



Patterns of Ossifying Fibromas in the Maxillofacial Skeleton: A Case Series and Literature Review.

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ABSTRACT: Ossifying fibroma is a benign fibro-osseous neoplasm of the jaws characterized by the replacement of normal bone with a well-demarcated mass containing fibrous tissue and varying amounts of mineralized material. Although slow-growing, these lesions may attain considerable size and produce facial asymmetry, tooth displacement, or functional impairment if not detected early. This case series highlights the clinical presentation, radiographic features, and surgical management of patients diagnosed with ossifying fibroma of the mandible and maxilla. All cases demonstrated well-defined, unilocular or multilocular radiolucencies with varying radiopaque foci, consistent with the lesion's maturation stage. Surgical enucleation or resection was performed based on lesion size, location, and cortical expansion. Histopathologic examination in all patients confirmed the diagnosis by revealing a fibrous stroma containing trabeculae of woven bone and cementum-like calcifications. Postoperative follow-up showed satisfactory healing without evidence of early recurrence. This series emphasizes the importance of correlating clinical, radiologic, and histologic findings for accurate diagnosis and appropriate treatment planning. Early intervention and complete surgical removal remain essential to minimize recurrence and preserve jaw function. A brief review of current literature is included to contextualize the evolving understanding of this lesion's behavior and optimal management strategies.

Introduction

Fibro-osseous lesions (FOLs) of the craniofacial region represent a diverse group of conditions characterized by the replacement of normal bone with a hypercellular fibrous stroma containing varying amounts and patterns of mineralized or osseous tissue [1]. Ossifying fibroma is a fibro-osseous neoplasm involving either jaw, characterized by the proliferation of fibroblasts along with the formation of mineralized tissue such as bone and

cementum-like material. Menzel first described a variant of ossifying fibroma, referring to it as a "cemento-ossifying fibroma" [3]. Later, in 1927, Montgomery proposed the term "ossifying fibroma," which remains the standard nomenclature. Subsequent authors have widely adopted this terminology. Before 1948, ossifying fibroma and fibrous dysplasia were often considered identical or simply different presentations of a single disorder. This view changed when Sherman and



Sternberg documented the key clinical, radiologic, and histopathologic characteristics of ossifying fibroma, establishing it as distinct from fibrous dysplasia.^[4,5,6]

The 1992 WHO classification of odontogenic tumours adopted the term cemento-ossifying fibroma and, for the first time, identified a distinct, rapidly growing variant termed juvenile aggressive ossifying fibroma^[21]. In the 2005 WHO classification, this subgroup was further delineated into juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF), based on differences in calcification patterns, age of occurrence, anatomical predilection, and recurrence tendencies^[22]. In the 2017 WHO classification, the terminology was revised again, reinstating the term cemento-ossifying fibroma (COF) to more accurately reflect its origin within the tooth-bearing regions of the jaws and its designation as a benign mesenchymal odontogenic tumour likely derived from the periodontal ligament^[23]. Accordingly, three distinct categories of ossifying fibroma were recognized: COF as an odontogenic tumour, and JTOF and JPOF as non-odontogenic variants classified under benign fibro- and chondro-osseous lesions. Cemento-ossifying fibroma (COF), previously classified in 2017 as a benign mesenchymal odontogenic tumor but discussed under fibro-osseous and osteochondromatous lesions, was fully reclassified in 2022 by the WHO as a benign mesenchymal odontogenic tumor^[9]. It is now clearly distinguished from the non-odontogenic, juvenile trabecular, and psammomatoid variants. In the 2022 classification, ossifying fibromas are divided into two main groups based on origin: odontogenic and craniofacial (non-odontogenic). The cemento-ossifying fibroma (COF) remains the variant considered to have odontogenic origin and is restricted to the tooth-bearing parts of the jaw^[8]. Accordingly, the term ossifying fibroma (OF) is applied when the lesion is primarily composed of bone, whereas cemento-ossifying fibroma is used for lesions containing both osseous and cementum-like components. Ossifying fibroma (OF) is a benign, non-odontogenic fibro-osseous lesion that emerges from the mesenchymal blast periodontal ligament cells, which consist of fibrous tissue, bone, and cementum-like substance. Unlike in the 2017 classification, juvenile trabecular ossifying fibroma (JTOF) and juvenile psammomatoid ossifying fibroma (JPOF) are now classified separately from odontogenic.

Notably, the term “juvenile” was included in the names of both variants in the 2017 edition^[8].

Although neither variant occurs exclusively in young patients, both show a marked preference for individuals in the first and second decades of life, with the trabecular form demonstrating a stronger predilection for younger age than the psammomatoid type. Consequently, the term “juvenile” was removed from the psammomatoid ossifying fibroma designation in the 2022 classification. (Figure 1)

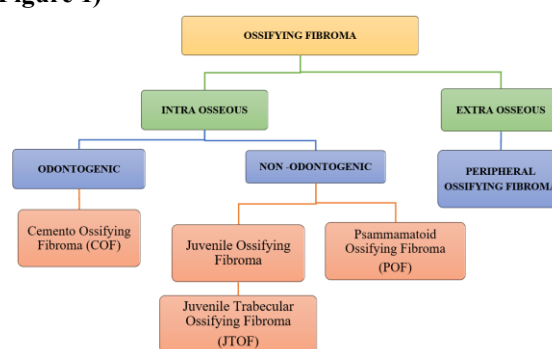


Figure 1 Classification of ossifying fibroma (WHO 2022; 5th edition)

Because these lesions share similar histologic patterns, an accurate diagnosis depends on carefully correlating the patient’s history with clinical presentation, radiographic characteristics, and microscopic findings^[10].

Here, we present four variants of ossifying fibroma (COF, juvenile Psammomatoid OF, juvenile trabecular OF, and Peripheral OF), along with their clinical, radiographic, histological, and management strategies.

CASE-1

A 14-year-old systemically healthy male patient presented to our department with a chief complaint of left facial swelling for 1 month. The extraoral examination revealed asymmetry caused by diffuse swelling in the left maxillary region, which was painless, nontender & hard in consistency. The skin overlying the swelling was stretched and usually coloured, with no secondary changes noted. The swelling started spontaneously and gradually increased to its current size. There was no history of trauma or associated dental pain. An intraoral clinical examination revealed a painless, diffuse, hard, sessile swelling in the upper left buccal vestibule extending anteroposteriorly from the left canine to the



second molar region (23 to 27 region). The mucosa overlying the swelling was smooth, similar to the adjacent mucosa, with no secondary changes seen. Clinically, no displacement of teeth was seen, and his mouth opening was standard, and no lymphadenopathy was found. Laboratory results were within normal ranges, and the family and medical history were otherwise normal.

Radiological investigations include cone-beam computed tomography. OPG/CBCT examination revealed a well-defined, large expansive radiolucency involving the left maxilla, measuring approximately 4×4centimeters (sagittal/axial/coronal planes). The lesion is extending from the roots of 13 to 16 regions, with superior extension into the maxillary sinus, complete erosion of buccal cortical plates, and thinning of the palatal wall, with gross displacement of adjacent teeth without any root resorption.

FNAC shows negative aspiration. An incisional biopsy was performed and sent for histopathological examination. The lesion consists of a well-defined fibrous connective tissue stroma, which is moderately cellular and contains plump spindle-shaped fibroblasts. The stroma comprises numerous basophilic, calcified deposits that resemble woven bone trabeculae with osteoblastic rimming. Based on clinical, radiographical, and histopathological findings, it was diagnosed as cemento-ossifying fibroma (COF). Surgical excision of the lesion was done under general anaesthesia along with the removal of the involved teeth 13 to 17, and the patient was under follow-up for recurrence (**Figure 2**).



Figure 2 Cemento Ossifying Fibroma (COF)
A) Extraoral B) Intraoral C) OPG D) CBCT
E) Surgical excision F) H/E staining

CASE-2

A 19-year-old female patient reported to our department with the chief complaint of swelling in her chin region for 2 years. The extraoral examination revealed asymmetry caused by diffuse swelling in the symphysis region, which was painless, nontender & hard in consistency. The skin overlying the swelling was stretched and was normally coloured with no secondary changes. Initially, the swelling was minor in size and gradually increased to its current size. There was no trismus, numbness, toothache, history of facial trauma, or previous similar swelling. An intraoral examination revealed a painless, nontender, diffuse, hard swelling in the lower anterior region extending from the 44 to the 36 areas with obliteration of the buccal vestibule. The mucosa overlying the swelling was smooth, similar to the adjacent mucosa, with no secondary changes seen.

Panoramic radiography showed an ill-defined, expansive, radio-opaque lesion extending from 44 to the mesial root of 36, with gross displacement of anterior & premolar teeth and thinning of the inferior border of the mandible. A radio-opaque mass was seen at the inferior border of the mandible within the lesion, suggestive of an impacted tooth irt 33. Cone beam computed tomography (CBCT) revealed that the size of this lesion was 6×6cm. The lesion expanded the inferior border of the mandible with thinning of cortical bone, but did not involve the right mental foramen.

An incisional biopsy was performed and sent for histopathological examination. The lesion consisted of numerous ossicles of psammomatous calcifications within a cellular, fibrous, spindle cell stroma; the psammomatous calcifications were basophilic and acellular, features suggestive of Psmmomatoid Ossifying Fibroma (POF). Surgical excision of the lesion was done under general anaesthesia, with the removal of involved teeth and the impacted tooth. The patient was under follow-up for recurrence (**Figure 3**).

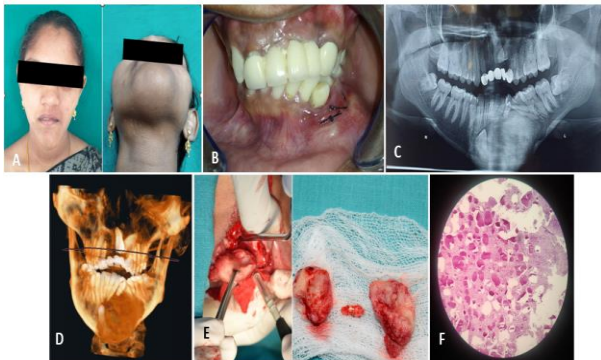


Figure 3 Psammomatoid Ossifying Fibroma (POF)

A) Extraoral B) Intraoral C) OPG D) CBCT
E) Surgical excision F) H/E staining

CASE-3

A 12-year-old male patient presented to our department with a history of swelling on the Right side of his face since 3months. Extraoral examination revealed a diffuse, painless, nontender & hard swelling in the right maxilla. The skin overlying the swelling was stretched and of normal colour, with no secondary changes. Initially, it started small and gradually grew to its current size. There was no history of trauma or associated tooth pain. An intraoral finding revealed a painless, diffuse, hard, sessile swelling in the upper right buccal vestibule extending from the right lateral incisor to the second molar region (13 to 17 region). Palatally, the swelling extends anteroposteriorly up to the right deciduous second molar region; medially, the swelling crosses the midline. The mucosa overlying the swelling was smooth, similar to the adjacent mucosa, with no secondary changes seen with standard mouth opening.

Computed Tomography scan revealed that a well-defined expansive radiolucency extending anteroposteriorly from 11 to 55 region with superiorly extending into the maxillary sinus and medially up to the nasal septum.

Incisional biopsy revealed a well-encapsulated lesion with numerous bony trabeculae. Peripheral areas show a dense collagen stroma with evidence of giant cells. Focal areas exhibit irregularly shaped immature bony trabeculae with entrapped osteocytes & osteoblastic rimming features, suggestive of a juvenile trabecular ossifying fibroma. Surgical excision of the lesion was

performed under general anaesthesia, and the patient was on follow-up. (Figure 4)



Figure 4 Juvenile Trabecular Ossifying Fibroma (JTOF)

A) Extraoral B) Intraoral C) 3D-CT E) Surgical excision F) H/E staining

CASE-4

A 58-year-old male patient reported with the chief complaint of swelling on his upper front teeth region since 5months. Initially, it started as a smaller size and gradually it increased with time to attain its present size. There was a history of bleeding on brushing, with no history of associated pain. His medical history was non-significant. He had a habit of smoking for many years. Upon examination, a solitary, firm, pedunculated growth of approximately 3 × 4 cm is observed, extending from regions 11, 21, and 22. On palpation, the lesion was painless, firm, non-pulsatile, non-reducible, and non-compressible. No mobility was noted in the involved teeth.

All the involved teeth were vital with no periapical pathology. Surgical excision of the lesion was performed by using electrocautery under local anaesthesia (Figure 21), and the excision was sent for histopathological examination. The cut section specimen showed radio-opaque calcification under radiovisio-graph. 10x view showed bony trabeculae with osteoblastic rimming in fibrocellular stroma, features suggestive of peripheral ossifying fibroma (Figure 5).

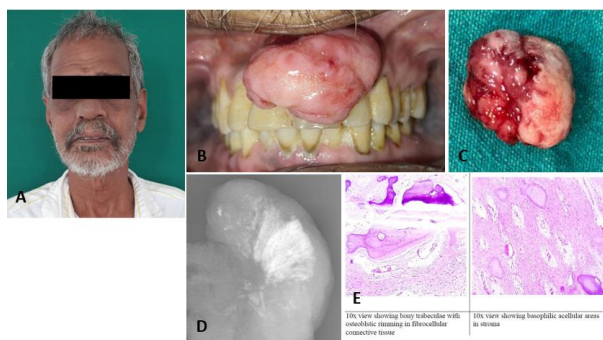


Figure 5: Peripheral Ossifying Fibroma

- A) Extraoral B) Intraoral C) Surgical Excision
D) Radiographic calcifications in the cut specimen
E) H/E staining

Discussion

Ossifying fibroma (OF) remains a diagnostically and therapeutically challenging benign tumour due to its variable biological behaviour and recurrence risk. It is the most common benign fibro-osseous lesion, defined by the replacement of normal bone with a fibrocellular stroma that produces mineralized tissue in the form of bone or cementum^[11]. Ossifying fibroma presents in two forms: a central variant originating from periodontal ligament cells near the root apex, and a peripheral variant confined to the soft tissues of the jaw. The central form is considered a true neoplasm, whereas the peripheral type represents a reactive, non-neoplastic soft-tissue proliferation^[12]. Ossifying fibroma typically occurs during the third and fourth decades, demonstrates a strong male predilection, and affects the mandible. It is observed more commonly in the tooth-bearing regions of the mandible than the maxilla, with the mandibular premolar–molar area being the most frequent site of occurrence^[13]. The lesion is usually asymptomatic in its early stages and may be identified incidentally on radiographs. With progressive enlargement, it can produce pain, cortical expansion, jaw deformity, and facial disfigurement. In our series, the mean patient age was 30.3 years. Studies indicate that ossifying fibroma in the maxilla tends to be more aggressive and symptomatic than in the mandible, likely due to anatomical differences. The mandible's dense cortical bone and loose medullary spaces restrict lesion growth, whereas the maxilla's thinner cortex and spongy cancellous bone allow easier expansion. The age distribution is similar to that in other studies, with a notable difference in gender distribution attributed to demographic factors. In our

research, the maxilla is predominantly affected compared to the mandible in 3 of 4 cases, which is inconsistent with other studies. On radiograph, in its early stages, OF may be completely radiolucent; as it matures, it becomes mixed; and at later stages, it becomes more radio-opaque^[11]. Ossifying fibromas may arise from abnormal mesenchymal cell differentiation into bone and cementum during odontogenesis, with disturbances in this inductive process contributing to their formation in the jaws^[15].

The neoplastic nature of the OF is attributed to the fact that larger lesions exhibit aggressive behaviour and cause significant bone destruction^[16]. JOFs occur at any age, from 3 months to 72 years^[15]. Juvenile ossifying fibromas (JOFs) are categorized into two distinct clinicopathologic types: trabecular and psammomatoid. Trabecular JOF is defined by the presence of trabeculae and woven bone, whereas psammomatoid JOF is characterized by small, uniform, spherical ossicles resembling psammoma bodies^[16]. The primary anatomical distribution distinguishes the two types of JOF. Psammomatoid JOF (JPOF) predominantly occurs in the fronto-naso-orbito-ethmoidal region, with approximately 75% of cases affecting the orbit, paranasal sinuses, and calvaria, and only 25% involving the jaw, mainly the maxilla. In contrast, trabecular JOF (JTOF) primarily affects the gnathic bones, with a slight predilection for the maxilla.^[8,19] Chrcanovic and Gomez^[24] reported mandibular involvement in 42% of JTOF and 22% of JPOF cases. While many studies indicate that JPOFs are more frequently observed in adults with a broader age range, JTOFs are typically present in younger patients, consistent with the cases reported here. In our series, one JTOF involved the maxilla and the other the mandible, aligning with findings from previous studies^[8]. The mean age of patients with JPOF (18.9 ± 12.0 years) was substantially higher than that of patients with JTOF (11.5 ± 6.0 years), as reported in a systematic review by Chrcanovic, findings consistent with ours. Studies indicate that fibro-osseous lesions can coexist with other giant cell lesions of the jaws, such as aneurysmal bone cysts (ABC) and central giant cell granulomas (CGCG), potentially due to stromal alterations within the primary lesion.

Studies reported that approximately 22% of jaw aneurysmal bone cysts (ABCs) are associated with other



bone lesions, including fibrous dysplasia, ossifying fibroma, or giant cell tumour. Lesions with secondary ABCs exhibit a higher recurrence rate compared to those without cysts, and such cystic changes are more frequently observed in JPOF than in JTOF^[15]. But in our case series, no such secondary ABC lesions were identified in any OF variant.

Management mainly depends on both clinical and radiographic findings and comprises one of the following options. Enucleation, Curettage, Enucleation +curettage/peripheral osteotomy & Resection with/ without continuity with reconstruction. (Figure 6)

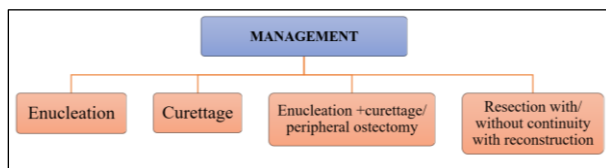


Figure 6: Management strategies of Ossifying Fibroma.

Enucleation is indicated for well-circumscribed, encapsulated lesions of limited size, while curettage is reserved for soft, poorly demarcated lesions that blend with surrounding bone or lack apparent radiolucency^[11]. A systematic review demonstrated that the use of enucleation or curettage as sole treatment modalities is associated with higher recurrence rates, regardless of whether the lesion is JTOF or JPOF or of its anatomical location. The combined technique of enucleation and curettage, supplemented with peripheral osteotomy, is regarded as the optimal strategy, as it effectively reduces recurrence while avoiding the functional and aesthetic deficits that may accompany more aggressive surgical resections. Additionally, resection is advised in situations involving recurrent disease, extension into neighbouring bony structures, or when preservation of the inferior mandibular border cannot be achieved.^[15] Resection resulting in a continuity defect is indicated when the lesion involves or lies too close to the inferior mandibular border. In the maxilla, resection is warranted when the tumour extends into the maxillary sinus or nasal cavity, particularly when its margins are diffuse or poorly defined. The literature suggests that most recurrences stem from incomplete removal of the lesion, likely due to the tumour's locally infiltrative growth pattern^[20,24]. Chrcanovic BR et al. reported that juvenile psammomatoid ossifying fibroma (JPOF) shows

recurrence rates of 46.2% following curettage, 37.5% after enucleation, 12.5% with combined enucleation and curettage or peripheral osteotomy, and 0% after marginal or segmental resection. For juvenile trabecular ossifying fibroma (JTOF), the recurrence rates were higher with curettage (62.5%) and enucleation (45.5%), while combined enucleation with curettage or peripheral osteotomy showed a recurrence of 33.3%. Marginal resection resulted in a 10% recurrence rate, and segmental resection demonstrated no recurrences.

In our cases, all lesions had well-defined margins from the surrounding bone, without any secondary cysts (ABCs/CGCG), and were treated by enucleation and curettage with peripheral osteotomy.

Conclusion

Ossifying fibroma (OF) is a benign fibro-osseous neoplasm comprising a diverse set of craniofacial lesions that exhibit considerable clinical and histopathologic variability. Except for their anatomical location, odontogenic and non-odontogenic subtypes share similar clinical presentations and may display either trabecular or psammomatoid architectural patterns. Consequently, a more streamlined classification system is needed to encompass the broad spectrum of clinical variations within this lesion group.

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