Cemento-ossifying fibroma occurring in an elderly patient. A case report and a review of literature

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Abstract: Cemento-ossifying fibromas (COF) are benign lesions affecting the jaws and other craniofacial bones. They commonly affect adults between the third and fourth decade of life. Radiographically, they appear as well-defined unilocular or multilocular intraosseous masses, commonly in the premolar/molar region of the mandible. The lesion is invariably encapsulated and of mixed radiolucent densities. The tumour may grow quite extensively, thus the term aggressive is some times applied. Their clinical, radiographical and histopathological features and those of fibro-osseous lesions are overlapping and may cause confusion in classification, diagnosis, and treatment. The histopathology is composed of fibrous tissues with calcified structures resembling bone or cementum. Surgical enucleation or resection is the treatment of choice. They are insensitive to radiotherapy and recurrences are uncommon. This case report presents a case of COF in 70 years old female patient that was asymptomatic. Clinically, there was an expansion of the buccal plate but not the lingual plate of the right mandible. The covering mucosa was normal and there was no tenderness or paraesthesia. Radiographically, the lesion extends superio-inferiory from the alveolar ridge to the area of inferior dental canal and mesiodistally from the premolar region to the retro-molar area. The lesion was of mixed radiolucent densities. The patient was followed up periodically for 5 years without any treatment. The patient continued to be asymptomatic with minimum changes. Occurrence of cemento-ossifying fibroma in patients over 60 years of age is unusual and had not been reported. The clinical, radiographic, histopathology and literature review are discussed.

Key Words: *Cemento-ossifying fibroma in elderly patient, Non-aggressive cemento-ossifying fibroma, Mandible.*

Introduction

Fibro-osseous lesions are a hetero-geneous group of benign lesions of unknown aetiology affecting the jaws and other craniofacial bones. Lesions in this category include fibrous dysplasia (FD), focal cemento-osseous dysplasia (FCOD) and cemento-ossifying fibroma (COF) [1]. The group often exhibit resemblance in clinical presentation, radiographic appearance and histological criteria, therefore, pose difficulties in classification, diagnosis and management [2]. COF is the most frequent fibro-osseous lesion encountered by oral pathologists and perhaps, it has more synonymous than any other jaw lesion [3]. Unlike FD, COF is considered as an osteogenic neoplasm manifesting as slow-growing, asymptomatic, well-defined unilocular or multilocular intra-osseous masses. The lesion is commonly seen in the premolar-molar region of the mandible. Infrequently, it may involve the jaws bilaterally or multiple quadrants [4-6]. Although, the tumour is of slow growing type, it may grow quite extensively and may even provoke mandibular fracture [7].

Radiographically, the COF presents as a well-defined unilocular or multilocular lesion with smooth contours. The maturity of the lesion will determine the degree of radiopacity. The immature lesion may present as completely radiolucent, whereas the mature lesion may appear completely radio opaque. Nevertheless, majority of the lesions demonstrate varying degrees of radiolucency.

Histologically, the COFs are well circumscribed, occasionally encapsulated, consisting of cellular fibrous tissues and thin isolated trabeculae of bones. The bone may show osteoblastic rimming and spherical deposits of calcified material, which are relatively acellular resembling cementum.

In some cases, the calcified materials predominate the tissue and such lesions are designated as psammomatoid ossifying fibromas, from a Greek psammos sand [8]. The histopathological features of COF are difficult to distinguish from those of FD, but the distinction is mainly relied on age and radiographic features. Discriminating FCOD from COF is important, as the former is a reactive lesion, whereas the later is neoplastic in nature. The pathologic nature of the two lesions is not yet clear and histopathologically difficult to differentiate. However, Su and associates [9] established a set of histopathologic features that can assist in discrimination, but still emphasizes on the significance of the clinical and radiographic features as an adjunct in making proper diagnosis.

The peak incidence of COF is the third and fourth decades and is more prevalent in white than black racial groups [9]. Female predilection has been reported as high as 5:1 [10-11].

Probably the close proximity and similarity to the periodontal ligament tissue have led to the assumption that the COF and FCOD are of periodontal tissue origin [4,10], thus the term periodontoma some times is applied [12]. There is, however, no proof to support this theory and their occurrence in areas distant from periodontal ligament remains unexplained [13].

In the literature, terms such as ‘aggressive', or ‘ juvenile’ are used to describe cases showing rapid and wide bone destruction [3,14]. However, at

present time, there are scanty evidences that may delineate the aggressive from non-aggressive COF cases. Complete removal of the tumour mass, using enucleation or surgical resection is the treatment of choice.

COF is usually well circumscribed and this facilitates its extirpation from the surrounding bone. In certain cases, bone grafting is essential to correct the defect. Radiotherapy has been proven ineffective and is contraindicated [15]. However, the treatment is governed by several factors, such as location, extent and size of the lesion. Recurrences are uncommon but it has been described [10, 16].

Case Presentation

A 70-year-old Caucasian, female patient was referred by her general dental practitioner for a bony swelling in the right mandible. The past dental history revealed presence of the swelling around 44-47 region of 3 years duration. Apart from the father who died of liver cancer, the medical, social and family histories were unremarkable.

Oral and maxillofacial examination revealed a large bony swelling of the right mandible. The covering skin showed no signs of inflammation. The regional lymph nodes were palpable but not enlarged. Intra-orally, there was a marked, bone expansion of the buccal plate extending from the position of the first premolar to the retro-molar region but no lingual expansion. The associated soft tissues were slightly swollen but there was no ulceration or fistula formation. On palpation, the swelling was bony hard in consistency but no tenderness or paraesthesia. Radiographically, the orthopantomograph showed a large radiolucent lesion (approx 5x3 cm) extending from the first premolar region to the third molar region. Interpretation of the recent and the past radiographs (approximately 3 years earlier) revealed gradual expansion of the lesion. The lesion was well demarcated with sclerotic border and heterogeneous in contrast. The lesion extended mesiodistally from the area of first premolar to the position of the third molar area and superioinferiorly from the crest of the edentulous alveolar ridge to the area beyond the inferior alveolar canal, just above the inferior cortex of the mandible (Figure 1A).

The superior alveolar ridge is slightly elevated. No evidence of cortical erosion was noticed, though difficult to judge in orthopantomograph view. The radiographic differential diagnosis included odontogenic keratocyst, odontogenic myxoma, cemento-ossifying fibroma, focal

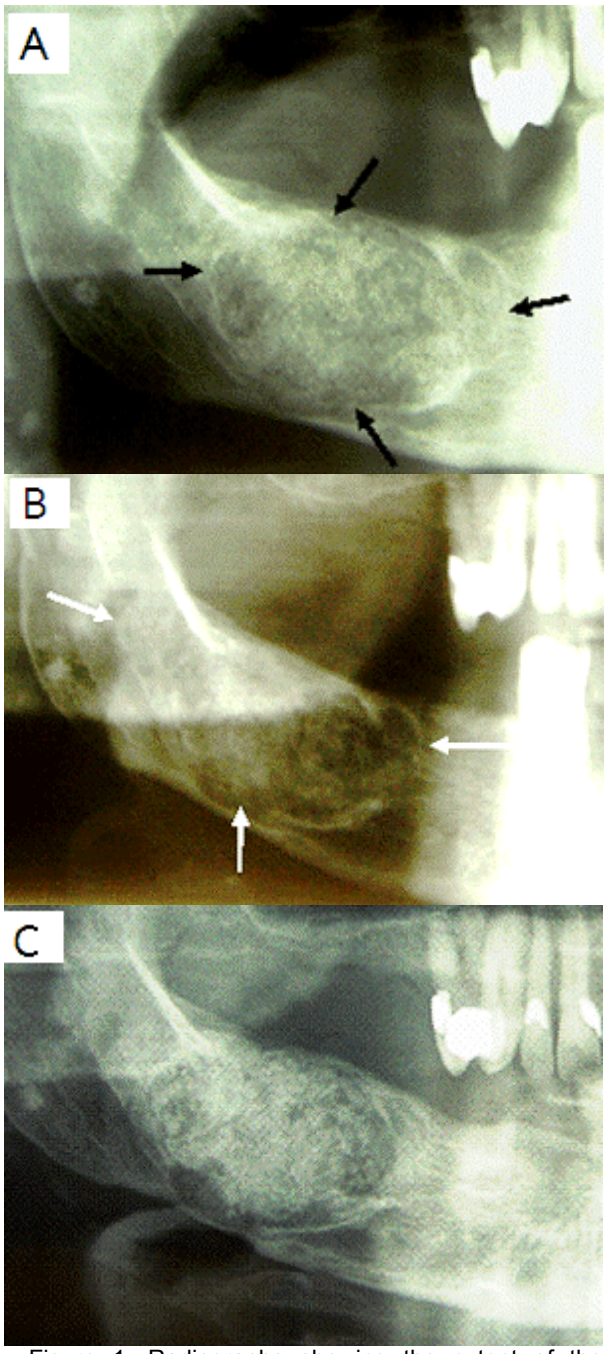

Figure 1: Radiographs showing the extent of the lesion (arrows). A, the lesion at the initial presentation; B, the lesion after 3 years, C, a recent view.

cemento-osseous dysplasia and central giant cell granuloma

A biopsy was performed under local anaesthesia and the specimen was sent for histopathology examination.

The histopathology report revealed presence of whorled fibrous tissue, containing calcified masses of bone/cementum and regular fibroblasts, which showed no mitotic figures (Figure 2, A&B).

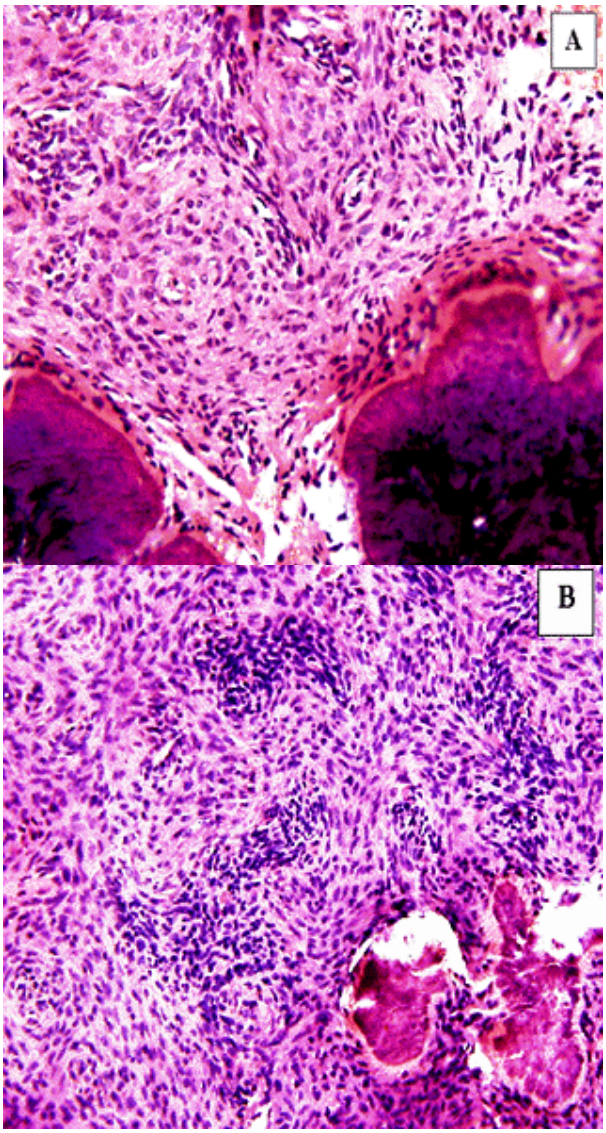

Figure 2: Photo-micrographs (A&B) showing whorls of fibrous tissue and calcified material (cementum) consistent with cemnto-ossifying fibroma (Original Mag. x40 & x25 respectively).

Cutting decalcified fragments caused artifact and provided no information. The clinical and histopathologic features were consistent with cemento-ossifying fibroma. Surgical removal of the lesion was advised, the patient, however, was not keen for its removal. Therefore, the patient was kept under periodic review with yearly x-ray to monitor the lesion expansion. Three years later, the radiographic examination demonstrated evidence of continued slow expansion of the lesion (Figure 1B). The radio-opacity of the lesion has markedly increased. There was buccal and slight lingual expansion of the bony plates. No soft tissue changes or paraesthesia were noticed. The patient continued to be asymptomatic and happy to live with the lump. On reviewing the case a year later, no major clinical or radiographical changes were noticed. However, visual comparison of the former and the recent orthopantomographs showed increased expansion in superior-inferior direction (Figure 1C), but the lesion remained well demarcated and no evidence of invasion.

Discussion

The cemento-ossifying fibroma is a benign osseous tumour that commonly affects adults of middle age, 30-40 years [17]. The mandible is the common site, though the lesion may involve multi-quadrants [18]. In some cases, initial symptoms are present [9]. The clinical scenario and age of the present case are of particular interest. Occurrence of COF at age of 70 years is unusual. Su and colleagues [9] reviewed clinical details of 75 cases of COF. The mean age was 32 years (range, 10-59 years). The authors reported that COF is not seen in patients over 60 years of age and is detected 10 years earlier than FCOD.

We had no reason why the patient was not referred to a specialist three years before attending our clinic. It appears that the lesion was quite small at the initial presentation and certainly, COF was not among the differential diagnosis list of the dentist. COF may present as small radiolucent areas close to the apices of teeth that could be mistaken for periodontal pathology [17]. What is common in both cases is the lack of symptoms and non-specific radiolucency (the absence of intra-lesional calcifications). COF may manifest as a lesion resembling stafne's bone cyst, central giant cell granuloma, residual cyst or odontogenic keratocyst. These lesions are relatively uncommon and unlikely encountered by the general dental practitioner. During the initial 3 years, before the correct diagnosis was made, the tumour had grown quite considerably. Nevertheless, the rapid growth was followed by a slow phase of growth in the following years. It is understandable that proper determination and monitoring of changes in a bony lesion would require different radiographic views, such as computerized axial tomography (CAT scan) as well as standardisation of the techniques.

However, due to absence of clinical signs and symptoms and reluctance of the patient to surgical treatment, it was not possible to justify invasive and expensive investigations. Our interpretation was subjective and based on visual comparison of the available orthopantomographs. The radiographic appearance of COF is invariably a mixture of radiolucency. Nevertheless, the recent orthopantomograph view revealed that the lesion was more radio-opaque in appearance. Possibly, the tumour was undergoing maturation and

ossification phase accounting for the increased density [19]. In addition, rapid followed by slow tumour growth may elucidate that the tumour was passing through a dormant phase. Nevertheless, this remains only a speculation. Since the actual lesion was not removed surgically, description of its histological characters would be difficult. Generally, COF characterised by a well-defined expansile bony mass and rarely associated with destruction or extraosseous soft tissue components [19]. Unlike the COF, the juvenile or aggressive (ACOFs) grow massively with extensive cortical expansion [3,10,20]. There are no histopathologic criteria that are predictive of aggressive behaviour or tendency for recurrence. Nevertheless, Zupi et al. [3] reported two features that may help in distinguishing ACOF from COF. Firstly, clinically, the ACOF occurs at a far lower mean age than the COF. Secondly, the histological pattern of the ACOF seems to be unique in being highly cellular with entrapped osteoblasts. Detailed histopathology of our case was unfeasible. However, considering the age, radiographic appearance and the growth behaviour, our case would clearly fit under the COF category, the non-aggressive type.

To the best of our knowledge and review of the literature, no COF cases of this age were reported.

The several years of follow up demonstrated a slowly, non-aggressive growing tumour, yet with no signs of soft tissue or vital structures involvement. Considering the age of the patient, this case might pose a dilemma to clinician and surgeon as whether to enucleate the tumour surgically or just monitor periodically would be enough. In fact, clinical and radiographical follow up of cases presented without disfigurement; dysfunction or pain has been advocated [21]. Long follow up of ossifying fibroma (more than 3 decades) without any problem or exacerbation has been reported [22]. Furthermore, the potential transformation of fibro-osseous lesions into sarcomas has only been suggested but not clearly reported.

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