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OSSIFYING FIBROMA – REVIEW AND REPORT OF THREE CASES.

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ABSTRACT

Lesions with fibrous and osseous components include fibrous dysplasia (FD), ossifying fibroma (OF), cementoossifying fibroma (COF), and cementifying fibroma (CF). Fibro-osseous lesions other than FD seem to arise from the periodontal membrane. Ossifying fibroma is a benign fibro osseous lesion of the jaw containing varying amounts of calcified deposits such as bone, cementum or both. It is characterised by presenting a deformed growth and an increased volume of slow evolution with a higher incidence in the mandible and provoke tooth displacement. In image examination, it presents diverse shades of radio opacity within the radiolucent lesion being radio opaque in rare occasions. Here in we report 2 cases of central ossifying fibroma and 1 case of juvenile ossifying fibroma with clinical, radiological and histological features with review of literature.

KEY WORDS: *Ossifying fibroma, Bone, Cementum, Mixed radiolucent - radioopacity*

INTRODUCTION

Cementoossifying fibroma (COF) is considered a benign osseous tumor, very closely related to other lesions such as fibrous dysplasia, cementifying periapical dysplasia or cemento-osseous florid dysplasia, however forming its own entity according to the 1992 classification of the WHO.[1]

Ossifying fibroma develops from the multipotential mesenchymal cells of periodontal origin which are able to form both bone and cementum.[2,3] Although the precise pathogenesis is still unknown, Wenig et al[4] has suggested that trauma induced stimulation may play a role.

In general, ossifying fibroma is an asymptomatic lesion until growth causes swelling and moderate deformation.[6-8] Displacement of the teeth[5,7] can be an early clinical manifestation.[6] The teeth associated with the lesion preserve their vitality and may present root resorption.[5,6] The lesion is relatively slow-growing,[6-9] as a result of which the overlying cortical bone layer and mucosa remain intact,[5,7] and thus the tumor may be present for a number of years before a diagnosis is made.[7]

Juvenile ossifying fibroma is a fibro-osseous neoplasm that arises within the craniofacial region in young individuals below 15 years of age. It is described in the World Health Oraganisation histological typing of odontogenic tumours as an actively growing lesion consisting of a cell rich fibrous stroma, containing bands of cellular osteoid without osteoblastic lining, together with trabeculae of more typical woven bone. Small foci of giant cells may also be present. The lesion is non – encapsulated but well demarcated from surrounding bone.[10]

The present paper describes three cases of which two are ossifying fibroma and one aggressive ossifying fibroma.

CASE 1

A 27 year old female was referred to our institute with a chief complaint of swelling since 12 months. There was no significant medical and dental history. On clinical examination it was 5 x 4 cm in size approximately extending in relation to 34, 35, 36 with bicortical expansion more on the lingual aspect with bulging of the lower border of the mandible (Fig.1 and 2). The swelling was non tender, bony hard in consistency with intact overlying mucosa. There was grade II mobility with 32, 33, and 34. There was no paresthesia or hypoesthesia. The associated teeth were vital. Laboratory findings were wit in the normal limits.

Orthopantomograph revealed mixed image with well defined borders extending from the right lower canine to lower left first molar with intact lower border of the mandible. Teeth associated with lesion were displaced (Fig.3).The patient was later subjected for CT scan and images were obtained in axial, coronal and sagital view with 3-D reconstruction. The images revealed large expan sile lesion about 3.8 X 3.3 cm with bicortical expansion and thinning of the buccal and lingual cortex with intact lower border

CASE I

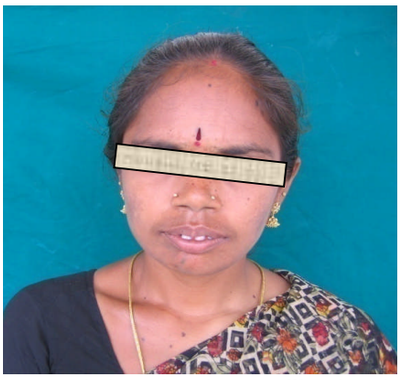

Fig.1. Extra oral view showing swelling involving left side of the mandible

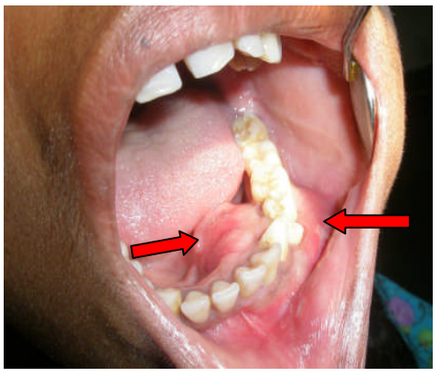

Fig.2.Intra oral photograph showing bicortical expansion

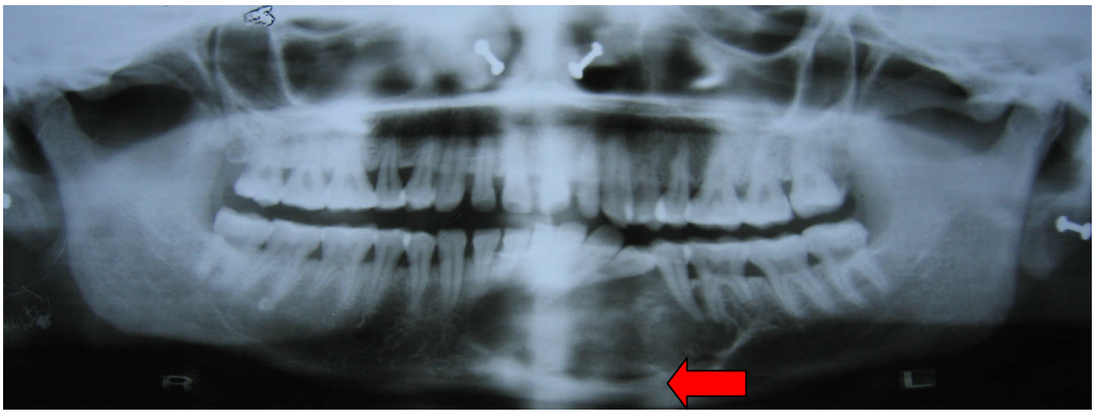

Fig 3. OPG revealing a mixed radiolucent radio opaque lesion involving left body of mandible

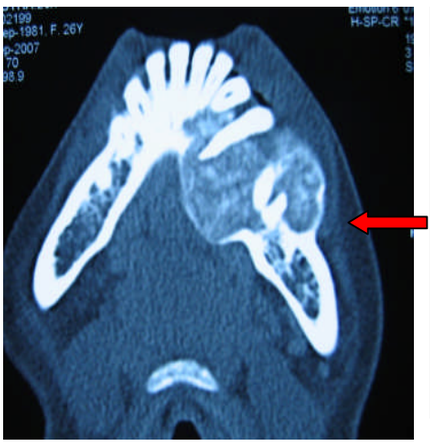

Fig. 4. Axial section showing an expansile lesion with bicortical expansion

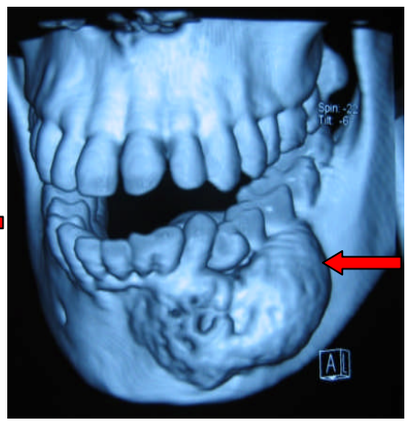

Fig. 5. Three dimensional view from buccal aspect

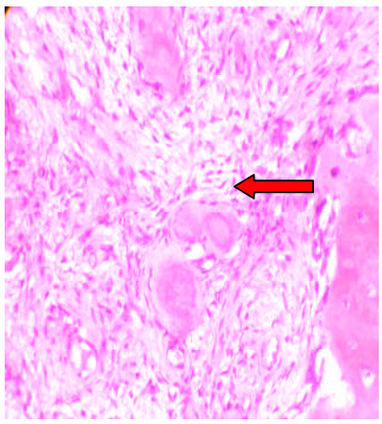

Fig 6. 10 X showing cellular connective tissue with numerous bony trabeculae

of the mandible. The lesion was well defined, non homogenous with radiopaque foci (Fig. 4 and Fig.5). Histopathology revealed cellular connective tissue stroma with numerous bony trabeculae. Stroma comprised of numerous spindle shaped cells with bony trabeculae showing osteoblastic rimming and osteocytes with lacunae. The result of the examination suggested a diagnosis of ossifying fibroma (Fig. 6).

CASE II:

A 35 year old female was referred to our institute with a chief complaint of swelling in the lower jaw since 11 months. There was no significant medical and dental history. On clinical examination it was 4 x 2 cm in size approximately, extending from 33 to 36 with bicortical expansion (Fig.7 and Fig.8). The swelling was non tender, bony hard in consistency with intact overlying mucosa. There was grade II mobility with 34, 35. There was no paresthesia or hypoesthesia. The associated teeth were vital. Laboratory findings were with in the normal limits.

Orthopantomograph revealed well defined radiolucent lesion extending from 32 to 36. The premolars were displaced. The lower border was intact (Fig 9). CT scan revealed expansile lesion with bicortical expansion. The cortical plates were thinned. There were no internal septas or radio opaque foci. 3 D construction revealed perforation of the buccal cortical plate (Fig.10 and Fig.11).

Histopathology revealed a highly cellular connective tissue stroma comprising of plump spindle shaped fibroblasts along with focal areas of ossification and calcification. The result of the examination suggested a diagnosis of ossifying fibroma (Fig. 12).

CASE III

A 18 year old lady presented to our out patient department with a chief complaint of swelling in the lower jaw since 7 months. No significant medical or dental history was present. On clinical examination the swelling was in the symphysis region measuring approximately 4 x 3 cm. intraorally the swelling was extending from 33 to 47 with bicortical expansion (Fig.13 and Fig. 14). The swelling was mildly tender, bony hard in consistency with intact overlying mucosa. There was no paresthesia or hypoesthesia.

Orthopantomograph revealed well defined mixed image extending from 33 to 46. The lower border was thinned but intact. There was no displacement of the associated teeth (Fig.15). CT scan revealed an expansile lesion with bicortical expansion and thinning of the cortices (Fig. 16).

Histopathology showed dense cortical bone surrounding a loose network of fibrous connective tissue stroma. Many areas with osteoid matrix and irregularly formed bony trabeculae were also seen. The result of the examination suggested a diagnosis of juvenile ossifying fibroma (Fig.17).

Discussion

Yih[11] et al. and Sciubba[12] et al. attributed the first description of this disorder to Menzel, in 1872. In 1927, Montgomery[13] first used the term ossifying fibroma, by which the lesion is currently known. COF is a benign, uncommon, monostotic well-defined unilocular or multilocular fibrous-osseous tumor, arising from the periodontal ligament composed of fibrous connective tissue with variable amounts of metaplastic bone and mineralization. The lesion is generally encapsulated – a fact that serves to distinguish it from fibrous dysplasia, which may exhibit similar clinicopathological features.[14,15,16] Although the lesion is preferentially located in the jaws, it can also be found elsewhere – including the frontal, ethmoid, sphenoid and temporal bones or orbit, as well as in the anterior cranial fossa.[17,18] Some authors have pointed to antecedents of trauma in the area of the lesion, the performance of tooth extractions, and the prior existence of periodontitis, as possible triggering factors.[19] COF most frequently occurs in female patients (age range 10–59 years, mean 32 years) with an incidence peak in the third and fourth decades.[20] Similarly two of our cases were in third and fourth decade and were female. The mandible, including the ramus, more commonly in the molar and premolar zone, is the region most commonly affected as seen in our cases.[21,22] It appears as a hard, localized and slow-growing mass that displaces the teeth, though the latter remain vital and the overlying mucosa is characteristically intact.[23] The size of the lesion can range from 0.2-15 cm[24]; in our case the first two cases measured 4 - 5 cm in diameter. Radiologically, the lesion appears well circumscribed, and is initially seen as an osteolytic image followed by gradual trans formation into a mixed lesion – in exceptional cases becoming radiopaque. [19, 23, 25] Some authors,[12,14] including Eversole[26] et al., have described two basic radiological patterns: a unilocular radiotransparency with or without radiopaque foci, and a multilocular radiotransparency.

CASE II

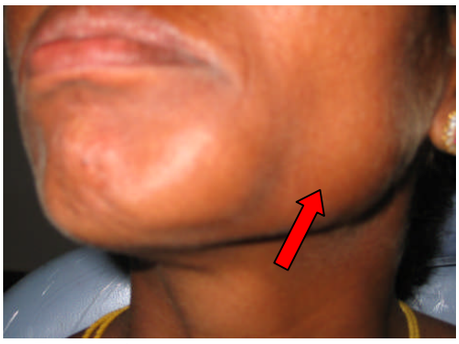

Fig.7. Extra oral view showing swelling involving left side of the mandible

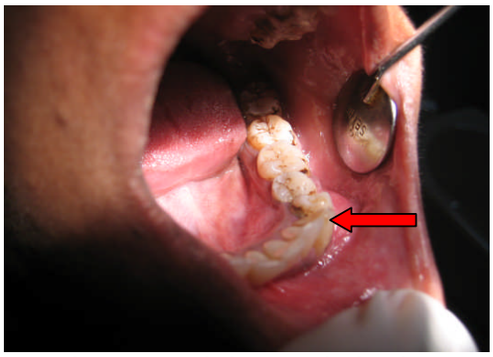

Fig.8.Intra oral photograph showing Swelling

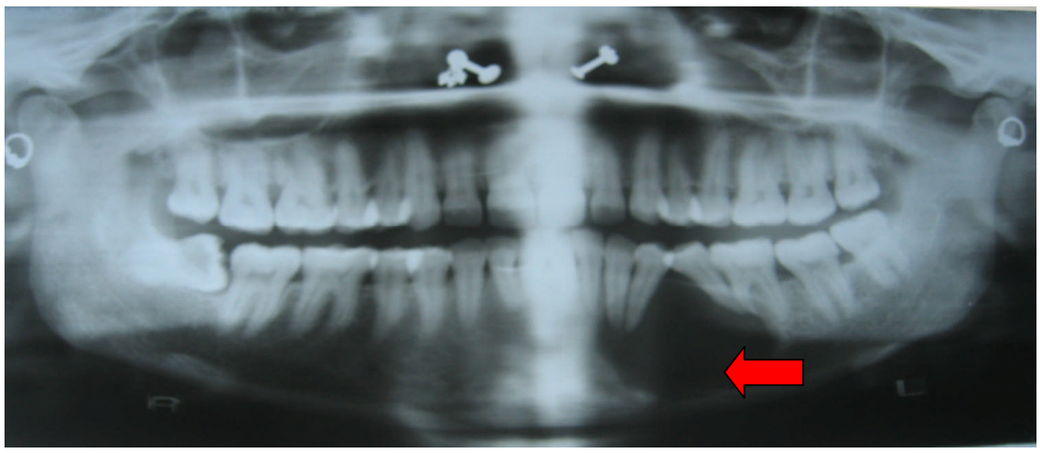

Fig 9. OPG showing well defined radiolucent lesion involving left body of mandible

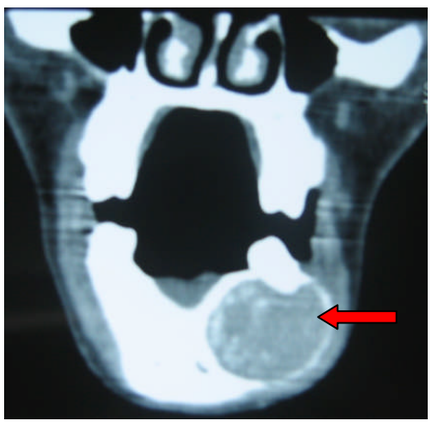

Fig. 10 coronal section showing an well defined expansile lesion with intact cortical plates

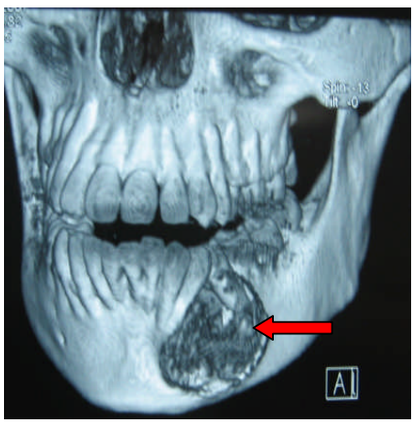

Fig. 11. Three dimensional view of the swelling

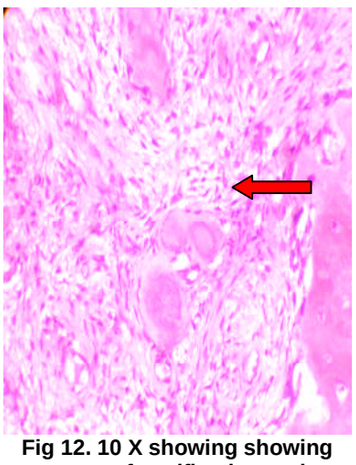

Fig 12. 10 X showing showing areas of ossification and calcifications in the stroma

Su[24] et al. commented that the radiological image most often associated with cementoossifying fibroma corresponds to a well defined radio transparency with or without a sclerotic margin, and often accompanied by cortical expansion. Ortho pantomography revealed the presence of a well defined unilocular lesion, with a central radio opacity zone and a more lucent peripheral area, in the first case while second case completely unilocular radiolucent and the third case revealed flocculent radio opacities. Computed tomography in turn afforded a clearer image of the lesions. Expansion of the vestibular cortical layer was also visualized, together with thinning and perforation of the cortices.

In the craniofacial region the lesion enlarges slowly and symmetrically resulting in bone expansion and facial deformity, but some lesions are asymptomatic and are discovered only during routine radiographic examination.[22, 27] Centrifugal growth will commonly cause bowing of the inferior border of the mandible but cortical perforation is rare.[20] However in our cases cortical perforation was present as seen on CT. The lesion may occasionally cause displacement of the mandibular canal.[28] Resorption and/or divergence of roots can occur with the continuous growth of the lesion, as in our cases although there was divergence of roots, there was no root resorption.[20]

Uncomplicated cases can be treated by simple enucleation of the lesion with curettage alone. Because the lesions are well circumscribed, they are removed easily from the surrounding tissue. On the other hand larger lesions require radical surgical resection.[20,28] It is also recognized that if there is no facial deformity and no evidence of concomitant osteomyelitis, a wait and see approach is an appropriate treatment choice.[29]

Juvenile active ossifying fibroma (JAOF) is rare. It is seen most often in patients between the ages of 5 years and 15 years (60% to 80% of cases).[30]Cases have however been reported in both older and younger patients,[31] some authors suggesting an age range of presentation from 3 months to 72 years.[32, 34] There is no clearly defined racial or sexual predilection. With its diverse histopathology, no clear-cut criteria exist for separating JAOF from the more common forms of ossifying fibroma but its behavior is markedly different. [32] The tumor can increase rapidly in size in a relatively short time as in our third case. The most common site of involvement for JAOF is the craniofacial skeleton primarily the maxilla, paranasal sinuses, orbital and frontoethmoid bones.[30,31] Among the 85% of facial lesions, 90% occur in the paranasal sinuses; most commonly the ethmoid sinuses 32,33. The remaining 10% arise from the mandible. Our case was 18 year old involving the mandible. Clinical manifestations usually begin with swelling of the affected site. The patient may have neurosensory disturbances ranging from pain to paresthesia in the involved area as in our case. The radiographic appearance of JAOF is of a unilocular, oval or spherical lesion with a distinct osteolytic density delineating it from normal bone. As the lesion enlarges, it can become increasingly radio dense, although still remaining less dense than the adjacent normal bone. Tooth displacement and partial destruction of the roots of teeth is well reported. The painless rapid growth demonstrated by this juvenile form of ossifying fibroma and the tendency to a locally aggressive growth pattern has justified the prefix ‘‘active’’ or ‘‘aggressive’’ in reference to these tumors. Histologically, JAOF shows some variance but typical features include a highly cellular stroma containing calcified components, which is usually fibroblastic and relatively avascular accounting for the lack of enhancement on imaging; woven and interconnecting lamellar bone with numerous eccentric rims of osteoblasts are also demonstrated. The highly cellular nature of the stroma reflects the aggressive nature of these tumours.[32,34,35] Complete surgical excision is necessary to obviate local recurrence. This can usually be achieved by local excision and curettage.[31,34] Adjunctive treatment is not required. Malignant transformation has never been recorded. JAOF, although a rare lesion, is worthy of inclusion in the radiological differential diagnosis of a rapidly expanding lesion affecting the craniofacial skeleton of an adolescent or young adult. Differentiation from other benign and malignant neoplasms is important in the correct therapeutic management of this tumor.

CONCLUSION:

Cemento ossifying fibroma is a benign fibro osseous lesion considered to be of periodontal ligament origin. Three such cases of cemento ossifying fibroma are reported here along with their clinical, histological features and methods of management. Among the three reported cases one is an aggressive juvenile type which is rare and its histological diagnosis is of importance as clinically it may resemble a malignant osseous tumor and Correct diagnosis is critical in its management.

CASE III

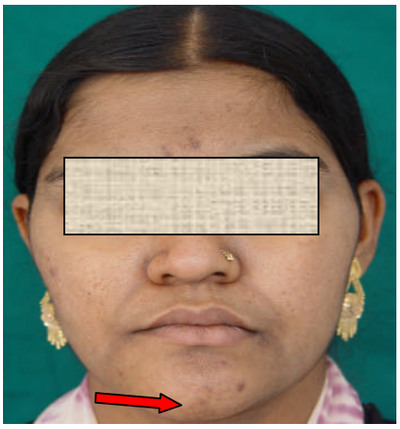

Fig.13. Extra oral view showing neglible swelling at the symphysis of the mandible

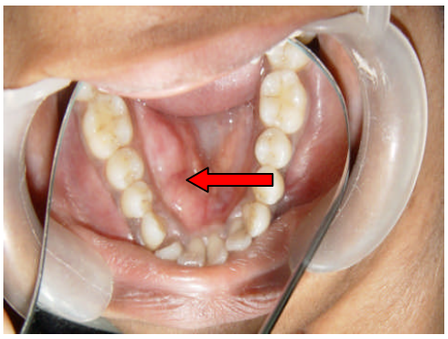

Fig.14 .Intra oral photograph showing Marked lingual swelling

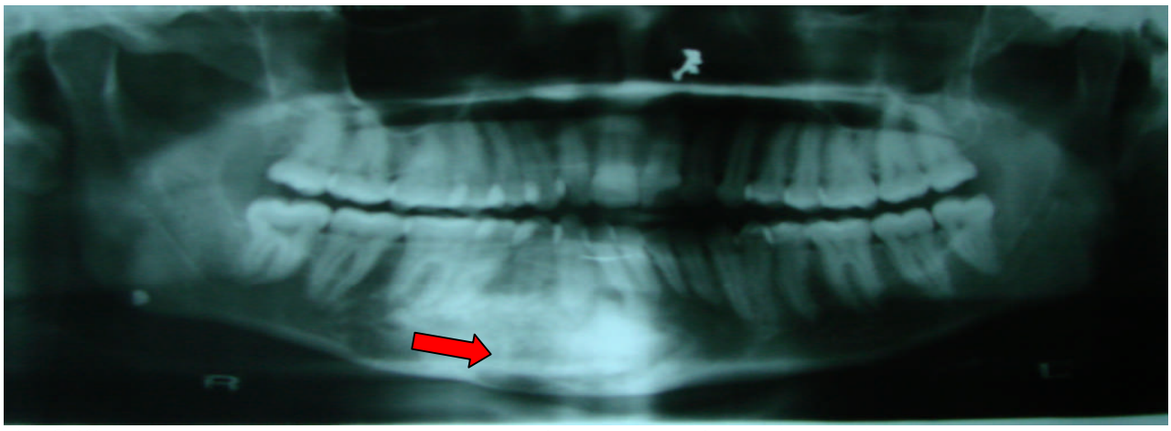

Fig 15. OPG showing a mixed radio opaque radiolucent lesion on right side of body of mandible

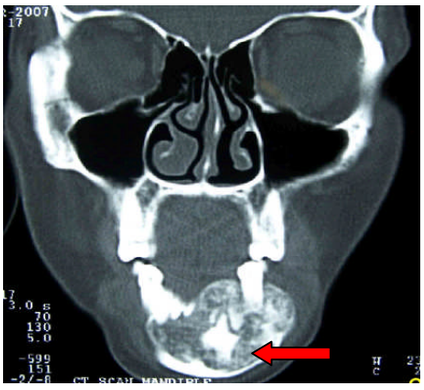

Fig. 16 coronal section showing a non homogennous image with bicortical expansion

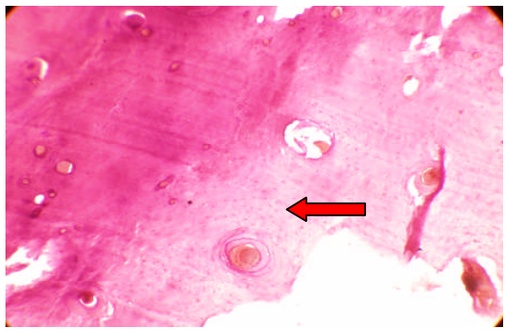

Fig 17. 10X showing osteoid matrix in connective tissue stroma

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