Juvenile Ossifying Fibroma of Mandible

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Abstract

Juvenile ossifying fibroma is a well defined clinical & histological entity that has been recently separated from the other fibroosseus lesions. The clinical & histological appearance bears a clear resemblance to the osteofibrous dysplasia of long bones.

The juvenile ossifying fibroma is a subtype of the ossifying fibroma, a benign neoplasm with substantial growth potential. The term juvenile ossifying fibroma is used in literature to name two microscopically distinct fibro osseous lesion of the craniofacial skeleton. The histologic pattern can be described as under two headings -

1. Trabecular juvenile ossifying fibroma (TrJOF)
2. Psammomatoid juvenile ossifying fibroma (PsJOF)

A rare & unusual case of Psammomatoid juvenile ossifying fibroma of mandible in a female patient aged 11yrs is reported here.

Key Words: Fibro-osseous lesion, Juvenile ossifying fibroma, Trabecular JoF, Psammomatoid Jof.

Introduction

Juvenile ossifying fibroma is an uncommon & rare fibro-osseous lesion that has been differentiated from the larger group of ossifying fibroma on the basis of the age of the patients, site of involvement & clinical behavior. The second edition of WHO classification of odontogenic tumours[5] defines juvenile ossifying fibroma as a lesion consisting of cell rich fibrous tissue containing bands of cellular osteoid without osteoblastic rimming along with trabeculae of atypical bones.

Case Report

A female patient aged 11 years reported to the Department of Oral Medicine and Radiology, Hitech Dental College and Hospital with a chief complaint of painless swelling of the right lower jaw for the past one month. The history of presenting illness revealed that the painless swelling started as a smaller one gradually grew to the present size over a period of one month. There was no secondary changes seen over the swelling.

The Extraoral Examination revealed facial asymmetry and single diffuse swelling present over the right body of the Mandible of approximate size (3x2) sq cm. The swelling extended anteriorly from the right parasymphysis to the anterior border of the ramus of the Mandible posteriorly, superiorly from a imaginary line joining the right commissure of the lip to the tragus of the ear and inferiorly upto the lower border of the Mandible. (Fig. 1, 2). On palpation, the swelling was non- warm, non-reducible, hard and mildly tender.

The Right Submandibular lymph-nodes are single, palpable, mobile, of size approximately (2×1) sq cm and are asymptomatic. The intraoral examination revealed a well defined oval shaped swelling w.r.t. the body of the mandible i.r.t 42, 43, 44, 45. Bicortical expansion of alveolar plates was elicited with obliteration of the Buccal vestibule. The Mucosa over the swelling was normal. On palpation the swelling seemed to be varying in consistency from firm to bony hard. The swelling was Mildly tender on palpation and there was no rise in temperature. (Fig. 3)

Based on the history & clinical findings a provisional diagnosis of ossifying fibroma was given. The differential diagnosis of Monostotic fibrous dysplasia central giant cell granuloma, cement- ossifying fibroma and odontogenic myxoma was kept in mind. The subject was put to radiological investigations. An IOPA, Mandible cclusal radiograph and OPG was taken. The radiographs revealed a well defined expansile mixed lytic tumour with bicortical expansion of the alveolar plates. There was near perforation of a lingual cortical plate and perforation of the buccal cortical plate. Displacement of 43,44 was also seen upon radiographic examination. (Fig. ,4, 5, 6)

A ct scan was advised, the axial and the coronal section obtained revealed a well defined expansile hypodense tumour of size (22×21) sq mm with intervening hyperdense focal spot. There were breaks in the buccal cortical plate. (Fig. 7, 8, 9)

The serum analysis revealed an elevated serum alkaline phosphatise level at 429 u/l (normal upto 190 u/l). The serum calcium level was 9.1 mg/dl. The patient was referred to the department of pedodontics and then to the department of oral & maxillofacial surgery, hitech dental college and hospital for further followup. A wide excision of the tumour along with the right submandibular lymphnode was performed. The surgical defect was replaced with fourteenholed miniplate. (Fig. 12)

The gross specimen was sent to the laboratory for the histopathological examination. On normal H/E section the tumour revealed stromal connective tissue with numerous hematoxyphillic calcification(psammomatoid bodies), cellular fibrous connective tissue with loose as well as dense arrangement of plump fibroblast without osteoblastic rimming. (Fig. 10)

Correlating the history, clinical features, radiological features, serum analysis and taking into consideration -

1. The tender age of the patient.
2. Benign but locally aggressive nature.
3. Growth pattern of the tumour.
4. Mixed lytic nature of the tumour on radiographs.
5. Elevated serum alkaline phosphatise level.
6. Histopathology report.

A final diagnosis of juvenile ossifying fibroma (psammamotoid variant) was given.

Discussion

Juvenile ossifying fibroma is an uncommon and rare fibro osseous lesion that has been deferentiated from the larger group of ossifying fibromas on the basis of age of the patient, site of occurence and clinical behaviour. The second edition of the WHO "classi

fication of odontogenic tumors" defines Juvenile ossifying fibroma as a lesion consisting of cell rich fibrous tissue containing bands of cellular osteoid without osteoblastic rimming along with the presence of trabeculae of atypical bone.[5] Lesion of this morphology have been reported as "young ossifying fibroma", "Juvenile active ossifying fibroma", "Aggressive ossifying fibroma", "Psquammous Desmoosteoblastoma"[4] and "Active fibrous dysplasia" It was first reported by Benjamins[1] in 1938 who described it as "osteoid fibroma with atypical ossification" later it was named as "Psammomatoid ossifying fibroma of nose and paranasal sinuses" by Gogl in 1949.[2] The same lesion was termed as Juvenile active ossifying fibroma by Johnson et al in 1952.[3]

Juvenile ossifying fibroma as a separate entity was given in the second edition of the World Health Organisation. "classification of odontogenic tumours". Recent studies suggest a genetic pattern of inheritance which can be demonstrated by the presence of Nonrandom translocation between break points at Xq26 and 2q33 chromosomes. The mean age of occurence of the trabeculae variant of Juvenile ossifying fibroma is approximately 11 yrs, where as that of Psammamatoid variant is 22yrs.

Makek et al[4] reviewed a total of 86 cases of PsJOF in which the age of the patients with PsJOF ranged from 3yrs to 49yrs with a mean age of 17.7 yrs. There was a slight male predominence with male to female ratio of 1.2:1) . This is contradictory to our case. The site of occurence of Psammamotoid variant is orbit ,frontal and paranasal bone.In 86 cases reported by Makek et al majority of these, 53 cases (61.6%) occurred in the orbital bones. Seventeen cases (19.7%) were in the maxilla, and only 7% of the cases were in the mandible. The case depicted here occurred in the mandible which adds to the rareness of the case.

The most common clinical manifestation of psammomatoid vaiant is proptosis and/or exolphthalmos. Other symptoms include nasal obstruction, headache and swelling over the involved bone.[7,8] The presence of cortical expansion with detectable facial enlargement adds to the complication which is secondary to the tumours. Radiographic features are seen to show well defined expansile & mixed lytic lesion. There may or may not be breakage of the overlying cortical bone. The characteristic histologic pictures is suggestive of presence of small mineralized bodies (Psammomatoid bodies) mixed with spindle shaped cellular stroma containing varying amount of fibrous tissue. Accentuated mitotic figures with areas of haemorrhage and small clusters of multinucleated giant cell are also seen.[6,8-10]

Conclusion

Clinical management & prognosis is uncertain. Although it shows slow progressive growth, it can enlarge rapidly with aggressive growth potential which is seldom seen in infants & young children. Recurrence rate is negligible. Malignant transformation in the tumours is not recorded. The surgical treatment of the tumour includes enucleation & curettage. Radiotherapy is contraindicated.[6,7,10,13,14]

References

References are available on request at editor@healtalkht.com

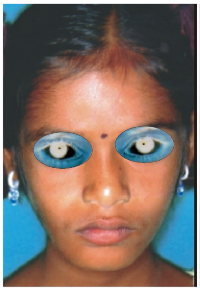

Fig. 1 : Front View

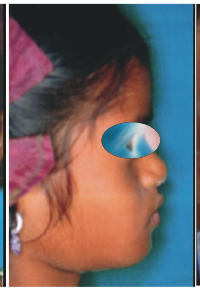

Fig. 2 : Lateral View

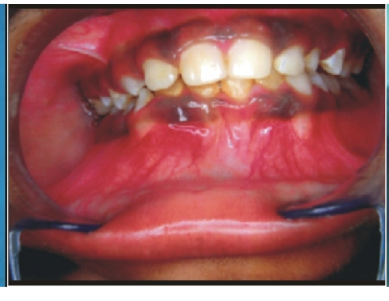

Fig. 3 : Intra-Oral

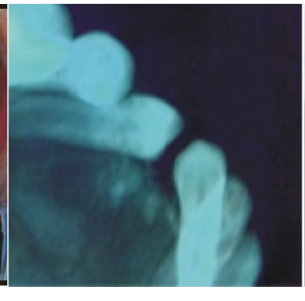

Fig. 4 : IOPA

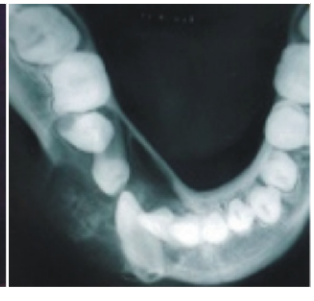

Fig. 5 : Occlusal

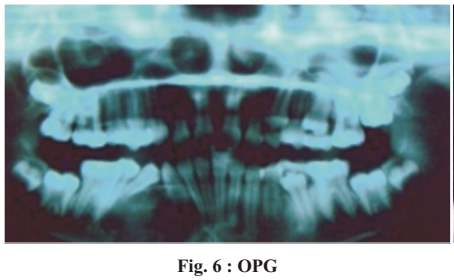

Fig. 6 : OPG

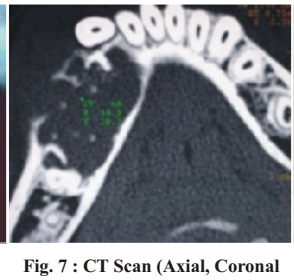

Fig. 7 : CT Scan (Axial, Coronal & 3D Reconstruction)

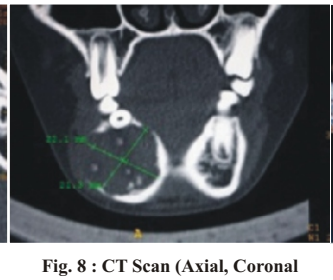

Fig. 8 : CT Scan (Axial, Coronal & 3D Reconstruction)

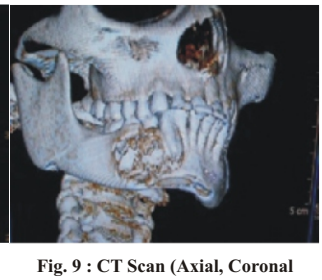

Fig. 9 : CT Scan (Axial, Coronal & 3D Reconstruction)

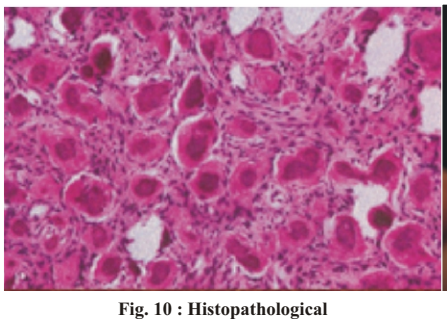

Fig. 10 : Histopathological

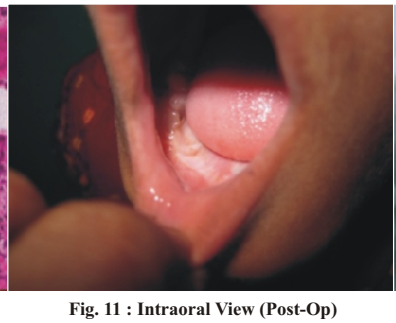

Fig. 11 : Intraoral View (Post-Op)

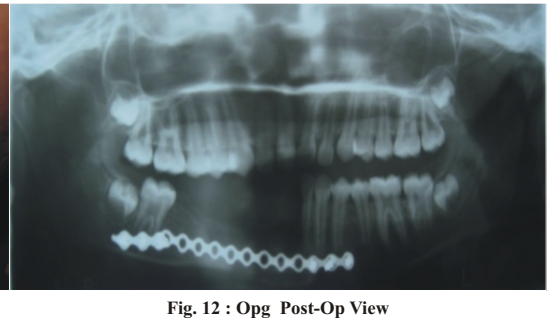

Fig. 12 : Opg Post-Op View