Fibro Osseous Lesions of The Craniofacial Structures – A Clinical Study

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Received: 15.11.17 Accepted: 17.03.18

Abstract:

Fibro osseous lesions area diverse group of disorders characterized by replacement of normal architecture of bone by a benign connective tissue matrix that displays various amount ofmineralizationin the form of woven bone or cementum. It includes developmental, reactive and neoplastic lesions. The different type of fibro-osseous lesions express a common clinical and radiological features. Soadequate knowledge and clinical observationare necessary for proper interpretation and appropriate diagnosis of these lesions.becausemanagement of patients with fibro-osseous lesions are case specificandindividualized. The aim of this study was to analyse the clinical, radiological and histopathological characteristics of fibro osseous lesions and provide a proper management system affected by this type of lesions. Materials and methods: The retrospective study was performed in the Department of Oral & Maxillofacial surgery, Dhaka Dental College and Hospital, Dhaka, Bangladesh from a period of January 2015 to January 2018. Patients were selected for this study based on clinical, radiological and histopathological confirmation of fibro-osseous lesion. The management of each case were plannedand follow-up data were also documented. Results: A total number of 30 patients were selected for this study. The most common fibro-osseous lesionsin this study were fibrous dysplasia 10 (33%) and ossifying fibroma 20 (67%). The mean age offibrous dysplasia were 17.4 years with an age range 12 to 33 years and at ossifying fibroma the mean age were 30.35 years with an age range 12 to 57 years. Female17 (57%) represented the majority of the affected patients. Fibrous dysplasia were more common in maxilla(70%) and ossifying fibroma were more common in the mandible(60%). Surgical recontouring and clinical observation were treatment of choice infibrous dysplasia and surgical resection, enucleation and curettage were treatment of choice in ossifying fibroma. Conclusion: The most common fibro-osseous lesion in our clinical study was fibrous dysplasia and ossifying fibroma which presents painless bony swelling and deformity in maxilla and mandible. Fibrous dysplasia presents as a homogenous, radioopacity,ill defined border and ossifying fibroma presents a mixed radioopacity and radiolucent lesion that is well demarcated from normal bone. Surgical recontouring and clinical observation was done in treatment of fibrous dysplasiaand ossifying fibroma wastreated enucleation and curettage, segmental resection completelyenucleatefromsurrounding bone.

Key words: Fibro-osseous, Fibrous dysplasia, Ossifying fibroma, Clinical study

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Introduction:

Fibro-osseous lesions are a diverse group of disorders characterized by replacement of normal architecture of bone by a benign connective tissue matrix that displays various amount of mineralization in the form of woven bone or cementum. It includes developmental, reactive and neoplastic lesions.^{1,2}. Fibroosseouslesionswere first described by Lichten- stein in 1936. Cooke classified these lesions as developmental, neoplastic, dystrophic and inflammatory.3 In 1993 Waldrondivided this type of lesionsinto fibrous dysplasia (FD), cement-ossifying fibroma (COF) and desmoplastic fi-broma (DF).4 In 2006 a simple classificationwas done based on the World Health Organization(WHO) by Speight and Carlos⁵ and divides these lesions into fibrous dysplasia (FD), ossifying fibroma (OF) and osseous dysplasia (OD). These lesions comprise fibrous dysplasia, ossifyingfibroma, periapical cementosseous dysplasia, focalcementosseous dysplasia, florid cementosseousdysplasia and cementossifying fibroma. Fibro-osseous lesions may be associated with significant aesthetic and functional disturbances or they may be completely asymptomatic localized lesions that are identified only on routine radiograph.2 Radiographically, fibroosseous lesions may manifest as solitary, multifocal or multiquadrant disease, itmay be ill defined or well defined, radiolucent, mixed radiolucentradiopaqueor ground glass appearance and may or may not be associated with the root apices of teeth. The gross appearance of fibro-osseous lesions may varydepending on the lesion. So most oral and maxillofacialsurgeons and pathologists would agree that definitive diagnosis of a fibroosseouslesion requires correlation of the histologic appearanceof the lesion with the clinical, radiographic and intra operative findings.^{2,8} In this article, we analysis and share our experience in the clinical, radiological, and histopathological characteristics of fibro-osseous lesions and provide a proper management system to patients affected by this type of lesions.

Materials and methods:

The retrospective study was performed in Department of Oral & Maxillofacial surgery, Dhaka Dental College and Hospital from a period of January 2015 to January 2018. A total number of 30 patients were selected for this study based on clinical, radiological

and histopathological confirmation of fibro osseous lesion. In this study we included patients who were clinically diagnosed with fibro-osseous lesions with radiological findings by experienced oral and maxillofacial surgeons and radiologist. We also included the patients who were diagnosed fibro-osseous lesions histopathologically despite a preoperative radiological diagnosis which did not indicate fibro-osseous lesions. Data were collected from data sheet provides age, sex, clinical presentation, anatomical location, radiological findings, histopathological findings and type of operation was done for a management of lesions. In all cases, imaging techniques orthopantom-ogram (OPG) and computed tomography (CT) were used for the evaluation of these lesions. All procedures were carried out under general anesthesia. An invasive surgerywas performed in all patients who were selected for surgery. After operation the specimens were submitted for histopathological analysis. Histopathological examination of each case was performed by experienced pathologists. After treatment the patients were regular evaluated for follow up at 1 month, 3 months and 6 months after the surgery.

Results and Observation:

A total number of 30 patients were selected for this study based on clinical, radiological and histopathological confirmation of fibro osseous lesion. The most commonfibro osseous lesions were documented including fibrous dysplasia 10 (33%) and ossifying fibroma 20 (67%).(Figure-1)

The mean ageof patientsat fibrous dysplasia were 17.4 years with an age range 12 to 33 years and at ossifying fibroma the mean age were 30.35 years with an age range 12 to 57 years.(Table-1)

There were 17 (57%) female and 13 (43%) were male patients. Female represented the majority of theaffected patients. (Table-2)

Anatomical location of fibrous dysplasia showed among 10 cases of fibrous dysplasia 07 (70%) were found in maxilla and 03 (30%) were found in the mandible. In the mandibular region body of the mandible is the commonest site, particularly premolar and molar region02 (67%).01(33%) cases were found body and whole ramus area of mandible.Among 20 cases of ossifying fibroma12 (60%) were found in the mandible and 8(40%) were found in the maxilla. In the mandibular regionbody of the mandible is the

commonest site, particularly premolar and molar region were 08(60%). 04 (40%) Cases were found body and whole ramus area. (Table-3) Most of the patients present with noticeable swelling and deformity. As the lesions were slow growing it is not possible to assess the age at which they first developed. For all patients average duration of illness at presentation was 2 years with a range of 1 year to 4 years. Fibrous dysplasia and ossifying fibroma presents apainless bony swelling on the maxillary and mandibular region inallmost all patients. Expansion of cortical plate was present both buccally and lingually or palatally which produce aesthetically visible deformities. some of patients developed headaches, nasal obstruction, epistaxis and symptoms like sinusitiswhen it was in maxilla. No one patients presents clinical manifestations related to compression or compromised of structures. A total number of 30 patients radiographical records were evaluation for study. In fibrous dysplasia most radiographic appearances were mixed typeradio opacity 06(60%) and radio opaque were 04(40%). In ossifying fibroma13(65%) radiographic appearances were mixed type13(65%), radio-opaque were 02 (10%) and radiolucent were 05 (25%). Fibrous dysplasia presented 08 (80%) ill-defined borders and 02 (20%) well-defined borders. Ossifying fibroma presented16(80%)well-defined bordersand 04 (20%)ill-defined borders. Common radiological features observed in both lesions were bone expansion and tooth displacement. Root resorptionwas observed inossifying fibroma. (Table-

The histopathological analysis of almost all fibrous dysplasia cases were composed of multiple small fragments of mineralized tissue with free haemorrhage. Bone trabeculae with large osteocytes within the lacunae were present in all cases offibrous dysplasia.Irregular mineralized mass (osteoid) were(29%)observed in fibrous dysplasia. The histopathological analysis of ossifying fibroma included bone trabeculae with large osteocytes within the lacunae (48%), free haemorrhage (50%), multiple curettage fragments (48%) and thick curvilinear trabeculae (20%),irregular osteoid masses (60%) were present. The common features observed in both fibrous dysplasiaandossifying fibromaincluding metaplastic woven bone in a fibrous stroma. Other common features were separate bony trabeculae, variable amounts of lamellar bone anddepositionof collagen. Among the 10 patients of fibrous dysplasia 05(50%) patients were treated by surgical recontouring and 05(50%) patients were still observation until the age of 18 years and were instructed for regular follow up. Out of 20 patients of ossifying fibroma 12(60%) patients had enucleation & curettage of lesions, 05(25%) patients had segmental mandibular resection and 03 (15%) patients had partial maxillectomy.(Table-5) In our study among 20 cases of ossifying fibroma 04 cases presented surgery-related complications; 01 case developed an infection in the treatment area. Pus cultures taken from infected area and treated effectively by sensitive antibiotics and were discharged with improved condition and 01 showed osteonecrosis signs which required an additional procedure involving curettage and bone remodeling. 02 patient had exposure of reconstruction plate intraoral 1 years after operationwhich were removed and reconstruction were done by fibula bone graft.In fibrous dysplasia recurrence was seen in 02 patients after 1 years of operation. The post-operative period of uncomplicated cases were excellent. Patients were discharged within 10 - 14 days after the operation was completed. The patient who developed complications stayed in hospital until complications resolved. There were no evidence of recurrence found in the post-operative follow up period. No malignant transformation from the lesions were observed.

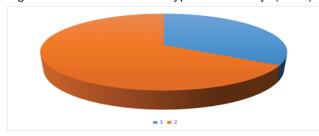
Table 1: Age distribution of the patients

Age	Fibrous dysplasia	Ossifying Fibroma
	Frequency &Percentage	Frequency &Percentage
10 - 20	07(70%)	05(25%)
21 - 30	03(30%)	09(45%)
31 - 40	00(00%)	02(10%)
41 - 50	00(00%)	02(10%)
51 - 60	00(00%)	01(05%)
61 - 70	00(00%)	01(05%)
Total	10(100%)	20(100%)
Mean (±SD)	17.4 (+/-5.07)	30.35 (+/-14.05)
Range	12 - 23	12 - 57

Table2: Gender distribution of the patients (n=30)

Gender	Fibrousdysplasia	Ossifying fibroma	Total
Male	05 (50%)	08 (40%)	13 (43%)
Female	05 (50%)	12 (60%)	17 (57%)
Total	10 (100%)	20 (100%)	30 (100%)

Fig 1- Fibro osseous lesion type in the study. (n=30)



1. Ossifying fibroma. 2. Fibrous dysplasia

Table 3: Anatomical location and distribution of the fibro osseous lesions(Fibrous Dysplasia &Ossifying fibroma) (n=30)

Fibrousdysplasia	Maxilla	Anterior	Body	Posterior
	07 (70%)	-	-	-
	Mandible			
	03 (30%)	-	02 (67%)	01(33%)
Total	10(100%)			
Ossifying fibroma	Maxilla	Anterior	Body	Posterior
	08(40%)	-	-	-
	Mandible			
	12 (60%)		08 (60%)	04(40%)
Total	20(100%)			

Table 4: Radiological features offibro osseous lesions(Fibrous Dysplasia &Ossifying fibroma) (n=30)

Patients &Percentages 05(25%) 02(10%) 13(65%)
05(25%) 02(10%)
02(10%)
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13(65%)
16(80%)
04(20%)
16(80%)
08(40%)
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Table5: Treatment options of fibro osseous lesions(Fibrous Dysplasia &Ossifying fibroma) (n=30)

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Fibro osseous lesions	Treatment	Percentages
Fibrous Dysplasia	clinical observation	05 (50%)
	Surgical Recontouring	05 (50%)
Total		10(100%)
Ossifying Fibroma	EnucleationHJDWWHblXtsions	12(60%)
	Segmental resection of mandible	05(25%)
	Partial maxillectomy	03(15%)
Total		20(100%)

Discussion:

Fibrous dysplasia and ossifying fibroma are the most common fibro-osseouslesionsoccured in the maxillo-facial region.9In this study the most common fibro-osseous lesions were fibrous dysplasia 10 (33%) and ossifying fibroma 20 (67%) which was comparable to Williams et al.¹⁰,Ajagbeet al.¹¹,Alsharifet al.¹²

The mean age of fibrous dysplasia was 17.4 years

with an age range 12 to 33 years and the mean age of ossifying fibroma was 30.35 years with an age range 12 to 57 years in this study which was comparable to Maki et al.¹³

Fibro-osseous lesions were more in females 17(57%) than males 13 (43%) in this study which is comparable to Ajagbeet al.¹¹ who reported 133 cases of Fibro-osseous lesions and noted 60.1% lesions in females and 39.9% in males. Alsharifet al.¹² reported equal gender predilection for fibrous dysplasia and ossifying fibroma in Chinese patients.

The maxilla or mandiblemay be involved but predominance of the maxilla hasbeen documentedinfibrous dysplasia^{5,14} which is similar to our findings. Williams et al.¹⁰ reported that 53.8% fibrous dysplasia occurred in the maxilla but did not indicate the predominant quadrant. Some other studies reported similar extents of maxillary andmandibular involvement.^{12,15,16} Mandibular premolar area (70 – 80%) and ramus areaparticularly in posterior region¹⁷ are the most common sites of ossifying fibromawhich was similar to our study.

Fibrous dysplasiamay be involveone (monostotic) or multiple bones (polyostotic). Monostotic fibrous dysplasia are less serious than polyostotic fibrous dysplasia.18Most patients present with the monostotic form of fibrous dysplasia. 10% of the patients with monostotic fibrous dysplasia have craniofacial involvement and subdivided into cranial (including frontal, parietal, sphenoidal and occipital bones) and maxillary regions are almost equally affected. Poliostotic form of fibrous dysplasia are less common and associated with skin pigmentation and endocrine disturbancesup to 3% of the total cases (McCune-Albright syndrome) with particular in young female patients.¹⁷ In this study most of the patients of fibrous dysplasia were monostotic form, only one patient we were found on polyostotic form.

Fibrous dysplasia manifests as a painless swelling to the affected bones and produce aesthetically visible deformities. The deformity of the jaw results from a progressively slow growing painless swelling, but growth often slows or become arrested at a time coinciding with the onset of puberty. The patient may experience a variety of symptoms, including headaches, loss of vision, proptosis, diplopia, loss of hearing, anosmia, nasal obstruction, epistaxis, epiphora and symptoms like sinusitiswhen the anatomical

spaces and foramina are constricted because of encroachmentof the lesions.¹⁹ Most of the patient present painless diffuse swelling in our study and some of patients develop a variety of symptoms, including headaches, nasal o-bstruction, epistaxis and symptoms like sinusitis. No one of the patients showed clinical manifestations related to compression or compromised of structures in this study.(Fig 1&2)



Fig 1-Fibrous dysplasia – clinical presentation (extra oral view)-A single diffuse non tender hard swelling-seen on the left side of the maxilla

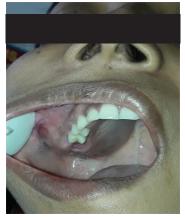


Fig 2-Fibrous dysplasia – clinical presentation (intra oral view)-Intraorally expansion of the buccal and palatalsite seen on the right side of the maxilla Ossifying fibroma isusually a painless slowgrowing and often asymptomatictumor; a rapid growth pattern with a malignant behavior is sometimes notedwhenthe tumor is located outside the mandible. When it is involve inthe mid-face and paranasal sinuses, patients commonly have apainless swelling of the

cheek, unilateral proptosis with diplopia, persistent nasal obstruction, rhinorrhea and epiphora, andrecurrent epistaxis and hemoptysis.20Painless bony swelling is the most common sign of ossifying fibroma^{2,12,17}and most of the patients present with painless bony swelling and deformity in this study.(Fig3&4)



Fig 3— Ossifying fibroma — Clinical presentation (Extra oral view) a painless hard swelling &facial deformity present on the right side ofmandible.



Fig 4– Ossifying fibroma – Clinical presentation (Intra oral view). Expansion of both buccal and lingual plate and displacement of toothpresent intraoraly.

Fibrous dysplasiapresents as a homogenous, groundglass, radiodensity that has no clear demarcation with the surrounding bone. Lesions usually appear as ill-defined, unilocular or multilocular radiolucent lesions with radiopacities on the inside due to the content of bone trabeculae. Larger lesions can cause cortical thinning and remodelingalthough they can rarely cause a breakdown. In our studythe

radiographs shows poorly defined mixed and radiopaque images. (Fig 5,6,&7)



Fig 5– Fibrous dysplasia – Poorly defined, radio opaque lesion on left maxilla



Fig 6 – Fibrous dysplasia on Poorly defined, radio opaque lesion on left maxilla.

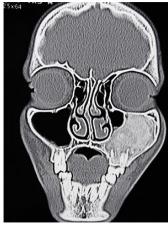


Fig 7– Fibrous dysplasia – CT scan showing complete obliteration of the left maxillary sinus.

Ossifying fibroma presents as a mixed radiodense and radiolucent lesion that is well demarcated from normal bone.²² In the early stages, the lesion presents as radiolucent areas in which bone densities

appear as the lesion matures, transforming the image into unilocular or multilocular masses of radiopaque tissue surrounded by less ossified tissue. Root resorption ofthe adjacent tooth is a pathological effect of ossifying fibroma.^{5, 23} Most ossifying fibroma present with mixed radiological images and well defined border in thisstudy. (Fig 8,9,10,11,12)



Fig8 – Ossifying fibroma – well defined, multilocular radiolucency on rightside ofmandible.



Fig9 – Ossifying fibroma – well defined, multilocular radiolucency, root resorption was seen on right side ofmandible.



Fig 10– Ossifying fibroma – well defined, multilocular radiolucency, root resorption was seen on left side ofmandible.

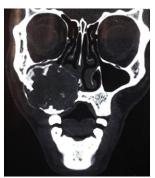


Fig 11 - Ossifying fibroma – well demarketed lesion on right maxilla.



Fig 12 - Ossifying fibroma – occulosal view showing expansion of both cortical plate on mandible.

The histological appearance of fibrous dysplasia usually exhibits a moderatelycellular fibrous stroma containing haphazardly arranged, spindle-shaped to ovoid fibroblastswhich are well differentiated and mature. The trabeculae tend to be delicate and curvilinear and have been linked to Chinesescriptwriting.8 Ossifying fibroma presents a relatively avascular fibrous stroma consist of fusiform cells intermingled with bone trabeculae and spheroidal calcifications resemble cement like structure.Focally scatteredmultinucleated giant cells also may be seen. The calcified materialmay consist of thin, irregularly shaped trabeculae of woven bone; scattered trabeculae of lamellar bone; deposits of basophilicstaining, round or ovoid, cellular or acellularcalcified depositsthat have been linked to cementum or any combination.24 which was comparable to our study.

Fibrous dysplasia is generally selflimiting and does not require treatment expect for cosmetic reasons, pain, discomfort or impaired function.¹⁸ Surgical procedures may be required for correction ofthe deformity, prevention of pathological fracture, anderadication of symptomatic lesions.8 The treatment consist of surgical recontouring or resection whichshould be postponed until after cessation of skeletal growth becauseearly treatment may accelerate growth of the lesion.6

Fibrous Dysplasia Case no - 01





Fig13

Fig14

Fig13- Fibrous dysplasia - clinical presentation, Fig14 - Fibrous dysplasia - post operative photographs. Recurrence occured one years after operation.

Ossifying fibroma - Case no - 01













Fig19

Fig 20

Fig 15- ossifying fibroma - post operative case. After surgical excision of lesion reconstruction was done by reconstruction plate. After 18 months later reconstruction plate was exposed. Fig 16- per operative case of ossifying fibroma. After removal of reconstruction plate, reconstruction was done by fibula graft. Fig 17-Ossifying fibroma - per operative photographs. Afterreconstruction by fibula graft micro vascular anastomosis was done. Fig 18- ossifying fibroma - final closure after reconstruction of fibula.-Fig 19- Ossifying fibroma - post operative radiographs after reconstruction. Fig 20- Ossifying fibroma – post operative photographs after 6 months later.

Ossifying fibroma - case no - 02





Fig 22



Fig 24



Fig 26



Fig16

Fig 23

Fig17

52

Fig 21– Ossifying fibroma – Mild sweeling on left maxilla. Fig 22– Ossifying fibroma - per operative photographs. A round well defined mass was seen and enucleation was done. Fig23–Ossifying fibroma - per operative photographs. After enucleation of surgical mass which was well defined. Fig 24– ossifying fibroma. After enucleation intact surgical specimen obtained at surgery. Fig 25– ossifying fibroma. After enucleation sectioned surgical specimen. Fig 26– ossifying fibroma – post operative photographs after 12 months later.

Lane et al.²⁵ treated fibrous dysplasia with oral or intravenous bisphosphonates and his therapy diminished pain, prevented fracturesand led to partial resolution oflesions. Chemotherapy is proved ineffective in retarding the progression of disease. Steroids have been used with partial success in treating painful lesions. Radiotherapy is contraindicated and has been associated with sarcomatous change.²⁶

In our study 05(50%) patients were treated by surgical recontoring and 05(50%) patients were still observation until the age of 18 years and were instructed for regular follow up. 01 patient of polyostotic form of fibrous dysplasia we used bisphosphonate therapy and the patient is under regular follow up.

Ossifying fibroma is often found to be well-encapsulated and easily enucleated. Treatment of ossifying fibroma is surgical enucleation of the lesion and curettage.21 The large ossifying fibroma which destroyed considerable bone may require surgical resection and reconstruction.In partial and incomplete excisions may cause a high recurrence rate due to locally aggressive behaviour in nature. In this study 12(60%) patients had local excision with curettage, 05(25%) patients had segmental mandibular resection and 03 (15%) patients had partial maxillectomy which is comparable to a study of Haider IAet al.27 The different type of fibro osseous lesions such as fibrous dysplasiaand ossifying fibroma can exibit similarclinical, radiological and histopathological features. As a result, distinguishing between them can be challenging. The clinical and radiological characteristics will help the diagnosis and therapeutic orientation of lesions and histopathological characteristics confirm the nature of lesion.

Conclusion

The most common fibro-osseous lesion in our clinical study was fibrous dysplasia and ossifying fibroma which presents painless bony swelling and deformity in maxilla and mandible. Fibrous dysplasia presents as a homogenous, radioopacity, ill defined borderandossifying fibroma presents a mixed radioopacity and radiolucent lesion that is well demarcated from normal bone. Surgical recontouring and clinical observation was done in treatment of fibrous dysplasiaand ossifying fibroma wastreated enucleationand curettage, segmental resection completely enucleate from surrounding bone.

This short time study reveals that a more detailed and longer duration of study is needed to clarify the present study for better management of these lesions.

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