

# Management of Giant Maxillary Cemento-ossifying Fibroma Invading the Orbital Cavity by Resection and Immediate Reconstruction Using Sagittal Coronoid - Ramus Graft: A Case Report and Literature Review

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**Abstract** A case of giant cemento-ossifying fibroma (COF) that is invading the maxilla, maxillary sinus, nasal nostril, inferior orbital floor (IOF) and inferior orbital rim (IOR) was resected along with total hemimaxillectomy and reconstructed immediately using mandible coronoid-ramus graft to the orbital rim, metal mesh to the floor, and an immediate maxillopalatalobturator to maintain the midfacial contour, cosmetics, speech, and function. The surgical and reconstructive technical details are discussed.

**Keywords:** *ossifying fibroma, fibro-osseous lesion, maxilla, ramus graft, giant, reconstruction*

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## 1. Introduction

COF is an unusual benign fibro-osseous tumor that replaces normal bony tissue with abnormal fibroblasts, collagen fibers, and osteoid and / or cementoid like deposits [1].

Menzel In 1872 was the first to use the term "cemento-ossifying fibroma, describing a large mandibular tumor in a 35 years old female [2]. In 1971, World Health Organization (WHO), categorized four types of cementum-containing lesions: fibrous dysplasia (FD), ossifying fibroma (OF), cementifying fibroma (CF), and cemento-ossifying fibroma (COF) [3]. Then the COF was classified as a benign osteogenic neoplasm [4]. However, the term (cemento-ossifying fibroma) was reduced to ossifying fibroma in the new WHO classification in 2005 [5].

There is still controversy regarding the origin of the COF, as claims to originate from the periodontal apparatus and invading the surrounding structures showing features of fibro osseous lesions exists, while the other way around was described as well [4,5].

Various subtypes of ossifying fibromas (OF) have been reported in literature, based on age (adult/juvenile), histological content (cementoid/ osteoid content), or histological pattern e.g. (Psammomatoid and trabecular) [1]. Juvenile ossifying fibroma (JOF), which also known as juvenile active ossifying fibroma or juvenile aggressive

ossifying fibroma, affects the craniofacial skeleton of children patients below age of 15 years old. It has two subtypes, the Psammomatoid Juvenile ossifying fibroma and trabecular Juvenile ossifying fibroma. However, it is not uncommon to find more than three subtypes in one single lesion such as Psammomatoid juvenile cemento-ossifying fibroma [6].

COF has a slight predilection in females of 30s-40s years old and it occurs in the mandible more common than in the maxilla [1]. However, it can be found in rare areas (such as: frontal, temporal, orbital and occipital bones, mastoid cavity, nasopharyngeal area, masticatory, parapharyngeal spaces and paranasal sinuses) [7-14]. Peripheral OF was reported in rare occasions as well. COF can be manifested as slowly growing, mild and asymptomatic jaw swelling, but it behaves as rapidly growing mass, especially in the maxilla, causing severe facial deformity [1].

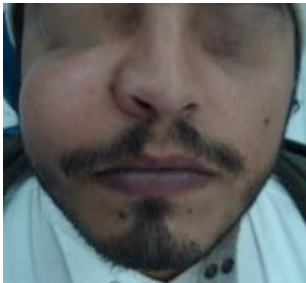
Usually, at the time of presentation, it presents as well-defined multilocular mixed radiolucent-radio-opaque mass with marginal sclerosis that differentiates it from fibrous dysplasia. But it can be seen also as unilocular radiolucent lesion, at the early stage, or more of a radiopaque mass at later stages. Surgical treatment of COF depends primarily on its clinical behavior, size, as well as its location and involvement of the surrounding tissues. Small and well-demarcated lesions can be treated with enucleation and curettage. However, surgical resection of the mass with 3-5mm healthy margins, always indicated in larger, more

aggressive ones, especially in the maxilla, or in recurring cases [1].

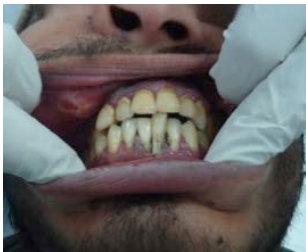
In our report we are presenting a case of large maxillary COF and the surgical details are discussed.

## 2. Case Report

A 28 years old healthy male patient walked into the oral maxillofacial surgery clinic at Aseer Central Hospital, King Khalid University Health Center complaining of a right maxillary swelling, right eye proptosis and nasal obstruction. The patient had noticed the swelling approximately six months before presenting to the clinic. On clinical examination, hard swelling of the right side of the face, mild elevation and proptosis of the right eye was clearly noticed with totally obliterated right nostril. Intra-orally, the right maxillary vestibule was obliterated and hard expansion at the right side of the palate and immobile teeth was noted. No motor norneuro-sensory deficit were detected (Figure 1 & Figure 2).



**Figure 1.** Clinical picture showing right maxillary swelling, causing proptosis



**Figure 2.** Intraoral picture showing swelling of the right maxillary vestibule extending from the midline to the 2<sup>nd</sup> molar area

Panoramic radiograph showed a homogenous mixed maxillary radiolucent mass with radio opacifications involving the alveolar bone, obliterating the right maxillary sinus and the right nasal cavity without crossing the midline.

The differential diagnosis of COF includes other lesions that contain radiopacities within a well-defined radiolucent mass such as fibrous dysplasia, ossifying fibroma, Calcifying epithelial odontogenic tumor, myxoma, Gorlin's cyst (calcifying odontogenic cyst), squamous cell carcinoma, chondrosarcoma and osteosarcoma. The well-defined borders of COF help differentiate it from aggressive sarcomas and carcinomas [4,5].

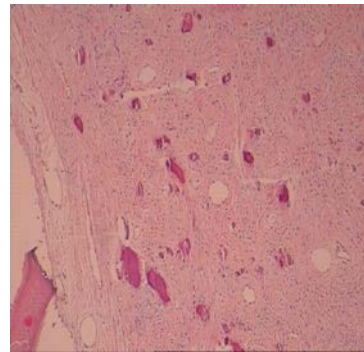
A CT scan of the head and neck area revealed approximately 5x5x7cm mass occupying the right maxillary alveolar bone, maxillary sinus, nasal nostril, and reaching the zygomatic maxillary junction, lateral orbital rim, the medial orbital wall at the anterior lacrimal crest

inferior orbital rim, and invading the inferior orbital floor leading to proptosis of 2cm with maxillary anterior expansion of 3.5cm (Figure 3). No signs of intracanal invasion of the mass was seen neither any abnormalities in the neck.



**Figure 3.** A coronal CT scan of facial aspect on soft tissue window showing approximately 5x5x7cm mass occupying the right maxillary sinus, alveolar bone, nasal nostril, inferior orbital rim, and invading the inferior orbital floor leading to displacement of the right eye globe

An intraoral incisional biopsy was done and the histopathological findings showed fibrous cellular background with osteoid and cementoid like bodies that is consistent with cemento-ossifying fibroma (Figure 4).



**Figure 4.** A histopathology specimen stained with Hematoxylin and Eosin, showing a fibrous background with collagen bundles running in a non-organized pattern. Deposits of osteoid and cementoid particles are dispersed with few small vascular bundles

The result was disclosed to the patient and the management plan was discussed that consisted of oral intubation, total right hemi-maxillectomy via Weber Ferguson incision with lateral extension, anterior mandible coronoid- ramus bone graft harvest to reconstruct the inferior orbital rim, orbital floor reconstruction using metal mesh, and finally insertion of maxilla palatal obturator. An option of anterior iliac crest bone graft harvest was discussed as well, but the patient refused it.

Accordingly, the patient was admitted and taken to the operating room. A corneal shield was inserted to both eyes and a tarsorrhaphy stay suture was applied. Next, The Weber Ferguson flap incision took place at the right aspect of the face with lateral lower lid extension and a supra periosteal dissection took place at the anterior maxillary wall (Figure 5). As the tumor borders identified, the osteotomies were performed at the zygomatico-maxillary boundary, infero-lateral orbital rim, inferomedial orbital junction, palatal, and pterygo-maxillary junction. The plane between the orbit and the

tumor hard expansion was carefully identified and dissected as the tumor got resected in toto. (Figure 6). The nasal extension was attached to the septo-vomerian lining that was carefully incised and inferior turbinectomy was done and sent with the specimen. Next, the attention was directed to the anterior coronoid – ramus part of the mandible, which found to be approximately 4.6cm length that was approximately matching the inferior orbital rim defect size 4.9cm. Hence, harvesting the ramus graft was attempted and reconstruction of the IOR was fixated using plates and screws. While titanium mesh was used to reconstruct the IOF (Figure 7). The maxilla-palatal defect was reconstructed using an immediate obturator. Next, copious irrigation of the surgical site was performed, hemostasis achieved, and flap closure was attempted in layers starting with re-suspending of the facio-orbital soft tissue envelope, subcutaneous approximation using 3-0 vicryl suture, and skin closure using 5-0 nylon suture.



Figure 5. Weber-Ferguson approach exposing the lesion from the different aspects



Figure 6. the resected tumor mass in toto



Figure 7. Intraoperative X-ray for the tumor showing periodontal ligaments invasion by the tumor

The patient follow up visits commenced at 1, 3, 6 and 12 weeks, that were uneventful (Figure 8 & Figure 9). A

stable recovery was noticed, up to three years and 6 months postoperatively. The patient showed satisfaction regarding his facial appearance, with favorable orbital symmetry, cheek and lip competence, as well as normal speech, feeding and sinus function (Figure 10 & Figure 11).

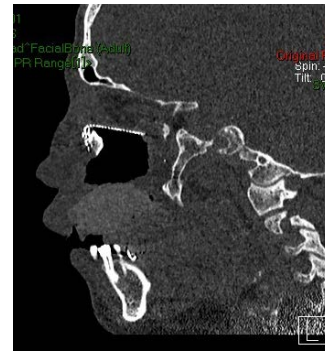


Figure 8. A postoperative parasagittal cut of CT scan of the face showing the reconstruction of IOR with ramus-coronoid graft that presents an acceptable thickness on site. The infrorbital metal mesh is then placed to reconstruct the floor



Figure 9. A one week postoperative clinical pictures showing the acceptable fullness of the midfacial area



Figure 10. A one week postoperative clinical pictures showing the acceptable fullness of the midfacial area



Figure 11. A three year follow up frontal and lateral photograph showing the patient satisfying results as fullness of the midface, cheek prominence, symmetrical IOR, non tender palpation, and normal masticatory function and speech. Mild retraction of the upper lip without lip incompetence is seen as well which was not of a major concern





**Figure 12.** a three year follow up frontal and lateral photograph showing the patient satisfying results as fullness of the midface, cheek prominence, symmetrical IOR, non tender palpation, and normal masticatory function and speech. Mild retraction of the upper lip without lip incompetence is seen as well which was not of a major concern

### 3. Discussion

Kramer *et al* [3] described the cemento-ossifying fibroma as an osteogenic neoplasm, while the fibrous dysplasia as a non-neoplastic bone lesion. The neoplastic nature is attributed to the significant osseous destruction produced by large aggressive COF lesions plus its recurrence that reached high percentage in some cases.

#### 3.1. Pathogenesis of COF

The Pathogenesis of COF still unknown. A number of authors have suggested that ossifying and/or cementing fibromas originating from the periodontal ligament which contains pluripotential cells, which under a variety of stimuli (such as traumatic extraction, inflammation) can produce lesions composed of cementum, lamellar bone, fibrous tissue, or any combination of these tissues [1].

However, there is controversy over such an origin, since tumors of similar histology have been reported in bone lacking periodontal ligament, such as ethmoidal and frontal bone [7,12].

Nasopharyngeal COF was reported and explained as it is originating from embryologic nests [11]. Brademann *et al* [9] explained that ectopic periodontal membrane differentiating from primitive mesenchymal cells in the petrous bone may serve as a cause of development of COF in this area, and trauma such as severe whiplash may be a factor in the induction of proliferation of COF. The ethmoidal location of COF may also be explained by incomplete migration of mesenchyme and its differentiation into periodontal membrane [12].

#### 3.2. Genetic Analysis of COF

There is no much cytogenetic analysis done for ossifying fibroma. Deletions were detected in 2q31-32 q35-36 in one case of mandibular COF [16]. While a study of 3 cases of JPOF of the orbit, that demonstrated a non-random chromosome break points at Xq26 and 2q33 resulting in (X;2) translocations [17]. Mutations of HRPT2 gene (tumor suppressor gene) which is associated with Hyperparathyroidism associated ossifying fibroma was detected in one study of OF suggesting that OF may arise due to haplo-insufficiency of the HRPT2 gene [18].

#### 3.3. Immunohistochemical Assays of COF

Although the histological similarities between cementifying fibroma (CF) and OF, differences have been found on immunohistochemical staining. CF has significant immunoreactivity for keratin sulfate and chondroitin-4-sulfate in contrast to OF [19]. Also, due to the ability of central OF to deposit hard tissue, it was found a dominant positive bone morphogenetic protein (BMP) in central OF and a higher osteopontin (OPN) and osteocalcin (OCN) staining, compared to those of peripheral OF [20]. Recently, Zhang *et al* [21] suggested that notch signaling molecules may participate in controlling cell differentiation and proliferation in normal bone and OF of the jaws. Notch signaling disorder may be a molecular incident in COF occurrence and development.

#### 3.4. Clinical Picture of COF:

COF shows predilection to the middle-aged females, 3rd to 4th decades with a female to male ratio of 2:1. However, Krausen [12] reported no particular sex predilection regarding the occurrence of OF. The age distribution varies widely, as in a Taiwan-Chinese population the range was between 16 and 62 years with a mean of 34 years [15]. COF affects craniofacial bones most commonly and rarely extra-gnathically [7-14]. It occurs more commonly in the mandible (62-89%) with (77%) in premolar-molar [14]. It is well capsulated, slow growing and asymptomatic intra-bony mass with a normal mucosal coverage, firm consistency and size ranges from 0.2 cm to 15 cm as reported in some cases [22]. Yet, they can grow aggressively leading to severe jaw expansion and displacement of the surrounding tissue [1]. Maxillary lesions usually behave more aggressive compared to mandibular ones, which is basically due to the medullary bone nature of the maxilla, the presence of the air filled spaces, the paranasal sinuses, and its tendency to involve the nasal septum, orbital floor, and infraorbital foramen, and usually they are large at time of presentation as seen in our case which reached 5 x 5 x 7 cm in diameter which among the largest in the literature according to our knowledge. These two characters of the maxilla make the surgical removal of maxillary COF more difficult than the mandibular lesions that usually "shell out" easily intraoperatively [14].

Usually, pain and neurosensory deficit are rare presentations, while patients with sinonasal lesions, might have different complaints (nasal obstruction, anosmia, hyposmia, headache, epistaxis, ocular symptoms such as diplopia, proptosis, epiphora and even visual loss) [1]. In our case, the patient was complaining of proptosis and nasal obstruction.

#### 3.5. Radiographic Picture of COF:

Radiographic evaluation of COF of a great value in the diagnosis as it helps in distinguishing between COF and other fibro-osseous lesions such as fibrous dysplasia. It is usually seen as a well-defined radiolucent lesion in the early stage, then as the lesion matures, bone densities appear so it takes the multi-locular mixed radiolucent-radio-opaque configuration, or complete radiopaque. Mostly seen as mixed radio-opaque-radiolucent, surrounded with a marginal sclerosis but a thin and intact cortex a characteristic downward bowing of the inferior cortex of the mandible seen with large mandibular lesions

while loss of lamina dura, root resorption and/or divergence, displacement of associated teeth, alveolar nerve, displacement of the surrounding organs and tissues may be noted [1]. In the rapidly expanded lesions it loses the regular border. COF has a centrifugal growth pattern and grows by expansion equally in all directions giving a spherical to egg-shape mass, while fibrous dysplasia expands the cortex linearly and the outline of the expanded mandible is not in continuity with the remainder of the outline of the lesion [1,22]. Aggressive maxillary COF lesions tend to show a greater degree of immaturity than that seen in mandibular lesions. There is a correlation between the amount of calcification seen in the surgical specimen and that seen on the CT scan [14].

### 3.6. Histological Picture of COF

Histologically, OF are well circumscribed and occasionally encapsulated lesions. Mainly, they composed of two components: fibrous stroma and bone elements which demonstrate various degrees of maturation. The stroma consists of fibroblasts and collagenous fibers and the bone elements consist of mineralized bodies (ossicles), osteoids, woven bone and lamellar bone. Ossicles connect to form bone trabeculae that usually is surrounded by osteoblasts and osteoclasts. Among that, rounded cementum-like masses may be seen. Because of the variation in the configuration of these calcified deposits, such tumors have been referred to as both ossifying and cementifying fibroma. The fibrous capsule can be lost in aggressive form of ossifying fibroma [1].

### 3.7. Treatment and Prognosis of COF:

The treatment of choice for OF is surgical excision. However, the decision on whether to enucleate or resect radically, depends on the size of the lesion and the involvement of the surrounding structures (inferior border of the mandible, soft tissues, the maxillary sinus and nasal cavity) [1,23].

Usually, small and well-demarcated lesions can be treated conservatively by enucleation and curettage, whereas larger aggressive lesions, especially in the maxilla, and recurrent lesions require resection with 5mm of healthy margins [1].

Recurrence rate found to be higher following curettage of COF lesions (0-28%), as well in the maxillary lesions because of the greater difficulty of their surgical removal and larger size at the time of presentation. So radical resection and continuous follow up, up to 10 years is usually recommended [1,2,14].

Once radical resection is chosen as the line of treatment, then reconstruction and rehabilitation of the lost defect should be taken into consideration. In fact, the maxillectomy defect presents a surgical challenge, particularly when the orbital floor is also resected, as in our case, which lies under the Brown's classification as class III defect (maxilla and orbital floor defect). A large number of techniques for the reconstruction of a class III defect have been described, including prosthetic obturator, pedicled local flaps and free flaps with or without bone grafts [24]. The use of titanium mesh and plates has been widely used to reconstruct orbital floor and inferior orbital rim. However, they still have the drawback of painful sensation at the IOR region, and eventual soft tissue

retraction. The anterior coronoid- ramusgraft was used for reconstruction of orbital and maxillary defects [25,26]. In some reported cases where large maxillary COF involved the orbital floor, the orbital floor was reconstructed using iliac bone graft, costochondral graft or titanium mesh [27,28].

In our case we used the anterior coronoid ramus graft to reconstruct the IOR, while the titanium mesh was used to reconstruct the orbital floor. The advantage of this technique was that the bone graft was harvested from regional area and hence saved the patient an additional surgical harvest site such as the calvarium or the iliac crest. In addition the curved nature of the anterior ramus graft made the recreation of the IOR of more resemblance compared to the iliac crest and the calvarium, and hence, faster and easier insertion. Also, it has the advantage of using the obturator, for sealing and social benefits as having an immediate new dentition with restoration of appearance.

Close follow up is necessary, as cases of synchronous occurrence have been reported in the same jaw or the opposing [29].

## 4. Conclusion

COF is a benign fibro-osseous lesion, where in some cases it can act aggressively to reach larger sizes occupying the maxillofacial region that make reconstructing such defects very challenging using hardware, osseous grafts, locoregional flaps, or / and vascularized flaps. And hence, the reconstruction technique should consider a lot of factors such as patient convenience, minimizing second surgical sites, minimizing second operative procedures, and meeting the functional and cosmetic objectives. Therefore considering the coronoid ramus graft technique can be a promising technique to consider in managing defects at the zygomatic maxillary orbital region.

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