ORIGINAL ARTICLE

A clinico-pathologic review of 56 cases of ossifying fibroma of the jaws with emphasis on the histomorphologic variations

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Abstract

Objective: This study aims at contributing to the definitive diagnosis of ossifying fibroma (OF) based on histomorphological features. The study also aims to determine some demographic features of OF, common sites of occurrence and to determine whether behavior correlates well with the histomorphological variations seen in the lesion.

Materials and Methods: A total of 80 patients who were diagnosed either as cementifying fibroma CF, OF and cemento-ossifying fibroma (COF) of the jaws from the files of the Oral Pathology Department of the University of the Witwatersrand Dental School were retrieved and the histology slides of each case were reviewed with the most recent diagnostic criteria for OF and the authors additional criteria. A total of 56 cases that met the set criteria were analyzed.

Results: The patients were clustered within the third and fourth decades of life (*n* = 39, 69.6%). Majority of the patients were black (83.93%), whereas the rest were whites (12.50%) and Asians (3.57%). There were 17 males (30.4%) and 39 females (69.6%), giving a male to female ratio of 1:2.3. Most of the lesions (70.3%) occurred in the mandible, involving the premolar molar region (56.7%). Scanty fibrous tissues in highly cellular lesions were found in 36 (64.3%) of the cases. There were globular, dystrophic or granular calcifications mixed with irregularly shaped trabeculae of lamellar or cellular woven bone or osteoid were found, in 36 (64.3%) cases.

Conclusion: Demographic data, clinicoradiologic features, combined with histopathology will continue to be relevant in the definitive diagnosis of OF and in predicting its behavior. Highly aggressive lesions with shorter duration in people below 15 years were called juvenile OF and treated as such, while OF applies to other conventional ones.

Key words: Clinical-features, histomorphology, jaws, ossifying fibroma

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Introduction

Recent studies into the understanding of fibroosseous lesions have led to some publications, in an attempt to clarify some unresolved problems of diagnosis which have been the bane of improper classification. These lesions have some peculiar similarities, although there has not been disagreement as to the mode of treatment of individual lesions within the group. Ossifying fibroma (OF) is the most common of the neoplastic component of the fibroosseous lesions.[1] Most studies on OF stress the similarities in the histomorphologic appearances of this lesion compared with others in the group without specifying features peculiar to OF only. The considerable overlaps in the clinical and sometimes radiologic presentations of these lesions are also stressed to the "detriment of some aspects of demographic studies on OF."

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Biochemical analyses of organic protein components of bone and cementum have shown no differences in the amino acid sequence.[2] Ultrastructural studies by Damjanov *et al*.,[3] and Mincer *et al*.,[4] have shown that the globular calcifications correspond to extracelluar calcium hydroxyapatite deposits with some having osteocytes. Studies by Giansanti[5] using polarized light have not shown any differences between cementum and bone in these lesions. These findings attest to the similarity in the cemento-osseous components of these lesions which were either cellular or acellular cementum-like bone in the form of globules or trabeculae within a cellular fibrous stroma. The cells arising from the pluripotential stem cells of the periodontal ligament are said to be capable of differentiating into osteoblasts or cementoblasts.[6,7] Finding bone or cementum in the same lesion is therefore not an uncommon phenomenon or expectation.

This study aims at contributing to the definitive diagnosis of OF through the establishment of specific and objective histomorphological features reliable enough to label OF as a distinct lesion. The study also attempts at determining some demographic features of OF, common sites of occurrence and to determine whether behavior correlates well with the histomorphological variations seen in the lesion.

Materials and Methods

A total of 80 patients who were diagnosed either as cementifying fibroma CF, OF and cemento-ossifying fibroma (COF) of the jaws from the files of the Oral Pathology Department of the University of the Witwatersrand Dental School were retrieved. The histology slides of each case were reviewed taking into consideration the most recent diagnostic criteria for OF and the authors additional criteria. These additional criteria included: (a) Assessment of stromal cellularity, arrangement of the cells, presence or absence of mitoses, presence or absence of reactive giant cells of the mononuclear-monocytic origin, (b) presence of different histomorphological calcifications (c) presence of small strands of immature cellular or acellular osteoid and trabeculae of woven bone, (d) absence of fusion of cementum or bone-like calcifications or sclerotic masses (e) absence of fusion of metaplastic bone or cementum at the surrounding normal bone (f) presence or absence of compressed periosteum or attenuated cortical bone (as fibrous capsule are not usually recognized histologically).

To reduce subjectivity, the reviewers completed the procedures before looking into the clinical and radiographic presentations of the lesions together with the biopsy report issued at the time of patient presentation. Joint reviews by at least two observers were carried out to achieve unanimity. Finally the assessment was based on features of the histopathological slides at the time of review combined with those of issued histopathology report. After the histopathologic review, the clinical records were reviewed for patients' race, age, sex, clinical presentation, site of involvement, radiographic appearance, treatment modality and the presence or absence of recurrence. A total of 56 cases that met the set criteria were analyzed.

Results

Clinical findings

The patients age range from 9 to 72 years with clustering around the third and fourth decades of life (n = 39, 69.6%) [Table 1]. Majority of the patients were black (83.93%), whereas the rest were whites (12.50%) and Asians (3.57%). There were 17 males (30.4%) and 39 females (69.6%), giving a male to female ratio of 1:2.3 [Table 2]. There were 5 (8.9%) females consisting of 4 (7.1%) blacks and one (1.8%) white who had multiple site occurrences. Most of the lesions (70.3%) occurred in the mandible, involving the premolar molar region (56.7%) and the symphysis (15.62%). The maxilla accounted for 29.7% of the lesions, with 17.2% in the anterior and 12.5% posterior regions [Table 3].

All the patients presented with painless facial swelling which was firm to bony hard. The lesions were slowly enlarging with no associated history of trauma or any other agent ascribed to its cause. Radiographic presentation varied from case to case. The lesions presented as unilocular or multilocular radiolucency or mixed radiolucent and radio-opaque mass with root resorption or displacement of adjacent teeth. The lesions were generally well circumscribed radiologically.

Table 1: Age distribution of 56 cases of ossifying fibroma

Age group (years)	Frequency	Percentage
0-9	1	1.8
10-19	11	19.6
20-29	25	44.6
30-39	14	25.0
40-49	2	3.6
50-59	1	1.8
60-69	1	1.8
70-79	1	1.8
Total	56	100

Table 2: Racial and gender distribution of patients

Race	Male	Percentage	Female	Percentage	Total	Percentage
Asian	1	1.8	1	1.8	2	3.6
Black	14	25.0	33	58.9	47	83.9
Coloured	-	-	-	-	-	-
White	2	3.6	5	8.9	7	12.5
Total	17	30.4	39	69.6	56	100

Table 3: Site distribution of ossifying fibroma

Site	Frequency	Percentage
Maxilla		
Posterior	8.0	12.5
Anterior	11.0	17.2
Total	19.0	29.7
Mandible		
Body (molar/premolar)	35.0	54.68
Angle	-	-
Ascending ramus	-	-
Symphysis	10.0	15.6
Total	45.0	70.3

Total number of sites in maxilla and mandible=64 (%) due to multiple occurrences in some patients, The five patients with more than one quadrant involvement are: Black, male, 21 years (left maxilla+left mandible), black, female, 20 years (left and right mandible, right maxilla), black, female, 62 years (left and right mandible+right maxilla), black, female, 37 years (left and right mandible)

Histomorphological variation in OF

Reports on surgical exploration showed that the lesions were relatively avascular and well demarcated from the surrounding bone making it easy to shell out. Surgical specimens of large lesions submitted for histopathology were usually covered by a thin layer of cortical bone. The histology of the lesion consists essentially of a benign fibro-osseous proliferation. The fibrocellular stroma consists of spindle shaped to polyhedral cells with prominent basophilic nuclear material. Fibrous tissues were scanty in highly cellular lesions representing 36 (64.3%) of the cases. Moderately cellular lesions were 14 (25.0%) cases, while lesions with poor cellularity (more fibrous tissue) were 5 (8.9%) cases. Highly cellular lesions also show cellular pleomorphism, but cellular atypia or anaplasia and tissue necrosis were not noticed. Some highly cellular lesions showed a few mitotic figures usually the normal prophase type signifying a higher turnover of cells in those lesions. The cellular stroma in 15 (26.75%) cases were arranged in fascicular, tangled or storiform pattern [Figure 1]. 4 (7.1%) of the cases with storiform pattern were associated with rnultinucleated foreign body giant cells.

The mineralised component of this lesion showed a range of histomorphologic variations and when these are juxtaposed with the stroma component, five distinct histologic presentations were encountered: (a) Mixed calcification in which globular, dystrophic or granular calcifications were mixed with irregularly shaped trabeculae of lamellar or cellular woven bone or osteoid. This was the most common presentation occurring in 36 (64.3%) cases [Figure 2]; (b) predominantly globular (small, oval, round) mineralized bodies also referred to as psammomatoid bodies. These were present in 27 (48.2%) cases [Figure 3]. Majority of this group also belong to the mixed calcification; (c) predominantly trabeculae of bone or cementum-like material seen in 14 (25.0%) cases was also part of the mixed calcification; (d) predominantly osteoid but focally located were 6 (10.7%) cases; and (e) predominantly dystrophic or granular calcification were 4 (7.1%) cases. The highly

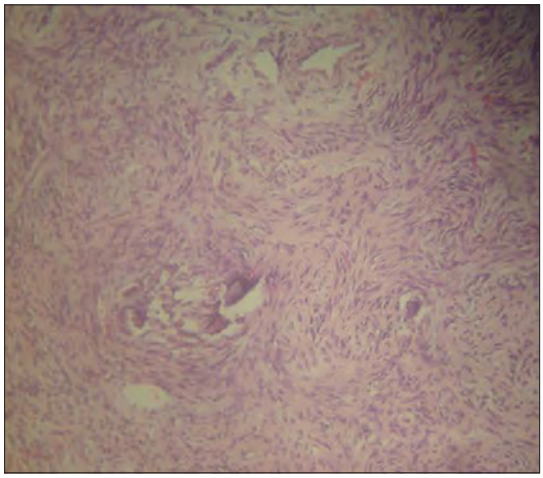

Figure 1: Ossifying fibroma with storiform stroma

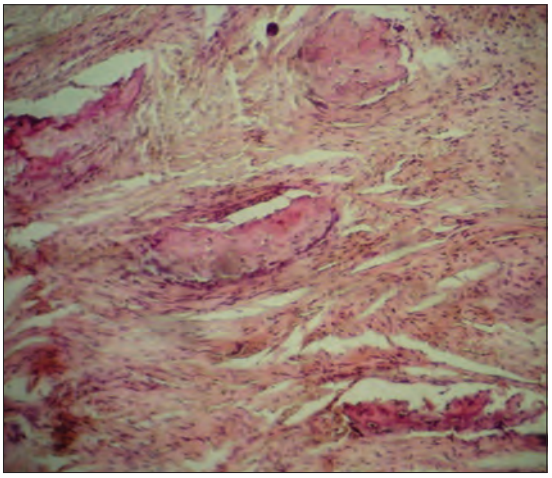

Figure 2: Ossifying fibroma with trabeculae of cellular woven bone

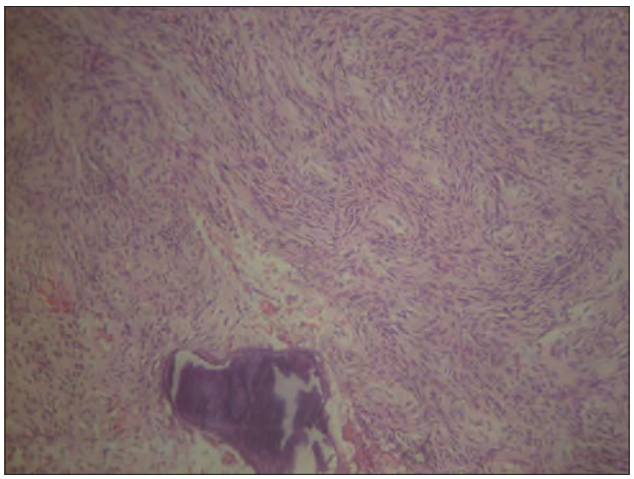

Figure 3: Ossifying fibroma with psammomatoid (globular) pattern of calcification

cellular lesions were seen commonly associated with globular calcifications and cellular woven bone. Twenty two (39.3%) of the 36 highly cellular lesions fell within the age group 20-29 years, which is the age range this lesion is commonly diagnosed, while 6 (10.7%) cases fell within 30-39 years and 5 (8.9%) cases fell within the age group 10-19 years. Of the latter, 3 (5.4%) cases fell within 10-15 years age bracket. Only one (1.8%) patient each fell within the age brackets 0-9, 40-49 and 60-69 years.

Average duration of cellular lesions before presentation ranged between 2 months and 1.5 years with an average of 8 months. Generally, stated duration of lesions for all cases ranged between 2 months to 4.5 years with an average of 1.5 years. Multinucleated giant cells were found in 19 (33.9%) cases and they were of two types. (1) Smaller diameter giant cells with fewer numbers of nuclei associated with trabeculae of bone, most of the latter with osteoblastic rimming; (2) Larger diameter giant cells with many nuclei, scattered or in focal collection in the stroma especially in lesions associated with aneurysmal bone cyst. There were 4 (7.1%) lesions associated with cystic degeneration. Three (5.4%) cases were associated with aneurysmal bone cyst found in 2 black females at ages 13 and 25 years and a white female at 15 years of age. One (1.8%) case with simple bone cyst was found in a black male at 20 years of age. Recurrences were recorded in 2 (3.6%) cases; a white female at 9 years of age and a black female at 20 years of age. The lesions were originally diagnosed as juvenile OF and the recurrent lesions had same histopathology as were originally diagnosed.

Discussion

There is considerable controversy surrounding the definitive diagnosis of fibro-osseous lesions and benign OF in particular because of confusion in terminologies and lack of acceptable criteria for discrimination between the lesions. OF is the most common benign neoplasia among the fibro-osseous lesions of the jaws.[1] The nature and proper nomenclature for this lesion have been controversial until recently. The nature of the calcified material in the lesion has been the real subject of controversy. This has led to several workers attempt at distinguishing between bone and cementum like material, the two main calcific materials in the lesion. The lesions were previously labeled cementifying fibroma or OF depending upon the predominant calcified material present in the lesions.

Biochemical analysis,[2] Ultrastructural studies[3,4] and polarized studies[5] did not show any differences between cementum-like material and bone in these lesions. The failure of these studies to show differences in the structure of bone and cementum-like material, coupled with the fact that such lesions occur in extragnathic bones,[6-12] led to the general agreement that fibro-osseous lesions containing bone and cementum-like material are a spectrum of the same lesion. Kramer *et al*.,[7] in previous World Health Organization (WHO) classification (1992) of Odontogenic tumors grouped lesions containing either cementum-like material or bone in a fibro-cellular stroma as COF. Current WHO classification (2003)[13] recognizes all variant of COF as OF thereby putting to rest the controversies.

Our study though limited has come out with some interesting findings in some aspects of demographic and histomorphologic outlook of OF which previous authors have not reported. Our patients' age range from 9 to 72 years with clustering around the third and fourth decades of life, which is similar to findings by most authors.[14-17] The majority of our cases were black accounting for 83.93% while the rest were whites (12.50%) and Asians (3.57%). This finding is different from those of Hauser *et al*.,[16] which showed a predilection in white Americans even though the percentages were not stated. Although patient turnover in our laboratory is skewed toward blacks, an adjustment for population differences may not produce a significant difference from the above figure.

The average waiting period before presentation was 8 months for cellular lesions and 1.5 years for all lesions put together. This delay in presentation may be due to the gradual growth of most of this lesion coupled with production of few signs and symptoms. The duration of these lesions and presumably the doubling time in terms of growth are variable and unpredictable. There was female predilection (male to female ratio of 1:2.3) for lesions in this study. This is similar to finding by Neville and Albenesium,[6] Waldron,[15] Wenig,[18] and Cawson *et al*.[19] 5 (8.9%) of the females had multiple site occurrences but no history of familial occurrence was elicited in any of the patients. Multiple OF should be differentiated from focal cemento-osseous dysplasia, which also occurs predominantly in black females in the 6th and 7th decades of life and histologically look alike at some stage in their development. Large coalescing trabeculae of cementum like material or osteoid when present precludes the diagnosis of the OF.[1,14,19-21]

Most of the lesions (70.3%) like findings elsewhere by Sciubba and Younai ,[22] and Wenig[18] occurred in the mandible of which majority occurred in the molar premolar region (56.7%), with the symphysis accounting for only 15.62% of the cases. The maxillary lesions (29.7%) occurred mainly in the anterior region (17.2%), while 12.5% cases were found in the posterior maxilla. Anterior maxillary occurrences were not commonly reported. No lesion was found in the angle and ascending ramus of the mandible in this study, attesting to the fact that lesions of the jaws are found in the tooth bearing regions.[12,19,22-24]

There are histomorphological variations in tissues submitted for diagnosis in our cases but essentially consist of a benign fibro-osseous proliferation. From the histological findings in this study, lesions of high cellularity were

found associated with less number and smaller globules of calcification (psammoma bodies) and cellular woven bone. These cellular lesions had shorter duration at presentation. Dehner[25] in his discussion on fibro-osseous lesions stated that lesions diagnosed as OF produced less bony destruction than the cementifying fibromas and that two cases of the latter required subtotal maxilloectomies to control the disease, The above agrees with our findings that cellular lesions which are much more associated with psammomatoid calcifications behave more aggressively than lesions associated with predominantly trabeculae of bone. Where psammomatoid cellular lesions now occur in weak and vascular maxilla compared with the mandible, bone destruction becomes more progressive. These factors must be borne in mind in the management of these lesions, OF's growth like all neoplasia will depend upon its own secreted growth factors which also depend upon its degree of cellularity, as the supporting fibrous stroma do not partake in tumor growth. It could be safely stated that lesions of high cellularity in both the fibro-cellular stroma and woven bone coupled with a history of short duration may be seen as reflective of a likely aggressive behavior and treated as such. OF may then be appropriately described as a benign lesion which are locally aggressive and show local invasion and may recur if not adequately excised but do not metastasize.

The histological pattern of spindle cell proliferation in a storiform, tangled or radiating cartwheel pattern characteristic of benign fibrous histiocytomas were noticed in 26.8% of our cases, although accompanying macrophages or histiocytes in fibrous histiocytoma. Unlike the latter OF are well defined lesions. Furthermore the presence of multinucleated giant cells in OF may lead to confusion with the malignant fibrous histiocytoma but unlike the latter inflammatory and angiomatoid cells are not encountered.[18]

In conclusion, demographic data, clinical presentation including radiology combined with histopathology will continue to be relevant in the definitive diagnosis of OF and in predicting its behavior. From our findings in this study, we associate with the thinking of Wenig *et al*.,[9] that the terms juvenile active OF and the so called aggressive active OF be jettisoned. We feel that all lesions continue to grow even though at different rates, as a result they are all active. There is no way a period of tumor inactivity can be accurately predicted. Those that behave aggressively in a shorter duration could still be called aggressive-OF irrespective of the age at presentation, since aggressive behavior is not age related, as it could develop in both young and old patients. This is consistent with the recent report of Cabibi *et al*.[26] that the "early" age of onset should not be included among the essential characteristics of OF with a high risk of recurrence. Similarly, highly aggressive lesion with shorter duration in people below 15 years could still be called juvenile OF and treated as such, while OF applies to other conventional ones.

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