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Case report

Cystic degeneration in cemento-ossifying fibroma: Diagnosis challenge and conservative management - Case report

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ABSTRACT

Introduction and importance: Cemento ossifying fibroma (COF) is an encapsulated lesion that is often found in the mandible of middle-aged females. Cystic degeneration may be associated to several osseous lesion. This particular presentation is not well documented in the jaws.

Case presentation: Patient presented was referred to oral surgery department for abnormal radiolucency in the right mandible on panoramic X-ray. The patient did not have any specific medical history and reported painful areas in right mandibular region. During diagnosis assessment, ameloblastoma was first considered due to the presence of cystic lesion clusters. But the mixed image in previous X-ray pointed to a fibro-osseous with a change in pathogenesis pathway. Diagnosis buildup was based on previous radiographs, incisional biopsy was proposed to identify the histopathological feature of the lesion before proceeding to further investigation (CBCT) or treatments (decompression or enucleation). COF of the jaws associated to non-specific cystic changes was the histopathological diagnosis. Delay in treatment showed an unexpected outcome with good clinical and bone healing.

Clinical discussion and conclusion: This case shows the importance of previous radiographs, when available, in diagnosis buildup. The fibro-osseous lesion with cystic change may show a good biological response with conservative management. A clinical and radiological surveillance after a conservative treatment may be a better option in treatment of these lesions.

1. Introduction

Non–epithelial-lined cysts occasionally occur in association with various benign and malignant bone lesions. These cysts vary in nature; some are aneurysmal bone cysts, some are simple bone cysts, and others are nonspecific cystic degeneration [1].

In the jaws this association has not been well documented, but non–epithelial-lined cysts have been reported in association with fibrous dysplasia [1–3], cemento-ossifying fibroma (COF) [4–7], and, more frequently, cemento-osseous dysplasia [1].

Through this paper we tend to highlight a case of COF associated to non-specific cystic degeneration. In literature, few cases have been reported with similar diagnosis [4]. This case appears as an additional case to highlight nonspecific cystic changes in COF, but more importantly to highlight the atypical imaging features along with diagnosis buildup and particular response to conservative management option. Usually, COF is a slow-growing benign neoplasm and is surgically excised conservatively, with no recurrence in most cases [8] however literature on COF with cystic changes has not a consensual standard of care.

This case report has been reported in line with the SCARE Criteria [9].

2. Presentation of case

A 40-year-old female patient was referred to the oral surgery department by her dentist for an abnormal radiolucency in the right mandible spotted in panoramic X-ray. She reported painful areas in right mandibular region.

The patient did not have any specific medical or family history, she was nonsmoker with no relevant social, surgical or familial history. According to her dental history she had previous dental treatment on the first right incisor five years ago, during this period she underwent a panoramic X-ray. There was no history of trauma.

Extraoral examination did not reveal any swelling, expansion,

Abbreviations: COF, cemento-ossifying fibroma; ABC, aneurysmal bone cyst; SBC, simple bone cyst; WHO, World Health Organization.

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cutaneous changes or cervical adenopathy and there was no motor nor sensory deficit. Intraoral examination revealed a normal dentition and occlusion, with expansion of the right mandibular buccal cortex to the anterior border of the right ramus, lingual cortical plate was normal. The region was painful with a bony-hard and non-tender effect during palpation and the margins were blended imperceptibly with the surrounding bone. The overlying mucosa was normal in color and texture. The tooth vitality test was positive for all the teeth in relation with the lesion.

Recent panoramic X-ray showed a multilocular extensive radiolucent lesion in the right mandible extending from the right mandibular canine to the right ramus, with radiopaque septa. The lesion respected the mandibular nerve path. No resorption or displacement of teeth was noted (Fig. 1).

Interrogation on previous dental procedures revealed that the patient underwent a radiographic examination (panoramic X-ray) five years ago. She was asked to bring it on her next consult. The panoramic X-ray showed well defined dome-aspect in retromolar area, "ground-glass" radiopaque lesion in the right ramus and posterior body of the mandible; more anteriorly mixed radiodensity was observed in periapical region from first right premolar to second right molar (Fig. 2).

During diagnosis assessment, ameloblastoma was first considered due to the presence of cystic lesion clusters. But the mixed image in previous X-ray pointed to a fibro-osseous lesion with a change in pathogenesis pathway. Which presented a diagnosis challenge.

An incisional biopsy was proposed to identify the histopathological feature of the lesion before proceeding to further investigations (CBCT).

The biopsy was performed, by oral surgery professor in university hospital, under local anesthesia and encountered a pellucid aspect in the buccal cortex. Aspiration of lesion content presented a yellowish color liquid. The incised specimen was a mix of a brittle hemorrhagic-like tissue and a fibro-osseous tissue (Fig. 3).

Microscopic examination showed a fragment bordered with a spongy bone tissue centered by a dense fibroblastic proliferation containing numerous bone structures. Fibroblast component had spindle-shaped mononuclear cells without noticeable atypia and without mitotic activity. The bone component was made of cemental-like calcified spans. These spans were often confluent in curvilinear structures and were often lined with regular osteoblastic cells (Fig. 4).

In liquid microscopic examination, smears contained a discretely hematic background some polymorphic inflammatory elements associated with a few histiocytes, without suspect cells.

Pathologist concluded to a discretely hematic and histiocytic intracystic fluid without suspect cells (Fig. 4).

In view of radiographical aspect of the lesion and the intraoperative finding of a soft, fibrous lesion with no plane of cleavage. Also, the absence of any history of trauma (although it should be noted that in approximately 50% of cases subsequently diagnosed as traumatic bone cyst, there has been no previous trauma to the area) and absence of blood content (diagnosis of ABC was ruled out). Basing on these elements, we concluded to an inhomogeneous lesion, associating a cystic degeneration surrounded by dysplastic bone corresponding to a cemento-ossifying fibroma.

Considering the benign nature of the lesion a surgical excision of the lesion was proposed through an intraoral approach. Management was postponed regarding pandemic covid-19 world situation.

Four months postoperatively, there was no increase in lesion size and the patient reported disappearance of pain. Radiographic evaluation (panoramic X-ray) showed reduction in lesion size as well as partial filling of the central cavity by mineralized tissue (Fig. 5). Clinically, the lesion did not evolve and the involved teeth kept their vitality. Regarding this positive outcome, a change in the planned intervention was suggested, together with the patient, we decided to schedule a regular radiological and clinical monitoring, first at six month rate then every year.

Seven months postoperatively, panoramic X-ray showed continuing good evolution with improved radiodensity in mandibular body and ramus and persistence of small radiolucency from first right premolar to first right molar periapical region (Fig. 6).

The patient is kept under close clinical and radiological monitoring, one visits a year, and expresses a relief about the positive outcome of this monitoring.

The outcome from a simple incisional biopsy along with aspiration of lesional content was unexpected as a surgical treatment for this large lesion, since literature always pointed that a good outcome is achieved by excision of the entire lesion.

3. Discussion

Cemento-ossifying fibroma (COF) is a distinct type of ossifying fibroma that occurs in the tooth-bearing areas of the jaws and is believed to be of odontogenic origin [8]. Cystic changes in this lesion is unusual, in our case report the fibro osseous origin of the lesion was suspected from previous radiograph, the modification of clinical features with

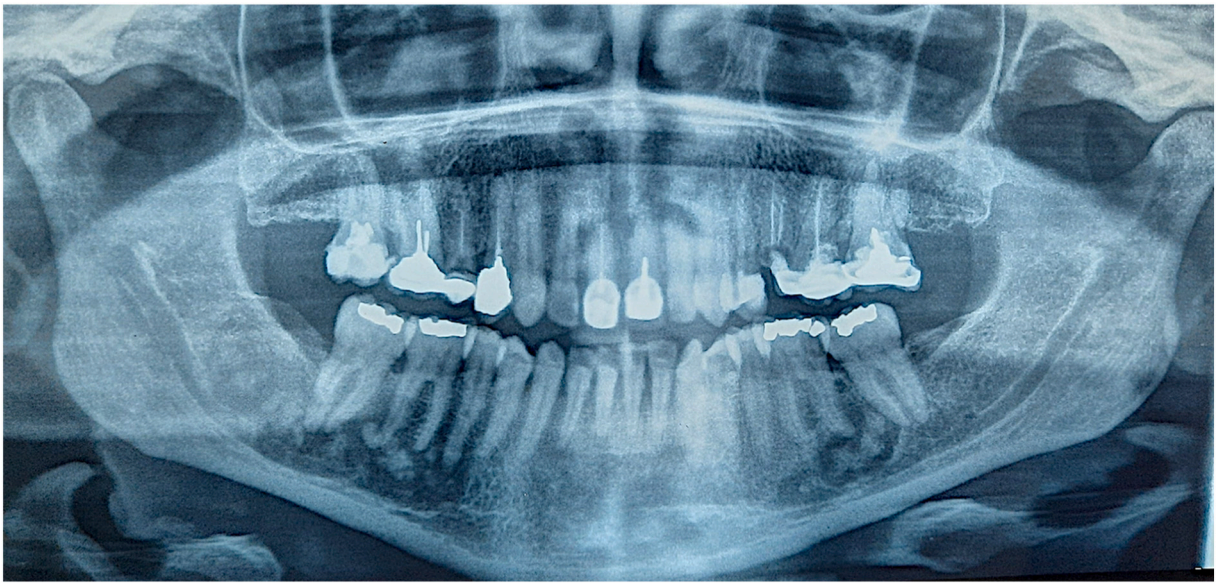

Fig. 1. 5 years ago panoramic radiograph: "ground-glass" radiopaque lesion in ramus and posterior body of the right mandible.

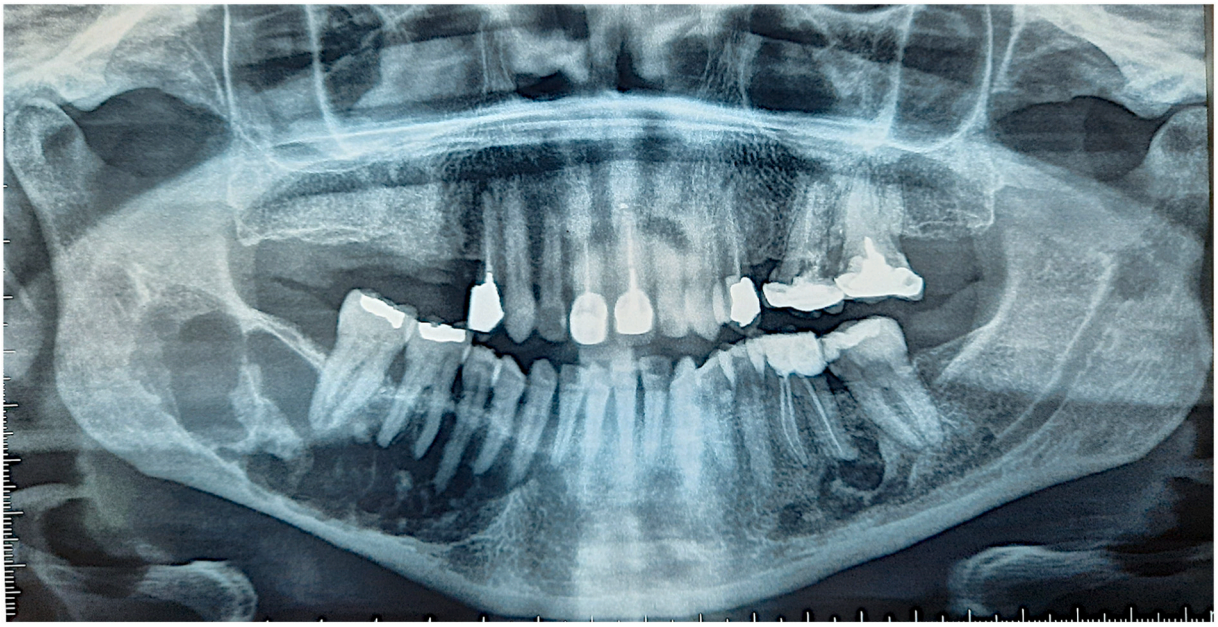

Fig. 2. Panoramic radiograph: well-defined, multilocular radiolucent image limited anteriorly by the right mandibular canine and posteriorly by the right ramus, radiopaque septa within the radiolucency. No root resorption is noted.

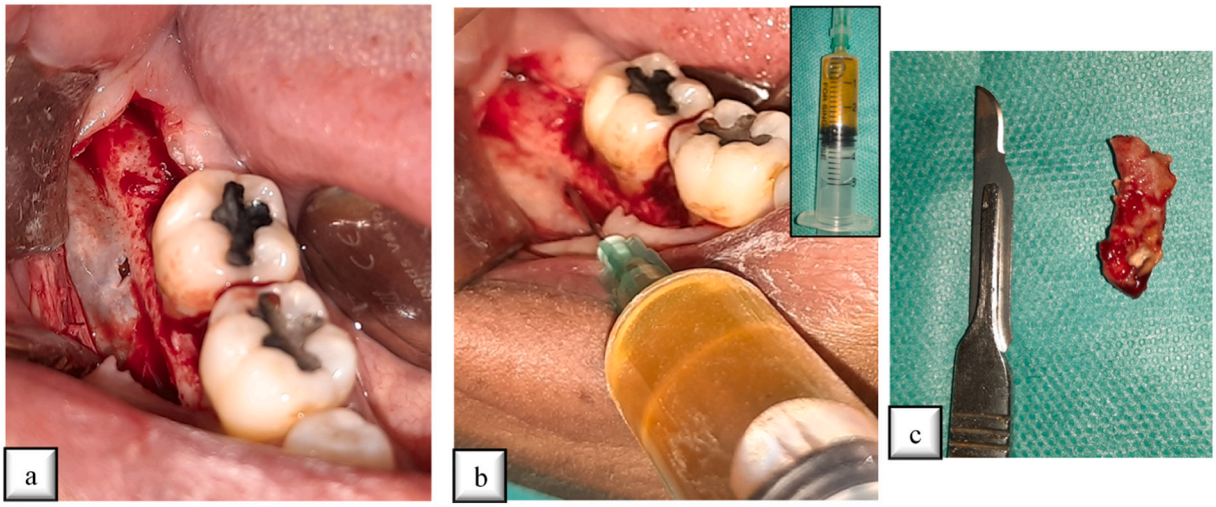

Fig. 3. Incisional biopsy: a. Full thickness flap: Clinical aspect of the lesion with pellucid aspect in the buccal cortex b. puncture lesion content: yellowish content recalling a cystic origin c. incised specimen with a fibro-osseous aspect.

buccal expansion and reported pain pointed to a change in the lesion pathogenesis. Management of COF alone or cystic lesion of the jaws alone is surgical and well documented, the association of these lesions however is rare and few cases reported various management techniques with various outcomes. In our case report a simple incisional biopsy along with aspiration of lesional content allowed unexpectedly a complete healing of the lesion, which may present a good treatment option, however conservative management in large lesion may have a potential risk of continuous growth or malignant transformation. For this reason, a close monitoring of the patient mandatory.

Cemento-ossifying fibroma has been variously called ossifying fibroma, cementifying fibroma, and cemento- ossifying fibroma. In the 2017 World health organization (WHO) classification, the latter is preferred because of its descriptive value, and more importantly, it was classified under odontogenic tumors to distinguish it from the juvenile types. However, it is still from a fibro-osseous origin [10].

COF is rare. The peak incidence is in the third and fourth decades of life. There is a definite female predilection with sex ratio as high as 5:1, COF occurs exclusively in the tooth- bearing areas of the mandible and maxilla. The mandible is far more commonly involved than the maxilla. The mandibular premolar and molar area are the most common sites [8]. In our case report clinical presentation matched sex and location predilection.

Early ossifying fibromas are small and may be radiolucent. As they enlarge and mature, they will become mixed radiolucent-radiopaque and then completely radiopaque. These tumors characteristically expand slowly and asymptomatically. Their expansion is symmetric from the epicenter of the tumor, creating a spherical or egg-shaped mass on plain radiographs and CT scans [11].

Non-epithelial lined cysts occasionally occur along with various bone lesions. These cysts vary from aneurysmal bone cyst (ABC), simple bone cyst (SBC) and non-specific cystic degeneration. In the jaws this association is not well documented but non-epithelial cysts have been described as secondary phenomena in many benign and malignant bone

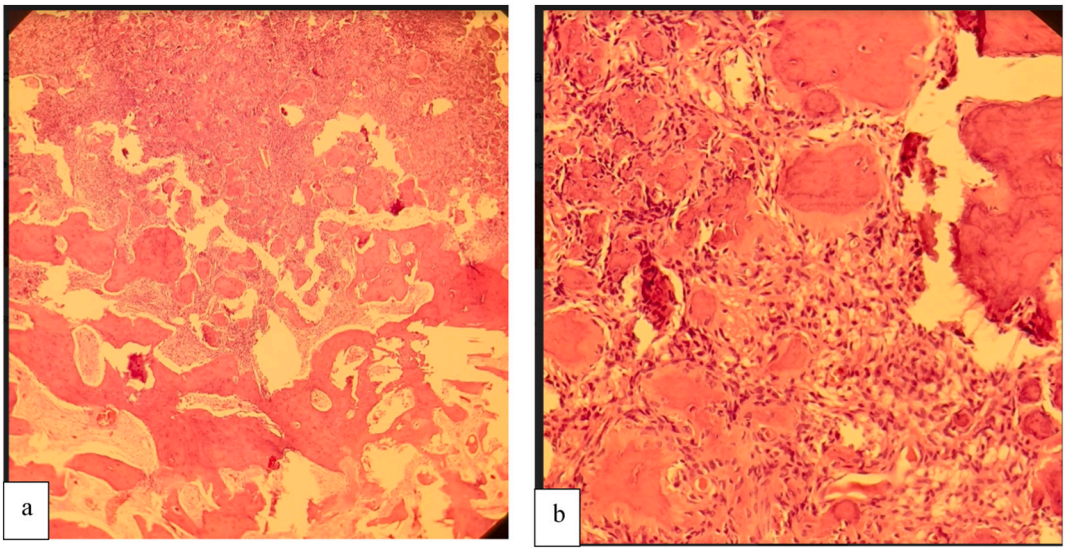

Fig. 4. a. H&E stain ×100: Fibroblastic proliferation associated with bone trabeculae and osteoid and cementum like clusters. b. H&E stain ×400: higher magnification showing little atypical fibroblast cells and cementum-like clumps.

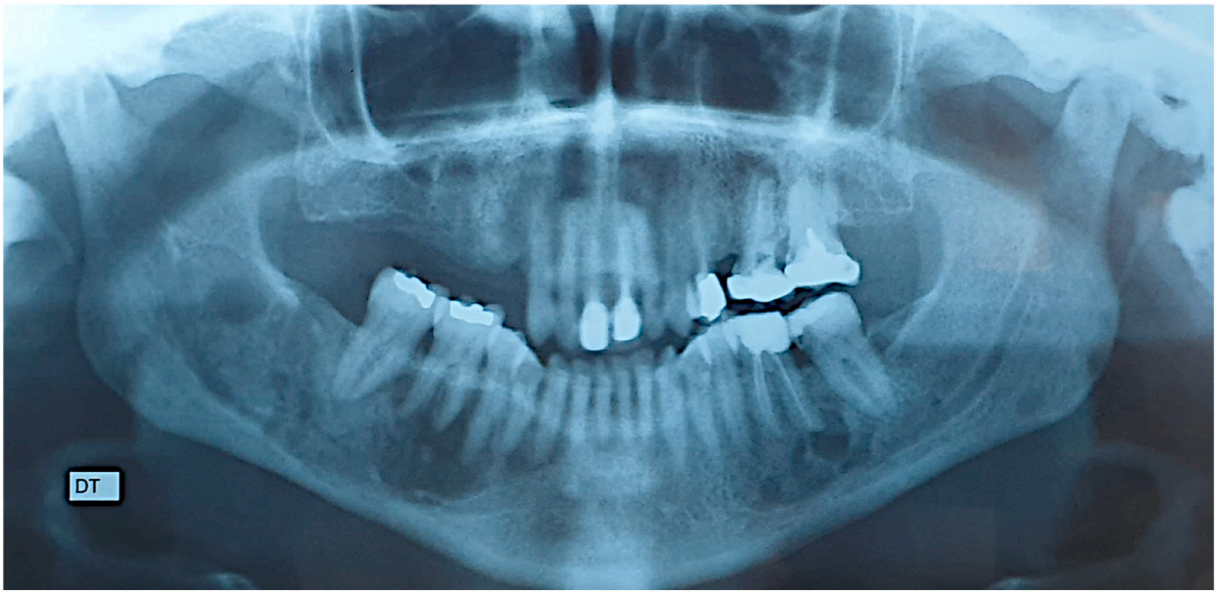

Fig. 5. Panoramic radiograph: 4 months follow up examination showing significant bone regeneration.

tumors and tumorlike lesions as fibrous dysplasia, OF, and cemento-osseous dysplasia. Cystic change may occur years after the initial diagnosis of fibro-osseous lesion and the mechanism of this change remains unknown [1].

In the same edition of WHO classification, it was pointed out that rapid tumor growth that has been observed in some cases is most likely caused by secondary aneurysmal bone-cyst formation [8]. Aneurysmal bone cyst (ABC) is a cystic or polycystic expansible osteolytic neoplasm composed of blood-filled spaces separated by fibrous septa containing osteoclast-type giant cells. There is enlargement, which is frequently painful. The teeth remain vital, but tooth mobility and displacement are common. Radiographically, there is expansion with well-delineated unilocular or multilocular radiolucencies. Perforation of the cortex can occur with extension to the adjacent soft tissues. Root resorption is seen. CT may reveal bone septa compartmentalizing the lesion. CT and MRI demonstrate fluid-fluid levels that are characteristic of (but not specific for) ABC. ABC-like areas (secondary ABC) can occur in a variety of other disorders of bone, including osteoblastoma, fibrous dysplasia, and ossifying fibromas [8].

In another hand, simple bone cyst (SBC) is an intraosseous cavity that is devoid of an epithelial lining and is either empty or filled with serous or sanguineous fluid. SBCs are generally asymptomatic, and are usually found incidentally during routine examination. Radiologically, they are well-defined radiolucencies that frequently extend between the roots of associated teeth, without resorption or displacement. Larger examples may be multilocular. A minority of cases may show bone expansion (17.6%), loss of the periodontal ligament space (2.9%), or effacement of the lamina dura (11.8%). Expansion of the cortical plates and loss of the lamina dura are more frequent in cases associated with osseous dysplasia [8].

Nonspecific cyst degeneration is a term for a lesion that fails to meet the histological criteria of either an aneurysmal or a simple bone cyst. The latter two entities consist of blood-filled cavities in bones, which are lined by a thick layer of fibrous tissue. The pathogenesis of such

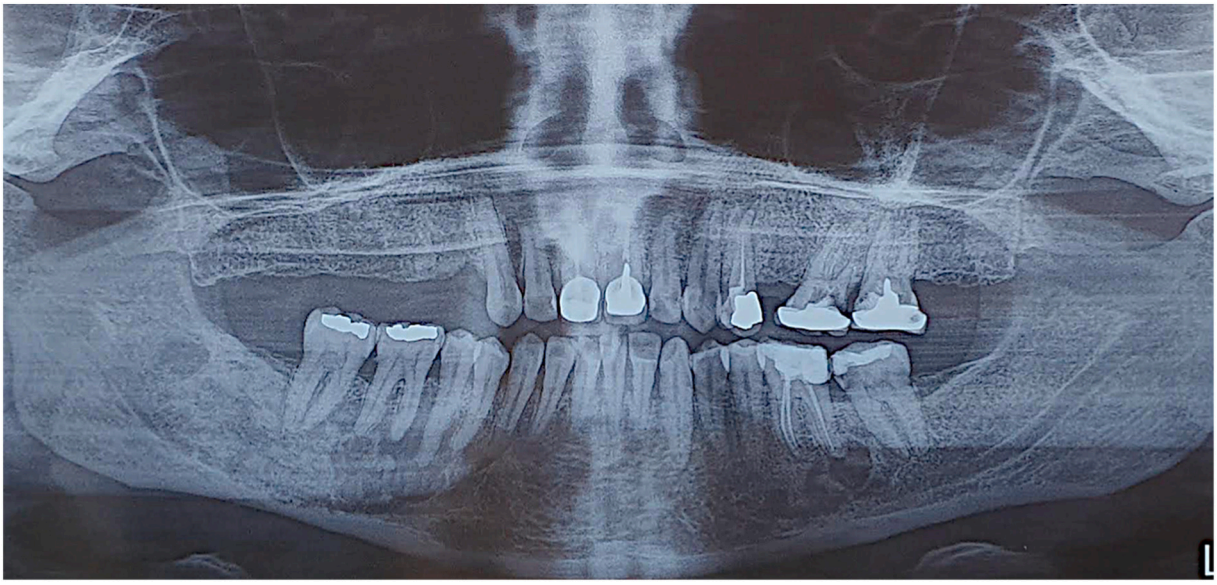

Fig. 6. Panoramic radiograph: 7 months follow up examination: improved radiodensity in mandibular body and ramus and persistence of radiolucency from first right premolar to first right molar periapical region.

nonepithelial cell–lined bone cysts is still unknown; however, it has been postulated that aneurysmal and simple bone cysts arise from related processes. It may be that the development of cyst degeneration does not depend on a single pathogenetic event, but that there are several potential mechanisms. Regardless of the mechanism by which COF undergoes cyst degeneration in a given case, all cases report that this degeneration is paralleled by an acute clinical change [3].

Although the ABC remains the most frequently reported type of cystic degeneration associated to COF, non-specific cyst degeneration might be observed [4]. As presented in our case report, the secondary changes, which although appeared filled with cystic liquid and lacked epithelial lining, does not meet the diagnostic need for SBC or ABC. Therefore, the present case could be classified as OF with non-specific cystic changes.

The development of cystic degeneration in COF can present a diagnostic and therapeutic dilemma since cystic changes were also reported with various other fibrous lesions. This might complicate the histological diagnosis of the primary disease and require careful scrutiny to reach a diagnosis. In the present case, evaluation of previous X-ray was the only way to have differential diagnosis.

Clinical presentation with rapidly enlarging mass can be alarming to both patient and physician. Such transformation should be kept in mind when cases of COF show sudden rapid enlargement. The swellings may be bony-hard or fluctuant. Moreover, the radiographic picture may fail to reflect the cystic nature of the lesion. CT is thus an essential aid in the diagnosis [1]. In our case panoramic radiograph objectified radiolucent feature consistent with cystic lesion nature.

The literature shows that depending on lesion nature and extent management modalities of fibro-osseous lesions vary from simple observation to surgical enucleation or resection [2,12]. COF is a slow-growing benign neoplasm. Without a cystic compound, COF can be surgically excised conservatively, with no recurrence in most cases. Untreated tumors can attain a massive size and may require bloc resection [8].

Cystic degeneration development may cause considerable expansion and concurrent functional and cosmetic problems that may compel surgery [6]. Some authors point that fibro-osseous lesions with concomitant SBC or cystic degeneration require aggressive management [7]. In several cases surgical resection was the management option, in these cases cystic degeneration consisted of ABCs [4,6,13] and non-specific cystic degeneration [7]. One case reported, considering the benign nature of the lesion, radiological and clinical monitoring as management option [5].

In our case report, surgical enucleation was decided after anatomo-pathological diagnosis, the delay of surgical management highlighted the very good outcome of only excisional biopsy along with content aspiration. Thereby it is interesting to note that minimal surgical intervention with close monitoring of fibro osseous lesions complicated by cystic degeneration may be a valid management option, allowing better surgical outcomes with less morbidity.

From patient perspective, a minimally invasive procedure allowing a good outcome presented a great relief especially compared to surgical enucleation that has been proposed and explained with all surgical morbidities of management of large lesions in the jaws.

4. Conclusion

Cemento-ossifying fibroma (COF) is a fibro-osseous odontogenic lesion that occurs in the tooth-bearing areas of the jaws [8]. Literature reported few cases of cystic degeneration related to fibro-osseous lesions [8], this modification in lesion pathogenesis must be kept in mind during X-ray evaluation. In our case report, previous panoramic radiograph allowed a better differential diagnosis. Regarding management, we think that incisional biopsy with cystic content puncture and postponing surgical management in cases with non-specific cystic degeneration may allow better outcomes and avoid extensive bone loss but clinical and radiological monitoring is mandatory to assess evolution. To our knowledge our case is the second reported case in literature pointing monitoring as management option in this clinical presentation. More literature cases are still needed to allow better understanding of pathogenesis of this lesion and objectify the suitable management decision.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Dr. Mouna Bouhoute: Data curation, Writing - Original draft preparation.
Pr. Bouchra Taleb: Surgical management, Reviewing and Editing of the article.

Declaration of competing interest

The authors certify that there is no conflict of interest with any financial organization regarding the material discussed in the manuscript.

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