Fibroosseous Lesions of the Craniofacial Region: A 14 Year Experience

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ABSTRACT

Introduction: Fibro-osseous lesion (FOL) is a broad term for a diverse group of jaw and craniofacial disorders that are often painless but grow progressively if uncontrolled. They are characterized by the replacement of normal bone by fibrous connective tissue matrix displaying varying degrees of mineralization in the form of woven bone or cementum-like structures.

Patients and Methods: Retrospective survey of the records of all patients seen between 2000 and 2013 at the department of Oral and Maxillofacial Surgery clinic, Ahmadu Bello University Teaching Hospital Zaria, Nigeria was undertaken.

Results: A total of 104 patients were seen over the period of study. Of these, males comprised 40 and female, 64 giving a male to female ratio of 1:1.6. The age ranged from 4-66 years, mean 25.8 (13.21) years. There was no gender difference in terms of age (t=1.78; P=0.078). Fibrous dysplasia was the most predominant lesion and accounted for 60 (57.7%) of the cases and this was followed by ossifying fibroma (n=39; 37.5%).

Conclusion: Due to late presentation, the size of the lesion, the outcome is usually not good esthetically especially for maxillary lesions.

Keywords: Fibro-osseous, fibrous dysplasia, lesions, ossifying fibroma.

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INTRODUCTION

Fibro-osseous lesion [FOL] is a broad term for a diverse group of jaw and craniofacial disorders that are often painless but grow progressively if uncontrolled. They are characterized by the replacement of normal bone by fibrous connective tissue matrix displaying varying degrees of mineralization in the form of woven bone or cementum-like structures (1, 2). These pathologic conditions can be categorized as developmental lesions, reactive or dysplastic diseases, and neoplasms (3, 4, 5).

FOL's may be associated with significant aesthetic and functional disturbances, or they may be completely asymptomatic lesions that are recognized only on routine radiographs (5). The diverse FOLs have common characteristics including clinical, histological and radiological features. However, several classifications have been proposed for these lesions, yet they remain confusing to clinicians, pathologists and surgeons (6).

Accounts of FOLs in black Africans indicate that they are common there, and frequently assume gigantic sizes due to late presentation (7). In view of changes in classification over the years and the paucity of recent reports from Northern Nigeria, we wish to present an analysis of 104 cases seen over 14years in Zaria with emphasis on the challenges of management.

PATIENTS AND METHODS

Retrospective survey of the records of all patients seen between 2000 and 2013 at the department of Oral and Maxillofacial Surgery clinic, Ahmadu Bello University Teaching Hospital Zaria, Nigeria was undertaken. Records with histological report of FOL based on WHO classification of FOL [2005] were selected for further study. Data collected included patient's age, gender, presenting complaints, location of lesion, histolopathologic classification of lesion, applied

treatment and result of follow up visits. Data were analyzed using SPSS version 13. Verbal consent were obtained for the photographs used in this study.

RESULTS

A total of 104 patients out of 1170 were seen over the period of study. Of these, males comprised 40 and female 64 giving a male to female ratio of 1:1.6. The age ranged from 4-66 years, mean 25.8 [13.21] years. There was no gender difference in terms of age [t=1.78; P=0.078]. Fibrous dysplasia was the most predominant lesion and accounted for 60 [57.7%] of the cases and this was followed by ossifying fibroma [n=39; 37.5%]. Table1 shows the gender distribution of various histopathologic types of FOLs seen at Zaria, Nigeria from 2000-2013. Although there was slight female predominance in the distribution of fibrous dysplasia, ossifying fibroma and cemento-osseus dysplasia, there was no statistically significant difference with respect to gender distribution [X2=3.38; df=4; P=0.574] shown in Table 1. The maxilla [n=56; 50.4 %] was the most common site of occurrence and this was closely followed by the mandible [n=41; 39.4%].

The distribution of the different types of fibro-osseous lesions was significant according to site [X2=83.94; df= 16; P= 0.002]. While fibrous dysplasia had a strong predilection for occurrence in the maxilla, ossifying fibroma was seen slightly more frequently in the mandible than in the maxilla. Table 2 shows site distribution of FOLs seen in Zaria, Nigeria. There was a wide age range of distribution for fibrous dysplasia and ossifying fibroma, but both lesions occurred most frequently around the 2nd and 3rd decades. Table 3 shows the age distribution of FOLs seen between 2000 and 2013. Table 4 shows the main clinical features of FOLs.

Of the 104 patients, 96 [92.3%] had surgical treatment with surgical excision as the most common mode of treatment and the least modality of treatment was subtotal mandibulectomy[Table 5]. About 7, 7% could not afford treatment.

DISCUSSION

FOL constituted 8.9% of all the tumours seen within the period. This was higher than 2.3% by Ajike et al (8) in Kano Nigeria and 2.47% by Butt et al (9) in Kenya but lower than the 13.0% by Bassey et al (10) in South-south Nigeria. Yamani et al (11) got 13.55% in Saudi Arabia.

FOL occurs over a wide age range. They were common within the second and third decades in this study [65.4%] while Ajagbe and Daramola (12) had the age range between 4 and 68 years with 56% occurring in the first two decade, Williams et al (13) with about 60% occurring within the first two decades with age range of 8 to 38 years [mean 21.3 years]. According to Makwana et al (14) some report claimed it is common within the first and second decade while other reports say more than 50% occur in the third and fourth decade. The age ranged from 4-66 years, mean 25.8 [13.21] years. Khattab et al (15) had age range 15 to 64 years with a mean of 37.56 years.

There were more female [61.5%] than male [38.5%] in a ratio of 1:1.6[male: female] while early report from William et al (13) got ratio 3:1[female: male]. Muwazi and Kamulegeya (16), got 65% female. Khattab et al (15) got 60% female. This was the same view with Lasisi et al(5) who found that fibro-osseous lesions of jaws were more in females [61.2%] than males [38.8%], giving a male to female ratio of 1:1.6. Kolomvos et al (17), found a male predilection

The grouping and classification of FOL's is dynamic and constantly changing, for example, Eversole et al (3), grouped FD under bone dysplasia but there are studies which classify FD as a neoplasm based on recent molecular findings. A classification system for FOL's by Waldron is widely accepted but even this has been slightly modified (5).

FOLs have been generally divided into two major groups: those originating from the medullary bone [FD as well as osteoblastoma, cherubism, aneurysmal bone cyst] and those originating from the periodontal ligament including OF, cement-ossifying fibromas and cement-osseous dysplasia (17).

Fibrous dysplasia [FD] was the most predominant lesion and accounted for 60 [57.7%] of the cases and this was followed by ossifying fibroma [n=39; 37.5%]. This agrees with Muwazi and Kamulegeya(16) who found that fibrous dysplasia was the most prevalent lesion [n = 87, 56.1%] followed by ossifying fibroma [n = 50, 32.9%] and osseous dysplasia [n = 17, 10.9%] in Uganda and Yamani et al(18),FD [n=10,47.6%] and OF[n=7,33.3%] in Saudi Arabia, Makwana et al(14) FD[39.3%] , OF[28.5%] in India and an earlier Nigerian study by Williams et al(13) , FD and OF was reported as constituting 37.1% and 22.9% respectively . This was how eve different from Lasisi et al (5) in South-west Nigeria who found ossifying fibroma [62%], fibrous dysplasia [37.2%] and florid cemento-osseous dysplasia.

There was however no statistically significant difference with respect to gender distribution. This agrees with Lasisi et al (5) in Southwest Nigeria. According to Nwizu et al (4), there is an equal sex distribution in FD except in polyostotic where there is female predilection.

There was a wide age range of distribution for fibrous dysplasia and ossifying fibroma, but both lesions occurred most frequently around the 2nd and 3rd decades. Makwana et al (14) found that

they occurred more in the first two decades of life. . According to Nwizu et al (4), there is an equal sex distribution in FD except in polyostotic where there is female predilection.

In the present study, the maxilla was the most common site of occurrence and this was closely followed by the mandible. This agrees with Makwana et al (14) but disagree with Kolomvos et al (17) who found the mandible to be mostly affected. While fibrous dysplasia [FD] had a strong predilection for occurrence in the maxilla, ossifying fibroma [OF] was seen slightly more frequently in the mandible than in the maxilla. The result, while it agrees with that of Nwizu et al(4), however contrasts that of Lasisi et al(5) who found OF more in the maxilla and mandibular predilection for FD.

FD can occur as monostotic or polyostotic forms, with or without a syndromic association. The monostotic type accounts for 80% of cases seen. Craniofacial FD is more commonly associated with the polyostotic types (4).

The lesion appeared just before puberty, and painless, progressive enlargement of the affected bones continued through adolescence to produce severe disfigurement [Fig 1] but surprisingly few symptoms. Disfigurement, though often tolerated for several years, becomes socially unacceptable and patients demanded treatment for both functional and aesthetic reasons (7).

Adekeye et al (7) in his review of literature observed that earlier authors attributed localized pain, headaches, anosmia, deafness and malnutrition through impaired mastication to progressive crania-facial fibrous dysplasia [Fig 2]. They found that changes such as orbital involvement with marked vertical displacement of the eye can be compensated for and will not always produce diplopia. However, the orbit may be filled with lesional tissue, and the displaced eye blind and atrophic. In our experience one female patient [Fig 3] with FD had blindness. The

maxillary bone may enlarge leading to obliteration of the buccal sulcus and elevation of the hard palate [Fig 4]. Massive tumours may protrude from the mouth [Fig 5], and part of the lesion may be extruded through the nostrils [Fig. 1]. Teeth are often displaced but remained firm despite bony enlargement.

The radiologic picture of an ill-defined lesion blending imperceptibly with the adjacent bone is said to be a defining characteristic of FD (4).

The radiographic appearances of these crania-facial fibrous dysplasias varied, and included 'ground-glass', lytic and sclerotic areas, usually with indistinct margins, an important diagnostic feature (7). These radiographic appearances were also noted in our patients.

Plain x-ray films and CT scans of OF reveal a circumscribed lesion that may be lytic, sclerotic or mixed. A significant feature of COF is its proclivity to shell out completely during surgery (4) .

According to Kolomvos et al (17), panoramic radiographs usually followed by a CT scan contribute to determine the lesion's margins and its relation to vital structures. An important radiologic diagnostic factor is the association of the lesion with the mandibular canal. COF, cementoblastoma of COD displace the mandibular canal downwards as they expand. Conversely, lesions arising and developing below the canal, displace it upwards, such as FD.

Treatment is surgical and ranges from conservative to extensive resection (17). It is also highly individualized. In our study surgical excision was the most common mode of treatment and the least was subtotal mandibulectomy.

The standard treatment of FD is surgical paring down of bone in symptomatic patients, since its growth tends to plateau after puberty and also for cosmetic reason. In asymptomatic patients, surgical intervention is traditionally discouraged for the same reason, though some surgeons choose to operate because growth sometimes continues unabated post-puberty (4).

Radical surgery may be indicated for extragnathic lesions of the head and neck.

Recurrence of FD is generally low, but rates as high as 25-50% have been reported in some cases treated by surgical recontouring (4). About 10% 0f our patients had recurrence.

A significant fact is that both syndromic and non-syndromic FD can undergo malignant transformation although the risk is quite low. Therefore, long-term follow-up is crucial for all patients diagnosed with FD irrespective of the particular variant (4). One patient [Fig.3] in our series had malignant transformation even though our follow up was not long for most patients are lost as soon as the tumour is removed.

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Table 1: Distribution of fibro-osseous lesions according to gender

Types	Female	Male	Total
Ossifying Fibroma	23	16	39
Fibrous Dysplasia	39	21	60
Cemento-osseous Dysplasia	2	0	2
Cherubism	1	1	2
Myositis ossificans	0	1	1
Total	64	40	104

X2=3.38; df= 4; P=0.574

Table 2: Site distribution of fibro-osseous lesions in 104 patients

Types	Cheek	Frontal bone	Mandible	Maxilla	Mandible-Maxilla	Total
Ossifying Fibroma	1	2	19	16	1	39
Fibrous Dysplasia	0	1	22	37	0	60
Cemento-osseus Dysp.	0	0	2	0	2	
Cherubism	0	0	0	1	1	2
Myositis Ossificans	1	0	0	0	0	1
Total	2	3	41	56	2	104

X2=83.94; df= 16; P= 0.002

Table 3: Distribution of fibro-osseous lesions according to age group (n=104)

Types	1-10 Years	11-20 years	21-30 years	31-40 years	41-50 years	51-60 years	61-70 years	Total
Ossifying Fibroma	4	10	13	7	4	1	0	39
Fibrous Dysplasia	5	21	19	7	5	2	1	60
Cemento-osseus Dysp.	0	0	2	0	0	0	0	2
Cherubism	0	2	0	0	0	0	0	2
Myositis Ossificans	0	1	0	0	0	0	0	1
Total	9	34	34	14	9	3	1	104

X2=12.72; df= 24; P=0.887

Table 4: Main clinical features in fibro-osseous lesions (n=104)

		Clinical Features*			
Tumour type	Duration	Swelling	Protruding	Displaced teeth	Pain
FD	1-25 yrs	60	36	56	12
OF	8mths-8yrs	39	18	22	3
COD	1-7 yrs	1	-	-	2
Cherubism	4-8yrs	2	-	-	-
Myositis ossif.	3yrs	1	-	-	1
Total		103	54	78	18

FD= Fibrous dysplasia
OF=Ossifying fibroma
COD= Cemento-osseous dysplasia

Table 5: Types of surgical treatment for fibro-osseous lesions in 56 patients

Surgical Treatment	Frequency	Percentages
Excision	46	82.1
Resection	3	5.4
Shaving	2	3.6
Resection + Disarticulation	2	3.6
Hemi-maxillectomy	2	3.6
Subtotal Mandibulectomy	1	1.8
Total	**18**	**100**

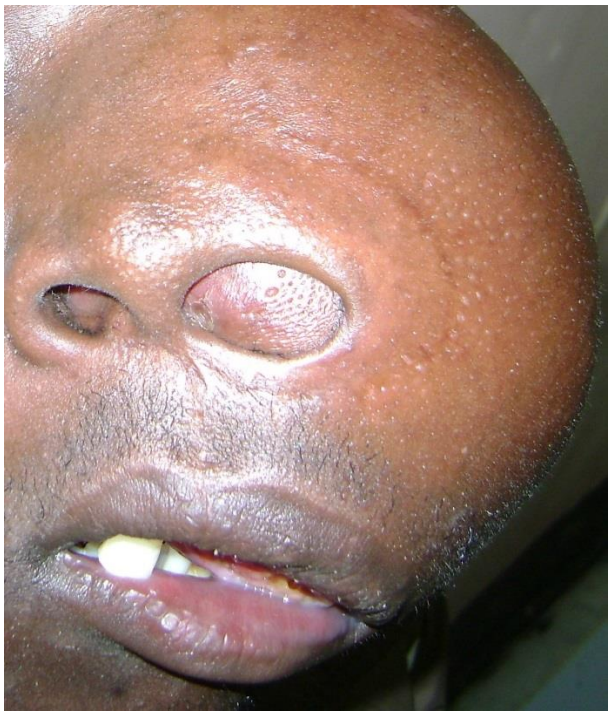

Fig. 1. Male patient with facial disfigurement

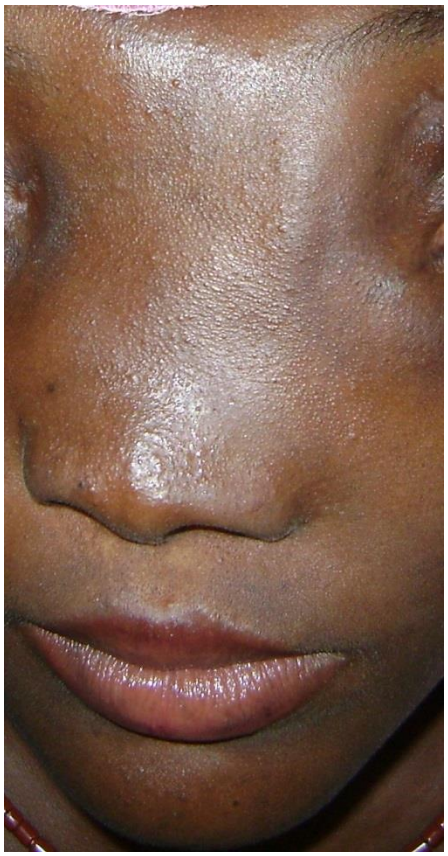

Fig. 2: Female patient with craniofacial fibrous dysplasia

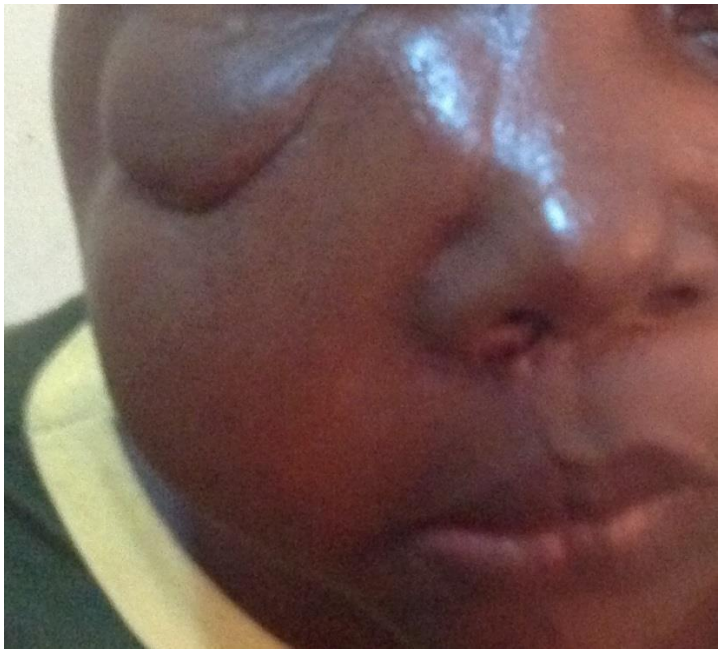

Fig. 3: Female patient with loss of vision on the right with previous history of surgery and malignant transformation.

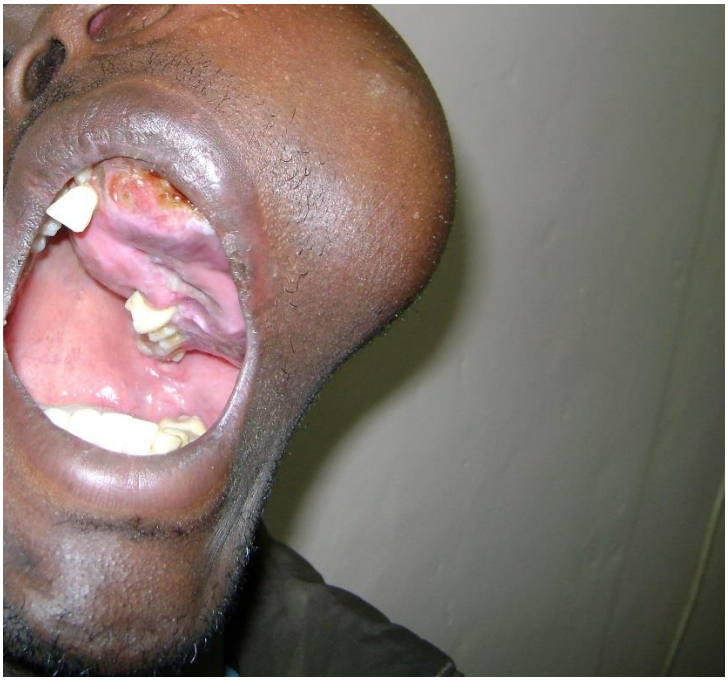

Fig. 4: Male patient with raised palate.

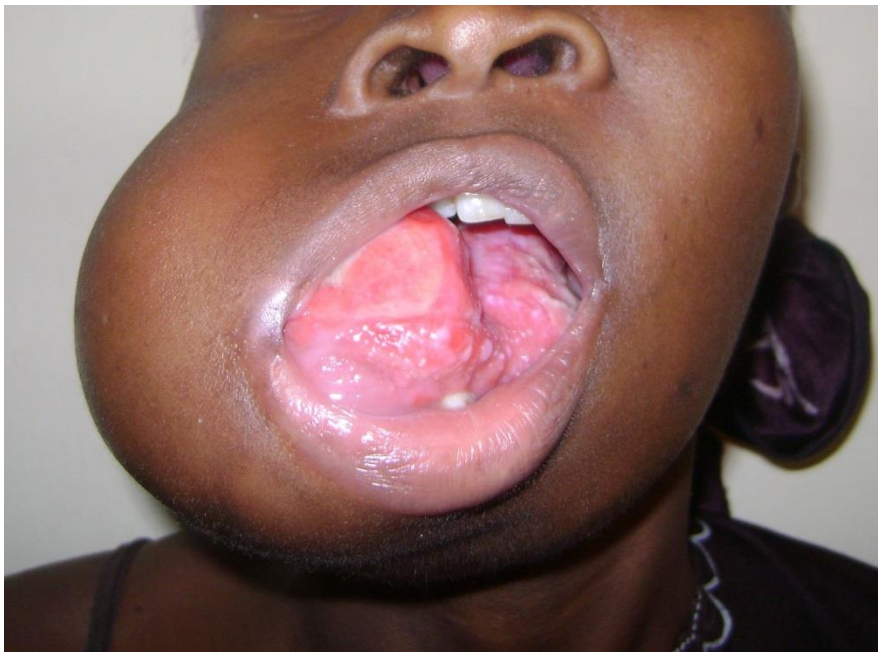

Fig. 5: Female patient with ossifying fibroma protruding from the mouth.