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Case Report

Juvenile Psammomatoid Ossifying Fibroma – A case report

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Abstract: Juvenile Psammomatoid ossifying fibroma (JPOF) is a rare, gradually progressive tumor, with a local aggressive behavior and tendency to recur. We report a case of JPOF of the mandible in a 24 year old male patient who had a slowly growing mandibular mass, causing severe facial asymmetry.
Keywords: ossifying fibroma, jaw tumors, mandible, fibro-osseous lesions.

INTRODUCTION

Ossifying fibroma (OF) is a well-demarcated lesion composed of fibrocellular tissue and mineralized material of varying appearances. OF most commonly occurs in the 2nd to 4th decades[1]. The histologic and clinical subtypes in the spectrum of ossifying fibroma include the rarer variants "juvenile active ossifying fibroma", and "juvenile aggressive or active or psammomatoid ossifying fibroma" [2].

The mean age of the histological subtypes varies[1]. The psammomatoid type is not restricted to children or adolescents. It has been reported in adults as old as 72 years, although the mean age is 20 years[3]. OF is mostly seen in the posterior mandible[1]. The use of the term juvenile reflects the average age of 20 years [2] and the cases of ossifying fibromas with prominent psammoma-like bodies have been given the qualifier psammomatoid[4], hence the name of juvenile psammomatoid ossifying fibroma is termed.

CASE REPORT

A 24-year-old male presented with a painless swelling on the left side of his lower face and chin region since 3 years. (fig 1). Extra-orally the swelling extended from the posterior border of the ramus of mandible on the left side to the inferior border of the body of mandible extending onto the right side. Overlying skin was normal. There was no complaints of dysphagia but slight difficulty in opening the mouth was present. There was no history of trauma. No cervical lymphadenopathy was noted.

On intra-oral examination, a swelling was seen distal of left mandibular third molar to the anterior aspect of the right second incisor teeth. On palpation, the swelling was hard in consistency. An orthopantomogram showed a expansile lesion on the right side of body of the mandible (fig 2).

Histopathological examination of incisional biopsy revealed hyperplastic squamous epithelium with subepithelial tumour composed of variable sized eosinophilic ossicles, with extensive areas of calcifications resembling psammoma bodies, with fibroblastic stroma. The stroma is scanty, mildly oedematous and shows lymphocytes with occasional foreign body giant cells. Correlating the clinical, radiological and pathological findings, the case was diagnosed as Juvenile psammomatoid ossifying fibroma.

The patient was operated, and the tumor was excised with reconstruction of the mandible. On gross examination, excised mass along with mandible and teeth measuring 8.5 x 4 x 4 cm of size showed a bulge over premolar and molar areas protruding outside of size 6 x 4 x 3 cm (fig 3). On histopathological examination of the excised tumor showed adjacent cortical bone and tumor composed of variable sized numerous Psammomatoid calcifications and moderately cellular dense fibrous stroma, (fig 4,5,6) confirming the diagnosis of Juvenile psammomatoid ossifying fibroma.

Regular follow-up has been done for one year since then and there is no evidence of recurrence so far.

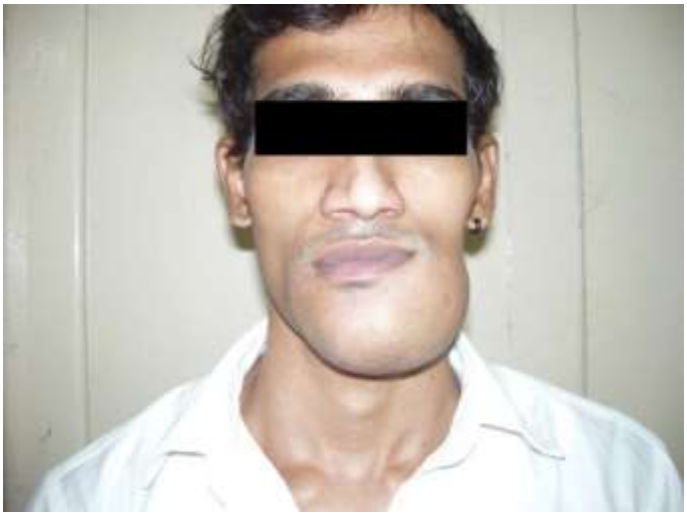

Fig-1: 24-year-old male with a painless swelling on the left side of his lower face and chin region.

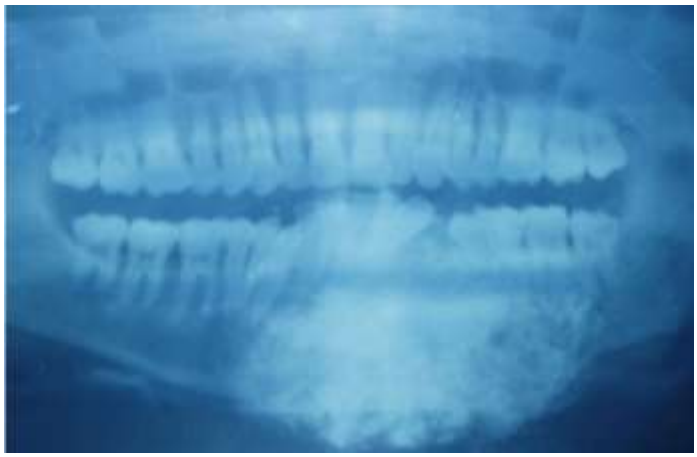

Fig-2: An orthopantomogram showing a expansile lesion on the right side of body of the mandible.

Fig-3: Excised mass along with mandible and teeth measuring 8.5 x 4 x 4 cm of size, inset showing cut section of the tumor.

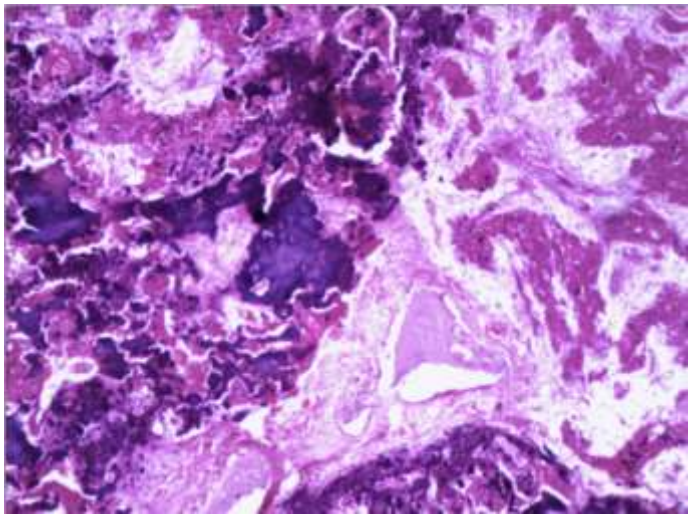

Fig-4: Microphotograph showing bony trabeculae and tumour composed of cellular fibrous stroma and psammomatoid calcifications. H & E 40x

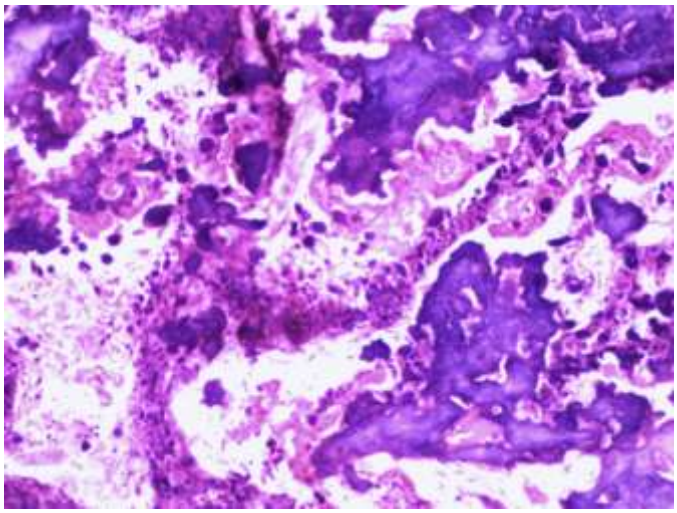

Fig-5: Microphotograph showing variably sized psammomatoid calcifications. H& E 100x

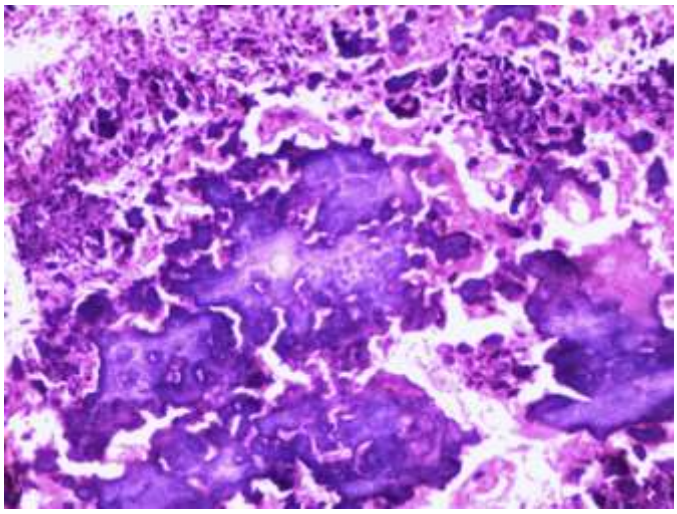

Fig-6: Numerous Psammomatoid calcifications H & E 100x

DISCUSSION

Ossifying fibroma is the name given to jaw tumors that may have a variety of histologic appearances but are composed of islands/trabeculae/reticulae of bone in a benign fibroblastic proliferation [2]. It seems likely that all of these lesions have a periodontal ligament origin[4].

Juvenile psammomatoid ossifying fibroma (JPOF) is characterized by a fibro -blastic stroma containing small ossicles resembling psammoma bodies[1]. However, unlike true psammoma bodies, these mineralized deposits range from acellular to sparsely cellular and may fuse to form trabeculae with reversal lines[2].

Differential diagnosis: Both being part of the spectrum of the fibroosseous lesions, the main differential diagnostic alternative for ossifying fibroma is fibrous dysplasia[5]. The most important distinguishing feature is the presence of demarcation and/or encapsulation in OF as opposed to the merging with its surroundings as shown by fibrous dysplasia[1]. In addition, the variation in cellularity as well as in appearances of mineralized material distinguishes ossifying fibroma from fibrous dysplasia[5].

To distinguish ossifying fibroma from osseous dysplasia, clinical presentation and radiographic appearance may be decisive[1]. Osseous dysplasia usually is asymptomatic, being an incidental finding on radiographs whereas ossifying fibroma shows the features of an expanding benign neoplasm both radiologically and clinically[5].

Juvenile trabecular ossifying fibroma (JTOF) consists of cell-rich fibrous tissue containing bands of cellular osteoid without osteoblastic rimming together with slender trabeculae of immature bone containing coarse lacunae with plump osteocytes and are lined by a dense rim of enlarged osteoblasts[1].

Treatment either by enucleation and curettage or by resection is tailored to tumor size, location, the desire to preserve vital structures, and the ability to remove the tumor without incurring a high risk of recurrence[2].

There is no standardized follow-up protocol in the literature. Because of high recurrence rate, immediate reconstruction is not advised. Secondary reconstruction may be undertaken sooner for slow-growing lesions, and be delayed for fast-growing lesions[6].

Radiotherapy is contraindicated because of the risk of malignant transformation and potential harmful late effects in children[7]. Thus appropriate recommended treatment is aggressive surgical approach followed by clinical and radiological follow-up.

CONCLUSION

A combined clinical, radiological and pathological correlation is mandatory to diagnose a case of Juvenile psammomatoid ossifying fibroma and a close followup should be ensured so as to detect these types of lesions early, before facial structures are destroyed by these benign lesions.

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