BENIGN FIBRO-OSSEOUS LESIONS OF THE FACIAL SKELETON: ANALYSIS OF 52 CASES SEEN AT THE KORLE BU TEACHING HOSPITAL

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SUMMARY
All cases of histologically confirmed fibrous dysplasia, ossifying fibroma or cementifying fibroma presenting for treatment at the Dental Department of the Korle Bu Teaching Hospital (KBTH) over a 14-year period (January 1989-December 31 2002) were reviewed to find out the characteristics of the disease in Ghana. A total of 52 cases of fibro-osseous tumors were obtained with an age range of 7-42 years. The mean age was 21 years (SD 7.1) and peak age range of occurrence was between 11-20 years. The commonest fibro-osseous lesion was ossifying fibroma (61.5%) followed by fibrous dysplasia (30.8%) and cementifying fibroma (7.7%). The overall male: female ratio was 2:3. Treatment was surgical curettage or excision of access bone with reshaping to resemble the unaffected side. The crude prevalence rate assessed in the Dental department of the KBTH was 2.46% of head and neck tumors. Treatment was generally successful with only two recurrences (3.8%) over a ten-year follow-up period.

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

INTRODUCTION
Benign fibro-osseous lesions of bone comprise tumor and tumor-like conditions with similar histological appearances but different clinical behaviour in patients[1-3]. These include fibrous dysplasia, ossifying fibroma and cementifying fibroma and also lesion with mixed histological features[1,2]. Fibrous dysplasias can be poly-ostotic or mono-ostotic with essentially the same microscopic appearance[4,5]. The poly-ostotic forms may be accompanied by pigmented skin lesions, and endocrine dysfunction as in Albrights syndrome, and as familiarly and hereditary forms[5,6]. Ossifying fibroma occurs most frequently in young adult women, and seems to have a predilection for the mandible[7]. Its relationship to fibrous dysplasia and other fibro-osseous lesions has been reported at length[1-3]. Cementifying fibroma is a condition also known as multiple cementoma and periapical fibrous dysplasia. In its most common form periapical areas of bone destruction appear, presumably as a result of proliferation of the periodontal membrane[8]. Fibro-osseous lesions may thus require extensive investigation for successful classification, and management requires careful interpretation of histological appearance along with radiographs and medical, family had dental history[9]. Adequate clinical history information on the tumor as seen at surgery and its behaviour after treatment are also important.

There is a lack of data on benign fibro-osseous lesions in Ghana and this paper seeks to provide data on these lesions and their management at the Korle Bu Teaching Hospital.

MATERIALS AND METHODS
The material studied were obtained from 52 cases histologically diagnosed as fibrous dysplasia, ossifying fibroma or cementifying fibroma presenting for treatment at the Dental Department of KBTH over a 14-year period, (January 1989-December 31, 2001). All the patients studied were Ghanaians. Thirty-eight of the patients were studied personally by one of us, whiles the case records and x-rays of the remaining fourteen patients were reviewed. Histopathology reports from the files of the Department of Pathology were retrieved along with slides and reviewed to confirm the diagnoses. Initial diagnoses of all the cases were based on their clinical presentations coupled with their radiological appearances while histological studies were based on biopsy and surgical specimens. All the specimens were examined grossly and fixed in 10% formalin. Routinely de-calcified paraffin em-

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bedded sections stained with haematoxylin and eosin were prepared and examined histologically.

RESULTS

During the 14-year period under review a total of 2,108 biopsies were recorded in the register for all types of lesions involving the head and neck region at the Korle Bu Teaching Hospital (KBTH). Fifty-two of these were confirmed histologically as fibrous dysplasia, ossifying fibroma or cementifying fibroma giving a prevalence of 2.46% of all lesions undergoing biopsy from the head and neck region. Sixteen cases (30.8%) of the fibro-osseous lesions were fibrous dysplasia, thirty-two cases (61.5%) were ossifying fibromas and 4 cases (7.7%) were cementifying fibromas (Table 1).

Sex, Age Distribution and Duration of Lesion

In this survey, all three variants of fibro-osseous lesions were found to be more common in females than in males (Table 1). Twenty-one cases (40.4%) of the study sample were male and thirty-one (59.6%) were females, thus giving a male to female ratio of approximately 2:3. This pattern of female preponderance was found to be the case in each of the three variants.

Table 1 Sex distributions of fibro-osseous lesions in the jaws of 52 patients

Lesion	Male	Female	Total (Percentage)
Fibrous dysplasia	7	9	16 (30.8)
Ossifying fibroma	13	19	32 (61.5)
Cementifying fibroma	1	3	4 (7.7)
All three lesions	21	31	52 (100)

The youngest patient was 7 years and the oldest 42 years (Table 2). Both patients were female and both presented with ossifying fibroma. However, considering the variants separately, the mean age for ossifying fibroma at the time of treatment was 19.9 years (SD 11.2), and the peak occurrence was between the ages of 11 and 20 years with 53.2% (17 out 32) of patients. Two of the four cases of cementifying fibroma were 14 years old while the other two were 15. The declared duration of ossifying fibroma by the patients prior to consultation varied from two months to fifteen years, while that of cementifying fibroma varied from four months to two years.

The youngest patient presenting with fibrous dysplasia in this study was a 12-year-old male while the oldest was a 38-year-old female. The mean age at the time of treatment was 21.75 years (SD 5.9), and the declared duration of the tumour, by the patients prior to treatment, varied from sin months to 22 years. The peak age range of occurrence was between 11 and 20 years.

Site Distribution

Table 3 shows the frequency of occurrence of the various fibro-osseous lesions in the facial skeleton of 52 patients. Fibrous dysplasia was located in the maxilla in 12 out of 16 cases (75%). In 3 out of 16 cases (18.75%), the lesion was located in the mandible, and only in 1 out of 16 cases (6.25%) was it located, outside the jaws, in the frontal bone. Both ossifying fibroma and cementifying fibroma were most frequently located in the mandible, in 87.5% (28 out of 32) of cases and 75% (3 out of 4) of cases respectively. Ossifying fibroma and cementifying fibroma were seen in the maxilla in 12.5% and 25% of cases respectively. None of these lesions were seen outside the jaws in the same patient. Considering all three disease entities as a whole fibro-osseous lesions in this survey affected the mandible in 34 cases (65.4%), the maxilla in 17 cases or 32.7% and the frontal bone in 1 case or 1.9%.

Table 2 Age distribution of the study sample at time of treatment

Age range (year) three	Fibrous dysplasia	Ossifying fibroma	Cementifying fibroma	All three lesions
0-10	0	5	0	5
11-20	7	17	4	28
21-30	8	3	0	11
31-40	1	5	0	6
41-50	0	2	0	2
All ages	16	32	4	52

Clinical features and diagnosis

The jaws were often enlarged, expanded and distorted in most patients, and this was the main presenting symptom in 92% (48 out of 52) of patients. In fibrous dysplasia, there was usually gross facial deformity from gross expansion of the bone in-

volved was usually the maxilla(Figure 1). In the case involving the frontal bone (Figure 2), expansion of the lesion into the orbit resulted in proptosis and downwards displacement of the eye.

Table 3 Site distribution of fibro-osseous lesion in the facial bones of 52 patients

Lesion	Mandible	Maxilla	Frontal bone	Total
Fibrous dysplasia	3 (18.75%)	12 (75.0%)	1 (6.25%)	16 (100%)
Ossifying fibroma	28 (87.5%)	4 (12.5%)	0	32 (100%)
Cementifying fibroma	3 (75.0%)	2 (25.0%)	0	4 (100%)
(Fibro-osseous lesions)	34 (65.4%)	17 (13.7%)	1 (1.9%)	52(100%)

Pain was not usually a significant factor, though it was present in some cases. This was often due to secondary infection arising from ulceration on the indented surfaces of the intra-oral expansion of the lesion where there was impingement of the teeth from the opposing jaws.

Figure 1 Fibrous dysplasia affecting the left maxilla in a 14-year old male patient. Note the upward displacement of left eye from expansion of the lesion into the orbit.

Figure 2 Photograph of a case of fibrous dysplasia of the frontal bone in a 12-year old male patient.

Radiographs largely exhibited a dense mass often with 'ground glass' appearance with ill-defined margins in the case of fibrous dysplasia. When it involved the maxilla, the lesion often expanded to obliterate the sinus on the affected side. In the mandible, the central portions of the largely 'ground glass' appearance often exhibited microcystic areas. With ossifying and cementifying fibroma, there was usually a clearly demarcated area of patchy radioluscency (Figure 3), appearing cystic at times. Forty-four (84.6%) of the fifty-two cases studied were correctly disgnosed by clinical and radiological finding while the remaining eight cases or 15.4% were diagnosed clinically as either osteoma or fibroma. In the list of differential diagnosis, benign osteoma topped with the highest index, followed by myxoma and ameloblastoma in the order.

Figure 3 Radiograph of the mandible affected with ossifying fibroma. Note the clearly demarcated area of patchy radioluscency in the left body of the mandible

Treatment

Of the 52 cases studied over the period, only 38 were treated and followed up by one of the authors; hence only the treatment of these patients is analysed in this paper. All the 38 patients were treated conservatively under general anaesthesia via an intra-oral approach. Teenage or younger patients presenting with fibrous dysplasia were often persuaded to defer surgery until they were about 21 years old, when skeletal growth was presumed to have ceased. In all such cases, surgery consisted of excising the enlarged diseased bone by paring it down and reshaping it to resemble that of the contra-lateral side. Where considerable bleeding was encountered during the surgery, the diseased bone was usually pared slightly lower than the normal side to allow for compensation by any organising haematoma following wound clo-

sure. Both ossifying fibroma and cementifying fibroma were essentially treated by curettage. Large volumes of normal saline were often used to wash the wounds thoroughly before closure. It was usual to achieve complete haemostasis before applying pressure dressing to minimize any haematoma formation. No drains were ever inserted.

Follow-up and results of treatment

The patients were usually hospitalized for periods ranging from 3 to 10 days post surgery. Review regimen started on the morning after surgery, and thereafter weekly for a month. The period was then extended to monthly for three months and then twice a year for as long as the patients could keep appointments. Most patients did not keep appointments beyond one year. Fourteen patients were followed-up beyond one year with one case of ossifying fibroma and another of fibrous dysplasia recurring nine years and eight months respectively after surgery. The recurrent lesion of fibrous dysplasia was larger and much more aggressive than the initial condition and thus necessitated complete removal of the affected maxillary bone, but sparing the palatal mucosa.

DISCUSSION

The overall incidence of fibro-osseous lesions of 2.4% of all head and neck tumors is comparable to that found in the literature[1-3]. In this study, ossifying fibroma was the commonest lesion forming in the 61.5% of cases. This is different from studies from advanced countries that found more fibrous dysplasia (67%)[1,2,11]. The reason for this difference in the incidence of the various types of lesions is not clear. The study showed a slight female preponderance even though the literature, from studies on larger samples, indicates equal incidence for males and females[10,11]. This finding is however, comparable to that reported by Ramsey et al (1968), who according to Killey et al[8], in their series of 47 patients, found 26 female and 21 male. The peak age for all lesions is reported as 25-35years[10]. This is higher than 11-20 years seen in this study and may be due to either the small number of cases reported on in this study, or possibly, to population differences.

The commonest site for fibrous dysplasia was the maxilla and the commonest site for cementifying fibroma and ossifying fibroma was the mandible. The main presentation of unilateral facial bone swelling is as reported by other studies[1-4]. There were no bilateral lesions seen. Though there are poly-ostotic forms of fibro-osseous lesions that may even have endocrine components[4-6], there was no such case in this study. Although the lesions may present difficulties with diagnosis, a combination of clinical findings and radiological appearances gave a high correlation with histological diagnosis. It is clear however that the best chance of correctly characterizing the lesions was a combination of clinical findings, radiological and histological appearances.

Fibro-osseous lesions are known to be benign and all who discuss it are virtually unanimous in recommending conservative treatment[7]. In view of the fact that areas of fibroma dysplasia continue to enlarge during the period of general skeletal growth it is advisable to defer surgery until growth has ceased[8]. Though, according to Killey et al[8], Seward (1970), reported cases where the lesion continued to enlarge after this time, no such observation was made here. In this series, most of the patients sought treatment only for cosmetic reasons, as there was very little disturbance of function. It was therefore easy to persuade most patients presenting with fibrous dysplasia to defer surgery until they were about 21 years old, when skeletal growth was presumed to have ceased.

This, in our view, accounted for the low rate of recurrence. In fact only one of the two cases that recurred in this survey was fibrous dysplasia. His deformity was so cosmetically unacceptable that earlier surgery was required. The patient was therefore, prior to the surgery, alerted of a possibility of recurrence. Indeed, the tumour recurred in eight months and required excision of the entire affected maxillary bone. There have been reports of malignant changes occurring in some cases of fibrous dysplasia, especially when radiated. No radiation was carried out in any of the patients studied, and there was no malignant change encountered in the series.

The ossifying fibroma and cementifying fibroma seldom recur and malignant degeneration has not been reported. For this reason, the sacrifice of large segments of bone is contraindicated. Cryotherapy has been reported to be very effective in treating tumours lying adjacent to or within bone, and has been effectively used in treating ossifying fibroma[7]. This facility is lacking in this hospital, and as such was never used. All the patients in this series were treated by simple curettage via intra-oral approach. This method was largely satisfactory with only one case of recurrence after a period of nine years. In all the patients, there was never the need to sacrifice or damage either the inferior alveolar or infra-orbital nerves during treatment.

CONCLUSION

Fibro-osseous lesions are benign conditions with similar histological appearances but different

clinical behaviour in patients. When surgical treatment is carried out on them at an early age, though both ossifying fibroma and cementifying fibroma seldom recur, in view of the fact that areas of fibrous dysplasia continue to enlarge during the period of general skeletal growth, it may recur with disastrous consequences. Their successful management therefore depends largely on the establishment of accurate clinical diagnoses aided by extensive investigation and careful interpretation of radiographs and medical, family and dental history. Most patients seek treatment for cosmetic reasons therefore, unless there is a significant disturbance of function, it is advisable to defer surgery until the cessation of general skeletal growth.

REFERENCE

1. Dehner LP. Tumors of the mandible and maxilla in children. I clinicopathologic study of 46 hsitologically benign lesions. *Cancer* 1973; 31: 364-384.

2. Eversole LR, Sabes WR, Rovin S. Fibrous dysplasia, a nosologic problem in the diagnosis of fibro-osseous lesions of the jaws. *J Oral Pathol* 1972; 1: 189-220.

3. Waldron CA, Giansanti LS. Benign fibro-osseous lesions of jaws. *Oral Surg Oral Med Oral Pathol* 1973; 35: 190-201, 340-350.

4. Reed RJL. Fibrous dysplasia of bone. *Arch Pathol* 1063; 75(1): 480-495.

5. Shmaman A, Smith I, Ackerman LV. Benign fibro-osseous lesions of the mandible and maxilla: A review of cases. *Cancer* 1970; 26: 303-312.

6. El Deeb M, Waite DE, Gorlin RJ. Congenital mono-ostotic fibrous dysplasia. A new possibly autosomal recessive disorder. *J Oral Surg* 1979; 37: 520-525.

7. William Sippel H, Emmings Fred G. Cryotherapy in the treatment of ossifying fibroma: report of a case. *J of Oral Surg* 27: 32-35.

8. Killey HC, Seward GR, Kay LW. An outline of oral surgery. Bristol: John Wright & Sons Ltd. 1975; (II): 111-112.

9. Pirce Am, Wilson DF, Goss AN. Inherited cranio-facial fibrous dysplasia. *Oral Surg Med Oral Pathol* 1985; 60: 403-409.

10. Su L, Weathers D, Waldron C. Clinical and radiological spectrum of focal cemento-osseous dysplasia and cemento-ossifying fibromas: A statistical study of 316 cases. *J Oral Med Oral Path* 1997; 84: 186.

11. Rosai J (Ed). Ackerman's surgical pathology (8^{th} Ed). 1999; 1: 259-260.