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Original Research Article

Spectrum of fibro osseous lesions of jaw: An institutional study

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ABSTRACT

Introduction: Fibroosseous lesions (FOLs) of craniofacial complex are poorly defined but pathologically diverse group of conditions. FOLs are represented by variety of disease processes mimicking clinically and histopathologically as neoplasms, developmental dysplastic lesions or inflammatory reactive process leading to difficulties in diagnosis and hence clinicopathological correlation is required.
Objective: To analyse fibro osseous lesions for demographic details and clinicopathological correlation.
Materials and Methods: 26 cases of clinicopathologically diagnosed FOLs such as fibrous dysplasia, ossifying fibroma and cherubism were studied from archives of the Department over a 14yr period (2009 to 2022) and clinicopathologically reviewed.
Result: In retrospective study 26 cases of FOLs showed that FOLs are more commonly seen in second decade of life with female predilection and mandible is most commonly affected. Amongst these ossifying fibroma is found to be most common in our study 46% (12/26) as compared to fibrous dysplasia 42% (11/26) and Cherubism 12% (3/26).
Conclusion: In the present study FOL were slightly more prevalent in females than male, affecting individuals below 30 years of age. Our data showed several overlapping epidemiological and clinicopathological features. Thus combination of different modalities seems to be necessary for an accurate diagnosis.

1. Introduction

Fibro-osseous lesions (FOLs) are defined as group of lesions affecting the jaws and craniofacial bones. All are characterized by cellular fibrous tissue replaced with the foci of mineralization suggestive of osseous matrix that vary in amount and appearance.[1] It comprises tumor and tumor like conditions with similar histologic appearances but different clinical behavior.[2] The FOL as a group has been introduced in the WHO classification (2017) of odontogenic and maxillofacial bone tumors. There are 3 recognized FOLs; fibrous dysplasia (FD), ossifying fibroma (OF), and cemento-osseous dysplasia (COD).[3]

The group of lesions always revelation a divergence of clinical behaviour, but share similar microscopic features consisting of a benign connective-tissue matrix and new bone formation by fibrous component.[4] Overlapping histologic features of entities within this group and anomalous features of individual lesions often make definitive diagnosis difficult.[1] The final diagnosis of FOLs of the jaws relies on careful correlation between the clinical presentation and radiographic appearances.[5]

Several retrospective studies lay out the differences in the relative frequency of the various FOLs. Hence, the aim of the study was to analyse FOLs for demographic details and clinicopathologic correlation.

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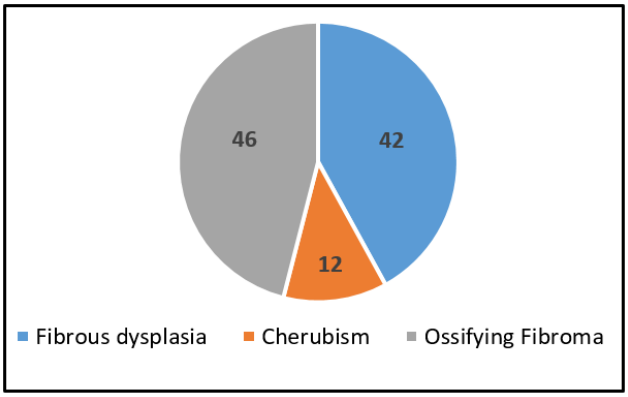
2. Materials and Methods

Clinicopathologically diagnosed cases of fibrous dysplasia, ossifying fibroma and cherubism reported at our institute over a period of 14 years were studied. Detail clinical records and radiographs of patients were reviewed. Initial diagnosis of all the cases were based on clinical presentation coupled with radiological appearances Histopathology reports were reviewed to confirm the diagnosis.

Patients with FOL frequently complained of a gradual enlarging swelling of the jaws which leads to facial asymmetry. None of the patients had significant family history or past medical history in our study. Out of 26 cases, 3 cases were diagnosed as cherubism.

3. Result

During the 14-year period under review out of twenty-six cases of FOL diagnosed 46% were ossifying fibroma, 42% were fibrous dysplasia and 12% cherubism. (Graph 1)



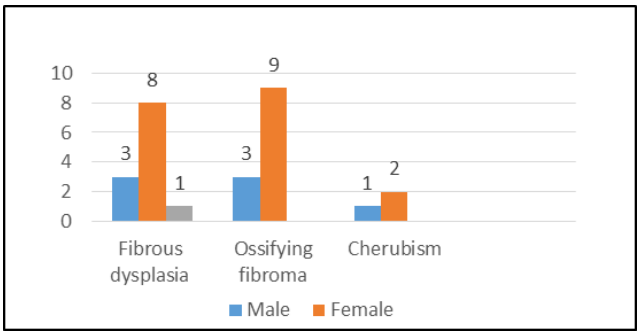
Graph 1: Demographic distribution of FOLs

3.1. Gender and age distribution of lesion

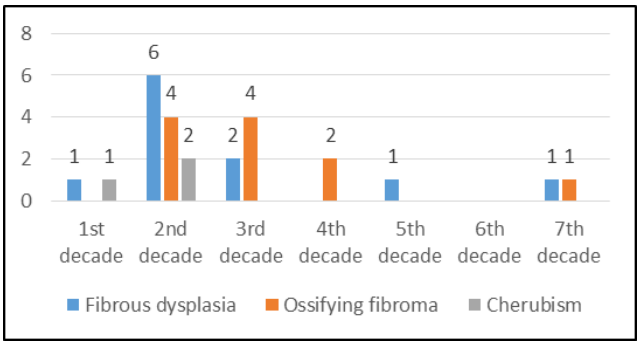
In this retrospective, observational study, all FOLs were found to be more common in females than in males (Graph 2). 73% cases were females and 27% were male, thus giving a female to male ratio of approximately 2.7:1. So female preponderance was noted. Fibro osseous lesions were common in 2^{nd} decade of life. (Graph 3)

4. Discussion

FOLs demonstrate the replacement of osseous trabeculae with fibroblasts, collagen fibers, with altering amounts of mineralized materials.[4] Several classifications have been proposed with aim to distinguishing these lesions into 3 different categories on the basis of causative factor such as developmental, reactive and neoplastic origins.[6] Presently 3 recognized FOLs; fibrous dysplasia (FD), Ossifying fibroma (OF), and cemento-osseous dysplasia (COD).[7]



Graph 2: Gender distribution of FOLs



Graph 3: Age distribution of FOLs

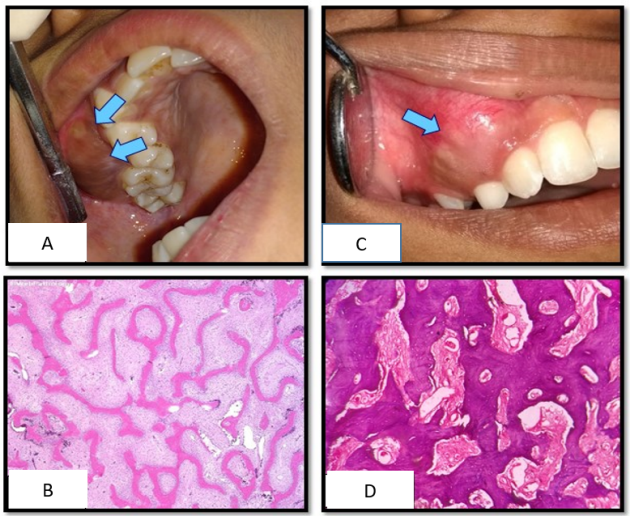


Fig. 1: A): Clinical examination of FD; **B):** Histopathological examination of FD; **C):** Clinical examination of OF; **D):** Histopathological examination of OF. (FD – Fibrous dysplasia, OF – Ossifying fibroma)

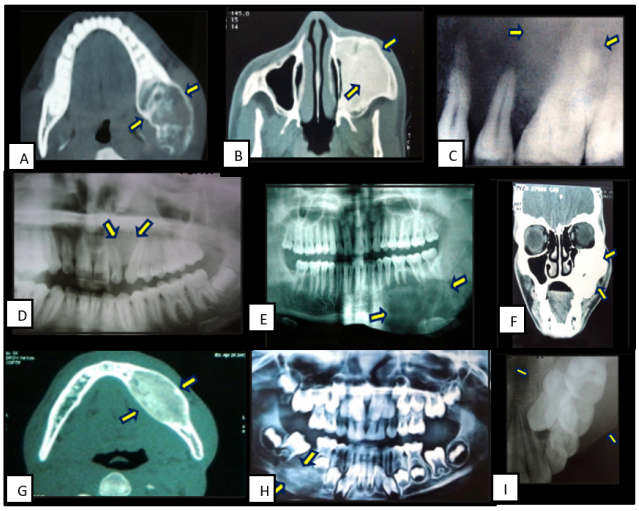


Fig. 2: Variation in radiographic finding of fibrous Dysplasia **A):** Mixed radiolucency extending from distal 36 to ramus; **B):** Matured lesion appeared with increasing radiopacity; **C):** Ground glass Appearance; **D:** Divergence of Root with 22 & 23; **E):** – Displacement of canal with inferior border of left side of mandible; **F):** Radiopacity of lesion extending upto orbital floor; **G):** Fusiform mixed radiolucency with ground glass appearance. **H& I):** poorly defined border which classically merge with adjacent bone

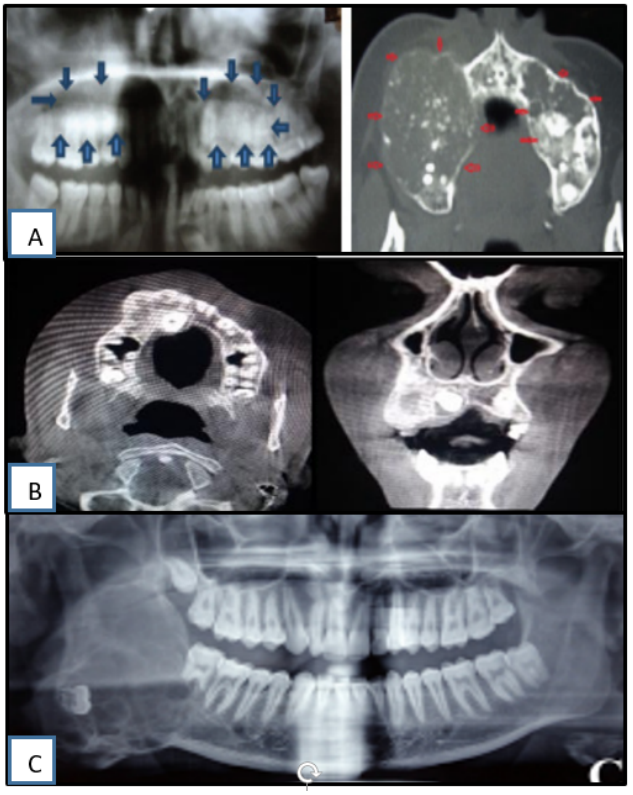


Fig. 3: Variation in radiographic finding of Ossifying Fibroma **A):** Bilateral Mixed radiopacity in maxillary region. **B):** Expanded and thin cortical outlines with easy separation from adjacent cortical bone. **D):** Multilocular radiolucency involving left ramus with distal root resorption 47

OF is a true benign neoplasm of the bone-forming tissues.[8] It can be classified as conventional and aggressive (juvenile).[9] According, WHO 2017 classification of FOLs, JOF histologically has two variants; Juvenile trabecular OF (JTOF) and juvenile psammamatoid OF (JPsOF).[3] Pluripotent mesenchymal cells capable of generating cementum, fibrous tissue and bone give rise to OF. Molecular studies have identified mutations in cell division cycle 73 (CDC73) / HRPT2, gene that encodes parafibromin protein.[5] The HRPT2 gene is mostly associated with multiple ossifying fibroma.[10] 46% of lesions in the study were OF. Amongst these 7.7% were JPsOF out of which one case of showed secondary aneurysmal bone cyst of left mandible and 11.5% were JTOF. One case showed bilateral involvement of maxilla.

FD is a disease of bone remodelling in which normal medullary bone and cortices are replaced by a disorganized woven bone.[11] GNAS gene mutation is underlying cause of FD. The clinical severity of the condition depends upon the time of GNAS1 mutation occurrence during fetal or postnatal life. If mutation occurs during the early embryonic life, which results McCune Albright syndrome. If mutation occurs in the later stages of embryonic life polyostotic FD. If mutation occurs during the postnatal life resulting monostotic FD.[12] Total 42% FD were noted in our study. According literature, 80% FDs are monostotic in nature[13] and in the present study 92.3% presented as solitary lesion in the jaws. Based on literature 22.3% FDs are involving craniofacial complex[13] and present study we found as 7.7% were extended up to infra-orbital margin and resulted in impaired vision. There was no case of polyostotic and syndromic FDs.

Previously Cherubism was considered as familial dysplasia and also named as "Familial Fibrous Dysplasia of jaw "but now it is considered as a genetic disorder of SH3BP2 gene mutation.[14] Current study we noted as 12% was of cherubism.

In existent study FOLs were common in 2^{nd} decade of life. Compared with previous studies, both OF and FD were predominantly occur in younger population with mean age of 21.1 yrs and 24.2 yrs respectively.[4] OF and FD showed a female predilection as favourable with previous literature reports.[2] FOLs commonly involve maxilla. In present study, Mandible was commonly involved site which is not in accordance with the previous literature.

The radiographic appearance of FOLs varies with the stage of development and maturation of bony matrix within the lesion.[15] On a radiograph, early lesions show radiolucency, but they become increasingly radiopaque to diffuse radiopacity.[14] Radiologically, OF are spherical to

egg shaped, expanded and thin cortical outlines, displace adjacent structures and shows features of easy separation from adjacent cortical bone.[10] In present study 91.66% of showed mixed radiopaque and radiolucent area, 8.33% of JPsOF with secondary aneurysmal bone cyst, showed a multilocular expansion with radiopacity. Lesion showing feature of endosteal scalloping and a narrow transional zone. (Figure 3) FD show fusiform expansion of bone and poorly defined lesion classically merging with adjacent bone with "ground glass appearance".[13,16] In present study 63.63% of FD showed mixed radiopaque and radiolucent area, 36.36% showed ground glass appearance, which is in accordance with the previous literature. (Figure 2)

Histologically, the FOLs mainly consist of two components - hard tissue and soft tissue.[9] OF is composed of fibrous connective tissue with well-differentiated fibroblasts.[17] Collagen fibers are arranged in a whorled or storiform pattern.[10] Bony spheroids or trabeculae are evenly distributed throughout the fibrous stroma.[17] Bone is surrounded with osteoblastic rimming. A similar histopathology was noted in present study, JTOF (25%) consisted of lamellar bone with osteoblastic rimming and fibrocellular stroma and bands of cellular osteoid trabeculae. (Figure 1 C and D) JPsOF (16.6%) were characterised by the presence of spherical ossicles/psammoma bodies in a variably cellular fibrous stroma. Gögl termed the spherical structures as "psammoma-like bodies."[3] Mature bone was viewed in 8.33% of our case and osteoblastic rimming was evident in few areas of 41.66% of cases. FD consists of a slight to moderate cellular fibrous connective tissue stroma that contains foci of irregularly shaped trabeculae of immature bone. A relatively constant ratio of fibrous tissue to bone throughout a given lesion is characteristic.[18] The fibroblasts exhibit uniform spindle-shaped nuclei. The bony trabeculae assume irregular shapes likened to Chinese characters, without any functional orientation. The predominantly woven type of bone and appears to arise directly from the collagenous stroma without prominent osteoblastic activity.[12,18,19] In present study, our histopathological finding was favourable with the literature. '(Figure 1 A and B)

FOLs usually represents a favourable prognosis, but literature suggests that some long standing FD, may have a risk for malignant transformation. The rate of malignant transformation of FD as reported in the literature ranges from 0.4% to 6.7%, it is estimated as 0.4% in monostotic FD and 4% in association with syndromic FD. Patients with Mazabraud syndrome show higher risk of malignant transformation, estimated to be about 8.3%.Malignant tumors resulting from FD were diagnosed primarily as osteosarcomas, some cases may diagnosed as chondrosarcoma, malignant fibrous histiocytoma and fibrosarcoma.[20] In present study, none of the case showed malignant transformation.

FOLs do not have pathognomic clinicopathologic features. Some bony lesions are considered in differential diagnosis to FOLs such as Osteopetrosis, Sclerosing osteomyelitis, Paget disease, Osteoblastoma, Hyperparathyroidism, Cementoblastoma and Low grade Osteosarcoma.[2,5,18,21] Serological investigations are helpful in diagnosis of FOLs such as level of alkaline phosphatase, acid phosphatase, calcium and Vit.D.[16] The alkaline phosphatase some time elevated in FD.[12] According to literature, No any serological changes are observed in OF and COD. The management of these lesions depending on the diagnosis, size, location of the lesion and patient status.[16] Complete surgical excision signifies the treatment for OF. In present study 46% of cases of OF were treated by complete surgical excision. The wait n watch with surgical remodelling is main approach to treat patients with FDs. In present study biopsy were taken for confirmatory diagnosis and patients kept under observation. COD usually do not require treatment, but require periodic follow up.

5. Conclusion

A number of diseases exhibit similar findings that closely mimic with FOL. Thus, a definitive diagnosis of a FOLs should always be based on demographic, clinical, radiologic and Histologic information. Their successful management depends on the precise histopathological diagnosis. Thus, a sound support from imaging and an experienced pathologist would be very essential in proper diagnosis and management of these lesions.

6. Source of Funding

None.

7. Conflict of Interest

None.

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