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Juvenile Ossifying Fibroma Versus Fibrous Dysplasia: A Dilemma Among Clinicians

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ABSTRACT
The dilemma of diagnosis between two fibro-osseous lesions that is fibrous dysplasia and ossifying fibroma is not uncommon as both fibro-osseous lesions can mimic each other's clinical, histopathological and radiological features. We came cross one such case which was clinically diagnosed as juvenile ossifying fibroma but the histopathologically it was diagnosed as craniofacial fibrous dysplasia.
Keywords: *Fibro-osseous lesions, Fibrous dysplasia, Ossifying fibroma, Pediatric, Dental*

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INTRODUCTION

The fibro-osseous lesions occur as a result of substitution of normal bone structure by fibrous matrix that contains variable amounts of mineralized material such as bone, osteoid, or cementum[1-2]. The dilemma of diagnosis between two such lesions that is fibrous dysplasia (FD) and ossifying fibroma (OF) is not uncommon as both fibro-osseous lesions can mimic each other's clinical, histopathological and radiological features. A diagnostic evidence for distinguishing these two lesions is that OF has well-demarcated margins whereas in FD the lesion blends into the normal bone without any differentiation but sometimes this can also be confusing[3-4]. Here, we present a case of fibro-osseous lesion, which on the basis of clinical and radiological features was diagnosed as juvenile ossifying fibroma(JOF) but later thehistopathologically report came as FD.

CASE REPORT

A 13 years old, male presented with a diffused swelling with respect to right middle third of face, which increased in size over five months. Extra-orally, the swelling was approximately 4X4 cm in dimension, extending supero-inferiorly from the right infraorbital margin to the horizontal line joining the right corner of the mouth to the ipsilateral tragus and antero-posteriorly from the perpendicular line drawn from medial corner of the right eye till the perpendicular line drawn from the lateral corner of the right eyebrow (Figure 1).

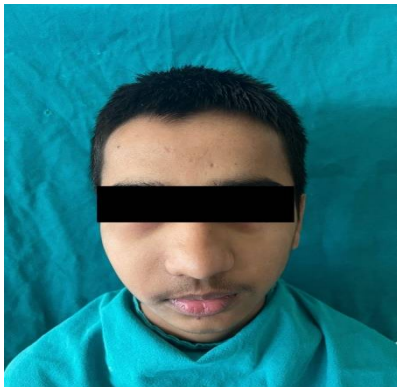

Figure 1. swelling involving the right moddle third of face.

There was no change in skin overlying the swelling. Obliteration of right nasolabial fold was seen along with slight dropping of the right corner of the mouth. On palpation, the swelling was found out to be hard in consistency without any tenderness; it was non-reducible, non-compressible and non-pulsatile. The overlying skin appeared normal with no rise in local temperature when compared to contralateral side. There was no paresthesia of the overlying tissues. Intra-orally, the bony swelling with respect to right maxillary alveolus extended from right maxillary canine till right maxillary second molar with obliteration of the corresponding vestibule (Figure 2).

Figure 2. Intraoral picture showing swelling obliterating the right upper vestibule

No ulcerations, draining sinuses or scars were seen. There was no mobility and tenderness with respect to the corresponding teeth, however canting of occlusion was seen with respect to the right maxillary arch along with posterior open bite on the contralateral side. Patient was examined for café au lait spots and endocrinopathies, both were absent. Cone Beam Computed Tomography scan revealed a hyperdense lesion involving right side of maxilla, of size 47.60 mm (medio-laterally) X 39.20 mm (antero-posteriorly) X 46.83mm (supero-inferiorly), with non-corticated but well defined borders. The radio-density of lesion was variable varying between 55HU to 386HU showing a ground glass appearance. The lesion fully obliterated the right maxillary sinus, ipsilateral nasal turbinates also seemed enlarged (Figure 3 and 4).

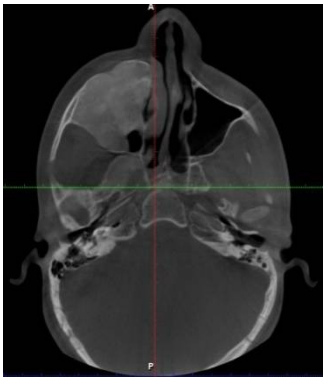

Figure 3. Axial section of CBCT showing lesion of right maxilla.

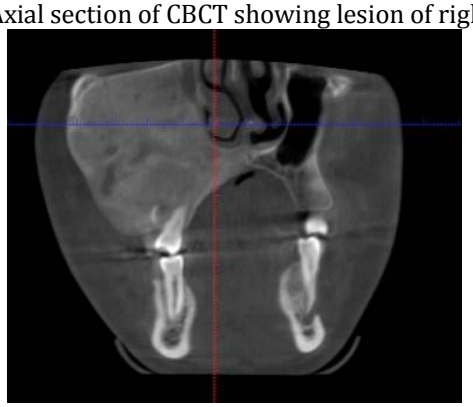

Figure 4. Coronal section of CBCT showing lesion of right maxilla, palatal expansion

In axial section expansion of buccal as well as palatal cortex can be seen with displacement of right upper canine and lateral incisor (Figure 5.).

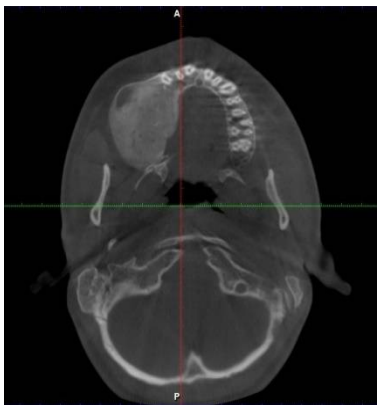

Figure 5. Axial section of CBCT showing expansion of buccal and palatal cortex

Serum calcium and alkaline phosphatase levels came out to be normal. The patient had an identical twin brother who did not have any such lesion. A clinical diagnosis of JOF was given as the swelling was well distinguished from the adjacent tissues unlike fibrous dysplasia as it has ill-defined margins merging into the normal adjacent bone.A tissue biopsy was taken which was reported as Chinese letter pattern of immature bony trabeculae in a loosely arranged fibrous stroma giving a hint of fibrous dysplasia, thus a final diagnosis of craniofacial FD was given. Since, the lesion was asymptomatic, wait and watch approach was suggested with review after every three months. Any cosmetic correction required would be done after the patient has completed his growth that is after 18 years of age.

DISCUSSION

The fibro-osseous lesions occur as a result of substitution of normal bone structure by fibroblasts and collagen, containing inconsistent amounts of mineralized material, these lesions include a diverse group of several benign entities which have recently been classified by Eversole. The dilemma of diagnosis between two such predominant lesions that is FD and OF is not uncommon as both these lesions can have overlapping features. A radiological evidence for distinguishing these two lesions is that OF has well-defined margins whereas in FD the lesion blends into the normal bone without any differentiation but sometimes this can also be confusing. Histopathological examination then becomes more reliable [5-6].

Both FD and JOF occur predominantly in maxilla. The polyostotic FD is typically seen in age less than 10 years, whereas the monostotic FD is found in somewhat elder age, whereas JOF is seen under the age of 15 years, FD shows no gender predilection but in JOF, it is debatable. Our patient presented with the lesion of posterior maxilla at the age of 13 years. Both lesions are painless, however FD can become symptomatic if adjacent vital structures are compressed. FD is slow growing and arrests with age where as JOF is rapidly growing and aggressive, it may cause displacement of teeth, in our case, the lesion was rapidly growing that is a period of five months[7].Radiologically, JOF is reported as a well-defined, expansive lesion with variable density of mineralization, perforation may be present whereas in FD, the lesion ill-defined with no clear cut borders. Both the lesions may be radiolucent in the initial stages but the radiopacity increases as the lesion progresses. Our lesion showed well defined margins with variable density which inclined our diagnosis towards JOF[8-9]

Histologically, JOF shows areas of condensed cellularity which may substitute with myxomatous regions. Bony trabeculae are unevenly distributed, giant cells are also seen. El Moftyidentified two histological types, that is trabecular JOF and psammomatoid JOF. The psammomatoid JOF commonly involves orbit and paranasal sinuses, whereas the trabecular type mainly involves the maxilla, mandible is rarely involved.The pathognomonic feature of the psammomatoid type is the presence of eosinophilic spherical structures termed as psammoma like bodies, dispersed in a fibrous stroma. FD on the other hand, consists of irregular bony trabeculae mixing into the adjacent normal bone lying within a cellular fibrous stroma, the diverse shapes of the trabeculae give a Chinese letter appearance[10]. Similar findings were there in our case, coming to a final diagnosis of craniofacial fibrous dysplasia.

Since JOF is aggressive and shows a recurrence rate of 36-50%, therefore adequate resection is required whereas, fibrous dysplasia is to be treated conservatively by cosmetic contouring and bone shaving in case of nerve compression, therefore proper distinguished diagnosis is required to facilitate the formulation of management protocol[11-12].

CONCLUSION

Differentiating FD and JOF is extremely necessary as both may have overlapping features but the management modalities are different for both as OF requires aggressive management whereas fibrous dysplasia requires conservative approach. We suggest thorough histopathological study to come to a final diagnosis after correlating with clinical as well as radiological features.

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