarium and very seldom (3%) extracranially.

In the light of a clinical case, we review the clinical, histological and radiological features of this tumor with treatment and follow up.

Case Report: We report a case of an 8-year-old male child who presented with a unilateral swelling of the left middle face. On evaluation of previous medical history, a surgical intervention in the same area for a similar type of swelling about 6 months ago was reported and a histopathological report of central giant cell granuloma was given at a different center. Clinical examination revealed an expansile swelling involving the left maxilla. CBCT scan was taken and showed the presence of calcific flecks along with gross expansile mass. Excisional biopsy was done and the mass was sent for histopathological examination, which revealed the presence of trabecular type juvenile ossifying fibroma of left maxilla. No recurrence noted.

Conclusion: It is generally agreed that juvenile ossifying fibroma behaves aggressively locally and has a high recurrence rate when not adequately treated, the correct treatment being en bloc resection with free surgical margins.

The juvenile form could be distinguished from ossifying fibroma by the following features:

1. Earlier onset (at childhood or adolescence),
2. Locally aggressive growth,
3. Osteoid trabeculae on histological examination.

Because of its aggressive and compressive nature, juvenile ossifying fibroma requires an early complete surgical excision. A long-term clinical and radiological surveillance is necessary to prevent recurrence.

Keywords: *Fibroosseous Lesion; Juvenile Ossifying Fibroma; Maxilla; Aggressive Lesion*

Introduction

Juvenile ossifying fibroma is a rare benign fibro-osseous lesion.

Case Report

An 8 yrs old boy who reported to the department of oral medicine and radiology with chief complaint of swelling in the left side of the face since 8 months. The swelling was initially small and gradually increased in size to attain the present size and associated with mild pain on applying pressure to the swelling. History of trauma to the same area 1 year ago (the boy got into a fight with his classmate and got punched on his face) was reported. Patient had visited another centre for the same complaint 6 months ago and after relevant investigations, underwent curettage and contouring of the left maxilla. The biopsy report was that of central giant cell granuloma. The swelling again came back 2 months after the surgery and has gradually attained the present size. No relevant medical and family history and no deleterious oral habits were reported.

n extra oral examination, diffuse swelling present on the left middle and lower third of the face (Figure 1), roughly measuring 7 cms x 6 cms in its greatest dimension, extending - anteriorly from the ala of the nose, posteriorly 2 cms anterior to the tragus. Superiorly from the infra orbital ridge, inferiorly about 3 cms superior to the inferior border of mandible. Surface and surrounding skin appeared to be normal. On palpation inspectory findings are confirmed with no local rise in temperature, firm in consistency and mild tenderness present on applying pressure. No altered sensation was reported in the region.

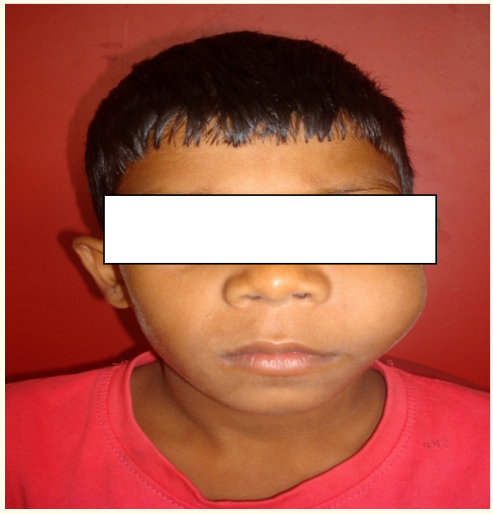


Figure 1: *A swelling noted in the left middle third of the face.*

On intraoral examination, diffuse swelling present in the left buccal vestibule with obliteration of the same (Figure 2). The swelling was Anteriorly extending from the distal aspect of 22, posteriorly till the distal aspect of 26, inferiorly from the attached gingiva and superiorly into the sulcus. A Surgical scar was also noted. Surrounding area appears normal. No abnormalities detected on hard tissue examination.

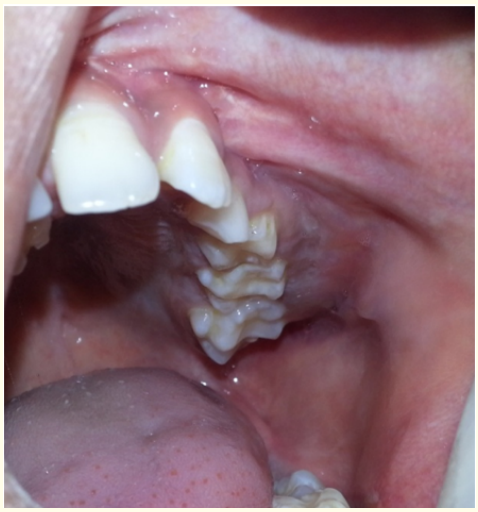


Figure 2: *Expansile lesion in left maxilla with obliteration of sulcus.*

Maxillary lateral occlusal radiograph revealed a well-defined homogenous, space occupying mass with radiopaque flecks and cortical expansion.

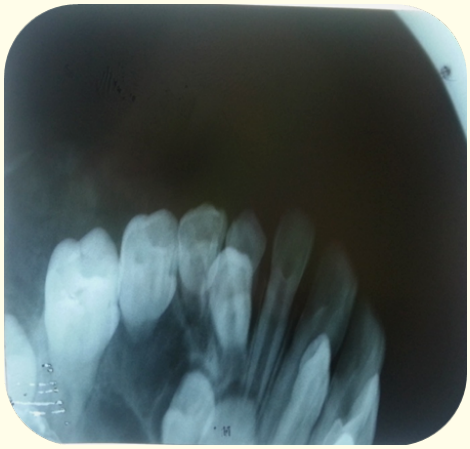


Figure 3: *Maxillary lateral occlusal radiograph revealing space occupying mass with radiopaque flecks and cortical expansion.*

Initial panoramic view showed well defined expansile mass in the left posterior maxilla with radiopaque flecks and haziness of left maxillary sinus with displacement of developing tooth buds (Figure 4A). OPG 6 months later shows the definite expansion of posterior maxilla with increased amount of radio-opacity associated within the lesion (Figure 4B).

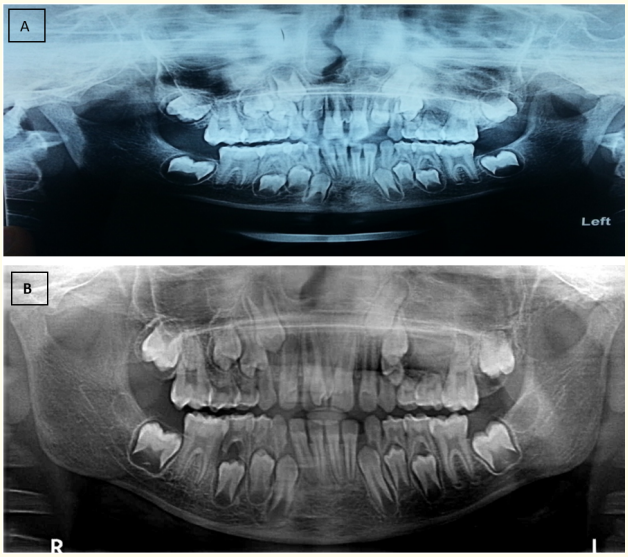
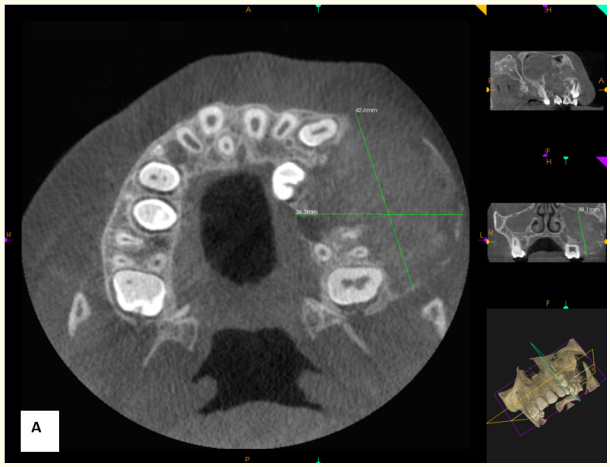


Figure 4A and 4B: *Well defined expansile mass in the left posterior maxilla with displacement of tooth buds. OPG A and B were taken 6 months apart and the increase in the size can be noted in the OPG B.*

CBCT findings confirmed routine radiographic features and further showed heterogeneous tumour mass with evidence of erosion of maxillary bony wall and extension into maxillary sinus and involvement of lateral wall of nasal cavity and in homogenous radiodense areas were evident within the lesion. Approximate maximum dimension in buccopalatally 36 mm, anteroposteriorly 42 mm (axial section) and superoinferiorly 39 mm (Figure 5A and 5B).



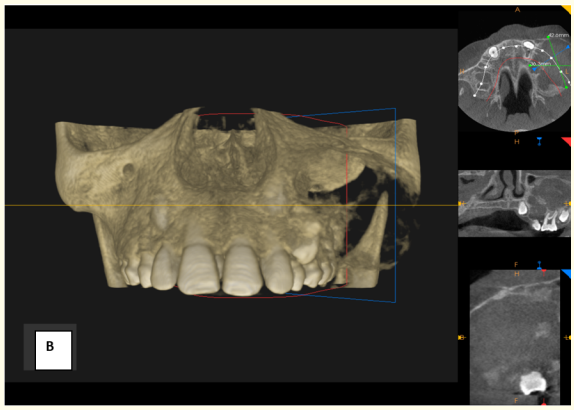


Figure 5A and 5B: *A) reveals the expansile nature of the mass along with the calcific flecks. B) reveals the buccal cortical plate destruction due to the tumour mass.*

Excisional biopsy done under GA and the tissue specimen sent for histopathological evaluation and Trabeculae of cellular woven bone were present associated with a densely cellular connective tissue (Figure 6A-6D). The results of histopathological examination were consistent with Trabecular type Juvenile ossifying fibroma of left maxilla.

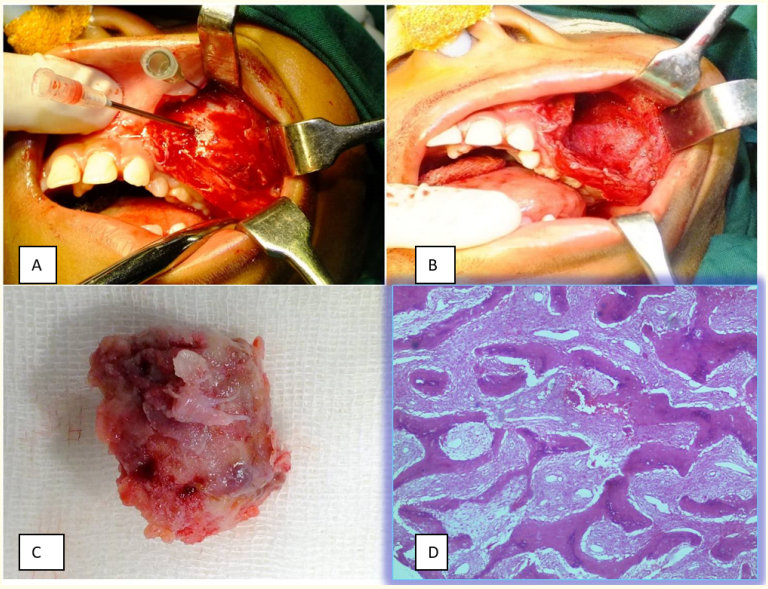


Figure 6A-6D: *A, B and C showing the excisional biopsy process and specimen. D showing the histopathological slide showing cellular woven bone with a densely cellular connective tissue consistent with trabecular type juvenile ossifying fibroma.*

Patients follow up is going on and reduction of bony defect of left maxilla noted (Figure 7). No recurrence noted.

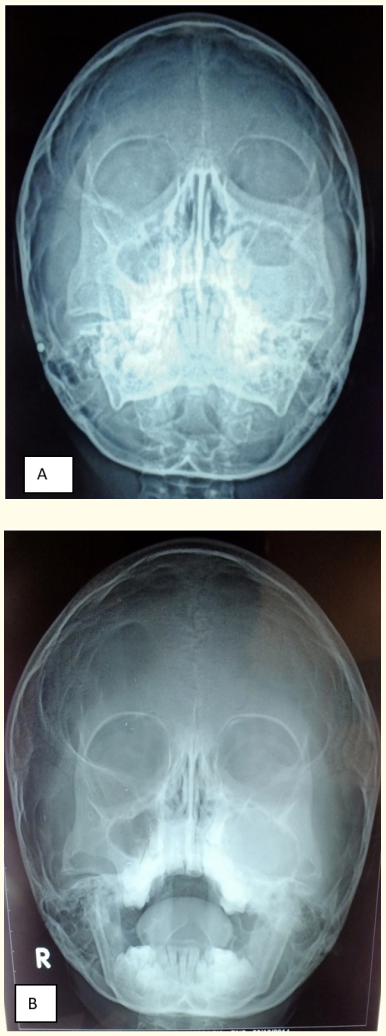


Figure 7: *A) PNS view post surgery. B) 4 months post op showing the marked reduction in the bony defect of left maxilla.*

Discussion and Conclusion

Juvenile aggressive ossifying fibroma (JAOF) is a rare benign and locally aggressive tumor with high recurrence. JOAF is a distinct clinical entity, often confused with other malignant entities because of its rapidly progressive and osteolytic nature [1]. It is a relatively rare fibro-osseous lesion of the jaws characterized by the early onset of age (usually less than 15), location of the tumor, radiological appearance, and high recurrence. JOAF may present as one of two histologic variants: Juvenile psammomatoid ossifying fibroma and juvenile trabecular ossifying fibroma [2]. The psammomatoid type of juvenile ossifying fibroma is more common than the trabecular variety, is more aggressive, and it has a strong tendency to recur [3].

Juvenile trabecular ossifying fibroma occurs in young individuals with peaks occurring in those who are below 15 years of age and is usually characterized by rapid growth and recurrence [4].

Ossifying fibroma arises in the jaw, facial bones and skull and juvenile ossifying fibroma involves most commonly the paranasal sinuses and periorbital bones in 90% of cases [5] mandibular lesions are seen in 10% of the cases.

Clinically, it presents as an asymptomatic tumor of aggressive appearance. The symptoms are variable and consist on facial swelling, sinusitis, nasal obstruction, teeth displacement, eye proptosis, visual disturbances, progressive blindness, airway obstruction, and progressive craniofacial deformities [6]. The local invasion may include pushing and displacement of adjacent bony limits or invasion through osseous delimiting walls with extension into adjacent anatomic compartments. It appears locally aggressive with cortical disruption and involvement of many adjacent anatomical structures. This lesion has predominating soft tissue consistency with variable amounts of internal calcification and/or linear or irregular focal bone.

Histologically, these lesions are always benign, composed of highly vascular and fibroblast-rich connective tissue with calcified substance shaped on bony trabeculae distributed throughout the fibrous stroma. The highly cellular nature of the fibrous matrix reflects the more aggressive behavior of the tumor [7].

Radiologically, JOF appears as a well circumscribed solitary lesion concentrically expanding, with bone density [8] and also presents well-defined, unilocular or multilocular expansile and well-circumscribed radiolucent lesions with cortical thinning The radiolucency of the lesion varies, depending on the maturation stage and amount of calcification. The demarcation of the tumor from the surrounding bone is well-defined by a radio-opaque border. This aggressive lesion can cause expansion, cortical thinning as well as perforation of surrounding bone [9].

Surgery is the primary mainstay treatment. The reference treatment consists of en bloc surgical resection. Various external approaches are used according to the location of the lesion. The tumor mass must be removed down to the level of normal bone with preservation of adjacent vital structures as much as possible. Immediate reconstruction is not recommended. Secondary reconstruction if needed may be undertaken sooner for slow-growing lesions (< 1 year) and be delayed for fast growing lesions (> 1 year).

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