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Case Report

Dentistry Section

Pericoronal Occurrence of Cemento-Ossifying Fibroma: An Unexemplified and Unusual Case Report with Review of Literature

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ABSTRACT

The Cemento-ossifying fibroma (COF) is an odontogenic neoplasm that is predominantly considered as a fibro osseous lesion of the jaw bone. The histogenetic origin of COF was thought to be derived from the periodontal cells, which have the ability to form fibrous, cementum and osseous tissue. Due to the periodontal cellular origin, the lesion often occurs in the radicular portion of the bicuspid and molar tooth region of the lower jaw. We present a case of Cemento-ossifying fibroma in a 43-year-old female in the pericoronal aspect of an impacted third molar mimicking an odontogenic lesion. The occurrence of COF in pericoronal region is an unusual site. This article also discusses about the various hamartomatous lesions occurring in the pericoronal region of the teeth.

Keywords: Cemento-ossifying fibroma, Pericoronal hamartomas, Multiple calcifying hyperplastic dental follicles

CASE REPORT

A 43-year-old woman visited our dental office for regular dental checkup. On intraoral examination, clinically missing third molars were found in the left and right sides of the lower jaw. The lesion was asymptomatic and there was no significant medical history. A provisional clinical diagnosis of impacted third molars in relation to the right and left sides of the lower jaw was made. An orthopantomogram (OPG) was advised to evaluate the unerupted lower third molar status. OPG was preferred instead of the Intra Oral Peri Apical (IOPA) radiograph due to the reason that, OPG can reveal a broader area of coverage at the pathologic site of interest. The OPG revealed a vertically impacted third molar in the right mandible region. The radiographic interpretation revealed a radiopaque area distal to the impacted tooth and is surrounded by a thin well defined radiolucent border. The radiopaque area was measuring about 0.7 x 0.6 cm approximately. [Table/Fig-1]. Internal structures that may be giving the appearance of mixed radiographic density are abnormal bone, dystrophic calcifications, cementum and tooth structures such as enamel and dentin. Based on the radiographic interpretation, the lesion was provisionally diagnosed as a Calcifying epithelial odontogenic tumor. Surgical removal of impacted teeth along with the lesion was planned. This was carried out under local anaesthesia and access was established by raising a mucoperiosteal flap. The tooth and the lesion were removed separately. The excised specimen was subject for pathology evaluation. Gross examination of the specimen revealed 0.6 x 0.6 cm approximately, which was brownish white in colour. During grossing the lesion, a gritty material was encountered, which gave a suspect of mineralized areas in the lesional tissue.

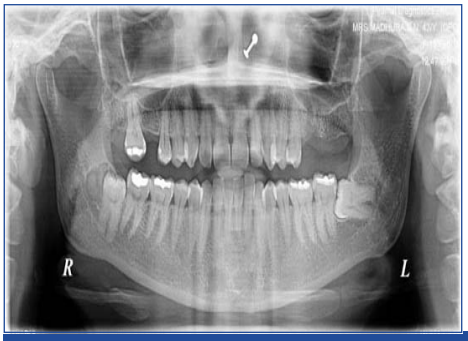
Microscopically, the lesional tissue showed the fibrocellular stroma. The stroma showed a hypercellular fibrous element. The collagen fibres were haphazardly arranged. The fibroblast observed in the lesional tissue was plump and intensely stained basophillic nucleus. The stroma showed focal areas of varying degree of mineralization [Table/Fig-2a]. The focal areas of homogenous, acellular, eosinophillic mass with peripheral basophillic stained areas resembled cementum like areas and suggesting cementoid apperance [Table/Fig-2b]. One such cementoid area showed the peripheral brush border appearance. Few focal areas showed homogenous eosiphillic areas with concentric appearance resembling osteoid areas [Table/Fig-2b]. The histopathological interpretation of the present lesional tissue suggested the diagnosis of Cemento-ossifying fibroma. The patient was followed post operatively for one year and there was no recurrence during this period [Table/Fig-3].

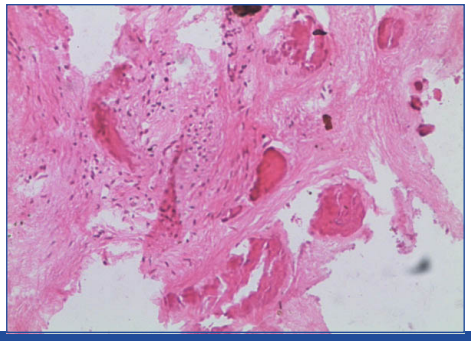
DISCUSSION

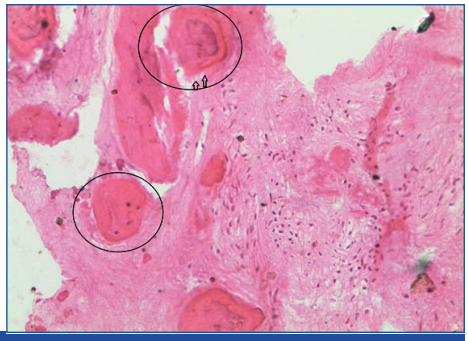
In 2005, the World Health Organization (WHO) categorized the COF as a fibro-osseous lesion derived from multipotent mesenchymal blast cells of periodontal membrane origin that have the ability to form fibrous tissue, cementum, bone or a combination of these elements [1]. The exact pathogenesis and stimuli is not well established still. However, few authors mentioned that trauma may play a role in pathogenesis [2]. To our knowledge, the COF had never been reported in the pericoronal region in the literature. The most common pathologic lesions occurring in the pericoronal region are represented in [Table/Fig-4]. Commonly seen pathologies in the pericoronal region and age distribution of the same are represented in the schematic chart [Table/Fig-5].

The present case, in which the lesion occurred in the pericoronal area of an impacted mandibular third molar, presented a challenge to clinician, radiologist and pathologist because the clinical and radiographic appearance had features common to a number of different pathologic entities. Histologically since it showed moderate dense bundles of collagen fibres with moderate cellularity and discrete masses of calcified deposits, a diagnosis of COF was made. Although a common differential diagnosis for the ossifying fibroma is fibrous dysplasia, the site of occurrence in the present case required consideration for other pericoronal lesions.

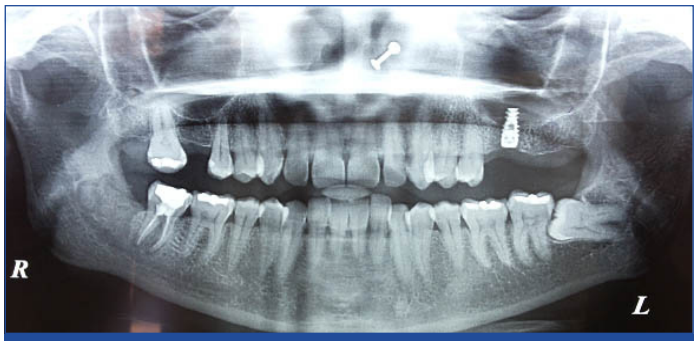
Pericoronal lesion is always a neglected part in case of asymptomatic impacted teeth. Impacted teeth with pericoronal lesion leaves different strategic opinion among the dental clinician, is that whether to consider the pericoronal excised tissue for histopathological evaluation or not. Conventionally, any pathologic conditions that are observed in the clinical practice had to be considered for the histopatholgoical evaluation. The importance of biopsy practice and histopathological evaluation are reinforced through the present case once again. A diagnosis of COF was established in the present case based on the histopathological examination solely. Although the differential diagnosis of radiopaque lesion is possible through radiographic interpretation, a final diagnosis is not established







[Table/Fig-1]: Orthopantomogram showing vertically impacted teeth in the right mandibular region with mixed radio-opaque and radiolucent area distal to the associated teeth. Central radiopaque area surrounded by a thin radiolucent area is observed in the associated teeth. **[Table/Fig-2a]:** (10x view) Hematoxylin and Eosin stained tissue showing fibrocellular stroma with focal areas of mineralization. **[Table/Fig-2b]:** (10x view) Hematoxylin and Eosin Stained tissue showing focal area of mineralization showing cementoid and osteoid area. The cementoid area showing homogenous eosiphillic area and peripherally basophilic stained, note the peripheral brush border and osteoid area showing the homogenous eosinophillic area with concentric arrangement



[Table/Fig-3]: Post-operative orthopantomogram of the patient. (after one year)

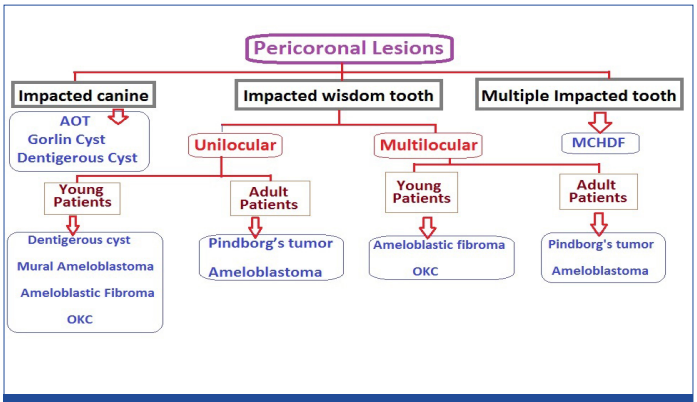
until the histopatholgical grounds. The present case suggests the evidence based dental practice and interdisciplinary approach.

Many pathoses have been reported in the pericoronal region namely: pericoronal hamartomas, odontogenic cysts and odontogenic tumors. Odontogenic hamartomas include the odontoma, pericoronal myxofibrous hyperplasia (PMH), adenomatoid Odontogenic tumor and infantile ameloblastic fibromatosis (IAF) [3]. Dentigerous cysts, Odontogenic keratocyst, paradental cysts, and calcifying odontogenic cysts are common odontogenic cysts associated with pericoronal lesions. A pericoronal involvement of odontogenic tumors reported in the literature includes ameloblastic fibroma, keratocystic odontogenic tumor, calcifying epithelial odontogenic tumor, ameloblastic fibroodontoma, odontogenic fibroma, adenomatoid odontogenic tumor and ameloblastomas [4]. Literatures about the pericoronal involvement in Odontogenic cyst and tumors are well-identified; however, pericoronal hamartomas are scant. The present manuscript attempts to highlight about the pericoronal hamartomas.

The concept of pericoronal hamartomas is gaining new attention in the oral medicine and oral pathology literature due to the higher incidence of pericoronal pathologies among the dental patients with impacted teeth. This supports us to emphasize on pericoronal hamartomas in the present manuscript. As mentioned in the [Table/Fig-1] pericoronal hamartoma includes: Odontoma, Adenomatoid Odontogenic Tumor, Pericoronal myxofibrous hyperplasia and Infantile ameloblastic fibromatosis. Odontomas are well-identified radiographically and histologically by the presence of definite dental structures. Adenomatoid Odontogenic tumor is although represented as benign Odontogenic ectodermal tumor, it is also described as a hamartomatous lesion. AOT is considered as a hamartoma due to the limited size, lack of recurrence and presence of metaplastic mineralization histologically [5]. Yonemochi et al., Proposed two additional lesions to pericoronal hamartomas and it includes the pericoronal myxofibrous hyperplasia (PMH) and infantile ameloblastic fibromatosis (IAF) [6]. PMH and IAF are suggested as the lesions that are induced by mesenchymal tissues; this is different with odontomas, where it is suggested as a result of epithelial and

Pericoronal hamartomas	Odontoma Adenomatoid Odontogenic Tumor Pericoronal myxofibrous hyperplasia Infantile ameloblastic fibromatosis
Odontogenic cysts	Dentigerous cyst Odontogenic Keratocyst Paradental cyst Calcifying odontogenic cyst
Odontogenic tumors	Ameloblastoma Keratocystic odontogenic tumor Calcifying epithelial odontogenic tumor (Pindborg's Tumor) Ameloblastic fibroma Ameloblastic fibroodontoma Ameloblastic fibro-dentinoma Odontogenic fibroma

[Table/Fig-4]: The most common pathologies in the pericoronal region



[Table/Fig-5]: Common pericoronal lesions according to site and age

ectomesenchymal interaction. PMH and IAF are diagnosed by their characteristic odontogenic ectomesenchymal tissues which contain varying calcified materials. PMH's are primarily characterized by the hyperplastic odontogenic mesenchymal tissues with the most prominent histologic feature being submucosal fibrosis. PMH stroma contains a delicate to thick collagen fibrils and multinucleated giant cells that are spindle or stellate-shaped with multiple bizarre and hyperchromatic nuclei. IAF is composed of definite nodular aggregations of cellular and myxofibrous tissue resembling dental papilla. Odontogenic epithelial islands consisting of peripheral palisading basaloid cells and inner polygonal cells with a stellate reticulum-like arrangement are seen in IAF. The dysplastic dental hard tissues are larger and more mature than those seen in PMH.

Multiple calcifying hyperplastic dental follicles (MCHDF), initially described by Sandler et al., [7] and the term given by Gardner and Raddin [8], a distinctive condition that warrants a separate designation, is characterized by multiple unerupted teeth with abundant calcifications and rests of odontogenic epithelium in enlarged dental follicles. Fibro-odontogenic dysplasia, a term introduced by Dominguez et al., [9], shows histologic similarity to MCHDF, but fibro-odontogenic dysplasia shows expansive osseous development and a familial trait.

In 2003, Onishi et al.,[10] proposed a new type of odontogenic pericoronal hamartoma: the "eruption mesenchymal calcified hamartoma" (EMCH). Although uncommon, the EMCH contains mesenchymal multinucleated giant cells and dysplastic dental

matrices such as osteodentin, cementum and pulp-like components.

The exact etiopathogenesis of the COF is unknown. Many authors hypothesized that COF arises from periodontal membrane, as it contain multipotent mesenchymal blast cells that are capable of forming cementum, bone and fibrous tissue [11-13]. Controversy exists for the aforementioned hypothesis since the COF has been reported in non-tooth bearing areas such as ethmoid bone, frontal bone or even the long bones of the body. COFs occurring in non-tooth bearing areas are thought to arise from embryonic nests. Ectopic periodontal membrane differentiating from primitive mesenchymal cells in the petrous bone has been hypothesized by Brademan et al., [14]. Chromosomal abnormalities have also been observed in the ossifying fibroma but the data are still too scarce to determine their patho-genetic significance [15].

The COF is usually reported in young and middle aged adults with a female predilection in a 2:1 ratio. When it occurs in children, it has been termed juvenile aggressive COF and is more aggressive clinically and histopathologically the lesional tissue shows highly vascularized stroma. The COF occurs most commonly below the roots in the premolar-molar area in the mandible, although cases have been reported in the other craniofacial bones. Based on our literature search, the present manuscript is the first to report the COF in the pericoronal region. The lesion usually enlarges slowly and symmetrically resulting in bone expansion with facial deformity, but some lesions are asymptomatic and are only discovered during routine radiographic examination. Radiographically it can present with different patterns depending upon the degree of mineralization, ranging from an immature, radiolucent and cyst-like lesion with scattered radiopaque foci to mature dense sclerotic lesions. The borders are well-defined and usually a thin radiolucent line representing a fibrous capsule separates the lesion from the surrounding bone.

Curettage and Surgical excision are the treatment modality for the COF. The prognosis of the lesion is usually good, since the lesion can be surgically excised without leaving remnants of lesional tissue, as the lesion has a good delimitation of the border. A one-year follow up of our case showed good healing with no complications.

CONCLUSION

A suggestion of cemento-ossifying fibroma in the list of pericoronal pathology is proposed through this manuscript. Although, many lesions are associated with impacted / unerupted teeth. Pathology associated with impacted teeth requires attention to investigation and appropriate treatment since a “wait and watch” approach, may result in the expansion of the lesion and destruction of the adjacent bone, due to the disease activity. This case report updates the knowledge of the dental clinician to include the cement-ossifying fibroma in the differential diagnosis panel of the pericoronal pathology with mixed radiolucent and radiopaque appearance. Further, it is emphasized that a lesion in the pericoronal region of an impacted tooth may in fact be a more unusual entity than expected. Hence, the present manuscript reinforces the concept of the histopathology requirement and evidence based dentistry.

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