Original Article

Spectrum of Fibro Osseous Lesions: A Retrospective Study

Sajitha K*, Kishan Prasad H L, Netra Sajjan and Jayaprakash Shetty K

Dept of Pathology, K S Hegde Medical Academy, Mangalore, India

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ABSTRACT

Background: Fibroosseous lesions (FOL) are a group of lesions which affect the jaw and the craniofacial bones and are a challenge to pathologists and clinicians in their diagnosis and treatment. It includes developmental lesions, reactive lesions and neoplasms. Many other lesions share the clinical, radiological and histopathological features of FOL. The identification of benign FOL and their sub classification is important because the therapeutic management varies depending on the actual disease process. The aim of this study was to analyse the spectrum of FOL and its mimickers that presented in our hospital and to study its clinicopathological aspects.

Methods: A retrospective analysis of 31 cases of benign FOL and its mimickers which presented between 2007 & 2013 was done in the Dept of pathology. Clinical data and X-ray findings were obtained from the medical records department.

Result: Among 31 cases studied, 15 cases were diagnosed as FOLs and 16 as fibro-osseous like lesions. FOLs were most commonly seen in females in 1st to 3rd decade with a predilection for facial bones. Commonest lesion was fibrous dysplasia - 11 cases (73.3%), followed by 2 cases each of cemento ossifying fibroma and osteofibrous dysplasia (13.3%).The commonest diseases among the mimickers included Aneurysmal Bone Cyst- 6 cases (37.5%), followed by osteoid osteoma - 4 cases (25%), 2 cases (12.5%) each of pagets disease of bone & osteoblastoma and 1 case (6.3%) each of brown tumour of hyperparathyroidism and cementoblastoma and these were most commonly seen in long bones. In histopathology, FOLs show densely collagenous fibroblastic tissue containing metaplastic bone where as fibro osseous like lesions exhibit less fibrous tissue in the stroma.

Conclusion: A definitive diagnosis of a FOLs and its differentiation from its mimickers requires correlation of histological features with the clinical, radiographic and intraoperative findings.

***Corresponding author:**
Dr Sajitha K, 4 C, KSHEMA Staff Quarters, Deralakatte, Mangalore, India
Phone: + 91 9741993622
Email: drsk29@hotmail.com

Introduction

The term Fibroosseous lesions (FOL) refers to a process in which the normal architecture of bone is replaced by fibrous tissue containing varying amount of foci of mineralization. These lesions are rare and predominantly affect jaws and craniofacial bones. This group comprises of tumours and tumour like lesions which are histologically similar but have different clinical **behavior**[1]

Many authors have put forth different classification systems to classify FOL. Various lesions demonstrate clinical, radiological and microscopic features that are similar to those encountered in recognized fibro-osseous conditions. [2] The differential diagnosis include osteoid osteoma, osteoblastoma, pagets disease of bone, brown tumour of hyperparathyroidism and cementoblastoma. However these lesions do not completely fulfil the histological criteria for FOLs as defined by Waldron. [3] A definite diagnosis may thus require extensive investigation for classification and interpretation of histological appearance along with radiological and clinical data.

AIM: To analyse the spectrum of fibroosseous lesions received in our department and to differentiate it from its mimickers.

Materials and Methods

A total of 31 cases of FOLs were retrospectively analysed from 2007-2013 in the Department of pathology of a tertiary hospital situated in the outskirts of Mangalore. The clinical data and radiological findings were obtained from case files. The diagnosis was based on the clinical findings, radiological appearances and histopathological examination.

Result

A total of 31 cases were studied, among them 15 cases were diagnosed as FOLs and 16 cases as fibroosseous like lesions. The commonest lesion among FOLs was fibrous dysplasia 11cases (73.4%) followed by 2 cases (13.3%) each of cemento-ossifying fibroma and osteofibrous dysplasia (Table 1). Majority of the fibrous dysplasia were mono ostotic. One case was polyostotic fibrous dysplasia in which patient presented with multiple lytic lesions in the proximal & distal tibia and distal radius. Histopathological examination showed fibrous dysplasia with extensive areas of cartilaginous differentiation and hence the diagnosis of fibrocartilagenous dysplasia was made. The fibro osseous like lesions were Aneurysmal bone cyst(ABC) – 6 cases (37.5%), followed by osteoid osteoma - 4 cases (25%), Paget disease and osteoblastoma -2 cases each (12.5%), and 1 case (6.3%) each of brown tumour of hyperparathyroidism and cementoblastoma (Table 2). FOL were most commonly seen in the 2st and 3rd decades and 40% of the lesions were in the 3rd decade. It was more common in females compared to males. The mimickers of FOLs showed a male predilection and majority of cases (43.7%) cases presented in the first decade. The most common site involved among FOL was craniofacial bones (56.6%) followed by long bones. Among the mimickers the most common site was long bones (62.5%) followed by craniofacial bones. Histopathologically, FOLs showed densely collagenous fibroblastic tissue containing metaplastic bone where as fibroosseous like lesions exhibited less fibrous tissue in the stroma.

Table 1 -Distribution of Fibro osseous lesions- (Total no of cases – 15)

Type of lesion	No of cases	Percentage
Fibrous Dysplasia	11	73.40
Cemento-ossifying Fibroma	02	13.30
Osteofibrous Dysplasia	02	13.30

Table 2 - Distribution of fibro-osseous like lesions (Total -16 cases)

Type of Lesion	No of cases	Percentage
Aneurysmal Bone Cyst	06	37.5
Pagets Disease	02	12.5
Osteoblastoma	02	12.5
Osteoid Osteoma	04	25
Brown Tumor	01	6.25
Cementoblastoma	01	6.25

Table 3 - Benign Fibro- osseous lesions -

FOL	Present study	Abdulai et al[1]	Ogunsalu et al[4]	Sharanya et al[16]	Langdon et al [17]
Fibrous dysplasia	11	16	17	20	15
Cementoossifying fibroma	02	36	10	60	19
Osteofibrous dysplasia	02	-	-	-	-
Cementoid lesions	-	-	05	-	05
Total	**15**	**52**	**32**	**80**	**39**

Table 4 –Comparison of Age and Sex incidence (M/F) for FOL in various studies –

BFOL	Present study		Abdulai et al[1]		Ogunsalu et al[4]		Langdon et al[17]		Hammer et al[15]	
	Age	Sex	Age	Sex	Age	Sex	Age	Sex	Age	Sex
FD	34.5	1/1.2	21.7	1/1.3	25.8	2/3	24	1/1.5	-	
COF	20	0/2	19.9	1/1.6	26.5	2/3	35	1/1.23	26	1.2/1

Discussion

Fibro-osseous lesions (FOL) of bones comprise a diverse group of conditions that include developmental lesions, reactive or dysplastic lesions, and neoplasms. Due to this diversity, they are a diagnostic challenge to the pathologist. Regardless of the subtype, FOLs demonstrate replacement of normal bone by fibrous connective tissue along with an admixture of mineralized product, which may be osteoid, mature bone, and/or cementum-like calcifications. A number of lesions mimic FOL histologically and the differential diagnosis includes aneurysmal bone cyst, osteoblastoma, osteoid osteoma, pagets disease of bone, brown tumor of hyperparathyroidism and cementoblastoma.

Before 1970, the term FOL included fibrous dysplasia, ossifying fibroma, fibrous osteoma and osteoblastoma. [4] Waldron in his classification suggested that the FOL originate from the periodontal ligament which contains multipotent cells which are known to differentiate into fibrous tissue cells, cementum and bone. [5] The latest classification given by WHO in 2005 considers these lesions as a spectrum of clinicopathological entities in which the diagnosis can only be made after taking into consideration the clinical, histological and radiological features. [2],[6] Under this classification, the major fibro-osseous lesions include fibrous dysplasia (FD), osseous dysplasia, ossifying fibroma with their various subtypes. More recently, Eversole (2008) classified the benign FOL based on the pathogenetic mechanisms and the lesions included neoplasms, developmental lesions and inflammatory or reactive processes. [3] A specific diagnosis is needed as the treatment varies from none to surgical removal. Radiographically, FOL vary from a simple radiolucent lesion, a radiopaque lesion or a mixed radiolucent/radiopaque lesion. These can be well defined or ill-defined. There may or may not be expansion of bone. [7]

Fibrous dysplasia, Osteofibrous dysplasia and Cemento-ossifying fibroma: FD is a benign FOL commonly seen in infancy and childhood or young adults. [8],[9] The femur, tibia, skull and facial bones, or ribs are most commonly affected. Females are more commonly affected than males. [4] When the disease is limited to the single bone it is called monoostotic and involvement of two or more bones is known as polyostotic fibrous dysplasia. When it is seen with pigmentation it is termed Jaffe-lichtenstein syndrome and if associated with multiple endocrinopathies and cafe au lait spots, it is termed as McCune Albright syndrome. Fibrous dysplasia also forms a component of Mazabraud syndrome where it is seen along with soft tissue myxomas. [10] Radiologically, the lesions are poorly defined and described as orange peel or ground glass in appearance. Microscopically, FD shows irregular bony trabeculae giving the appearance of the chinese letter pattern. These bony trabeculae do not have any osteoblastic rimming (Fig 1A). However, a few lesions may show osteoblastic rimming. [10] In the present study, one case of fibrous dysplasia showed bony trabeculae which resembled cementum like material (Fig 1B).

Osteofibrous dysplasia is similar to FD on histopathology except for the presence of osteoblastic rimming. (Fig 1C & 1D). The other differentiating feature is the location of FD in medullary cavity and osteofibrous dysplasia in the cortical region and the presence of cytokeratin positive epithelial cells in osteofibrous dysplasia. [8]

Ossifying Fibroma is commonly seen in the jaw bones and is a true neoplasm with a potential for continued growth if not excised. Radiologically, in contrast to FD, ossifying fibromas are well circumscribed and present as expansile mass with a sclerotic rim. Histology shows fibrous tissue arranged in a storiform pattern and variable amount of bony trabeculae or cementum like spherules. [5],[9] The

bony trabeculae show a uniform pattern of mineralization throughout the lesion in fibrous dysplasia while the mineralization pattern varies from place to place in ossifying fibroma. Whenever the cementum like material is predominant, it is termed as cementifying fibroma. Radiologically, it is seen associated with tooth and have to be distinguished from Cemento Ossifying Dysplasias (COD). Ossifying fibromas tend to present at a slightly younger age than COD. A diagnosis of COD is favoured when histology shows thick, curvilinear trabeculae resembling ginger roots and cavernous vascular spaces. These lesions are less than 2 cm in size and less cellular. [10],[11]

Paget disease and brown tumor of hyperparathyroidism: Osteitis deformans or Paget disease of bone is characterized by rapid turnover and remodelling of bone throughout the skeleton. Unlike FD, osteitis deformans is a disease of the elderly. [12]

Brown tumor of hyperparathyroidism is common in diaphysis of long bones, the jaw, or the skull, which on microscopic examination shows many clustered giant cells in a fibrovascular cellular stroma. It mimics giant cell tumor on histopathology. [13] In the jaws, it closely resembles central giant cell granuloma (CGCG). However, CGCG is most commonly seen in the younger age group. Paget disease and brown tumor of hyperparathyroidism also share the similar features and have to be differentiated correlating with the laboratory findings. Calcium and parathyroid hormone levels are normal in Paget disease of bone whereas they are increased in brown tumor of hyperparathyroidism. [13] There is marked elevation of alkaline phosphatase in Paget's disease. In our case, a similar picture was seen with normal calcium and PTH levels in the Paget's disease and an increased levels of the same in hyperparathyroidism.

Osteoblastoma, Osteoid osteoma, Cementoblastoma and Aneurysmal Bone Cyst: Osteoblastoma (OB) and osteoid osteoma are benign neoplasms which resemble FOLs. It is difficult to differentiate OB and osteoid osteoma on histotology alone. Osteoid osteoma is seen commonly in males and presents with intense pain relieved by analgesics and small well circumscribed radiolucent nidus of <1.5 cm. Osteoblastomas are usually more than 2 cm and in contrast to osteoid osteoma, the pain associated with OB is not relieved by analgesics.

Cementoblastoma (CB) is an odontogenic neoplasm representing <1% of all odontogenic tumors. A cementoblastoma is characterized by the formation of cementum like tissue in connection with the root of a tooth [6] (Fig 2A). Histologically, CB and OB have the same appearance including peripheral spiculae rimmed by plump osteoblasts (Fig 2B). This histologic similarity between OB and CB indicates that the diagnosis of cementoblastoma should be made only when the lesion is connected with a tooth. [14],[15] Radiologically, the tumor appears as a radioopaque mass that is fused to one or more tooth roots and is surrounded by a thin radiolucent rim. [2],[14]

ABC is seen predominantly in the first two decades of life and comprise of blood filled cystic spaces separated by connective tissue septa containing fibroblasts, osteoclast-type giant cells and reactive woven bone. It is rare in craniofacial bones. Histologically, interconnected, ossified woven bone rimmed by osteoblasts is seen. Fibrous tissue, vessels and multinucleated giant cells are also identified. Radiologically, ABC presents as a lytic, eccentric, expansile mass with well defined margins.

Comparison of our study with other studies: [1],[4],[15],[16],[17] In our study, the predominant FOL was fibrous dysplasia which correlated with the findings of Ogunsalu et al. [4] In other studies, the predominant lesion was ossifying fibroma.[1],[16],[17] The average age of presentation of fibrous dysplasia was 34 years in our study while in other studies the mean age was slightly lower.[1],[4],[16],[17] For ossjfying fibroma, the mean age of presentation was 20 and this correlated with the other studies. The FOL showed an increased incidence among the women in our study as well as the other studies. Out of the eleven (11) cases of fibrous dysplasia, ten (10) were mono-ostotic while the one case was polyostotic and was reported as fibrocartilagenous dysplasia.

In our study, we also evaluated the lesions which histologically mimick the major FOL. Sixteen of our cases were classified as fibro-osseous like lesions and their parameters were compared with those of FOL. The commonest mimicker was aneurysmal bone cyst followed by osteiod osteoma. Although osteoid osteoma has a rare rate of recurrence, we had one case of recurrent osteoid osteoma. The age of presentation was lower than that of FOL and majority of the patients were in the adolescent age group. The predominant site of these lesions was the long bones, femur and tibia followed by the cranio-facial bones.

In this study, we encountered difficulty in distinguishing fibrous dysplasia from osteo-fibrous dysplasia in cases where the thin trabeculae of woven bone in FD showed focal osteoblastic rimming. The final diagnosis was made based on the radiological findings. The histological findings in cementoblastoma were identical to osteoblastoma. A diagnosis was made based on the direct connection with the surface of the tooth. The clinical parameters like increased serum calcium levels and parathormone levels were useful in distinguishing brown tumor of hyperparathyroidism from Paget's disease.

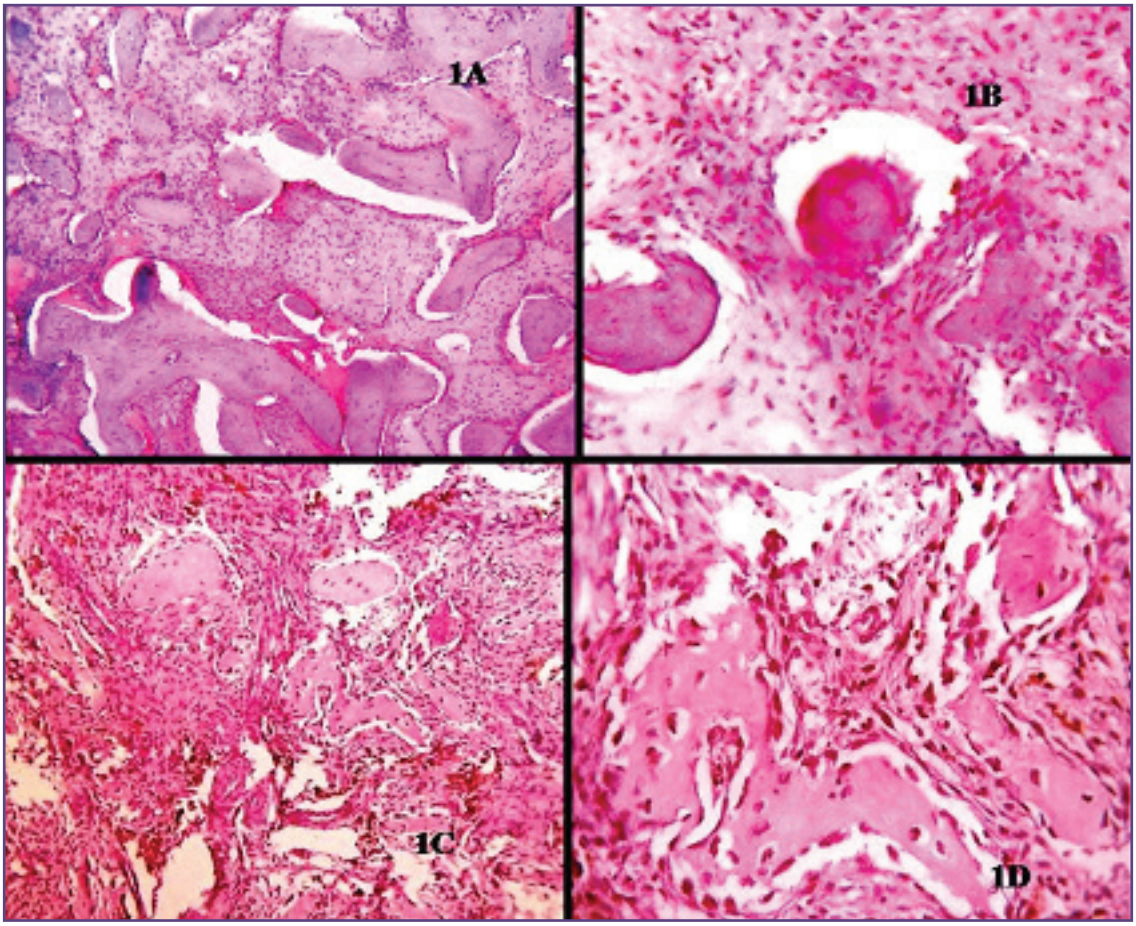

Fig. 1A: Histopathology of Fibrous Dysplasia (H & E ×100) showing cellular fibrous tissue containing irregular bony trabeculae.; Fig 1B – Focal areas in Fibrous Dysplasia showing cementum like material (H&E × 100).; Fig 1C – Histopathology of osteofibrous dysplasia (H&E × 100).; Fig 1D – Osteofibrous dysplasia (H&E × 400) showing bony trabeculae rimmed by osteoblasts.

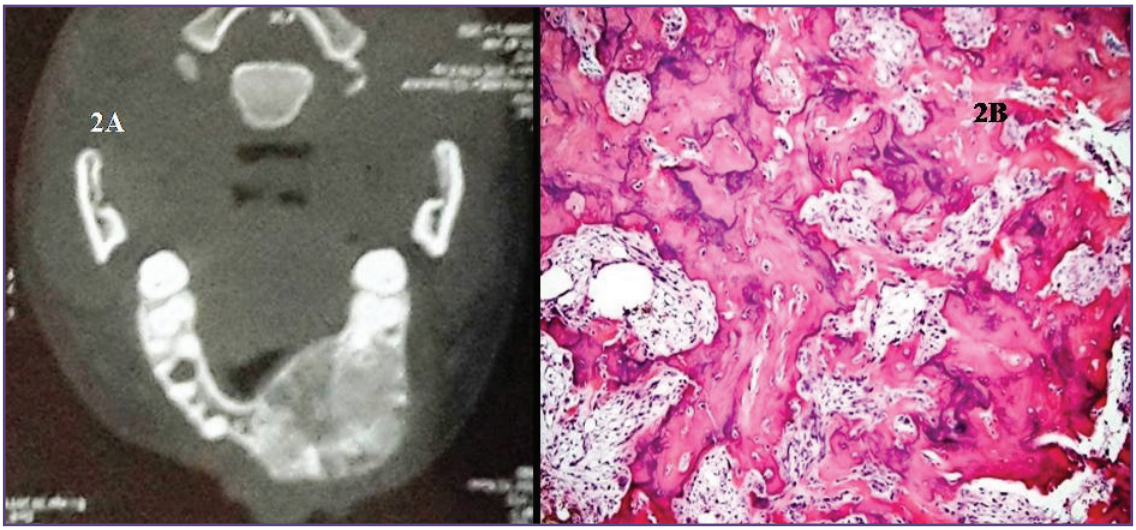

Fig. 2A: Cementoblastoma – CT scan – Showing association with tooth.; Fig 2B – Histopathology of Cementoblastoma (H & E × 100) - Peripheral trabeculae rimmed by plump osteoblasts.

Conclusion

A number of diseases exhibit findings that closely mimic those seen in FOL. Thus, a definitive diagnosis of a FOL and its differentiation from its mimickers requires correlation of the histologic features with the clinical, radiographic, and intraoperative findings. Their successful management depends on the accurate histopathological diagnosis. Histologically, FOL show more fibrous tissue with metaplastic bone and slender thin bony trabeculae whereas the mimickers exhibit less fibrous tissue.

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Competing Interests

Nil

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