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Clinical management of cementifying fibroma: A case report and pertinent review of the current literature

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Abstract

Cementifying fibroma is an uncommon neoplasm composed by varying amounts of cementum, bone and fibrous tissue. As a results of having similar histolological features based on inactive-looking odontogenic epithelium embedded in a fibrous stroma, it is often hard to differentiate from other fibro-osseous lesions such as fibrous dyplasia and calcifying odontogenic tumor. At this point, it is undoubtedly clear that proper radiological and clinical diagnosis play a great role together in identification. We present a rare case of this entity along with a number of clinical and radiographic features that set it apart from other pathologies mimicking fibro-osseous lesions.

Key words: cemento-ossifying fibroma, odontogenic tumor, neoplasms, cone-beam computed tomography, pathology

Introduction

Fibro-osseous lesions (FOLs) were first included as a lesion group among odontogenic and maxillofacial bone tumors in the *Classification of Head and Neck Tumours*, published by the World Health Organization (WHO) in 2017. FOLs of the craniofacial complex comprise a subgroup of benign tumors that differ morphologically, clinically, and radiographically. There are three recognized types of FOLs; fibrous dysplasia (FD), cemento-ossifying fibroma (COF), and cemento-osseous dysplasia (COD). While FD can occur anywhere in the skeleton, COF and COD are found solely in the maxillofacial bones [1].

Cemento-ossifying fibroma (COF) is uncommon, benign, mesenchymal odontogenic tumors growing slowly out of the periodontal ligament, comprising of a layer of fibrous connective tissue that encircles the root part of the tooth [2]. The lesion is characterized by multipotential cells that are prone to forming cementum next to lamellar bone and fibrous tissues. Histologically, it exhibits variable quantities of cement clusters embedded in the fibrous tissue with regions of splintered and disorganized bone fragments [3].

Reported and prevalence series data indicate that women specifically aged between 30 and 40 years are more likely to have the lesions than men. COF is more common in the mandible (70% frequency) than in the maxilla and posterior regions are reportedly exposed to higher risks than anterior ones [3]. Clinicians occasionally identify a lesion via an orthopantomogram during a routine dental checkup. The lesions can be defined with various degrees of opacification depending on multilocular or unilocular mixed radiolucent and radiopaque masses with marginal sclerosis [4].

A COF is generally a spherical or egg-shaped, slowly expanding mass that affects the teeth-bearing areas, resulting in root resorption of varying degrees on adjacent teeth. Although the origin of COF is still debated, it is considered that traumatic injuries or local irritants are possible reasons for their occurrence [5]. In the present study, we discuss the case of a 23-year-old male who was referred to the oral surgery department just before he was about to be conscripted into the military. The report also aims to discuss COF as a distinct type of ossifying fibroma and rare tumor and conduct a literature review on the current discussion about whether a new category of ossifying fibroma of non-odontogenic origin should be designed. The objective of this case report was to guide the clinicians in diagnosing and managing of a COF case.

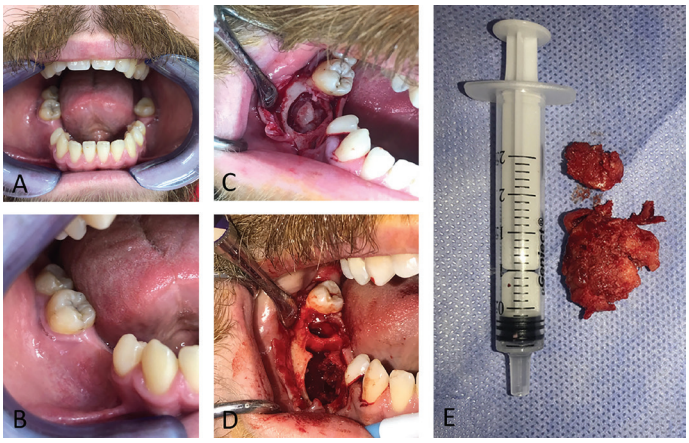
Case presentation

A 23-year-old male attending a private dental clinic for a routine dental check-up was referred to the oral surgery department for an abnormal radiolucency in the right mandible. He did not describe any of the five cardinal signs of inflammation, such as rubor (redness), calor (heat), tumor (swelling), dolor (pain) and functio laesa (loss of function). There was a history of a prolonged and painful healing period following the extraction of tooth no. 46

dating back to 4 years ago. He had no drug allergies or systemic disease, and denied any history of bad habits such as smoking tobacco, vaping or heavy alcohol consuming.

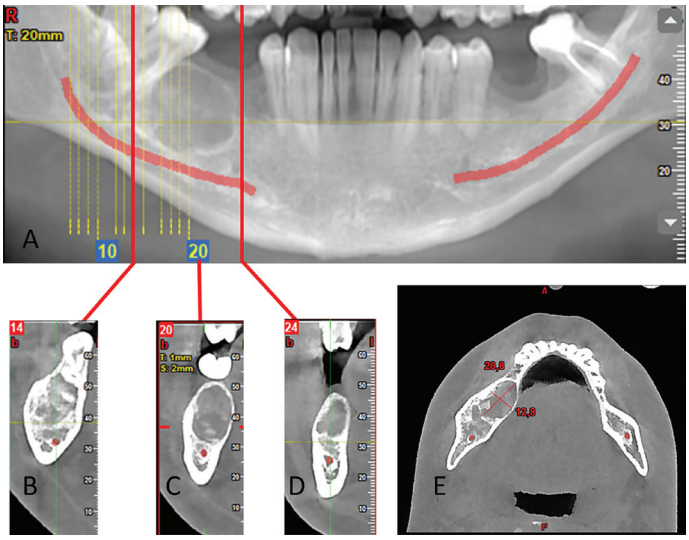
Extra-oral examination did not reveal either a firm or a mobile mass in the right lower jaw; however, intra-oral clinical examination revealed firm, non- tender moderate swelling around the alveolar ridge and on the buccal side of the right posterior mandible (Figure 1A-1B). On palpation, the swelling was firm in consistency. There was no pus discharge and bleeding on

Figure 1 - Intraoral photograph before operation (A, B). Preparation of bone window (C). After removing the mass (D). Specimen (E).



provocation. In the mandible, the swelling was extending from distal side of the second premolar to the second molar tooth region obliterating the buccal and the alveolar ridge corresponding to the region of extracted first molar tooth. There was also no complaint of facial or labial numbness. Orthopantomogram also revealed a lesion well-defined, expansive radiolucent mass with a number of scattered radiopaque calcified spots extending from the region of the previously extracted first molar to the roots of the second molar with a diameter of approximately 2×3 cm (Figure 2).

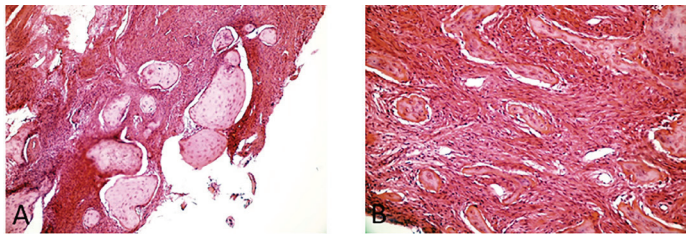
Figure 2 - Panoramic view (A). Cross-Sections on CBCT scans of the tumor (B-D). Axial CT image (E).



After the clinical examination, the mandible was scanned using a cone-beam computed tomography (NewTom Vgi evo, CeflaGroup, Verona, Italy) for further evaluation. Multiplanar images (Figure 2) provided detailed information on relevant roots of the second molar, cortical expansion and internal calcification. Examination of proximity to vital anatomical structures was also made prior of surgical intervention.

After informing the patient about the pertinent clinical findings, upcoming treatment and potential diagnosis of the lesion, the patient provided consent for the necessary operation. He underwent surgery under local anesthesia in the outpatient clinic. The regional nerve block was initiated with an inferior alveolar nerve (IAN) block using 2 ml of 40 mg/ml articaine with 12 mcg epinephrine. Subsequent buccal nerve block was also applied. A mucoperiosteal flap was raised from the region of the second premolar to the retromolar area. After flap elevation, the surgeon set about to preparation of a window by cutting into the thinned bone region seen over the translucency. The lesion was then completely removed in two pieces and the bony walls around the pertinent region curetted until sound tissue was macroscopically visible (Figure 1C-1D). The smaller specimen was labelled as "a small bony-like piece within the center" and the larger one was labelled as "other fully part of the lesion". The dimensions of smaller and larger specimens were 1×0.7×0.4 cm and 2.5×2.5×1.7 cm in aggregate, respectively (Figure 1E).

Figure 3 - Semento-osseous nature surrounded by lymphoplasmacytic infiltration (H&EX100) (A). Small and larger concentric cement foci and new bone trabeculae in a fibroblastic tissue (H&EX200) (B).



Microscopically (Figure 3), the decalcified smaller specimen demonstrated compact bone tissue and bone marrow fibrosis with angiogenesis in between. Microscopy of the larger specimen indicated that the lesion was characterized by spindle cell proliferation arranged in bundles and swirls. There were various sizes of calcifications most of which were in the form of bone trabeculae anastomosing with each other while some of which were resembles oval to round cementum-like structures. Based on all these observations, the lesion was diagnosed as COF.

The patient was prescribed amoxicillin/clavulanic acid 500 mg/125 mg po bid for a week and ibuprofen 600 mg po bid for 5 days. No adjuvant therapy was given. The patient had the checkup appointments at every week just for a month, and then he was informed about upcoming appointments arranged every 6 month for the next 2 years. Recurrence was not observed after one-year follow-up period.

Discussion

FOLs are generally defined as an uncommon condition in which normal bone tissue is replaced by a fibrous tissue containing a newly formed, mineralized substance. Although the definition of FOLs is fairly well established, there seems to be disagreement in the literature on definition of the different subtypes of FOLs. Charles Waldron proposed the first classification of FOLs of the jaws in 1985 [6]. In the Waldron classification, "ossifying and cementifying fibromas" were among "fibro-osseous (cemental) lesions presumably arising in the periodontal ligament" [7]. From a historical standpoint, while there were many amounts of proposals in classification, the WHO shed light on the current nomenclature and terminology of FOLs with their classifications in 1992, 2005, and 2017.

In 1992, the WHO grouped FOLs under osteogenic neoplasms and non-neoplastic bone lesions in the jaw. In this classification, both cementifying fibroma and ossifying fibroma lesions were listed under COF, as they overlapped with different histological variants of the same type of lesion [8, 9].

In the 2005 classification of odontogenic neoplasms, the WHO replaced the term "COF" with ossifying fibroma on the grounds of observation of cementum-like material of odontogenic origin in fibromas in extragnathic cases. It was clear that cementum and bone were actually the same tissue, to be differentiated according to their association with the tooth roots [8, 9].

Advances in classification have continued. The recent WHO (2017) edition of the classification of odontogenic and maxillofacial bone tumors introduced, for the first time, a lesion group of "fibro-osseous and osteochondromatous lesions". This edition re-added the prefix "cemento-" reintroducing cemento-ossifying fibroma (COF) as both a benign mesenchymal odontogenic tumor and a fibroosseous lesion (FOL) [1]. COF was therefore relocated into the benign mesenchymal odontogenic tumors category. It was also defined as a distinct type of ossifying fibroma that occurs in the tooth-bearing areas of the jaws and is believed to be of odontogenic origin [1, 5]. Nevertheless, the acronym COF was used throughout the relevant section on ossifying fibroma except for juvenile ossifying fibroma (JOF) variants: juvenile trabecular ossifying fibroma and juvenile psammomatoid ossifying fibroma [1]. Therefore, there is a proposal to reconsider conventional ossifying fibroma as comprising odontogenic and non-odontogenic subtypes because of several extragnathic cases reported [5].

The origins of COF and the other recognized FOLs are actually complex and not well understood. Most cases reported in the literature were associated with a previous trauma history [9, 10]. Another concept related to its origin is based on developmental causes. A few reports have pointed out extragnatic cases originating from embryologic nests [11, 12]. In the present case, the patient had a history of delayed healing, with concominant purulent discharge, following the extraction of tooth no 46. Considering that there were no previous radiological records either electronic or in hard copy format, we could not be sure whether the presence of the lesion was overlooked by the clinician. From a clinical standpoint, if there is prolonged or unusual healing following dental treatment, suspicion for any kind of lesions is crucial as early diagnosis is important for an effective cure [13]. Clinicians should consider that delayed diagnosis of any kind of lesion can result in more advanced disease at the time of treatment, potentially leading to the patient's frustration and loss of confidence in the healtcare system, greater cost and higher morbidity [13, 14].

The COF is one of the most recognized FOLs along with to FD and COD. Although histologically similar, they can be distinguished by their clinical and radiological features. Clinically, a COF shows a slow- growing pattern, and evolves in young and middle-aged adults with a predilection for women. Traditionally, COF has an ovoid shape owing to a centrifugal growing pattern resulting in growth from the center to the periphery and presenting as a round tumor mass. Looking more closely at demographic factors, COF is prone to occur in the second to fourth decades of life, with a noticeably higher proportion among women, at a ratio of 5:1 [9, 15]. In addition, a COF may occur in any tooth-bearing areas of the mandible and maxilla, most commonly in the posterior side of mandible. In our case, while clinical presentation matched location predilection, there is a contradiction about the sex predilection [16].

Radiographically, a COF appears as a generally well-demarcated and unilocular lesion, which displays radiolucency with variable radiopaque foci depending on the amount of mineralization. In a retrospective study conducted by Titinchi et al. [17], it was reported that approximately 15.9% of lesions would be radiolucent and multilocular. Nevertheless, if there is an impacted tooth, it is difficult to distinguish a COF from either a calcifying epithelial tumor or a calcifying odontogenic cyst based on its radiographic appearance [9, 18]. Moreover, the differential diagnosis of COF involves not only both a calcifying epithelial tumor and a calcifying odontogenic cyst, but also ameloblastic fibroodontoma, cementoblastoma, odontoma, and fibrous dysplasia [9]. In the present case, the radiologist made a provisional diagnosis of ameloblastic fibroodontoma or cemento-ossifying fibroma. On the radiographic examination, displacement of teeth and root resorption may be seen in COF [6, 9]; however, in the present case, radiographic and CT scan images did not reveal any displacement or resorption related to the neighboring tooth. Thus, our diagnosis was made on the basis of the histopathological report.

Microscopically, a COF represents a hypercellular fibroblastic stroma with many delicate bundles of spindle shaped collagen fibers, proliferating fibroblasts, cementoblasts and variable amounts of calcified structures [6]. The latter consist of osteoid bone and hypocellular basophilic structures of cementum-like tissue resembling the cementicles that are normally seen in the periodontal membrane [19].

The mainstream treatment of COF depends on its clinical and radiological features and includes one of the following methods: enucleation, curettage, or surgical resection. While enucleation and primary closure can be performed for smaller lesions, moderate lesions require a more cautious approach with local excision and curettage up to a point at which sound tissue becomes macroscopically visible. A larger COF will need a bone resection as a radical approach to guard against the high tendency to relapse following incomplete removal [17]. In such cases, the resection has to be performed in alignment with bone reconstruction, either by an iliac crest nonvascularized bone graft or a free fibula flap. The most common complications in such operations are flap dehiscence and graft exposure, which may be treated with a second surgery at a time when soft tissue maturation occurs [17-20]. In the present case, the patient was treated thoroughly with local excision/curettage of the lesion regarding a little cleavage margin on radiographic examination.

Conclusion

In conclusion, distinguishing between COF and other lesions and benign tumors is a challenge and a decisive factor in the proper diagnosis. On this basis, oral surgeons should be aware of different aspects of radiographic findings and should regard the pathologists' report due to the definitive diagnosis being mostly based on them. Besides prompt evaluation, accurate diagnosis and proper treatment of the lesions, long-term follow-up is crucial to evaluate recurrence for all patients, regardless of the type of surgical management.

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