Cemento Ossifying Fibroma of the Maxilla - A Case Report

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Abstracts: Cemento ossifying fibroma is a uncommon benign fibro osseous lesion commonly occurs in the young adults in the age group of 20 to 40 yrs, most commonly in the females with an female to male ratio of 5:1.It usually occurs in the craniofacial bones and rarely in long bones, of the craniofacial bones, posterior mandible in the region of premolar molar area is commonly seen. The occurrence COF in the maxillary, ethmoid, frontal, and sphenoid sinuses are rarely reported in the literature. A rare case of COF affecting the maxilla in a young male patient is discussed. [Surname + name char. NJIRM 2014; 5(6):108-112]

Key Words: Cementeo - Ossifying Fibroma (COF), Fibro Osseous Lesion, Maxilla

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Introduction: Cemento ossifying fibroma is a well-demarcated benign fibroosseous lesion occurs in the patients of a wide age range, the greatest numbers of cases are encountered during the third and fourth decades of life. There is definite female predilection, with female-to-male ratio as high as 5:1. Several rare cases involving the maxilla, nasal bones, orbit, ethmoid sinus, sphenoid sinus, maxillary sinus, occipital, temporal bone, and nasopharynx have also been reported. The lesion is generally asymptomatic until the growth produces a noticeable swelling and mild deformity; displacement of teeth may be an early clinical feature. [1, 2]

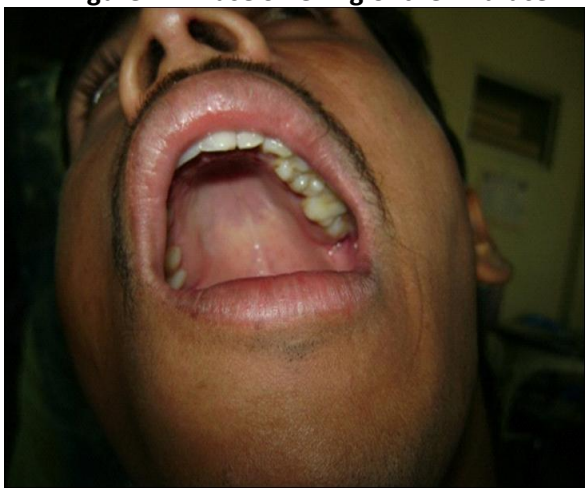
Radiographically, Cemento ossifying fibroma of the head and neck is described as a well-circumscribed, expansile bony lesion with calcified matrices in the mandible and maxilla. In the early stages, the Cemento-ossifying fibroma appears as a radiolucent lesion with no evidence of internal radiopacities. 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. Displacement of adjacent teeth is common. One additional important diagnostic feature 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.[3,4]

Histopathology reveals many delicate interlacing collagen fibers, seldom arranged in discrete bundles, interspersed with large numbers of active, proliferating fibroblasts and cementoblasts. As the lesion matures, the islands of calcification increase in number, enlarge, and ultimately coalesce. The increase in degree of calcification, accounts for the increase in the radiopaqueness of the lesions on the roentgenograms.[5]

Case Report: A 22 year – old male reported with the complaint of swelling in the left side of the face. The swelling was present from the childhood, but 2yrs back patient noticed another swelling in the same region which was initially small in size and it slowly increased to the present size and not associated with pain. The medical, social and family histories were unremarkable.

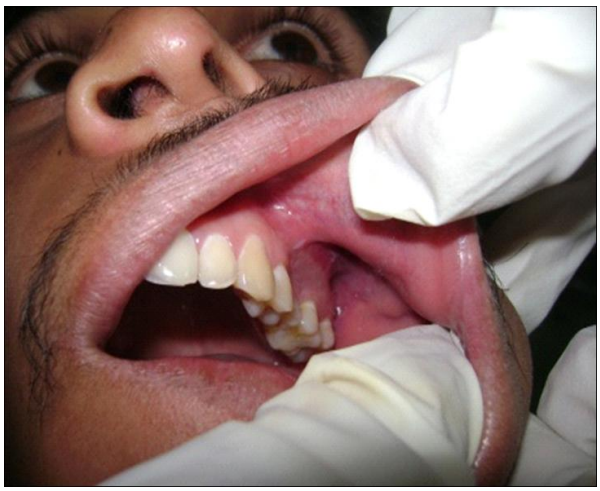
On examination, the diffused swelling present over the left mid-face measuring about 3x2cm with the over lying normal skin. The swelling was firm, fixed and smooth surface with distinct borders (Fig 1).

Figure 1: Difuse Swelling of the Midface



Intraorally, a diffused swelling present over the upper left vestibule extending from 23 to 27 with slight reddish appearance of overlying mucosa and with no palatal expansion. On palpation the swelling was firm, tender, fixed and smooth surface with distinct borders (Fig 2).

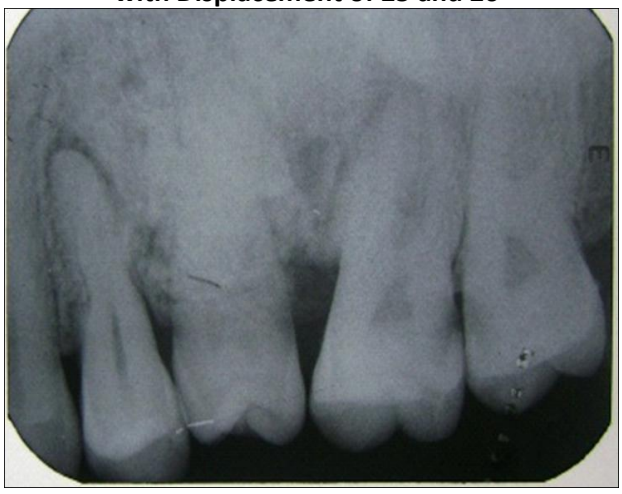
Figure 2: Difuse Swelling in the Upper Left Buccal Vestibule



Based on history and the clinical examination, the provisional diagnoses of central ossifying fibroma was considered and central giant cell granuloma, fibrous dysplasia, odontogenic tumor and cysts are considered as differential diagnosis.

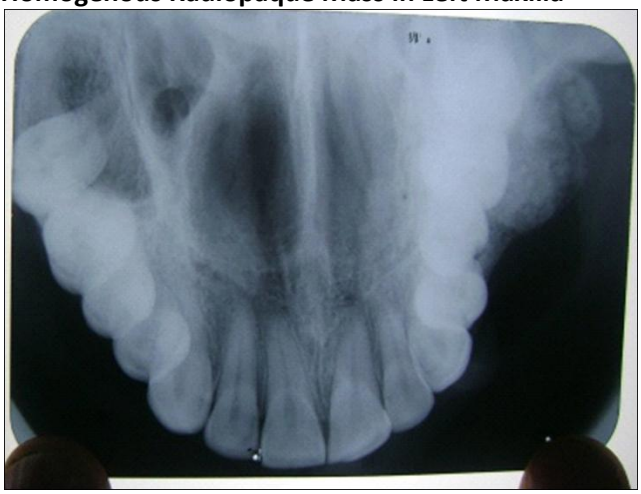
IOPA Radiograph shows alteration in trabacular pattern and the rotation of 25 with the displacement of roots of the 25- 26 and widened periodontal ligament space irt 24 (Fig 3).

Figure 3: IOPA Show Altered Trabacular Pattern with Displacement of 25 and 26



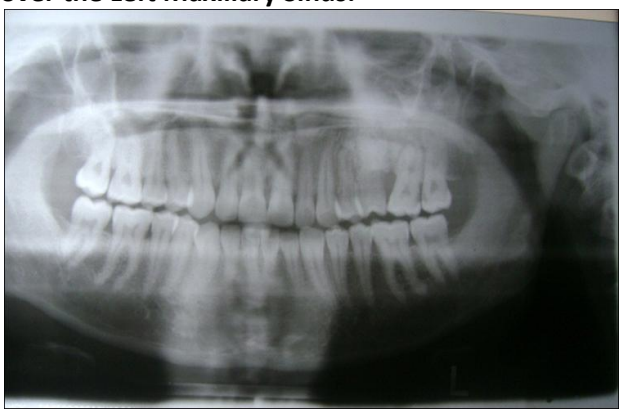
Maxillary Occlusal view shows a well-defined homogenous radiopacity on the buccal aspect of left maxilla in the region of 26-27 and the radiopacity involving the left nasal fossa (Fig 4).

Figure 4: Occlusal Radiograph Show Well Defined Homogenous Radiopaque Mass in Left Maxilla



OPG shows a homogenous radiopacity in the left maxilla over the apices of 25, 26, 27 extending in the left maxillary sinus and displacement of roots of 25 - 26 (Fig 5).

Figure 5: Well-Defined Homogenous Radiopacity over the Left Maxillary Sinus.



PNS view shows a homogeneous radiopacity involving the left maxillary sinus (Fig 6). Axial and Coronal CT scans shows the well-defined irregular exophytic sessile bony space occupying lesion involving the left maxillary alveolus, malar eminence and the anterior wall of the maxillary sinus (Fig 7, 8).

Figure 6: PNS View Show a Homogeneous Radiopacity Involving the Left Maxillary Sinus

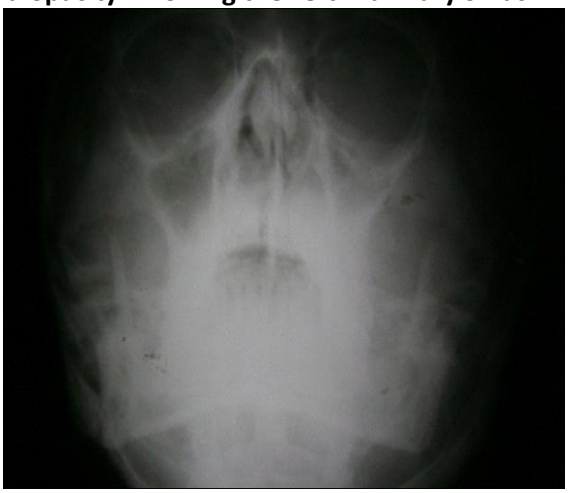


Figure 7: Axial CT scan show the well-defined irregular exophytic sessile bony space occupying lesion

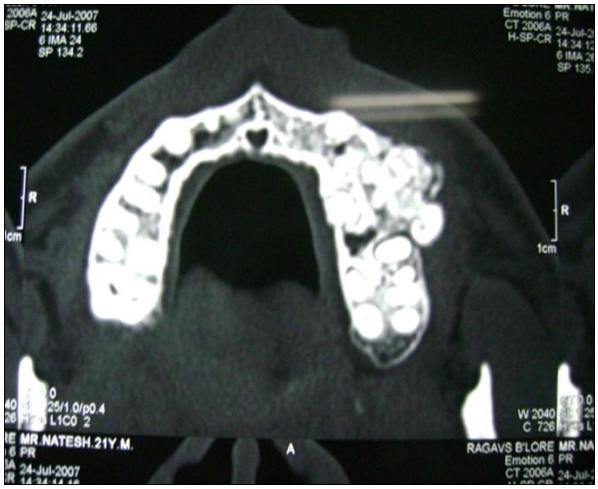
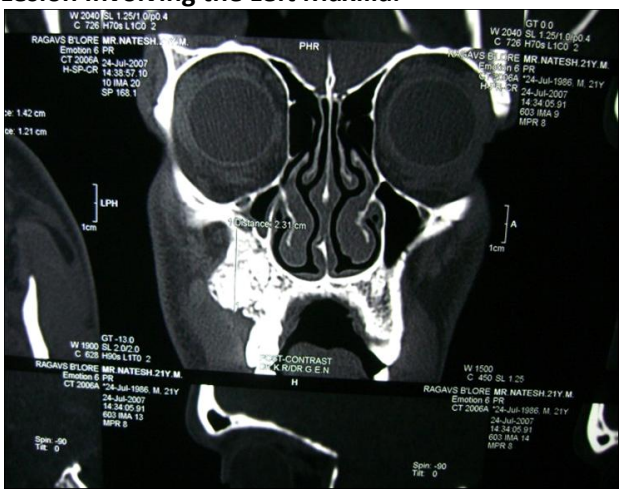
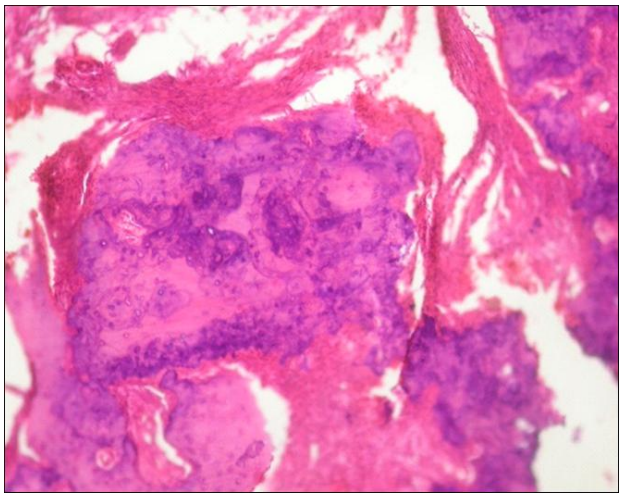


Figure 8 Coronal CT scan Show the Well-Defined Irregular Exophytic Sessile Bony Space Occupying Lesion Involving the Left Maxilla.



Incisional biopsy was done from the left buccal vestibule and sent forhistopathological examination. The histopathology revealed presence of whorled fibrous tissue, containing calcified masses of bone and cementum like substances and regular fibroblasts. Mitoticfigures were notrevealed. Based on radiographic and histopathological examination the final diagnosis of cemento ossifying fibroma was confirmed (Fig 9).

Figure 9: under High Power Show the Calcified Bone and Cementum like Substance



Discussion: The Fibro-Osseous lesions are the hetero-geneous group of benign lesions of unknown etiology affecting the jaws and other craniofacial bones.The peak incidence of COF is the third and fourth decades. It is more prevalent in white than black racial groups; Female predilection has been reported as high as 5:1.[6]. The Lesions in this category include, Fibrous dysplasia (FD), Focal Cemento-Osseous dysplasia (FCOD) and Cemento-Ossifying fibroma (COF) [1]

In 1971, The World Health Organization classified all cementum-containing lesions into 4 groups: Benign Cementoblastoma, PeriapicalCemental dysplasia, Gigantiform Cementoma, and Cementifying Fibroma. COF as a subclass of the Cementifying fibroma group.[2] In the nomenclature by Kramer *et al*, the cemento-ossifying fibroma is described as an osteogenic neoplasm and the fibrous dysplasia as a non-neoplastic bone lesion. Liechtenstein and Jaffe also classified ossifying fibroma as a histologic variant of fibrous dysplasia, a concept that persisted in the literature until

1963, when Reed separated the lesions into distinct clinicopathologic entities. But Reed viewed fibrous dysplasia as an arrest of bone maturation at the woven stage of development, whereas ossifying fibroma was believed to be a benign neoplasm of bone, in which normal bone architecture was replaced by a tissue composed of collagen fibers, fibroblasts, and various amounts of calcified tissue with the potential for unlimited and destructive growth. A close relationship exists between the central cemento-ossifying fibroma and the central ossifying fibroma. It is based on the marked similarity between the two regarding predilection of age of occurrence, sex, race, location, roentgenographic appearance, and clinical behaviour, 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. Hamner*et al* advocated the periodontal origin of ossifying fibroma[3]. The periodontal ligament has been shown to be capable of producing cementum and osteoid, both of which are characteristically found in ossifying fibromas. Eversole*et al*. reported that the production of these cementum-like structures may be associated with membranous bone and may not only be related to cementogenesis.

COF is the most frequent fibro-osseous lesion encountered by oral pathologists and perhaps, it has more synonymous than any other jaw lesion[4]. In 1872, Menzel gave the first description of a variant of ossifying fibroma called Cemento-ossifying fibroma, in a 35 year old woman with a long-standing large tumor of the mandible.

The COF shows three distinct stages during its development. In the initial osteolytic stage, the tumor is composed of cellular tissue only with no deposition of calcified material. This represents an immature cementoma and appears radiolucent. The second stage is the cementoblastic stage. Cementum is deposited in the fibrous mass and becomes calcified with flecked opacities. Finally, in the mature inactive stage the entire mass becomes calcified and encapsulated by remnant connective tissue [5]. It is almost exclusively found in the cranial bones, with only a rare case reported in the long bones. Of the cranial bones, the mandible is the most common site (75% in some series), followed by rarer reports of the maxillary, ethmoid, frontal, and sphenoid sinuses, as well as the orbit, occiput, and temporal bone in an extragnathic variant.[7 8]

Although recurrences and disfigurement occur, death from these lesions is rare and usually results from intracranial extension and meningitis.[9]

Radiographically, the COF presents as a well-defined unilocular or multilocular lesion with smooth contours. The maturity of the lesion will determine the degree of radiopacity. The immature lesion may present as completely radiolucent, whereas the mature lesion may appear completely radio opaque as seen in this case. Nevertheless, majority of the lesions demonstrate varying degrees of radiolucency.[10] One additional important diagnostic feature 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, which is true in this case.

Histologically, the cemento ossifying fibromas are well circumscribed, occasionally encapsulated, consisting of cellular fibrous tissues and thin isolated trabeculae of bones. The bone may show osteoblastic rimming and spherical deposits of calcified material, which are relatively acellular resembling cementum. In some cases, the calcified materials predominate the tissue and such lesions are designated as psammomatoid ossifying fibromas, from a Greek psammos sand.[11] The histopathological features of cemento ossifying fibroma are difficult to distinguish from those of FD, but the distinction is mainly relied on age and radiographic features [12].The pathologic nature of the two lesions is not yet clear and histopathologically difficult to differentiate. The differential diagnosis includes other lesions that contain radiopacities within a well-defined radiolucent mass: chondrosarcoma or osteosarcoma, odontogenic cysts like calcifying odontogenic cysts (Gorlin cysts), and calcifying epithelial odontogenic tumors (Pindborg tumors). The well-defined border of the cemento ossifying fibroma helps differentiate it from aggressive sarcomas and carcinomas. Fibrous dysplasia has a characteristic “ground glass” appearance not seen

in the cemento ossifying fibroma. The radiologic differentiation of cementoossifying fibroma from Gorlin cyst and Pindborg tumor is difficult; the final diagnosis is based on histologic appearance. Pindborg tumor has a high association with impacted teeth, which was not present in this case. The COF is usually well circumscribed and this facilitates its extirpation from the surrounding bone. The treatment COF is governed by several factors, such as location, extent and size of the lesion. Recurrences are uncommon but it has been described.

The Fibro-Osseous lesions are the hetero-geneous group of benign lesions of unknown etiology affecting the jaws and other craniofacial bones. In craniofacial region, COF occurs commonly in the premolar' molor region but rarely it may arise in the nasomaxillary complex. There is definite female predilection, with female-to-male ratio as high as 5:1. On radiograph it appears as radiolucent lesion in the initial stage than to mixed radiolucency and in later stage as homogenous radiopaque lesion. Histopathologically, it reveals variable amount of cemental and osteiod substance. The COF is usually well circumscribed and this facilitates its extirpation from the surrounding bone. Recurrence is rare and Death resulting from this tumor is very rarely reported.

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