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Case Report

Management of Cemento-Ossifying Fibroma: A Report on Two Clinical Cases

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Abstract: Cemento-Ossifying Fibroma (COF) is a rare benign tumor of the jaw characterized by the formation of cementum and bone-like tissue, often arising from the periodontal ligament. This article presents two clinical cases that illustrate the complexities of diagnosing and managing COF. The first case involves a 30-year-old male with a painless, progressively enlarging swelling in the left mandible, diagnosed through panoramic radiography and cone-beam computed tomography (CBCT). Surgical intervention included mandibular resection and reconstruction using an iliac graft. The second case features a 26-year-old female presenting with a non-painful swelling in the right mandibular premolar region, which was also confirmed via imaging studies. Complete surgical resection was performed, and histopathological examination validated the diagnosis of COF. Both cases highlight the importance of accurate diagnostic imaging and histological analysis in confirming COF, as well as the need for individualized surgical approaches based on lesion characteristics. Despite its benign nature, COF can lead to significant morbidity due to local invasion and recurrence potential. The article emphasizes that thorough follow-up is essential to monitor for recurrence after surgical management. These cases contribute to the understanding of COF's clinical presentation and reinforce the necessity for a multidisciplinary approach in managing this condition effectively. Further research is needed to explore the underlying pathogenesis and optimize treatment strategies for COF in clinical practice.

Keywords: Cemento-Ossifying Fibroma, Surgical Excision, Histopathology, Fibro-Osseous Lesion, Maxillofacial Surgery, Conservative Management.

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INTRODUCTION

Cemento-Ossifying Fibroma (COF) is a rare benign tumor of the jaw characterized by the formation of cementum and bone-like tissue. This lesion typically arises from the periodontal ligament or the jawbone itself, manifesting as a slow-growing mass. Despite its benign nature, COF can cause significant morbidity due to its propensity for local invasion and recurrence. Understanding the clinical features, diagnostic modalities, and management strategies of COF is crucial for accurate diagnosis and optimal treatment outcomes. In this article, we delve into the intricacies of COF through two clinical cases, exploring its pathogenesis, clinical and radiological presentation, diagnostic challenges, and therapeutic interventions.

CASES PRESENTATION

Case 1

A 30-year-old male patient reported to the Oral surgery and medicine Department of Fattouma Bourguiba hospital with a painless, progressive, slow growing swelling on the left side of the lower jaw for 3 months.

Extraoral examination showed a diffuse, round shaped 5cm.2cm swelling over the left body of the mandible extended along the inferior border up to the submandibular region (Fig 1/A). The swelling was non-tender and bony hard in consistency, had a normal skin color with no paresthesia.

Intra-orally, the swelling was located in the buccal side and was extending from 36 to 38 and approximately measured 3 cm *1 cm. The overlying mucosa was apparently normal (Fig 1/B).

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The Panoramic radiograph showed a well-defined unilocular radiolucency in the left body of the mandible, extending from 6 to the left angle of the mandible, with polycyclic contour, measuring 5cm.2 cm and extending to the mandible inferior border (Fig 1/C).

Cone-beam computed tomography (CBCT) images revealed a large well-defined expansile mixed lesion containing central intralesional calcifications, measuring 46,3mm*30,6mm*43,7mm, in the left side of the mandible (Fig1/D).

Buccal, lingual plates and inferior border were blowed, thinned and interrupted.

Comparing clinical and radiological data, the diagnosis of a cemento-ossifying fibroma or an ameloblastoma was evoked.

The patient was referred to the Maxillofacial Surgery Department at HPE SAHLOUL for surgical management under general anesthesia. The treatment plan consisted in a mandibular interrupted resection associated with a reconstruction by an iliac graft (Fig 2).

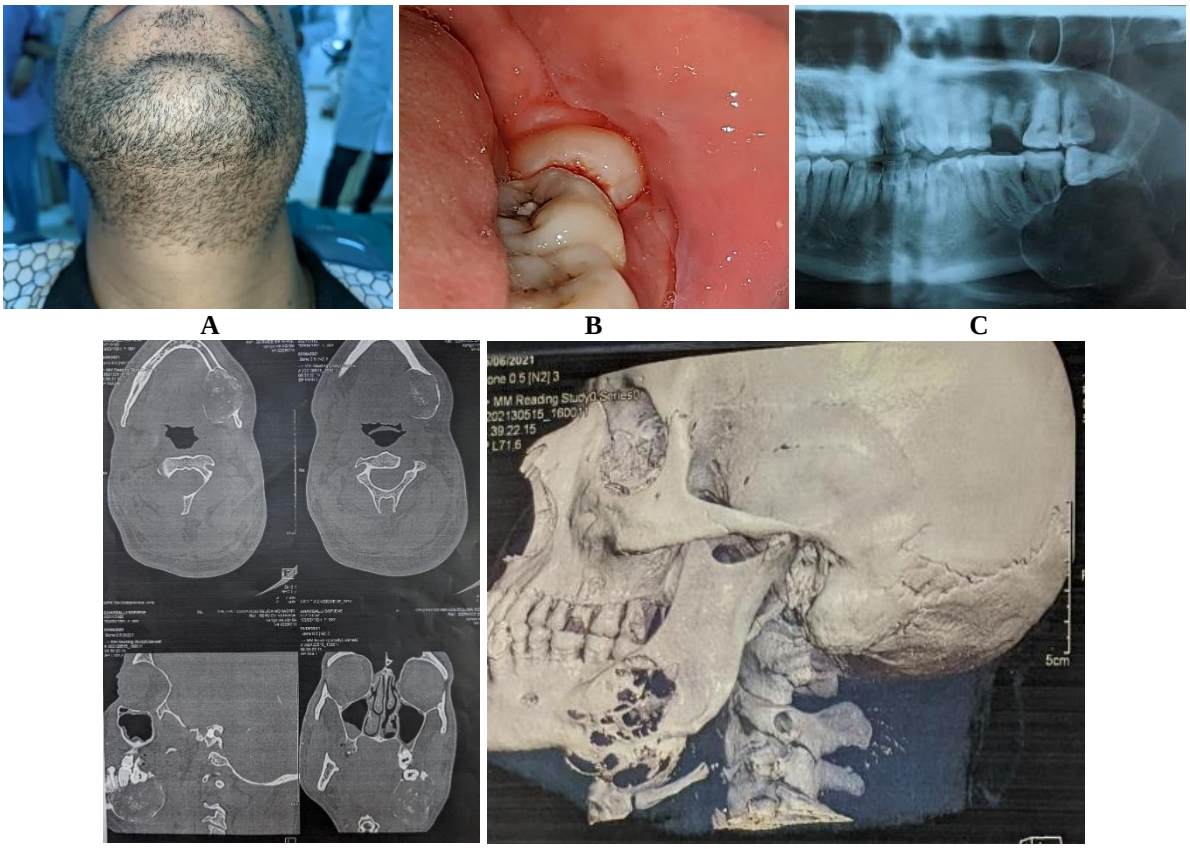
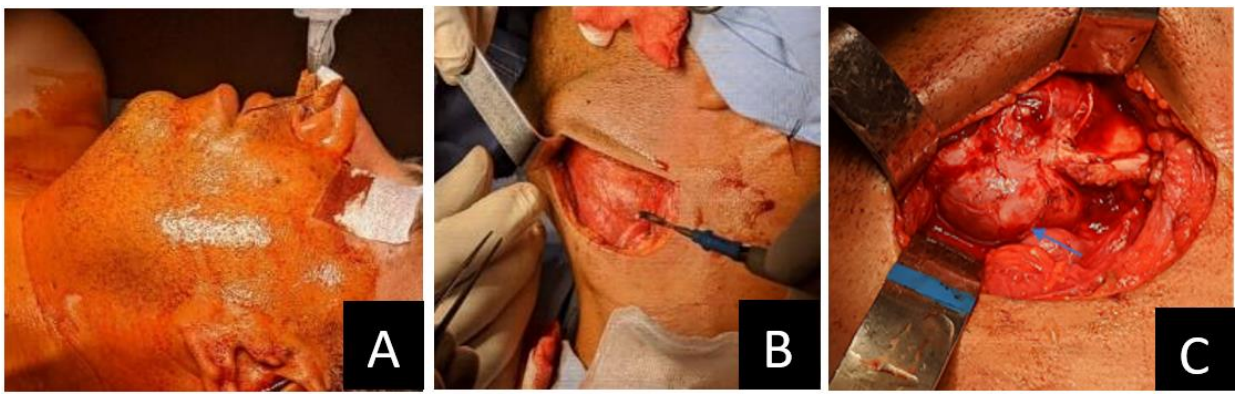


Fig 1: Clinical observation:
A: Extra-oral view/B: Intra-oral view/ C: OPG radiograph
D: Preoperative CBCT / E: 3D reconstruction



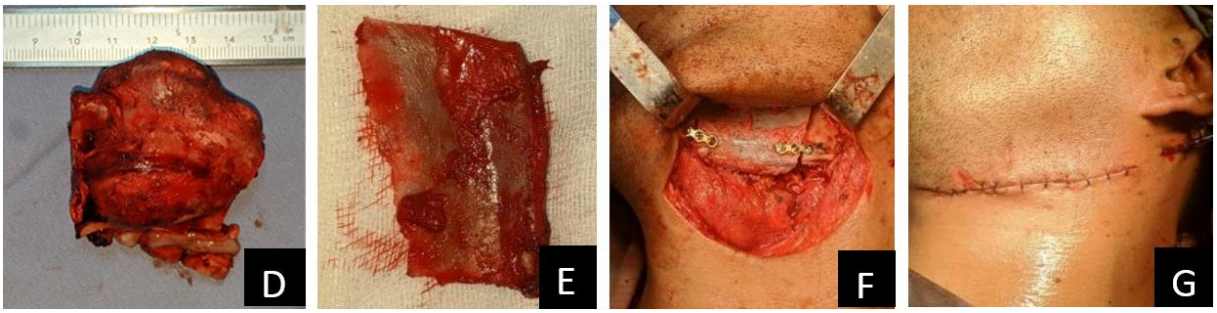


Fig 2: Surgical procedure
A: General anesthesia and patient intubation / B: Cervical cutaneous approach / plane by plane dissection / C: Tumor exposure / D: Surgical specimen after interrupted resection / E:Iliac graft harvesting / F: Fixation with osteosynthesis plates / G: Sutures of the different plans + Drain installation

Case 2

A 26 year-old-female was referred to the Oral surgery and medicine Department of Fattouma Bourguiba hospital for a 1 month non painful growing swelling located in the right mandibular premolar region.

She was non-smoker, and the medical history did not reveal any significant systemic diseases.

Extra oral investigation showed no specifities.

Intra orally, the swelling was located in the buccal side from 44 to 46 and approximately measured 1,5cm x 1,5 cm (Fig 3/A).

The overlying mucosa was normal.

Panoramic radiograph showed a well-defined unilocular radiolucency, extending from 4 to 6 in the right mandibular region, with a 2 cm intralesional calcification measuring diameter (Fig 3/C).

CBCT images showed a well-defined expansive mixed lesion containing central intralesional calcifications, in the right mandibular premolar region. Buccal plate was expanded and thinned (Fig3/D).

A complete surgical resection of the lesion was decided

Intra-sulcular incisions were made with a mesial discharge incision and a mucoperiosteal flap was elevated (Fig4/A-B).

The lesion was removed and sent for pathological examination, which confirmed the diagnosis of a cemento-ossifying fibroma (Fig3/C-F).

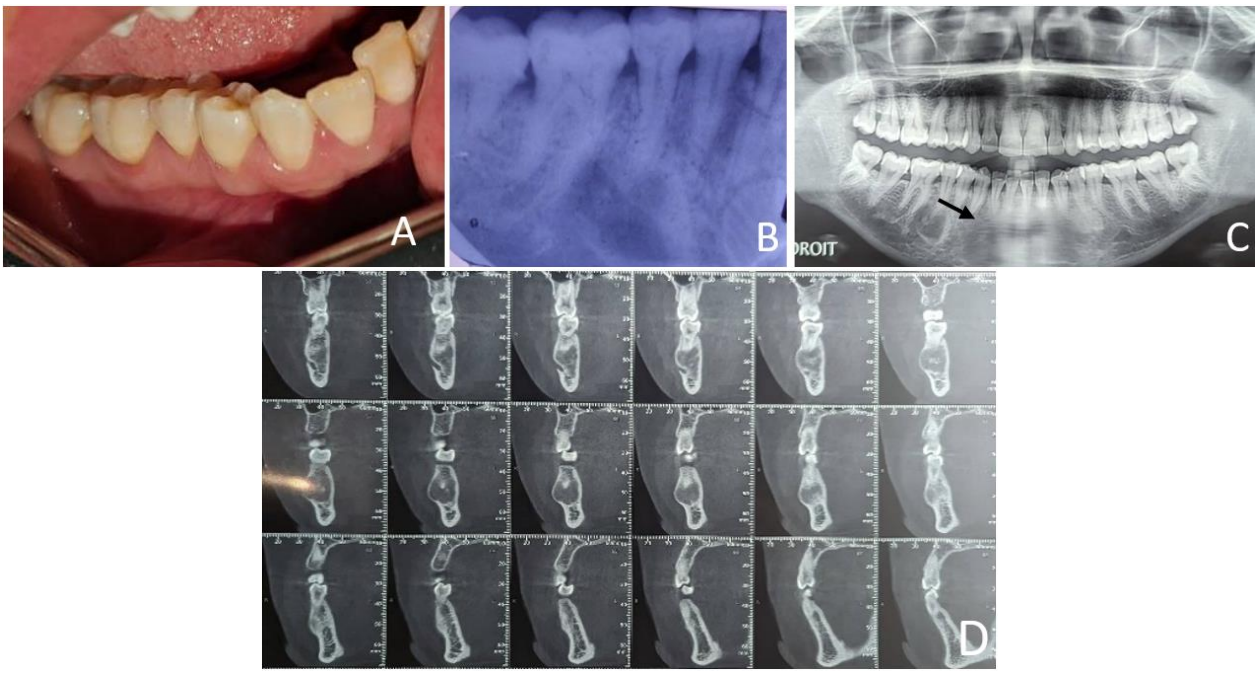
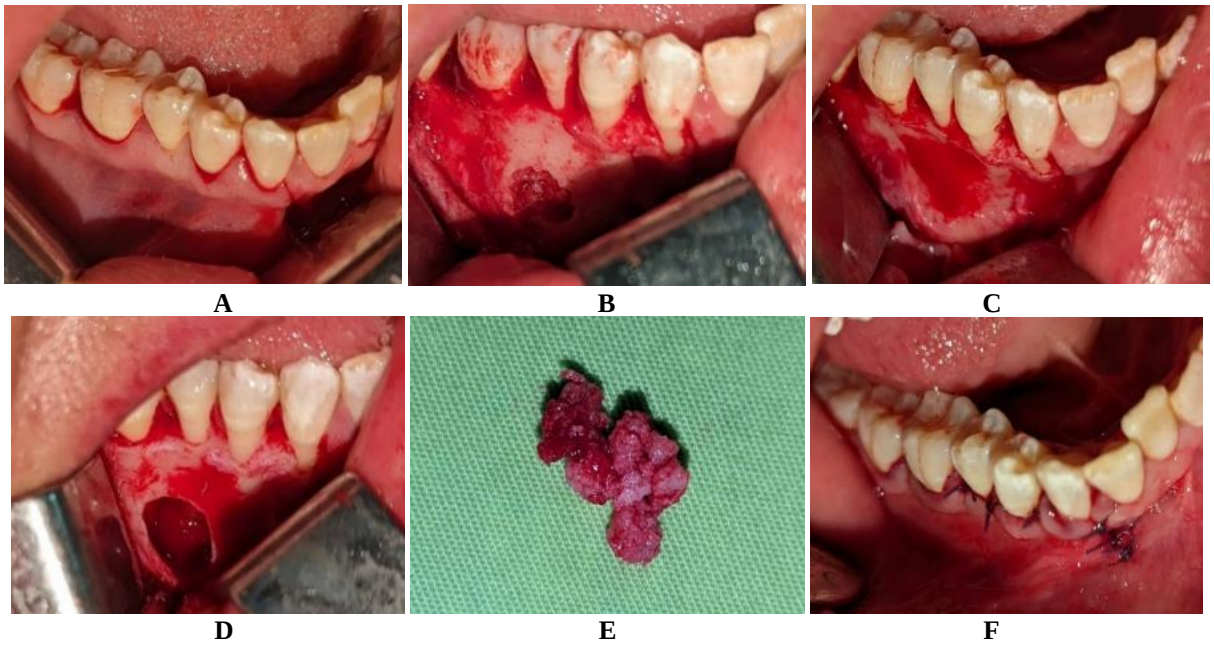


Fig 3: Clinical observation:
A: Intra-oral view /B: Standard X-Ray / C: OPG radiograph
D: Preoperative CBCT



A **B** **C**

D **E** **F**

DISCUSSION

Historic of terminology

Ossifying fibroma along with fibrous dysplasia are the most common fibro-osseous lesions (FOLs) occurring in the maxillofacial region. They have distinct patterns of disease progression, but the various similarities in histomorphological and radiographic features pose difficulties in their classification and management.

In 1971 (Pindborg J, Kramer I (1971)), the authors included two separate entities: cementifying fibroma as a type of "cementoma" and ossifying fibroma as a type of osteogenic neoplasm. The histological description of cementifying fibroma was what we now consider as an ossifying fibroma(« Zegalie N, Martin L, Speight PM (2015)(« El-Mofty SK (2014)).

In 1992 (Kramer I, Pindborg J, Shear M (1992)), both lesions were called cemento-ossifying fibroma.

In 2005 (Barnes L, Eveson JW, Reichart P, Sidransky D (2005)), the terminology changed again, and all the "cemento-ossifying fibromas" were regarded as ossifying fibroma. This was because cementum and bone are essentially the same tissue and can only be distinguished by their relationship to the tooth root.

In 2017, the consensus group felt that the term cemento-ossifying fibroma is suitably descriptive and indicates that these lesions are specific to the tooth-bearing areas of the jaws and can be distinguished from the two juvenile variants of ossifying fibroma. The newest classification therefore classifies cemento-ossifying fibroma as a benign mesenchymal odontogenic tumour(El-Naggar AK, Chan JKC, Grandis JR, Takata T, Slootweg PJ (2017)). This clearly distinguishes it from ossifying fibroma that are non-odontogenic and are classified under benign fibro- and chondro-osseous lesions.

The three variants are therefore defined as cemento-ossifying fibroma, juvenile trabecular ossifying fibroma and juvenile psammomatoid ossifying fibroma(« El-Mofty SK, Nelson B, Toyosawa S (2017)).

Etiology and risk factors

Cemento ossifying fibroma (COF) is associated with numerous genetic and epigenetic factors, trauma, hormonal disturbances, and local factors resulting in disorganized molecular signaling. Lastly, chief nodal molecular mediators such as bone morphogenetic proteins and Notch signaling are involved. Neurotrophic factors, especially the nerve growth factor, may also play a vital role in the overall pathogenesis of this rare but significant neoplastic entity. No clear correlation has been established regarding the exact etiological factor inciting the onset of the neoplasm, and conflicting results exist. A comprehensive insight into the etiology and pathogenesis of COF has been discussed, and areas of potential future research have been suggested.(Toferer A, Truschnegg A, Kashofer K, Beham-Schmid C, Beham A,(2021))

Numerous genetic, environmental, and biological etiological factors have been suggested contributing to COF pathogenesis. They include factors such as gene-environment interactions, local factors, hormonal disturbances, and trauma; these factors have been discussed in detail. Few factors may also act as catalysts to predispose or initiate COF, which are yet to be discussed completely. Trauma, as such, may not initiate benign neoplasms in the head and neck region despite local influences. Modifications in existing receptor states due to trauma may, in turn, start

neoplasms of local projection, e.g., pleomorphic adenomas. Taking the same theory forward, hormonal factors acting with or without trauma also need a comprehensive overview as a part of the etiological standpoint. Lastly, extensive transcriptional changes must also be studied in detail. It is a challenge in clinical medicine to state that any one causal or risk factor initiates any neoplasm. The multifactorial etiology of COF has been highlighted, and these aspects present themselves as potential subjects of future research. A wholesome study focusing on all etiological initiators of COFs should ideally be undertaken in the context of its occurrence, treatment, and periodicity, especially from a clinical standpoint.(Agustí A, Melén E, DeMeo DL, Breyer-Kohansal R, Faner R,(2022))

Pathogenesis and Histopathology

Cemento ossifying fibroma (COF) is a fibro-osseous lesion affecting the maxillofacial region. The etiology of COF is poorly understood, though COF is believed to arise from multipotential cells of the periodontal ligament, possibly due to persistence of odontogenic epithelium following tooth extraction or chronic infection. Accumulating cellular and molecular changes that are involved in the development of COF are believed to represent reactive changes. The main cellular activity in COF is mineralized tissue formation, presumably by fibroblasts. This mineralized tissue formation occurs in associations of spindle-shaped fibroblasts, possibly representing active cells, and inactive, polygonal osteoblasts. At histopathological examination, COF shows three features: immature, mature, and inactive layers. Varying mixtures of these layers are typically found. The immature layer is solidly composed of spindle-shaped fibroblasts with nuclei distending the cytoplasm, and is at least in part surrounded by inflammatory cells. The mature layer contains bony spicules surrounded by osteoblasts, a more abundant fibrovascular stroma, and larger blood vessels. The inactive layer is densely stellate in shape, consists typically of one layer of osteogenic cells and is relatively hypocellular. The thicker this last layer is, the less chance of recurrence there is. Many patients are treated and definitively diagnosed in non-specialized hospitals. In these hospitals, COF is diagnosed exclusively by means of a histopathological examination. Innovations in immunohistopathologic and molecular techniques could, however, provide histopathological diagnostic criteria that will contribute to the differential diagnosis of this enigmatic lesion.(Gennaro P, Gennari L, Latini L, Cavati G, Vannucchi M, Giovannetti F, Cascino.F, (2024))

Clinical Presentation and Diagnosis

The COF may occur in a wide age range, albeit it is predominantly diagnosed in the third and fourth decades of life, with a two-fold higher predilection for females. Clinical signs usually include increasing asymptomatic swelling or asymmetry, and the symptoms may vary between dental movement, diastema appearance, root resorption, dysphagia, trismus, and infection. Swelling may involve the mandibular and/or maxillary divisions and usually causes a slow and progressive facial swelling; an increase needs further examination through imaging. When clinical and radiographic outcomes are correlated, a definitive diagnosis may be attempted. COFs frequently show the following radiographic features: well circumscribed; ground-glass or homogeneous patterns; minimal to no surrounding sclerosis; an internal calcific/sclerotic central core; and may displace or resorb the surrounding dentition. Bone expansion and perforation, together with thinning of the cortical bone, have also been reported and may relate the COF to benign fibro-osseous lesions. Radiographically, COF and other relatively common jaw pathologies share numerous features, which may drive necessary differential diagnosis.(Pick E, Schäfer T, Al-Haj Husain A, Rupp NJ, Hingsammer L, Valdec S,(2022))

Following the correlation of clinical and imaging, a tentative diagnosis may be attempted. To confirm the final diagnosis, however, histopathological examination is mandatory. In the majority of cases, diagnostic discrepancy between differential diagnosis and pathologic diagnosis was not reported. The typical appearance and the expression of COF usually allow an uneventful diagnosis. Nevertheless, some COFs display atypical clinical-radiographic features, hampering the suitable diagnosis establishment. Moreover, in these cases, the COF may be misdiagnosed with other jaw pathologies, resulting in inadequate clinical management. A multidisciplinary assessment and expertise are essential for managing these patients.(SOLUK-TEKKEŞIN, Merva et WRIGHT, John M., (2017).)

Treatment Strategies and Prognosis

The treatment of COF is an essential part of its management. Depending on the lesion type, numerous strategies from conservative to radical surgery are available. COF with no symptoms and intact tissue between the tumor and important anatomical structures might be managed with observation and follow-up. Surgical removal is recommended in symptomatic cases or in tumors with the penetration of the tumor into vital anatomical structures. In such cases, the surgeon can choose from options of conservative curettage followed by cauterization, enucleation, and marginal osteotomy, as well as more radical solutions, including segmental osteotomy or even composite resection of the mandible or maxilla. The choice should be tailored to the independent clinical and radiological circumstances for every patient and lesion. The follow-up appointment is suggested because of the potential for COF to recur within a few years after the operation.(Kaur T, Dhawan A, Bhullar RS, Gupta S.,(2021))

Various prognostic factors have been suggested as possibly having an impact on prognosis: lesion size

and patient age, but only the extension of the lesion had statistically significant meaning regarding the recurrence of COF. After a 3 to 5-year follow-up, no case of recurrence has been reported. In the 10-year follow-up, the recurrence rate varies between 3.3% and 19.5% after surgical intervention. The maxilla and only maxilla locations present higher rates of recurrence after a long-term follow-up. If the lesion diameter is ≥6.9 cm on preoperative 3D CT, it has an intense relationship with recurrence. Although a limited number of studies have a long-term follow-up and a large sample size, all the above-mentioned numbers should be carefully reviewed. Thus, case-specific considerations are needed for young patient diagnosis, lesion size, and symptoms. The majority of patients present a favorable prognosis after treatment.(Lan YT, Chang SC, Lin PC, Lin CC, Lin HH, Huang SC, Lin CH, Liang WY, Chen WS, Jiang JK, Yang SH., (1983).)

CONCLUSION

In conclusion, the management of Cemento-Ossifying Fibroma (COF) presents unique challenges due to its benign yet locally invasive nature. The two clinical cases discussed highlight the importance of accurate diagnosis and tailored treatment strategies to ensure optimal outcomes. Surgical intervention remains the primary approach, with techniques ranging from conservative curettage to more radical resections depending on the lesion's size and location. The histopathological examination is crucial for confirming COF, as its clinical and radiographic features can overlap with other jaw pathologies. While the prognosis for COF is generally favorable, with low recurrence rates following appropriate surgical management, ongoing monitoring is essential due to the potential for recurrence, particularly in larger lesions. Future research should focus on elucidating the multifactorial etiology and pathogenesis of COF, as well as exploring innovative treatment modalities that may enhance patient outcomes.

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