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Case Report

## Cemento-ossifying fibroma of the jaw

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#### **ABSTRACT**

Background: Cemento-ossifying fibroma is a benign neoplasm characterized by replacement of normal bone by fibrous tissue and varying amounts of newly formed bone or cementum-like material, or both. The cemento-ossifying fibroma has caused considerable controversy because of confusion regarding terminology and the criteria for its diagnosis. In addition, the cemento-ossifying fibroma often shows variations in clinical, radiographic, and histopathologic features, hence require different treatment options. Purpose: This paper attempts to elaborate the classification and terminology of cemento-ossifying fibroma of the jaw, the clinical characteristic, radiographic, and histopathologic features, the different tumor behaviors, and the surgical treatment modalities required. Case: Two patients diagnosed with cementifying fibroma and two patients with ossifying fibroma were reported, presenting their clinical presentation, diagnostic imaging, and histopathology reports, as well as their surgical treatments. Classifications of fibro-osseous lesion of the jaws and characteristics as well as variations in several aspects of cemento-ossifying fibroma of the jaws are discussed. Conclusion: The diagnosis of cemento-ossifying fibroma of the jaw can be established quite consistently based on clinical, radiographic, and microscopic features. However, these tumors may exhibit variations in their neoplastic behaviors. It is therefore important to take into account the individual tumor behavior when one is planning a proper surgical treatment. The behavior of the tumor governs the required surgical treatment which may range from simple curettage of the tumor to radical resection of the jaw.

Key words: cementifying fibroma, ossifying fibroma, tumor's behaviors

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#### INTRODUCTION

The ossifying fibroma is a benign neoplasm characterized by the replacement of normal bone by fibrous tissue and varying amounts of newly formed bone or cementum-like material, or both. As a result of histological similarities, ossifying fibroma, fibrous dysplasia, and cemento-osseous dysplasia are classified together as benign fibro-osseous lesions. The diagnosis of benign fibro-osseous lesions is based on clinical, radiographic, and histopathologic correlation.<sup>1</sup>

Ossifying fibroma is a benign neoplasm usually presented as a painless, slow-growing, expansile lesion which is believed to be confined to the jaws and craniofacial complex.<sup>2</sup> There are numerous similarities between this lesion and the cementifying fibroma, a fibroosseous lesion arising from the periodontal membrane, regarding predilection of age of occurrence, sex, location,

roentgenographic appearance, and clinical behavior. Therefore, the term cemento-ossifying fibroma is now more widely used.

The cemento-ossifying fibroma is odontogenic in origin, whereas ossifying fibroma is of bony origin. Cemento-ossifying fibroma is a fibro-osseous lesion that arises from the periodontal membrane.<sup>3</sup> It contains multipotential cells that are capable of forming cementum, lamellar bone, and fibrous tissue.<sup>4,5</sup>

The cemento-ossifying fibroma has caused considerable controversy because of confusion regarding terminology and the criteria for its diagnosis. In addition the cemento-ossifying fibroma often shows variations in clinical, radiographic, and histopathologic features depending on the nature of the tumors. Majority of the lesions grow slowly and unidentified by the patient until swelling of the face is noted while in other cases some tumor may grow rapidly and cause symptoms. Inadequate surgical treatment may

cause recurrence of the lesions, therefore proper diagnosis and treatment plan are required to achieve good result in the management of this tumor.

In this paper, four patients diagnosed with ossifying fibroma and cemento-ossifying fibroma of the jaw who subsequently underwent surgical treatment are presented.

### **CASES**

Case #1: A 21-years-old male patient came to our clinic with chief complaint of large swelling of left mandible. It was first noted by the patient 8 months ago, has been growing slowly and was not associated with

pain. Clinically, a large swelling was noted on the left buccal region measuring approximately 8 cm in diameter, which was firm and non-tender on palpation (Figure 1-A). Intraorally, a large smooth-surface mass was found on the left mandible extending to the left buccal mucosa. It was firm and non-tender on palpation (Figure 1-B).

Panoramic x-ray showed round shape radiolucency on the left mandible extending from the distal aspect of #34 towards the region of #37. It has sharp and sclerotic margin, showing multiloculation and spots of radiopacity in the centre of the lesion. The teeth #35, 36, and 37 were missing (Figure 1-C). The incisional biopsy of the lesion revealed the microscopic diagnosis of cementifying fibroma.



Figure 1. (A) A large swelling on the left buccal region; (B) The intra oral mass on the left mandible extending to the left buccal mucosa); (C) On panoramic a round shape radiolucency noted in the left mandible showing sharp, sclerotic margin, multiloculation and spots of radiopacity in the centre of the lesion; (D) The tumor appeared as a round, encapsulated mass with multiple foci of whitish component in the centre of the lesion reflecting the tissue with calcified material; (E) Microscopic view showing stroma of fibroblastic proliferation with foci of cementum-like material

The tumor was removed together with the adjacent bone with segmental resection of the left mandible. Upon removal of the bone segment it was found that the tumor was a round, encapsulated mass with multiple foci of whitish component in the centre of the lesion reflecting the tissue with calcified material (Figure 1-D). The defect in the mandible was reconstructed by placement of bridging plate using Coen's stainless steel reconstruction plate. The histopathologic result shows that the tumor consists of fibroblast proliferation with foci of cementum-like material (Figure 1-E) which confirm the diagnosis of cementifying fibroma.

Case #2: A 29-year-old female patient came to our clinic with complaint of painless swelling of the left maxilla. It started as a small mass six years ago which

grew slowly and did not give any symptoms. The mass, however, grew relatively faster in the past six months causing noticeable facial deformity which rendered her to seek medical consult. Clinically, a large swelling was noted over her left cheek and malar region which was firm and non-tender on palpation (Figure 2-A). Intra orally, a rounded, smooth-surface, well-circumscribed mass was found on the buccal side of maxilla from the region of #21 through #26. The mass was also found on the posterior part of the left hard palate extending nearly to the midline (Figure 2-B).

Panoramic x-ray show a large mass in maxilla extending from #11 region towards that of #26 and involves the nasal cavity as well as the left maxillary sinus. The tumor consists of a blend of radiopaque and radiolucent components and

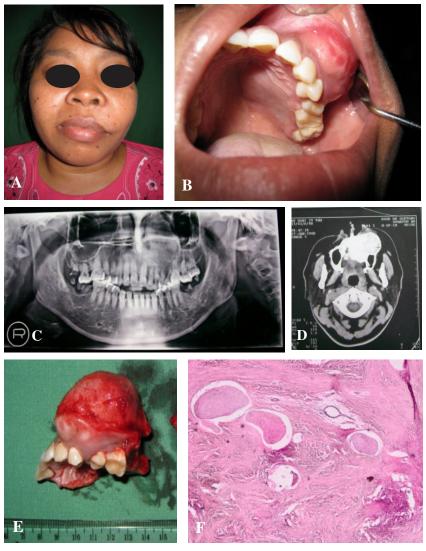


Figure 2. (A) A large swelling on the left cheek and malar region; (B) Intra orally, a rounded, smooth-surface, well-circumscribed mass found on the buccal side of maxilla and over the posterior part of the left hard palate extending nearly to the midline; (C) Panoramic x-ray showing a blend of radiopaque and radiolucent components with defined boundaries showing sclerotic margin; (D) CT scan showing the extension of the mass to nasal cavity as far as *middle nasal conchae* and towards medial part of the left maxillary sinus; (E) Macroscopically, the tumor was well encapsulated on its buccal part which was easily shelled out from the surrounding bone whereas the medial part had relatively poor margin requiring maxillectomy; (F) The microscopic view of the lesion showing proliferation of spindle-shape cells with multiple formation of bony trabeculae showing osteoblastic rimming at the periphery of the trabeculae.

has defined boundaries showing sclerotic margin (Figure 2-C). Computed Tomogram scan shows a round tumor mass at the buccal side of the left maxilla which extends to hard palate, nasal cavity as far as middle nasal conchae, and towards medial part of the left maxillary sinus (Figure 2-D). Incisional biopsy of the lesion revealed cementifying fibroma.

The surgery was done using Weber-Fergusson approach, followed by complete removal of the whole tumor from the left maxilla. The tumor was found to be rounded and well encapsulated on its buccal part but has a relatively poor margin on its medial part, therefore resection of the left maxilla, or hemimaxillectomy, was done (Figure 2-E). The defect in the maxilla after resection was reconstructed by packing it up with vinyl polysiloxane impression material (putty type, Exaflex<sup>TM</sup>, GC, Japan) supported with acrylic surgical obturator serving as a base plate. The histopathology examination of the lesion shows proliferation of cellular

fibrous connective tissue with multiple formations of bony trabeculae showing osteoblastic rimming at the periphery of the trabeculae (Figure 2-F). These findings support the diagnosis of ossifying fibroma.

Case #3: A 17-year-old female patient came with a large swelling of the right mandible which had been growing from a small mass in the gum since 10 years previously. The mass never caused pain except that there had been episodes of bleeding from the tumor recently. Clinically, a large swelling was noted over her right mandible which was firm and non-tender on palpation (Figure 3-A). Intra orally, the tumor was found on the buccal side of the mandible obliterating the mucobuccal fold over the region of #45 to the right retromolar area. The lower right molars were severely displaced lingually. The mass was firm and not tender on palpation. The lingual side of the mandible was normal (Figure 3-B).

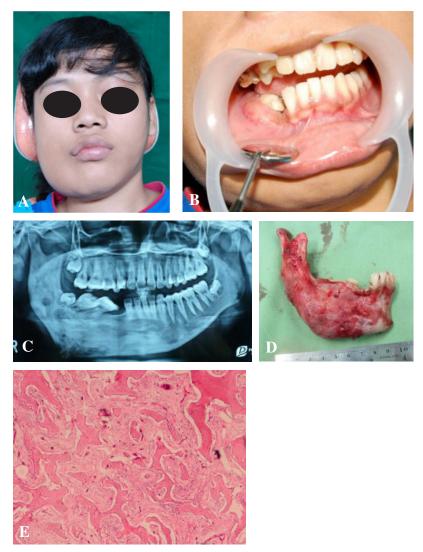


Figure 3. (A) A large swelling over the patient's right cheek involving the lower border of the mandible. (B) Intra orally, the tumor noted on the buccal side of the mandible obliterating the mucobuccal fold; (C) On panoramic the lesion noted as irregularly-shaped, intrabony lesion with ill-defined margins showing flecks of radiopacity within the radiolucent area indicating increased calcification of the tumor; (D) The resected mandible showing a well encapsulated mass involving the body and ascending ramus of the mandible; (E) Microscopic view showing formation of bone trabeculation within fibrous connective tissue with osteoblastic rimming being strongly evident at periphery of the trabeculae.

Panoramic x-ray showed irregularly-shaped intrabony lesion with ill-defined margins extending from the region of tooth #33 to the right ascending ramus; flecks of radiopacity were seen within the radiolucent area indicating increased calcification of the tumor (Figure 3-C). The incisional biopsy of the tumor showed the microscopic features of ossifying fibroma.

The surgical treatment performed in this case was hemimandibulectomy of the right mandible extended to the left parasymphysis distal to tooth #33. Following resection of the mandible it was found that the tumor was a well encapsulated mass involving the body and ascending ramus of the mandible and causing destruction of the lower border of the mandible (Figure 3-D) but the lingual part of the mandible was normal. Reconstruction of the mandible was performed using autogenous rib graft which was supported with stainless steel reconstruction

plate. The histopathologic examination shows formation of bone trabeculation within fibrous connective tissue with osteoblastic rimming being strongly evident at periphery of the trabeculae (Figure 3-D). These findings confirmed the diagnosis of ossifying fibroma.

Case #4: A 30-year-old female patient came with a swelling on her right mandible of six months duration. There had been no pain or chewing problem except for facial deformity caused by the swelling. Clinically, there was a marked swelling on the right parasymphyseal region and body of the mandible which was firm and non-tender on palpation (Figure 4-A). Intra orally, the tumor appeared as obliteration of the vestibulum over the region of #43 to #46, slightly displacing tooth #45 (Figure 4-B). The mass was firm and not tender on palpation and the lingual side of the mandible was normal.

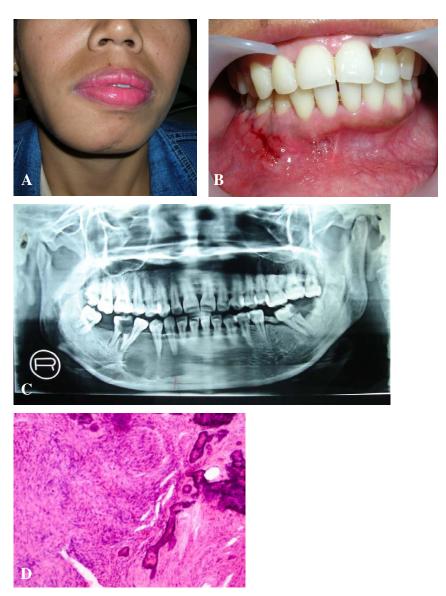


Figure 4. (A) An oval-shaped swelling on the right parasymphysis region and body of the mandible; (B) intra orally, the tumor appeared as obliteration of the vestibulum oris; (C) panoramic x-ray showing oval-shaped radiolucency at the body of the mandible which has distinct boundaries but lacks sclerotic margins, flecks of radiopacity is seen within the lesion; (D) The microscopic examination showing cellular fibrous connective tissue with multiple foci of mineralized component resembling the features of cementum.

Panoramic x-ray showed that the tumor appeared as radiolucency at the body of the mandible from #42 to mesial of #46, which was somewhat oval in shape, had distinct boundaries but lacks sclerotic margins. The anterior extension of the lesion was not very clear in the panoramic x-ray due to its superimposition with the cervical bony structure. A small amount of radiopaque component was noted in the center of the lesion (Figure 4-C). The incisional biopsy of the tumor revealed the microscopic features of cementifying fibroma.

The surgical treatment performed in this case was en bloc resection of the right mandible followed by ostectomy of the surrounding bone using large round surgical bur, leaving the lingual plate of the mandible intact. Coen's stainless steel reconstruction plate was subsequently placed across the defect, serving as a stabilizing plate, to prevent the mandible from pathologic fracture (Figure 4-D). The histopathology examination of the lesion shows cellular fibrous connective tissue with multiple foci of mineralized component without osteoblasting rimming, the feature resembling that of cementum (Figure 4-d). These findings confirmed the diagnosis of cementifying fibroma.

#### DISCUSSION

Maxillofacial fibro-osseous lesions comprise a group of face and jaw disorders characterized by the replacement of normal bone by a benign connective-tissue matrix with varying amounts of mineralized substances.<sup>7</sup> The designation "fibro-osseous lesion" is not a specific diagnosis and describe only a process. Fibro-osseous lesions of the jaws were initially classified by Waldron<sup>8</sup> into three main categories namely, fibrous dysplasia, fibro-osseous (cemental) lesions such as ossifying and cementifying fibroma, and fibro-osseous neoplasms such as juvenile active ossifying fibroma. The concept of 'fibro-osseous lesions' of bone has evolved over the last several decades and now includes two major entities: fibrous dysplasia and ossifying fibroma, as well as the other less common lesions such as florid osseous dysplasia, periapical osseous dysplasia, focal sclerosing osteomyelitis, proliferative periostitis of Garre, and ostitis deformans. 6 In recent years, these lesions were reclassified into fibrous dysplasia, reactive (dysplastic) lesions arising in the tooth-bearing area, and fibrous osseous neoplasms such as cementifying and ossifying or cemento-ossifying fibroma. 9,10 In contrast, based on nomenclature by Kramer et al.11 the cementoossifying fibroma is described as an osteogenic neoplasm and the fibrous dysplasia as a non-neoplastic bone lesion.

A neoplastic etiology of ossifying fibroma is supported by examples of lesions that achieve a large size, exhibit aggressive behavior, and produce significant osseous destruction.<sup>2</sup> Additionally, recurrences, though rare, have been described in some studies of ossifying fibroma. Chromosomal translocations have been identified in a few cases of ossifying fibroma, however, the molecular mechanisms that underlie the development of this tumor remain unknown.<sup>2</sup>

The cemento-ossifying fibroma has caused considerable controversy because of confusion regarding terminology and the criteria for its diagnosis. The cemento-ossifying fibroma is odontogenic in origin, whereas ossifying fibroma is of bony origin. Cemento-ossifying fibroma is a fibro-osseous lesion that arises from the periodontal membrane.<sup>3</sup> It contains multipotential cells that are capable of forming cementum, lamellar bone, and fibrous tissue.<sup>5</sup> A close histogenetic relationship exists between the central ossifying fibroma and the central cementifying fibroma. It is based on the marked similarity between the two regarding predilection of age of occurrence, sex, race, location, roentgenographic appearance, and clinical behavior, these two lesions represent the same basic neoplastic process. The only difference between the two being in the type of cell involved and its end product-cementum in one case and bone in the other. This has prompted many to use the term cemento-ossifying fibroma.<sup>2</sup>

Its occurrence in anatomical regions, not associated with periodontal membrane, is unexplained. It was supposed that pluripotential mesenchymal cells could differentiate, as does the periodontal ligament, to produce calcified material resembling bone and cementum, as well the presence of ectopic periodontal membrane has been hypothesized. Despite its origin in the periodontal membrane, the factors that stimulate this structure to produce cementum in an aberrant anatomical site remain controversial. Inflammation secondary to either infections or trauma has been proposed as a causative agent. 13

Clinically, the cemento-ossifying fibroma presents as a painless, slowly growing mass in the jaw where displacement of teeth may be the only early clinical feature. The lesion is therefore frequently ignored by the patient until the growth produces a noticeable swelling and facial deformity. The tumor is well-circumscribed from its surrounding bone and will continue to grow bigger, slowly or actively, until it is removed surgically. 11 These seem to be the case in all of our patients considering that they have ignored the masses in their jaws as they had been asymptomatic and that the lesions had all grown considerably big when they first came to our clinic. All of the three cases in the mandible above showed only buccal bony extension with the lingual bone being normal. This is in contrast to Sapp et al. 14 who suggest that cementoossifying fibroma often exhibit marked buccal and lingual bony expansion.

Cemento-ossifying fibroma has a marked predilection for female sex, the female: male ratio being 2:1.<sup>6,14</sup> Central cemento-ossifying fibromas are more commonly found in the mandible than in the maxilla<sup>6</sup> some reports indicate 90 percent of all cases are located in the mandible.<sup>10</sup> In mandible, it occurs particularly in the premolar-molar region.<sup>1,2,6,15</sup> These characteristic clinical features of cemento-ossifying fibroma support several facts in the our cases where three of the four cases occurred in females,

three of the four cases were found in mandible, and all of the tumors in the mandible occurred in the premolar-molar region. Regarding the age of onset, one case was found in the second decades of life, one case in the early third decades and the other two cases in their late third decades of life. This is in accordance with the majority of literatures<sup>3,11</sup> which show that cemento-ossifying fibroma occur mainly in the second to the fourth decades of life.

The radiographic appearance is of utmost importance in the diagnosis of cemento-ossifying fibroma because it is often needed to separate it from other fibro-osseous lesions. The lesions may be either unilocular or multilocular. <sup>14</sup> All of the tumors in the mandible above radiographically appeared as multilocular lesion and the tumor in maxilla appeared as a unilocular lesion. In the early stages, the cementoossifying fibroma appears as a radiolucent lesion with no evidence of internal radiopacities. As the tumor matures, there is increasing calcification so that the radiolucent area becomes flecked with opacities until ultimately the lesion appears as an extremely radiopaque mass. The cementoossifying fibroma presents a radiolucent appearance in 53%, a sclerotic radio density in 7% and mixed or mottled appearance in 40% of the cases. 12 Variation in the amount of radiopacities and radiolucency are seen in our reported cases. The first and fourth case show small amount of radiopaque component whereas in the second and third case much larger amount of radiopacities are found in the centre of the lesions. Interestingly, the differences in the amount of opacities within the lesion seem to have correlation with the duration of tumor themselves. The tumors in the first and fourth case were of eight and six months duration respectively, whereas those found in the second and third case were six and ten years old respectively. These facts strongly suggest that the older the tumor the larger the amount of the calcified material found within the lesion.

One additional important diagnostic feature radiographically is that there is a centrifugal growth pattern rather than a linear one and therefore the lesions grow by expansion equally in all directions and present as a round tumor mass. This characteristic rounded-shape is reflected in three of the cases reported herein, only one case in the mandible exhibiting irregular shape.

There are three different patterns of radiographic borders of cemento-ossifying fibroma which are: defined lesion without sclerotic border (40%); defined lesion with sclerotic border (45%); and lesion with ill-defined border (15%) indicating a rapidly growing tumor. <sup>12</sup> These variations in the tumor borders are also evident in our cases. The first two cases showed defined margins with sclerotic border, the fourth case exhibited defined margin but lacked sclerotic border, while the third case showed ill-defined border. The case with ill-defined border has, actually, a history of ten year old tumor which seemingly does not support the theory which stated that ill-defined border indicated a rapidly growing tumor. However, since there was a history of bleeding episodes experienced by the patient over the past few months it is possible that the tumor

may have grown rapidly within the bone so that it has made the border of the tumor became less distinct.

The characteristic macroscopic features of this tumor is replacement of normal bone by a benign connective-tissue matrix with varying amounts of mineralized substances, however, there are some variations in microscopic features of this tumor. The microscopic findings mirror the radiographic findings. The more radiolucent lesions are composed of cellular fibrous connective tissue, frequently in a whorled pattern. 14 Collagen fibers are often arranged haphazardly, although a whorled, uniform pattern may be evident. Calcified deposits are noted throughout the fibrous stroma. The nature of the hard tissue is generally quite variable within a given tumor as well as between lesions. Irregular trabeculae of woven bone or lamellar bone are most consistently noted in these tumors. Additional patterns of calcified material include small, ovoid to globular, basophilic depostis and anastomosing trabeculae of cementum-like material.<sup>2</sup> These variations in hard tissue configuration make no difference to the clinical behaviour of the tumour. However, recognition of these structures is important in establishing its diagnosis. 16 Osteoblast may or may not be evident at the periphery of the bone deposits. A thin outer zone of fibrous connective tissue is usually present, separating the fibro-osseous tissue from the surrounding normal bone. 14 The microscopic examination of all of the cases presented above show characteristic calcified material within fibrous connective tissue which are the indicative features of cemento-ossifying fibroma. In the first and fourth case the foci of calcified material observed show the characteristic feature of cementum hence the term of cementifying fibroma. In the second and third case, however, the foci of calcified material appear as bony trabeculae with evidence of osteoblastic rimming at the periphery of the trabeculae therefore the term ossifying fibroma is applied as the histopathologic diagnosis.

Treatment of cemento-ossifying fibroma generally has been by conservative enucleation or curettage or radical surgery<sup>1,2,11</sup> depending on the size and location of the individual lesion.4 They are characterized by easy shell out from the surrounding bone.<sup>2</sup> Conservative surgery is therefore recommended even if the tumour is large with bowing and erosion of the inferior border of the mandible and radical treatment of the tumour such as an en bloc resection should only be considered if there are recurrences due to its aggressive nature. 11 Slootweg and Muller 18 reported that there was no difference in outcome between patients treated in a more limited way and those treated by major surgery. Other authors, however, advocate more extensive surgery for more aggressive lesions and lesions involving craniofacial bones in light of the potential for recurrence. 19,20 Sakoda et al. 17 described the procedure of a segmental resection of an extensive ossifying fibroma with the replacement of the excised segment with immediate reconstruction. Eversole and his coworkers<sup>19</sup> in a study of 64 cases of cemento-ossifying fibroma reported a recurrence rate of as high as 28 per cent following surgical curettage of these lesions.

It is interesting to note that all of our current cases were treated radically comprising en bloc and segmental resection of the mandible in the fourth and first case respectively, hemimandibulectomy with autogenous bone graft in the third case, and surgical excision combined with radical resection of the maxilla in the second case. In the author's opinion they were reasonable sort of treatment since most of the patients came with relatively large tumors and had histories of rapid increase in size which might indicate increase aggressiveness of the lesion. Moreover, it is almost impossible to accomplish complete excision of the tumor in cases when the size of the tumor is extensively large only with surgical curettage through intra oral approach, not to mention the higher risk for mandible fracture following curettage procedure especially if the inferior border of the mandible had been involved in the tumor. The radical surgical treatments in the above cases were, therefore, aimed at eliminating the risk of tumor recurrence as well as the risk of pathological fracture of the jaw following tumor exicision. It is useful to note, however, that since ossifying fibromas do not display infiltrative patterns into bone Booth<sup>2</sup> suggest smaller margins than the 1 cm typically required for ameloblastoma, odontogenic myxoma, or calcifying epithelial odontogenic tumor.

En bloc resection followed by surgical ostectomy performed in the fourth case was the least aggressive surgical treatment in this case series since there was no history of rapid tumor growth, clinically and radiographically it was relatively not aggressive, and there still remained sufficient amount of bone in the inferior border of the mandible after excision of the tumor.

As the conclusion of this paper, although it is relatively not difficult to establish the diagnosis of cemento-ossifying fibromas from clinical, radiographic, and microscopic features, these tumors may exhibit variations in their neoplastic behaviors. It is, therefore, important to take into account the individual tumor behavior when one is planning a proper surgical treatment in order to eliminate the tumor completely and avoid tumor recurrence and at the same time improve the patient's cosmetic and functional problems.

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