CASE REPORT

Maxillary Cemento-Ossifying Fibroma In A Male Patient With End-Stage Chronic Renal Disease

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Abstract

Aims & Objectives: The cemento-ossifying fibroma (COF) is a benign fibro-osseous lesion with controversial features that occurs most frequently in the mandible of female individuals. The presence of chronic renal disease (CRD) is a predisposing factor for the occurrence of soft and hard tissue lesions in the oral cavity. The aim of this report was to describe clinical, radiological and histopathological characteristics of an unusual case of COF who was also presented with CRD. **Case Description:** A 41 year-old male patient undergoing chronic hemodialysis was referred because of the painless, slow growing, firm mass in the maxilla. Cone Beam Computed Tomography disclosed a well-circumscribed lesion containing numerous ring-shaped and curvilinear ossified nodules in the left premolar-molar region. Patient was operated under local anesthesia. The histopathological examination revealed irregularly shaped bone and cementum-like hard tissue calcifications contained within hypercellular fibrous tissue stroma, leading to a final diagnosis of cemento-ossifying fibroma. **Conclusion:** The clinical characteristics and radiological appearance of maxillary COF is unspecific and its relapse potential is unpredictable. A reliable classification at the cellular level is required to support evidence based clinical decisions, particularly in the presence of systemic conditions such as CRD.

KEYWORDS: cemento-ossifiying fibroma, chronic renal failure, maxilla, cone beam computed tomography, fibro-osseous lesions

Introduction

The oral cavity is a common site for the occurrence of bone-related lesions with different histopathological features. The cemento-ossifying fibroma (COF) is an uncommon benign fibroosseous lesion composed of fibrocellular component and calcified materials like cementum and woven bone [1]. This lesion can be diagnosed accidentally in radiographs or may present itself as a slow growing sub-periosteal mass, occasionally causing pain, swelling and teeth displacement. The true pathogenesis still remains unclear. COF is more frequent in the mandible and definite female predilection has been reported [2].

The soft and hard tissues of the oral region may become susceptible to the development of pathological growths in case of some particular systemic conditions. As such, the chronic renal disease (CRD) can be defined as the progressive and irreversible decline in kidney functions and up to 90 % of patients having renal insufficiency may present with oral signs and symptoms in soft and hard tissues either as a result of the disease itself or the treatment protocol [3]. Halitosis, altered taste and sensation, changes in salivary secretion, bone demineralization, loss of lamina dura, deep periodontal pockets and poor oral hygiene are often associated with this condition, based on individual predisposition and the severity of the disease [3,4].

The knowledge of bone related oral lesions in CRD patients is limited. The aim of this report is therefore to present a case of COF who was also suffering from CRD and to evaluate clinical characteristics of these conditions.

Case Description

In January 2010, a 41 year-old male patient has been referred to the Faculty of Dentistry from a nearby private nephrology and hemodialysis clinic for focal infection screening procedure and he was redirected internally to the Department of Oral Surgery. Patient's history revealed that he has been suffering from end-stage CRD for six years and he was a kidney transplant candidate from cadaveric donor. He has been undergoing hemodialysis three times a week and he had hypertension secondary to the declining kidney functions, which is controlled by constant medication. He was a heavy smoker consuming up to 35 cigarettes per day and he reported having previous licensed boxing career lasting for 15 years from which he had quitted eight years before the appointment. Written consultation and informed consent forms were obtained from the physician and the patient respectively, before surgical procedure.

The patient's extraoral features were noncontributory, apart from slightly yellowish skin tone which can be considered characteristic in individuals with CRD. In the intraoral examination, the oral hygiene status was subjectively classified as poor; based on the calculus depositions, deep periodontal pocket formations, recessions and the presence of untreated caries. There was a prominent soft tissue enlargement with no visible attachment on the left maxillary segment which is localized on the facial aspect of the premolar and molar regions. The patient had noticed the lesion six months before the appointment. The teeth in the area were moderately mobile. The lesion measured approximately 3 cm in the anterior-posterior direction and it was palpable for 4 cm vertically. The color of the mucosa was normal and no fistula or purulent discharge was noted. It had a rubbery consistency and was tender to firm pressure (Fig. 1). Radiographic examination with orthopantomogram and cone beam computed tomography (CBCT) revealed a well-delineated hard tissue mass localized on the facial aspect of the left maxillary canine to the first molar teeth. The lesion's border inside the maxilla was not discernible from the neighboring hard tissues. In contrast, its outer contours presented a well-circumscribed, thin and semi-circular radioopacity which has similar density to the bone. Internal composition of this hard tissue mass was heterogeneous, consisting of both radiolucent areas and numerous ring-shaped and curvilinear ossified nodules (Fig. 2). Based on these unusual radiographic and clinical findings, incisional biopsy was performed revealing histological characteristics consistent with benign fibro-osseous lesion.

The surgical intervention was scheduled the day after the dialysis session in the morning hours. The systolic and diastolic blood pressures were measured 130 mmHg / 80 mmHg immediately before the surgery. The patient was operated under local anesthesia. Following the incision and mucoperiosteal flap removal, the lesion was revealed as tiny irregular granulated fibrous tissue and pieces of bone-like immaturely calcified material on the facial aspect of maxillary cortex (Fig. 3). Teeth associated with the lesion were extracted and curettage was performed to remove all fragments which were firmly attached to the underlying bone structure (Fig. 4). The wound was closed with silk sutures. Antibiotics and non-steroidal anti-inflammatory drugs were prescribed by taking account of patient's CRD. The histological examination of the specimen revealed irregularly shaped bone and cementum-like hard tissue calcifications contained within hypercellular fibrous tissue stroma consisting of immature mesenchymal cells (Fig. 5). The definitive diagnosis was COF. The healing period was uneventful and ten days later the sutures were removed. The patient was clinically and radiographically symptom-free at the 12 months follow-up.

Discussion

The COF is a benign fibrous neoplasm that contains different types of calcified materials such as bone and cementum that occurs more frequently in women with reported male to female ratios ranging from 1: 1.6 to 1: 5 [2,5,6]. Both jawbones can be affected, although 62 % to 89 % of the lesions arise in the mandible, involving premolar and molar regions in 77 % of cases [5]. They are mostly diagnosed between second and fourth decades of life [1]. The lesion in our case can be considered as rare and atypical based on its anatomical location and gender occurrence. The histopathological and clinical similarity to other fibro-osseous lesions such as fibrous dysplasia, periapical cemental dysplasia, cemento-osseous florid dysplasia often creates confusion in terminology. A typical COF consists of multiplied fibroblasts in storiform arrangement that produce collagen fibers. The stroma of the tumor contains bone spherules as well as cementum granules in varying size and number. The bone in COF is the woven type with peripheral lamellar maturing. On the surface of the bone spiculae, there are inactive osteoblasts focally [1,7-9].

The pathogenesis of COF is controversial. Earlier concepts relied on the cementum, lamellar bone and fibrous tissue producing capacity of the pluripotent cells of periodontal ligament [10]. However, lesions with similar histopathological features have been reported in other craniofacial structures and in long bones which do not contain cementum [11]. In addition, bone and cementum are both originated from mesenchymal stem cells. Accordingly, the classification of the World Health Organization suggested the term ossifying fibroma and considered it as a non-odontogenic neoplasm [12]. The connective tissue reaction triggered by local trauma had also been speculated among possible causes for the development of COF [13]. In our case, although the boxing career reported by the patient can be considered as a source of chronic trauma to the maxillofacial bones, it was not possible to identify a major predisposing factor because of the unexplained nature of this lesion and the simultaneous presence of CRD. Renal secondary hyperparathyroidism is a common complication of CRD

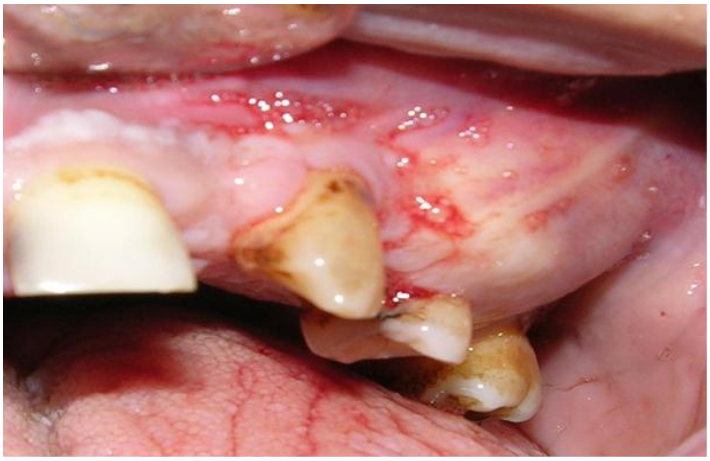

Figure 1. Intraoral view of the lesion in the left maxillary segment, note the normal color of the mucosa and the firm mass without soft tissue attachment.

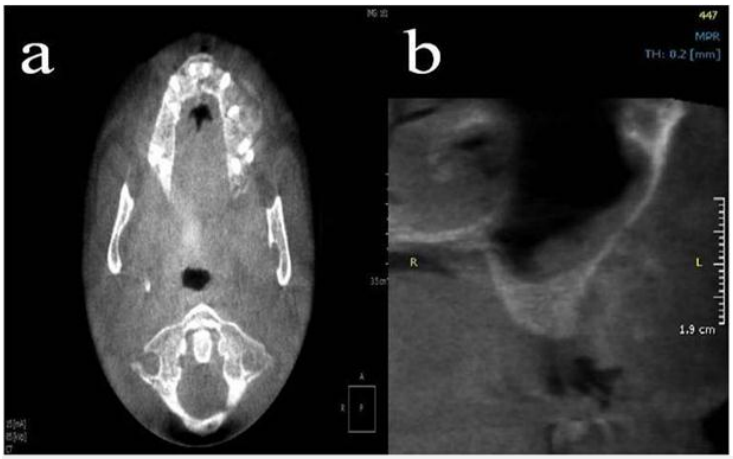

Figure 2. Cone beam computed tomography from the axial (a) and coronal (b) planes showing well-defined borders of the lesion that contains numerous ring-shaped and curvilinear ossified nodules.

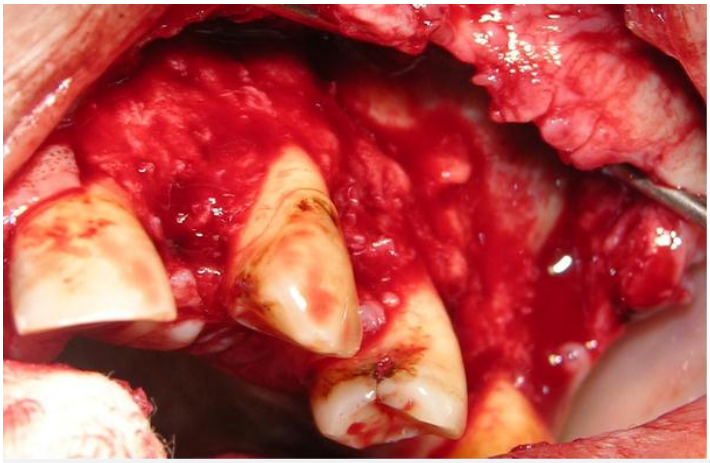

Figure 3. Intraoperative view of lesion: irregularly shaped calcified tissue fragments on the facial aspect of the premolar region.

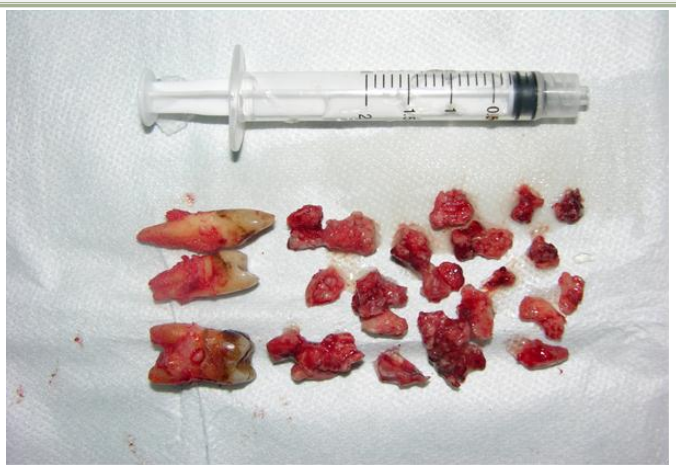

Figure 4. Surgical specimen collected during the enucleation and curettage.

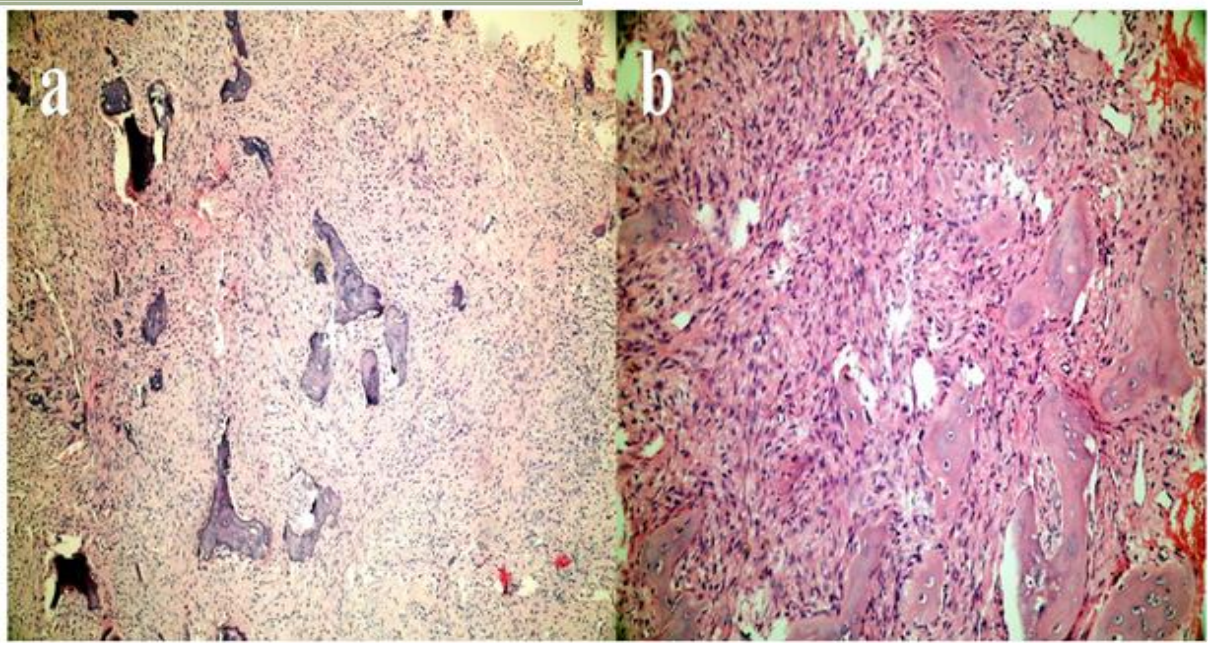

Figure 5. Bone spherules and cementum-like hard tissue calcifications contained within hypercellular fibrous tissue stroma consisting of immature mesenchymal cells (H&E staining X100 (a) X200 (b)).

characterized by increased endogenous levels of parathyroid hormone (PTH) [14]. The ossifying fibromas and hyperparatyroidism can also be observed in hyperparathyroidism-jaw tumor syndrome, in which the patients suffer from familial parathyroid adenomas, ossifying fibroma of the jaws, renal cysts and Wilms tumors [11]. The HRPT2 gene has been found to be mutated in this syndrome and it inactivates the parafibromin protein which has anti-proliferative properties. More recent studies have found perturbations in this gene among non-syndrome associated ossifying fibromas of the jaws [15]. Our patient's PTH levels were normal and no sign of paratyroid adenoma was found. Therefore, a possible relation between CRD and the COF remains to be elucidated.

A well-differentiated osteolytic image with easily distinguishable borders from the surrounding bone and solitary radiolucency with radiopaque foci are the most common radiological aspects of the COF [6,7,9,16]. Root displacement and resorption may be observed, probably related to the size and location of the lesion [16]. Growing COFs in the mandible often cause displacement of the inferior alveolar canal without destruction, thereby leading to the thinning or bowing of both vestibule and lingual cortices [17]. Trijolet et al. [2] suggested a relation between the maturation process and radiographic appearance of the lesion. According to this, the COF can be radiolucent at its earlier stages and it can even present a pseudocystic appearance. As the lesion grows, thin opacities having lower density than the surrounding bone which may be discovered at the center of the lesion becomes more irregular and they are usually circumscribed by dense spherical borders comparable to an eggshell. This process may further develop to the total opacity of the lesion in fully matured COFs. Because of this transition pattern the COF has been attributed a variety of descriptions such as "well-circumscribed radiolucency", "mottled radioopacity", "cotton wool" and "ground glass" [6,9,16]. On the other hand, Kuta et al. [5] have pointed out that the maxillary lesions tend to display a more immature appearance than those found in the mandible and no reliable pattern had been defined to distinguish between maxillary and mandibular lesions. In addition, the COFs occurring in the maxilla may be more aggressive and progress unnoticed, particularly when their growth pattern is not hampered by anatomical structures as in the mandible. Although the overall pattern of calcification in our case is consistent previous studies, we should note that the lesion had protruded laterally from the maxillary bone and this component was easily recognizable whereas its intrabony part shares similar density with the surrounding bone making it difficult to distinguish its borders. Majority of the reports on COF consist of cases occurring in the mandible which had been examined with CT. In contrast, our case is a rare example of COF visualized by CBCT that uses a cone shaped beam source and low dose of radiation. Therefore, the differences with medical CT scanners in defining the limits of the lesion in the maxillary segment could be related to the reduced X-ray dose and its subsequent absorption [18,19]. In addition, since the lesion was more prominent in the initial axial images when compared to multiplanar reformatted coronal slices, the usage of minimal secondary reconstruction intervals should be considered for the evaluation of COFs and similar fibro-osseous lesions in CBCT.

Complete removal of the lesion either by enucleation, curettage and resection with or without bone grafting is the preferred method of treatment for COF. A wide range of overall recurrence rates have been reported such as less than 1 % to 63 % [7] and 0 % to 28 % [2,5,20] in mandibular lesions. The relapse potential of maxillary COFs is largely unknown but it is suspected to be higher because of the greater difficulty of their removal and larger size at the time of presentation; in contrast to mandibular lesions which usually "shell-out" from their bony bed during the surgery. Fortunately, even in the larger cases which require radical excision of the neighboring structures, this lesion does not usually represent infiltrative pattern into bone; therefore, minimal surgical margins less than 1 cm has been suggested in these cases [7]. Such findings emphasize unusual features of the COFs and also raise questions concerning possible presence of different entities under the same classification.

Although being treatable by surgery, the clinical characteristics and radiological appearance of maxillary COF is unspecific and its relapse potential is unpredictable. Pathological examination is the only valid method for definitive diagnosis. Therefore, further studies should be focused on establishing a reliable classification at the cellular level to support evidence based clinical decisions, particularly in the presence of systemic conditions such as CRD.

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