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Cemento-ossifying fibroma of the left mandible: A case report

Christian INWS *et al.* COF of the left mandible

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Abstract

BACKGROUND

Ossifying fibroma is a type of benign fibro-osseous lesion. Most lesions affect the mandible region, particularly the molar and pre-molar areas. It predominantly affects females between the second to fourth decades of life. Larger ossifying fibroma tumors require more extensive resection.

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CASE SUMMARY

A 39-year-old female complained of occasional pain and tumor enlargement on her left jaw for the past three years. Intra-oral examination revealed a firm swelling on her left lower gum. Extra-oral examination revealed swelling on the left mandible body with no erythema and superficial ulcer. Computed tomography scan revealed a circular-shaped lesion on the patient's left mandible body with a well-defined radiolucent border, sized $3.2 \times 2.8 \times 0.9 \text{ cm}^3$. The tumor was removed by marginal mandibulectomy. Biopsy from the resected tumor suggested cemento-ossifying fibroma (COF).

CONCLUSION

COF is often unnoticed, but this slow-growing tumor can cause significant symptoms regarding its distortion into adjacent structures.

Key Words: Cemento-ossifying fibroma; Ossifying fibroma; Mandible; Marginal Mandibulectomy; Biopsy; Case report

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⁷ **Core Tip:** Ossifying fibroma is a benign fibro-osseous lesion that predominantly affects the craniofacial region. It is considered a rare tumor, with most lesions affecting the mandible region, particularly the molar and pre-molar areas. It is asymptomatic in most cases, but it can slowly grow until it is involved with the adjacent structures, leading to symptoms. The treatment is surgical excision, and it can be safely performed due to its well-demarcated nature. Recurrences can be avoided by extensive removal of the tumor. This case highlights a case of a rare tumor with surgical excision and a good outcome.

INTRODUCTION

Ossifying fibroma is one of the benign fibro-osseous lesions based on Waldron's classification, which predominantly affects the craniofacial region^[1]. It is considered a rare tumor, with most lesions affecting the mandible region, particularly the molar and pre-molar areas. It predominantly affects females with an incidence of 5:1 ratio compared to males and is commonly encountered between the second to fourth decade of life. It has characteristics of a slow-growing tumor, sometimes asymptomatic and expansive lesion, which, if left untreated, may lead to signs and symptoms caused by enlarged mass and significant involvement to the adjacent vital structures^[2]. During its early stage, the tumor may appear small and well-demarcated, which can be safely enucleated. However, larger tumors require more extensive resection, in which reconstructions are sometimes needed^[3]. In this study, we intend to report a clinical presentation of the cemento-ossifying fibroma (COF) on the patient's left mandible, which was treated with lip-splitting incision and tumor excision through marginal mandibulectomy. This study aims to share our experience in dealing with mandible COF and associated surgical approach.

CASE PRESENTATION

Chief complaints

A 39-year-old female patient complained of occasional pain in her left jaw for the past three years. Shortly after, the patient noticed a small tumor on her gum, precisely below the first pre-molar tooth on her left mandible.

History of present illness

The tumor was slowly growing in size, and the pain frequency had increased more than before. The patient had no other comorbidities. The patient had undergone a panoramic radiographic examination, which revealed a singular, well-demarcated, circle-shaped lesion in the left mandible body below the second molar and first pre-molar teeth. Based on patient's perspective, she felt that the tumor did not increase in size since her latest visit to the dentist.

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History of past illness

The patient had no history of trismus and difficulty in masticating, although pain occasionally emerged during eating.

Personal and family history

Patient also did not have any previous medical conditions and other family history.

Physical examination

Intra-oral examination revealed a firm swelling on her left lower gum, precisely below her previously drilled first pre-molar tooth. Mild tenderness was observed on percussion, but no tooth mobility was encountered. There was minimal swelling on the left mandible body with no erythema, superficial ulcer, and no other facial asymmetry on the extra-oral examination. When palpated, the tumor was fixated with hard consistency, similar to bone (Figure 1).

Laboratory examinations

A routine blood test was taken, and the results were within normal limits.

Imaging examinations

A computed tomography (CT) scan with 3D reconstruction revealed a circular-shaped lesion on the patient's left mandible body with a well-defined radiolucent border between the lesion and surrounding normal bone, sized $3.2 \times 2.8 \times 0.9 \text{ cm}^3$ (Figure 2) fine-needle aspiration biopsy from the tumor suggested COF of the mandible. A routine blood test was taken, and the results were within normal limits. Based on these results, the patient was scheduled to undergo tumor removal with marginal mandibulectomy.

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FINAL DIAGNOSIS

The final diagnosis of the presented case is COF of the mandible.

TREATMENT

The patient was then admitted two days prior to the elective surgical procedure. On the operation day, ⁴ the patient was positioned supine with her head slightly tilted to the opposite side. The patient then underwent general anesthesia with nasotracheal intubation. Lip-splitting incision was performed and extended towards the patient's left angulus mandibulae. After the left mandible and the bony tumor were completely exposed, marginal mandibulectomy was performed. Marginal mandibulectomy was the preferred option in order to achieve 0.5 cm tumor margins. As we preserved the periosteum, we hoped the remaining periosteum will naturally construct the bone defects, and reconstruction won't be necessary in the future. Mandible plate fixation was not performed, as we maintained the integrity of remaining mandible bone so it will not cause soft tissue collapse, which we observed in the next upcoming follow up examinations. The wound was then washed with normal saline solution, minor bleeding was controlled, and drain insertion and wound closure using a subplatysmal flap were performed (Figure 3). The excised tumor was sent for pathology examination and described as an ossifying fibroma. From 10 × 10 magnification, it showed the bone trabecular component and fibrous connective tissues along with fibroblast cell proliferation, and from 10 × 40 magnification, it showed as spindle-shaped cell with cigar-shaped nucleus, eosinophilic cytoplasm and smooth chromatin (Figure 4).

1 OUTCOME AND FOLLOW-UP

The patient had an uneventful postoperative clinical course with no postoperative complications. The drain was removed in three days postoperative and showed no further rebleeding. At a follow-up visit in three months after the surgery, the pain was resolved and she was asymptomatic. Panoramic radiographs were performed every 6 mo and CT scan with 3D images were taken one year after surgery with satisfactory results without any recurrent lesions.

DISCUSSION

COF are benign neoplasm that primarily affect the mandible and other craniofacial areas. COF consists of fibrous tissue, bone, and cementum in different proportions^[1]. COF is one out of three variants of ossifying fibroma; the other variants are juvenile trabecular ossifying fibroma and juvenile psammomatoid ossifying fibroma^[2]. These other variants reflect different demographics, their tendency to progress into malignant lesions, and their overall prognosis. The World Health Organization classifies COF as one of the fibro-osseous neoplasms, part of non-odontogenic tumors originating from periodontal ligament mesenchymal blast cells. It can progress into fibrous tissue, bone, cementum, or a combination of all three, as mentioned before^[3]. However, this theory is still debatable, pointing to recent microscopic studies of COF arising from frontal, sphenoid, temporal, and ethmoid bones^[4].

COF is asymptomatic in most cases, starting as a small unidentified tumor, but slowly grows until it becomes noticeable as a swelling on the patient's face and causes symptoms associated with its disturbance towards other structures. Due to its well-demarcated nature, enucleation and curettage of this lesion can be safely performed. However, larger lesions require more radical approaches, and inadequate surgical excision may result in recurrences^[5]. One case report in 2021 by Guddadarangiah *et al*^[6] presented a large mandible COF infiltrating parapharyngeal and infratemporal spaces, which required a hemi-mandibulectomy procedure. Another report in 2015 presented COF in a similar area to our case report but larger in size and treated with segmental mandibulectomy^[7].

COF predominantly affects the mandible and commonly occurs in the molar and pre-molar areas. However, there were cases of COF found elsewhere other than the mandible area, such as in maxilla, orbital and ethmoidal regions. From a case series of 16 COF cases, 50% of these cases were found in the maxilla and the other 50% in the mandible region. There was one case of COF found in the right zygoma region with clinical presentation of diffuse swelling extending toward the upper vestibule^[8]. Some COF cases in the maxilla region can cause facial deformities, sinus obstruction, and intra-

orbital and intracranial discomfort due to their involvement in adjacent structures. A recent report in 2020 presented a large neglected COF of the maxilla region with a complaint of significant pressure on her left eye^[4].

COF, regardless of their variant, location and size, are neoplasms that require surgical excision. Depending on its size and location, treatment can be divided into conservative or radical surgery. While some authors reported no significant outcome between conservative and radical excision, others had suggested extensively removing the tumor, particularly aggressive lesions, to avoid any recurrences^[5]. We preferred to extensively remove the tumor to avoid any recurrences in the future. A marginal mandibulectomy is a suitable option to remove the tumor adequately and safely. Lip-split incision is also preferred as a method of choice to expose the mandible adequately, as there are no significant differences in postoperative complications compared with the visor approach based on a retrospective review in 2018^[9]. Marginal mandibulectomy was considered a feasible procedure, as the tumor had approached mandible without infiltration toward tooth sockets and dental rehabilitation wasn't planned after surgery^[10].

In other studies, all mandible COFs are treated locally. Kaur *et al*^[8] in 2019 reviewed 16 maxillofacial COF cases, half of them affect mandible region. Ten out of 16 cases were treated with enucleation and curettage, five cases were resected locally and 1 case was resected en bloc with other involves structures. All patients did not develop any recurrences from follow up observation. There were other studies who managed this case extensively either by segmental or even hemimandibulectomy due to its massive size and mandible plate reconstructions were performed^[4-7]. One study reports a similar approach from our study, in which the tumor was safely excised with preservation of inferior mandible border and without any reconstruction. Unfortunately, the outcome and follow up reports were not mentioned^[11].

One study interestingly preserved the mandible periosteum after large COF enucleation. They used periosteum osteogenesis potential to induce bone regeneration with satisfactory result and no COF recurrence after 3 years of follow-up. It is based on

periosteum ability as barrier to prevent soft tissue migration, periosteum bone cells and its rich vascular supplies to support adequate bone growth. With periosteum preservation, we can expect bone restoration process from post-excision defect area in our patient and reconstruction won't be necessary, although Shirafkan *et al*^[12] and associates did reconstruct with mandible plate and screws.

CONCLUSION

COF is a benign neoplasm, usually asymptomatic, predominantly affecting the mandible with a molar and pre-molar tooth as its predilected site. It is often unnoticed, but this slow-growing tumor can cause significant symptoms regarding its distortion into the adjacent structures. COF requires surgical excision and larger tumor needs a more extensive approach. Recurrences after removal are usually rare.

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