ORIGINAL ARTICLE

Fibro-osseous Lesions of the Craniofacial Region: A 14-year Experience

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ABSTRACT

Introduction: *Fibro-osseous lesion 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.*

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 females comprised 64 giving a male to female ratio of 1:1.6. The age ranged from 4 years to 66 years and mean was 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 common 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 aesthetically, especially for maxillary lesions.*

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

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 (3–5).

Fibro-osseous lesions 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 14 years in Zaria with emphasis on the challenges of management.

SUBJECTS 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 the WHO classification of FOL (2005) were selected for

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 DOI: 10.7727/wimj. 2016.150

further study. Data collected included patient's age, gender, presenting complaints, location of lesion, histopathologic classification of lesion, applied treatment and result of follow-up visits. Data were analysed using SPSS version 13 (IBM Corp., Armonk, NY, USA). Verbal consent was 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 females comprised 64 giving a male to female ratio of 1:1.6. The age ranged from 4 years to 66 years and mean was 25.8 (13.21) years. There was no gender difference in terms of age (t = 1.78; $p = 0.078$). Fibrous dysplasia (FD) was the most common lesion and accounted for 60 (57.7%) of the cases and this was followed by ossifying fibroma (OF) (n = 39; 37.5%). Table 1 shows the gender distribution of various histopathologic types of FOLs seen at Zaria, Nigeria from 2000 to 2013. Although there was slight female predominance in the distribution of FD, OF and cemento-osseous dysplasia, there was no statistically significant difference with respect to gender distribution (Chi-square = 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 FOLs was significant according to site (Chi-square = 83.94; df = 16; $p = 0.002$). While FD had a strong predilection for occurrence in the maxilla, OF 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 FD and OF, but both lesions occurred most frequently around the second and third decades. Table 3 shows the age distribution of FOLs seen between 2000 and 2013. Table 4 shows the main clinical features of FOLs.

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	65	39	104

Chi-square = 3.38; df = 4; $p = 0.574$.

Of the 104 patients, 96 (92.3%) had surgical excision as the most common mode of treatment and the least common modality of treatment was subtotal mandibulectomy (Table 5). About 7.7% could not afford treatment.

DISCUSSION

Fibro-osseous lesion 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*

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-osseous dysplasia	0	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

$\chi^2 = 83.94$; 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-osseous dysplasia	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

Chi-square = 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 years	60	36	56	12
OF	8 months to 8 years	39	18	22	3
COD	1–7 years	1	–	–	2
Cherubism	4–8 years	2	–	–	–
Myositis ossificans	3 years	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	56	100.1

(9) in Kenya, but lower than the 13.0% by Bassey *et al* (10) in South–south Nigeria. Al Yamani *et al* (11) got 13.55% in Saudi Arabia.

Fibro-osseous lesion 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 decades, Williams *et al* (13) found about 60% occurring within the first two decades with age range of 8–38 years (mean 21.3 years). According to Makwana *et al* (14), some report claimed that it is common within the first and second decades, while other reports say that more than 50% occur in the third and fourth decades; the age ranged from 4 to 66 years and mean was 25.8 (13.21) years. Khattab *et al* (15) had age range of 15–64 years with a mean of 37.56 years.

There were more females (61.5%) than males (38.5%) in a ratio of 1:1.6 (male:female), while early report from Williams *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 FOLs of jaws were more common 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 FOLs 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 FOLs by Waldron is widely accepted but even this has been slightly modified (5).

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

Fibrous dysplasia was the most common lesion and accounted for 60 (57.7%) of the cases and this was followed by OF (n = 39; 37.5%). This agrees with Muwazi and Kamulegeya (16) who found that FD was the most prevalent lesion (n = 87; 56.1%) followed by OF (n = 50; 32.9%) and osseous dysplasia (n = 17; 10.9%) in Uganda, Waldron and Giansanti (18) found FD (n = 10; 47.6%) and OF (n = 7.33; 3%) in Saudi Arabia, Makwana *et al* (14) who found FD (39.3%) and OF (28.5%) in India and Williams *et al*, who in an earlier Nigerian study (13), reported FD and OF as constituting 37.1% and 22.9%, respectively. This was however different from Lasisi *et al* (5) in Southwest Nigeria who found OF (62%), FD (37.2%) and florid cemento-osseous dysplasia.

There was, 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 FD and OF, but both lesions occurred most frequently around the second and third decades. Makwana *et al* (14) found that they occurred more in the first two decades of life.

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 disagrees with Kolomvos *et al* (17) who found the mandible to be mostly affected. While FD had a strong predilection for occurrence in the maxilla, 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), contrasts with that of Lasisi *et al* (5) who found OF is more common in the maxilla and mandibular predilection for FD.

Fibrous dysplasia 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 with painless, progressive enlargement of the affected bones continued through adolescence to produce severe disfigurement (Fig. 1), but surprisingly few other symptoms. Disfigurement, though often tolerated for several years, becomes socially unacceptable and patients sought treatment for both functional and aesthetic reasons (7).

Fig. 1: Male patient with facial disfigurement.

Adekeye *et al* (7) in his review of the literature observed that earlier authors attributed localized pain, headaches, anosmia, deafness and malnutrition through impaired mastication to progressive craniofacial FD (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

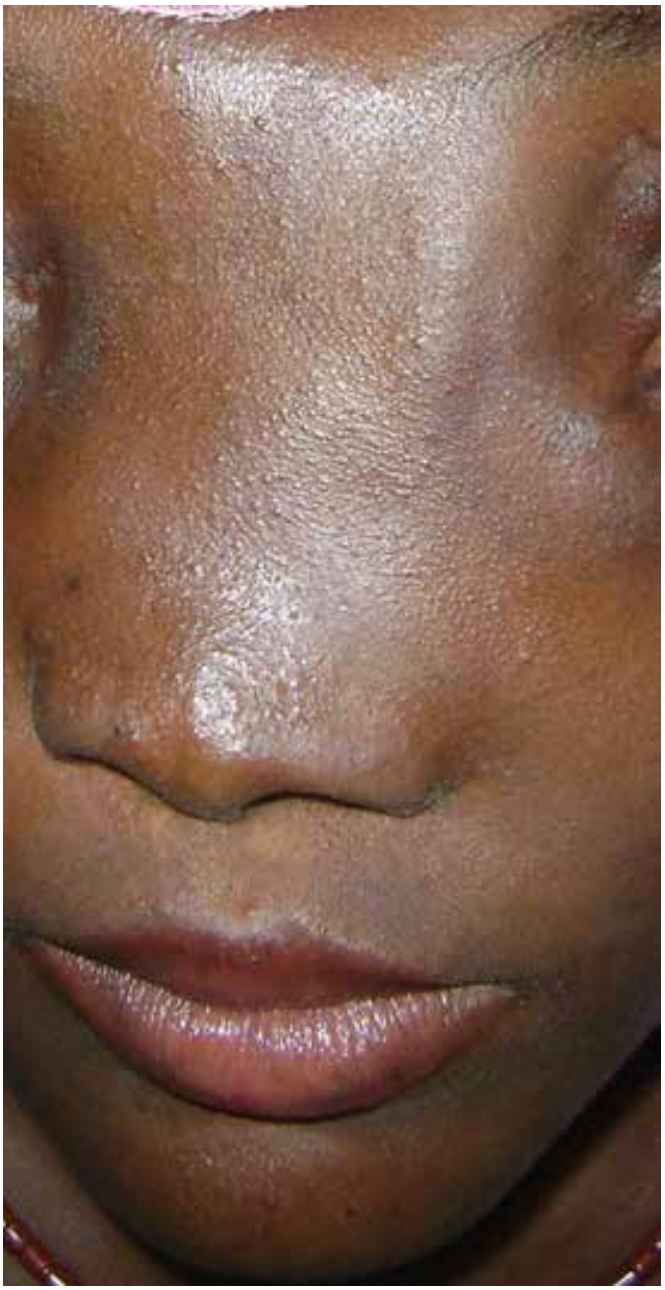

Fig. 2: Female patient with craniofacial fibrous dysplasia.

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).

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.

The radiographic appearances of these craniofacial FDs 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 cemento-ossifying fibroma (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 and cementoblastoma of cemento-osseous dysplasia displace the mandibular canal downwards as they expand. Conversely, lesions arising and developing below the canal displace it upwards as does 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 reasons. In asymptomatic patients, surgical intervention is traditionally discouraged for the same reason, though some

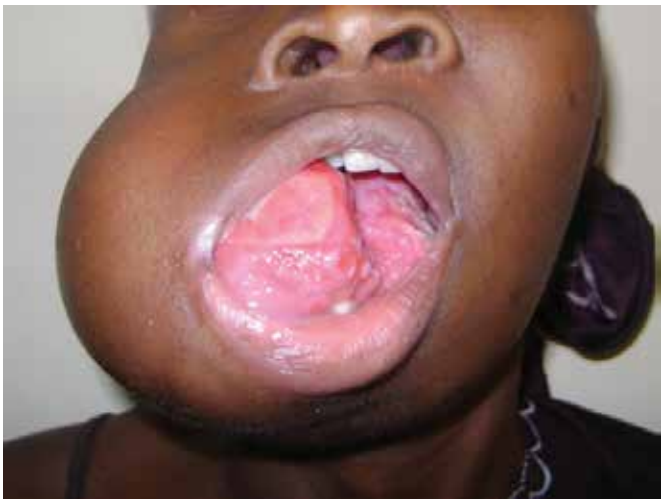

Fig. 5: Female patient with ossifying fibroma protruding from the mouth.

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% of 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 was not long; most patients are lost to follow-up as soon as the tumour is removed.

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