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Diagnostic challenging fibro-osseous lesion of the jaw - A case report

[1]Dr. Maumita Bhattacharya, Reader**,** Department of Oral Pathology, K.S.D. Jain Dental College and Hospital, Kolkata**.**

[2]Dr. Subhalakshmi Sen**,** Reader, Department of Oral Pathology, KSD Jain Dental College and Hospital.

[3]Dr. Rakshith Shetty, Professor, Department of Oral Pathology, KSD Jain Dental College and Hospital.

[4]Dr. Abhishek Rathi, Reader, Department of Oral and Maxillofacial Surgery, IDST, Kadrabad, UP.

Corresponding Author: Dr. Maumita Bhattacharya, Reader**,** Department of Oral Pathology, K.S.D. Jain Dental College and Hospital, Kolkata**.**

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Abstract

Fibro-osseous lesions are relatively common bone disorders and represent a clinically diverse group of disorders that share similar histopathologic features.

Although the general concept of these lesions is relatively well known, but specific diagnostic interpretation of individual cases is often challenging.

This case report presents the diagnostic challenges review is presented to update the surgical pathologist on the various entities comprising the spectrum of fibro-osseous lesions and to examine the criteria for their diagnosis.

Keywords: Fibrous dysplasia, ossifying fibroma, mutation, classification, differential diagnosis.

Introduction

Fibro-osseous lesions are assorted jaw disorders characterized by the replacement of normal bone into fibrous connective tissue matrix and in secondary attempt at new formation of dense bone.[1,2] Ossifying fibroma (OF) and fibrous dysplasia (FD) are quite a common fibro-osseous lesions (FOLs). There are major differences in the clinical and radiological presentation of these two lesions and in some cases there are major overlaps in findings causing difficulty in final diagnosis.[3]

In histological aspect, so much similarity in both the lesion, diagnosis of the either one them by only histological basis is very difficult.[4] Till date basis for final diagnosis and clear objectivity is therefore called into question, with overlapping clinical and histological presentation, final diagnosis can only reached by the further investigation.[4]

Comprehensive classification has been suggested by Eversole et al in 2008 suggests that the classification of

this disease is likely to evolve still further. This classification includes neoplasm, developmental dysplastic lesions and inflammatory/reactive processes. The basis of this classification is that definitive diagnosis can rarely be rendered on the basis of histopathological features alone rather; procurement of a final diagnosis is usually dependent upon assessment of microscopic, clinical and imaging features together.

The various Classifications systems proposed by authors are enumerated as below.[5]

- Charles Waldron Classification of The Fibro-Osseous Lesions of The Jaws (1985)
- Working Classification of Fibro-Osseous Lesions by Mico M. Malek (1987)
- Peiter J. Sloot weg & Hellmuth Muller (1990)
- WHO Classification (1992)
- Waldron Modified Classification of Fibro-Osseous Lesions Of Jaws (1993)
- Brannon & Fowler Classification (2001)
- WHO Classification of Fibro-Osseous Lesions of Jaws (2005)
- Paul M. Speight & Roman Carlos Classification (2006)
- Eversole Classification (2008)

Many New concepts and controversies have arisen over the past 10 to 15 years regarding classification and diagnostic criteria. However, among the new theories and contentions, there is now essential agreement that the osseous dysplasias represent a single disease process, while the so-called “juvenile active ossifying fibroma” and other “aggressive,” “active,” “psammomatoid” ossifying /cementifying fibromas remain controversial.

Advance diagnostic tools are used for the final diagnosis by genetic variants like Polymerase chain reaction for mutations at Arg-201 codon of the alpha subunit of the transmembrane stimulatory G protein gene (GNAS-1) has been shown to be a marker for jaw FD.[6] Pimenta FJ et al, 1996 found that the mutation of the tumor suppressor gene HRPT2 in ossifying fibroma and results showed that haplo insufficiency of the HPRT2 gene.[7]

In Immunohistochemical test author found that in some area of bone matrix proteins non-significant changes in both the lesions expect osteocalcin have strong expression in the calcified region of fibrous dysplasia but in there less and no expression of osteocalcin in ossifying fibroma case.[8]

Distinguishing between FD and OF using either the genetic or immunohistochemical tests in each case of a FOL may not be feasible in all cases for all laboratories due to overhead costs and or patients inability to afford such tests. [9]

Case report

A 25 years old male patient reported to Department of Oral pathology in KSD Jain Dental College and Hospital, West Bengal with the chief complaint of discomfort during chewing and speaking due to a growth in the left lower posterior region of the jaw, which started as a small growth of approximately 6-8 months ago and gradually increased in size with time to attain present size. There was no associated history of bleeding or pain and medical history was non-significant and no history of any medication at that time.

Detailed medical, family, dental and personal habit history was taken and no relevant history was found. Intraoral examination was performed and it revealed an approximately 2 cm × 2 cm sessile, non-tender, firm, pale pinkish growth present on the interdental gingiva in relation to the mandibular left premolar region [Figure-1]. No extra oral swelling was evident. Intra-orally the lesion was firm on palpation, no bleeding or exudation and discharge on percussion was noted.

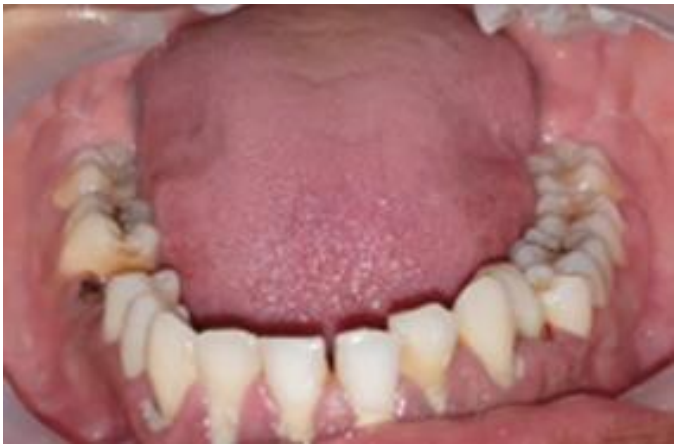

Figure 1: Clinical presentation of the lesion on left lower back tooth region.

The clinical differential diagnoses for the case was given were pyogenic granuloma, traumatic fibroma, peripheral giant cell granuloma, and peripheral ossifying fibroma with respect to the 32, 33, and 36 region.

Radiographically, there was angular bone loss in relation to mandibular left lateral premolar and molar region with displacement of mandibular first premolar on measurement on intraoral periapical and occlusion radiograph and 3x3x6 cm.

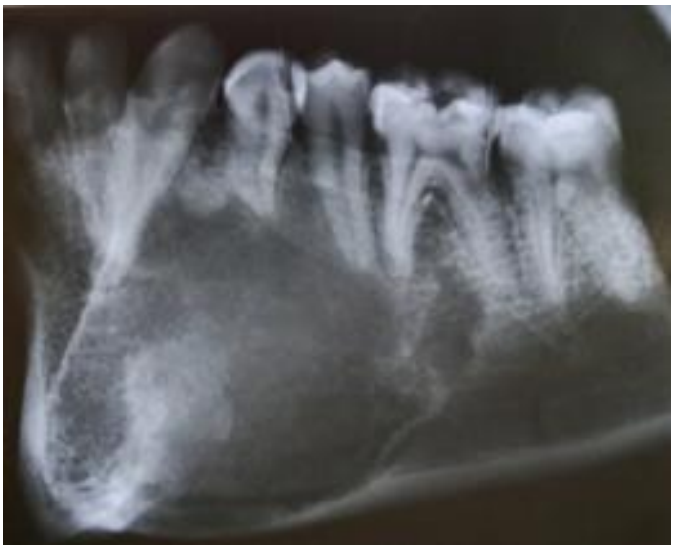

Figure 2: Intraoral periapical radiograph showing radiolucent lesion involving roots of lateral incisor to mesial root of mandibular first molar.

Thorough blood investigation was performed and all the reports were found to be in the normal reference range. The growth was then excised conservatively. Adjacent teeth were scaled to remove the local irritants. Tissue was sent for histopathological examination.

The gross tissue was oval, 6.5x6x6 cm in size, brownish to blackish in colour, and firm in consistency. [Figure-3]

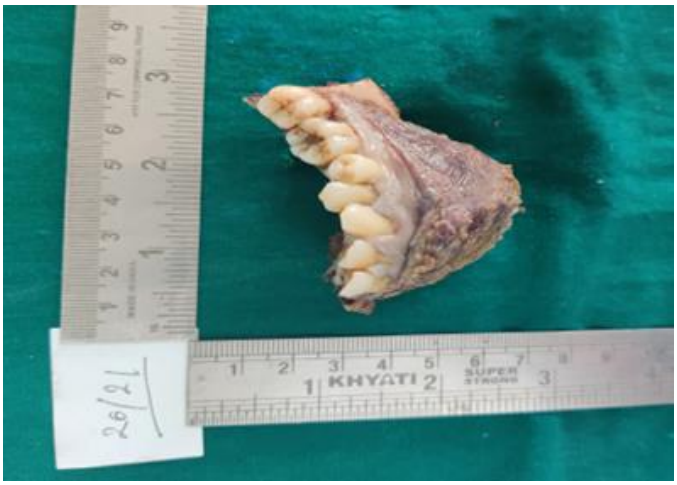

Figure 3: Gross specimen

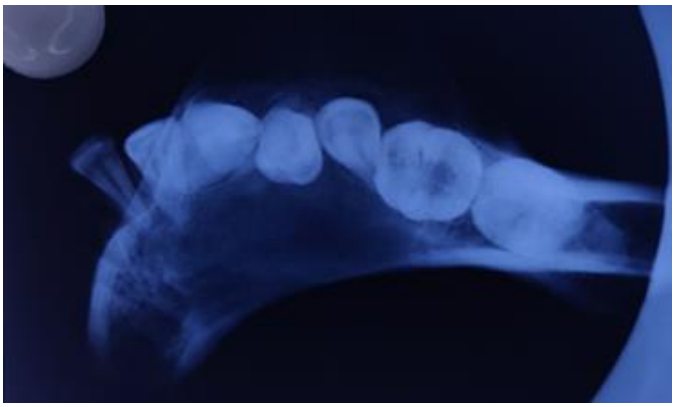

Figure 4: Occlusion radiograph showing intact cortical plates.

Specimen radiograph was performed for the case. The occlusal radiograph shows expansile swelling, radiolucent lesion with intact cortical plates and teeth displacement. Also, there was an angular bone loss in relation to mandibular left lateral premolar and molar region with displacement of mandibular first premolar. Radiograph shows expansile swelling, radiolucent lesion with intact cortical plates, teeth displacement.

Methodology

The tissue was stained with routine Hematoxylin and eosin stain and slide was viewed with a Lawrence & Mayo, LYNX research microscope (MODEL: LM- 52-RS) and photomicrographs were recorded with a connected digital camera DS-Fi1 together with a Nikon digital sight control unit (Nikon, Tokyo, Japan).

Since most common fibro- osseous lesions like fibrous dysplasia and ossifying fibroma have clinical, histological and radiographic characterization very similar to each other. So for further investigation tolu dine blue staining was performed.

Toluidine blue[10]

Udeabor, 2018 described the method of staining in his study we use in this case for toluidine blue. The sample is received from the surgery department in formalin. The formation of fixed paraffin embedded (FFPE) blocks for staining process. Each tissue was passed through xylene and descending grades of ethanol then rinsed in distilled water for 3 min and placed in nuclear fast red solution for 30 min, rinsed twice under running water and once in distilled water. The sections were deparaffinized, hydrated to distilled water and stained with toluidine blue solution for 1–2 min. They were then washed in 3 changes of distilled water and dehydrated quickly through 95% and absolute alcohol, cleared in 2 changes of xylene for 3 min and a cover slip was placed with DPX.

Histopathology

On histopathological evaluation, the section showed fibro cellular connective tissue stroma composed of large number of plump proliferating fibroblast. The connective tissue is arranged in discrete bundles interspersed with few chronic inflammatory cell infiltrates. There are areas of mineralization of varying pattern dispersed throughout the connective tissue. The mineralized masses are in the form of bony trabeculae of irregular shapes and sizes with peripheral osteoblastic rimming. Few muscle and proliferating endothelial lined blood vessels are also seen in the stroma. (Figure 5).

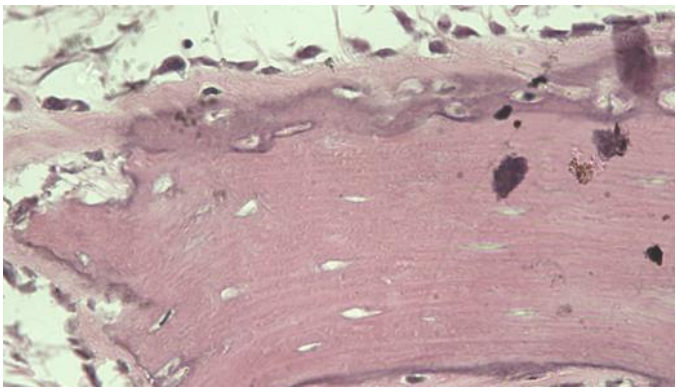

Figure 5: Hematoxylin & Eosin-stained Section

Ox Talan fibres have been reportedly seen in fibro-osseous lesions and they were found more in ossifying fibroma than fibrous dysplasia. In this case, tolu dine blue was found to be strongly positive in ossifying fibroma. (Figure 6). The above histopathological features were found to be suggestive of Ossifying Fibroma

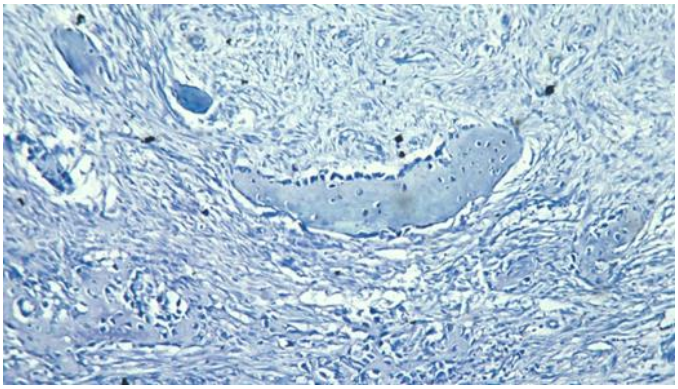

Figure 6: Toluidine Blue stained section of Ossifying Fibroma

Discussion

Ossifying fibroma (OF) is a benign, non-odontogenic tumor of the jaw, a type of fibro-osseous lesion. Traditionally, this type of lesion was sub classified histologically into ossifying fibroma and cementifying fibroma according to the hard tissues formed, but both

types are now known by the unified term, ossifying fibroma. It is generally accepted that the histological subclassification of these two lesions is of academic interest only since differential diagnosis is often arbitrary and their biological behaviour seems to be identical.

Central ossifying fibromas are typically well-circums cribed, solitary radiolucencies with scattered radiopaque foci. The lesion commonly occurs at apices of vital teeth in premolar-molar region. There is spherical expansion without cortical perforation, and may lead to divergence of adjacent teeth. [19] In our case the lesion was spherical in shape and expansile but the lesion was not well-circumscribed as mentioned in literature time and again. Also there was no displacement of teeth. The nasal septum, infraorbital foramen and orbital floor may be involved, if the tumor is large.

Surgical therapy is decided by the extent of tumor.[1,2] Histo pathological features include proliferation of irregularly shaped calcifications within a hypercellular fibrous connective tissue stroma. The calcifications are extremely variable in appearance and represent various stages of bone and cementum deposition. Histological differentiation between osteiod and cementum is difficult. Additional biochemical studies such as ultrastructural studies and polarized studies have been utilized for differentiating between cementum-like material and bone in these lesions with no definite results.

Females are more commonly affected than males, and the anterior maxilla is the most common site of occurrence. POFs are diagnosed at any age. Clinically, POF usually presents as a solitary, slow-growing, pedunculated or sessile, nodular mass. To the best of our knowledge, a unique case of multicentric manifestation has been reported. The surface mucosa can be smooth or ulcerated and pink to red in color.[2] Migration of teeth with interdental bone destruction has been reported in some cases. POFs usually measure <1.5 cm in diameter even though lesions of 6 cm and 9 cm in diameter are recorded. On the clinical level, differential diagnoses include peripheral giant cell granuloma, pyogenic granuloma.[3]

The presence of ox Talan fibers interspersed among the calcified structures, the almost exclusive occurrence on the gingiva, and the age distribution inversely correlating with the number of lost permanent teeth support the hypothesis of an origin from the periodontal ligament. Moreover, the fibro cellular response of POF is similar to that observed in other reactive gingival lesions originating from the periodontal ligament (e.g. fibrous epulis).[1,2] A possible hormonal influence has also been considered mainly because POFs are rare in prepubertal patients. However, a recent study failed to demonstrate the expression of estrogen or progesterone receptors in the proliferating cellular component. Regezi et al. found a large number of $XIIIa^{+}$ cells, a subset of monocyte/macrophages, in POF and in other oral fibrovascular reactive lesions; it was hypothesized that these dendrocytes could play a distinct pathogenic role. An important clinical aspect of POF is the high recurrence rate, which ranges from 8% to 45%.

Menzel first described the lesion ossifying fibroma in 1872, but its terminology was given by Montgomery in 1927.[3] Peripheral ossifying fibroma occurs mostly in craniofacial bones and categorized into two types central and peripheral. The central type of ossifying fibroma arises from the endosteum or the periodontal ligament (PDL) adjacent to the root apex and expands from the medullary cavity of the bone, and the peripheral type occurs on the soft tissues overlying the alveolar process.[5-9] POF is a solitary, slow growing nodular mass

that is either pedunculated or sessile. Most often it is located in the gingival papilla between adjacent teeth.[10]

Though the etiopathogenesis of POF is uncertain, origin from cells of periodontal ligament has been suggested. The reasons for considering periodontal ligament origin include excessive occurrence of POF in the gingival interdental papilla, the proximity of the gingival to periodontal ligament, the presence of ox Talan fibres within the mineralized matrix of some lesion, and the fibro cellular response in periodontal ligament [2, 8]. Migration of teeth with interdental bone destruction has been reported in some of the cases.[12-14]

In vast majority of cases, there is no apparent underlying bone involvement visible on the roentgenogram. However, superficial erosion of bone is noted occasionally.[17]

Peripheral ossifying fibroma has to be differentiated from traumatic fibroma, peripheral giant cell granuloma, pyogenic granuloma, and peripheral odontogenic fibroma. [18]

Peripheral odontogenic fibroma is an uncommon neoplasm that is believed to arise from odontogenic epithelial rests in periodontal ligament or attached gingiva itself. Traumatic fibroma occur on buccal mucosa along the bite line. Pyogenic granuloma presents as soft, friable nodule, small in size that bleeds with tendency to hemorrhage and may or may not occasionally or do not show calcifications but tooth displacement and resorption of alveolar bone are not observed. Peripheral giant cell granuloma has clinical features similar to POF however POF lacks the purple or blue discoloration commonly associated with peripheral giant cell granuloma and radiographically shows flecks of calcification.[19]

It is possible to histologically differentiate PGCG and peripheral odontogenic fibroma from POF as PGCG contains giant cells, whereas peripheral odontogenic fibroma contains odontogenic epithelium and dysplastic dentine; all the features are not seen in POF.[20-21]

Conclusion

Ossifying fibroma is a slowly progressing lesion, the growth of which is generally limited. Many cases will progress for long periods before patients seek treatment because of the lack of symptoms associated with the lesion. A slowly growing pink soft tissue nodule in the anterior maxilla of an adolescent should raise suspicion of a ossifying fibroma. Discussion of the differential diagnosis should be done tactfully to prevent unnecessary distress to the patient and family. From our experience and other reports noted, the diagnosis of OF and FD by Histopathology alone is mostly incorrect and thus should be supported by relatively simple histomorphometric analysis such as Toluidine blue staining in the absence of more advanced techniques like immunohistochemistry and genetic analysis.

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