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Management of Juvenile Ossifying Fibroma by Aggressive Curettage – A Case Report

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ABSTRACT
Juvenile ossifying fibroma is a benign, but potentially aggressive, fibro osseous tumour of the craniofacial bones[1]. The authors describe a case of a juvenile ossifying fibroma of mandible presenting in a 17-year-old girl who was managed by enucleation of the tumour and followed up for a period of 10 months with no signs of recurrence. This paper also reviews the histology, clinical behaviour and management of this uncommon but disfiguring lesion.

INTRODUCTION

Fibro-osseous lesions (FOL) are a poorly defined group of lesions affecting the jaws and craniofacial bones. They are characterized by the replacement of bone by cellular fibrous tissue containing foci of mineralization that vary in amount and appearance. The World Health Organization (WHO) describes Juvenile Ossifying Fibromas (JOF) as a benign, potentially aggressive, fibro-osseous tumour of the craniofacial bones affecting children under 15 years of age[2]. It was first described by Benjamin in the year 1938 as "osteoid fibroma with atypical calcification" (Khoury et al., 2002). Later in 1952 Johnson coined the term "juvenile active ossifying fibroma" (Neville et al. 2002) [3]

The lesions are well-circumscribed radiolucencies that in some cases contain central radiopacities. JOF may appear as one of the two histologic variants: Juvenile Psammomatoid Ossifying Fibroma (JPOF) or Juvenile Trabecular Ossifying Fibroma (JTOF). Although both patterns reveal similar radiographic features and growth patterns, the trabecular form is diagnosed initially in younger patients. The mean age of trabecular juvenile ossifying fibromas is approximately2 years. Whereas the age of patients diagnosed with the psammomatoid variant approaches 22 years. Both patterns occur in either jaw but reveal a

maxillary predominance. Although many of these tumours are initially discovered upon routine radiographic examination, cortical expansion may result in clinically detectable facial enlargement.

CASE REPORT

A 17-year-old female reported to the Department of Oral and Maxillofacial Surgery, Vokkaligara Sangha Dental College and Hospital, Bangalore with a chief complaint of swelling in the left side of lower jaw since 2 years (Fig. 1). The patient had visited a local dentist where a definitive diagnosis was not made. The lesion had been slowly increasing in size since it was first noticed over the past 2 years. No history of trauma, pain, paraesthesia or lymph adenopathy was elicited. Physical examination revealed a healthy, normally developed young girl in no apparent distress. Extra orally the swelling extended from the corner of mouth till the angle of mandible. Facial asymmetry was noted. On palpation the swelling was hard, non-tender and non-adherent to the overlying skin. Bruits or pulsations were ruled out. Intra-oral examination revealed expansion of the left buccal cortical plate, which was hard on palpation. There was no evidence of tooth mobility, abscess formation, dehiscence or malocclusion.

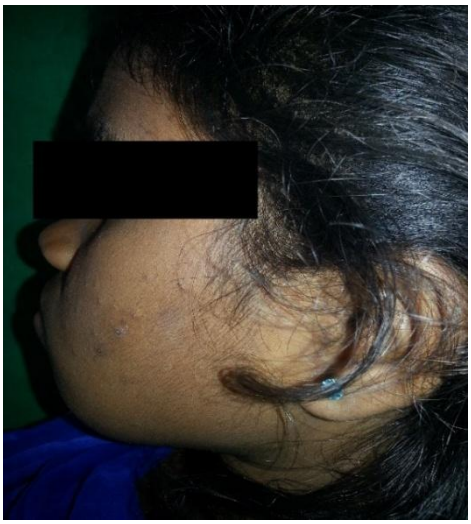

Fig. 1

A panoramic radiograph revealed a well circumscribed radiolucency measuring 5x3 cm present in the left body of the mandible region extending from 34 to 37 region. 38 was missing and no history of extraction of the same was elicited. Root resorption in relation to 35 &36 could be appreciated. Cone Beam Computed Tomography (CBCT) scan of the left mandibular region showed bucco-lingual cortical plate expansion and the inferior alveolar nerve passing through the lesion (Fig. 2).

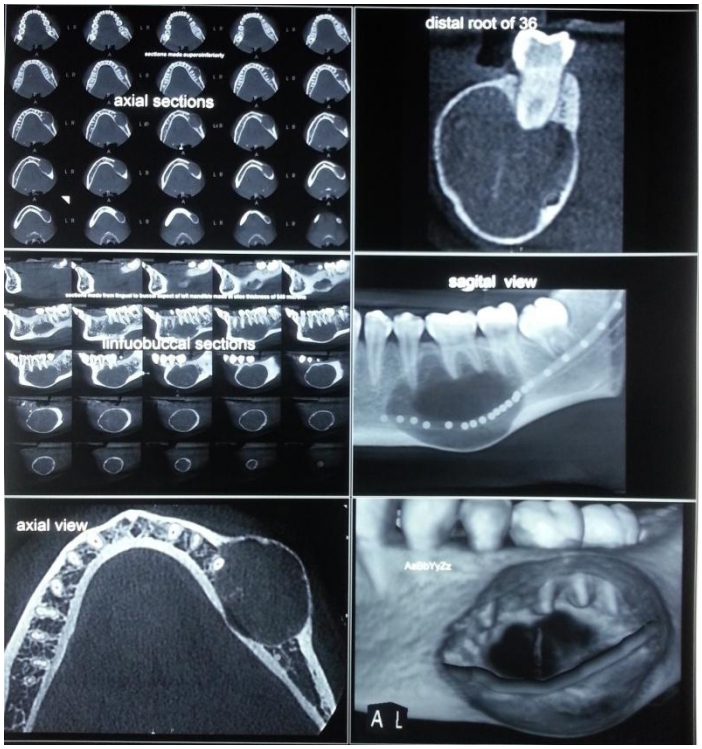

Fig. 2- CBCT findings

Laboratory values were within normal limits. On wide bore needle aspiration, blood was drawn from the lesion and was sent for histopathological examination where only RBCs with few inflammatory cells were seen without any proper diagnosis. The provisional diagnosis was made to be anuresmyal bone cyst and the differential diagnosis to be unicystic ameloblastoma. Surgical exploration of the lesion was done under general anaesthesia. Through an intra oral approach, crevicular incision from 34 to 37, full thickness mucoperiosteal flap was raised and a bony window was created through buccal trephination. Enucleation of the lesion was done which revealed solid firm tissue mass which was sent for histopathological examination (Fig 3). Inferior alveolar nerve was identified and mobilised to free

it from the mass around it. The lesion was not involving the nerve. Extraction of 36 & 35 was carried out. Haemostasis achieved and the lesion closed primarily. Inter-maxillary fixation was done with arch bars to prevent pathological fracture of the mandible.

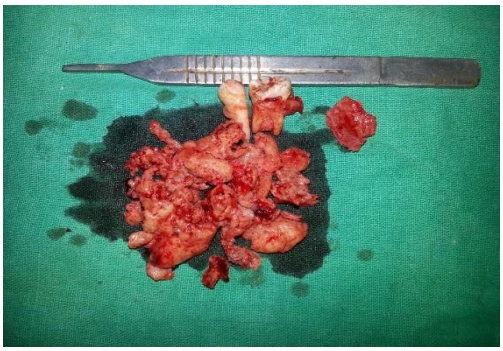

Fig. 3- curetted tumour mass

Microscopic examination with H & E staining showed fibrocellular connective tissue stroma interspersed with plump and spindle shaped fibroblasts. Few fibroblasts with angular and hyperchromatic nuclei with minimal mitotic activity. Multinucleated giant cell was present. Trabeculae of immature bone with osteocytes in lacunae were present. Cementum like calcification with dystrophic calcification were also present. Blood vessels lined by endothelial cells along with areas of haemorrhage were also seen. All these histologic features led to a final diagnosis of trabecular form of juvenile ossifying fibroma of the mandible.

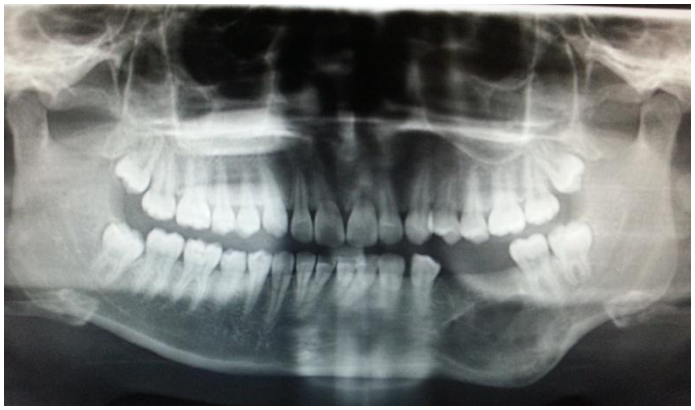

Fig. 4 – 6 months post op OPG

Post-operative panoramic radiographs were taken at the intervals of 1, 3, 6 months (Fig 4) to evaluate mandibular fracture and healing of the bony cavity. The patient was closely monitored for 10 months without any signs of recurrence and is under constant follow up.

DISCUSSION

Human mandible is inflicted with variety of lesions which vary from radio opaque to radio lucent lesion and even mixed, and also vary from one age group to other even according to sex. One of them are the fibro-osseous lesions affecting the mandible. They encompass AOF and JOF. Adult ossifying fibromas are thought to originate from periodontal ligament; whereas, JOF arise as a neoplastic myxoid tissue from cartilage and bone precursors[9]. JOF is a relatively rare fibro-osseous lesion of the jaws characterized by the early age of onset i.e., under 15 years of age, the location of tumour, and the radiological appearance and the tendency to recur[4]. Though it has no gender predilection, it is stated that mandibular tumours are more frequently common in girls between the age of 8-12 or during the second to fourth decades of life[1]. In present paper 17 years old girl was presented. The designations ossifying fibroma, cemento-ossifying fibroma, and cementifying fibroma are all appropriate for this tumour and continue to be used by many[7]. In spite of this, however, it is agreed that these are the same lesion and are classified best as osteogenic neoplasms. JOF is thought to arise from mesenchymal cell differentiation of the periodontal ligament-which is a precursor to cementum, fibrous tissue and osteoid[4]. Mutations of the tumour suppressor gene HRPT2 has been documented in these lesions by Pimentaet al[6]. JOF may also be due to mal development in tissues between roots of molar teeth which help in generation of bony septa.[8]

The most frequent location of JOF is in the paranasal sinuses[4], accounting for about 90% of cases, whereas mandibular lesions account for approximately 10% of the facial JOF cases making it a rare finding. On contrary, Neville mentioned mandible to be involved far more often than the maxilla with mandibular premolar and molar area being the most common site as in our case[3].

Clinically, JOFs are mostly asymptomatic with slow or rapid development leading to facial asymmetry. As for the radiological aspect, JOFs can be seen as unilocular or multilocular radiolucencies/mixed radiolucencies. Root resorption can also be observed as seen in our case.

Ossifying fibromas tend to grow in a concentric, expansive pattern. They encompass AOF and JOF. Cone-beam computed tomography assessment may show well-defined sclerotic borders with a variable amount of calcifications.

Histologically, JOFs are lesions characterized by cell-rich fibrous tissue with giant cells and bands of cellular osteoid trabeculae.[5]

The treatment of choice for such lesions is usually en block recession, but in this case through curettage and enucleation of the lesion was done and no signs of recurrence was observed.

Since JOF in patients below 15 years the need for reconstruction is also greatly increased. The child face keeps growing until the age of 16 – 18 years on an average. Hence, many authors prefer the late reconstruction of the defect in growing age group irrespective of the lesion being recurrent or non-recurrent in behaviour.

In cases where the lesion is small in dimension along with no involvement of the adjacent bone, curettage and enucleation of the lesion can be chosen as a better treatment option. There are also case reports of adequate healing and regeneration of the bone even in cases of juvenile ossifying fibroma. The theory postulated for such regenerate formed is the preservation of the periosteum. However, the recurrence rate of such treatment ranges between 30% to 58 % owing to which some authors prefer the treatment of this lesion with extensive surgery that is wide excision with adequate margins especially in cases where adjacent structures are also involved.

CONCLUSION

Juvenile ossifying fibroma is a locally aggressive lesion with high tendency to resorb adjacent bony structures and recur even after following a conservative treatment of the pathology. Treatment of JOF is either conservative or aggressive according to the behaviour of the lesion. Conservative treatment of non-aggressive forms of JOF includes curettage, local excision. Aggressive lesions, which show rapid growth, cortical bone thinning or perforation, tooth displacement or root resorption can exhibit early recurrence. Therefore an en-block resection is justified when extremely aggressive behaviour is observed.[10] On the contrary this case report shows that conservative treatment with aggressive curettage of this aggressive lesion may be considered as a treatment modality looking at the age of the patient. Although juvenile ossifying fibroma is not so common entity, its aggressive local behaviour along with high recurrence rate mean that it is important to make an early diagnosis and to apply the appropriate treatment along with long term follow-up. However further studies with good sample size and long term follow up are required to establish as the treatment of choice.

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