Central Odontogenic Fibroma (WHO Type): A Case Report and Review of Literature

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ABSTRACT

We report a case of a 15-year-old boy presented with painless swelling of right side of lower jaw since 2 years. A provisional diagnosis of dentigerous cyst involving an impacted lower second premolar was concluded after clinical examination. Radiographs revealed an expanding, well demarcated radiolucency with thin, fine, straight septae, causing root resorption and displacement of adjacent teeth and the impacted tooth was pushed to the inferior border of mandible. Histologically pronounced fibroblastic hypercellularity with epithelial rests and focal areas with dentinoid or cementoid material was seen, suggestive of Central Odontogenic Fibroma (WHO Type). Only eight such cases have been reported in English literature and present one is 9th case.

Keywords: Odontogenic fibroma, Mandibular swelling, WHO type.

INTRODUCTION

The central odontogenic fibroma (COF), as the name suggests, is a benign odontogenic neoplasm occurring within the jaw. Based on its anatomic location and histological appearance, this neoplasm is considered to be a tumor originated from proliferation of mature odontogenic mesenchyme. It is sufficiently rare that little is known about the neoplasm and is been infrequently reported in the literature. According to the new World Health Organization (WHO) classification, it is defined as a fibroblastic neoplasm that contains varying amounts of apparently inactive odontogenic epithelium. Some lesions may contain varying amounts of hard tissue that resembles dysplastic cementum or bone.

REVIEW OF LITERATURE

It was included in WHO classification of benign tumors in 1971, since then there has been a controversy as to the concept and definition of this lesion because the term COF has been applied to various types of lesions. The odontogenic fibroma (OF) is classified as a benign lesion derived from ‘odontogenic ectomesenchyme with or without odontogenic epithelium’. Wesley et al in 1975 suggested a set of criteria for diagnosing odontogenic fibroma as follows:

1. Clinically, the lesion is central in the jaws and has a slow persistent growth that results in painless cortical expansion.
2. Radiologically, its appearance varies, but, like the ameloblastoma and odontogenic myxoma, most examples are multicocular radiolucent lesions that involve relatively large portions of the jaws in the later stages. In some instances, they may be associated with unerupted and/or displaced teeth.
3. Histopathologically, the most consistent feature is a tumor composed predominantly of mature collagen fibers with numerous interspersed fibroblasts. The presence of small nests and/or strands of inactive odontogenic epithelium is a variable feature.
4. The lesion is benign and responds well to surgical enucleation with no tendency to undergo malignant transformation.

Gardner (1980) attempted further clarification of lesions previously described as odontogenic fibroma and classified them into three different, yet probably related lesions:

1. First, the hyperplastic dental follicle.
2. Second, a fibrous neoplasm with varying collagenous fibrous connective tissue containing nests of odontogenic epithelium—COF (simple type).
3. Third, a more complicated lesion with features of dysplastic dentine or cementum-like tissue and varying amount of odontogenic epithelium (WHO type) which was later designated as COF (WHO type). Current WHO publication mention the COF (WHO type) by either ‘odontogenic fibroma complex type’ or ‘fibroblastic odontogenic fibroma’.
Handlers et al were of the opinion that separation into simple and WHO types was arbitrary and inconsistent and proposed that all lesions meeting the criteria be classified solely as COF.

Sepheriadou-Mavropoulou et al (1985) believed it to be the counterpart to the peripheral odontogenic fibroma that arises from the periodontal membrane. Indeed, the COF may also arise from partially induced somatic mesenchyme otherwise destined to become periodontal membrane. However, this origin would not be from the odontogenic apparatus per se; instead, it could be said that it arises from the true odontogenic mesenchyme influenced by the odontogenic apparatus.

The alternative theory (Heimdal A 1980) was that it arises from the true odontogenic mesenchyme of the dental papilla, like the odontogenic myxoma, but that it differs from the myxoma by the maturity of the mesenchyme and its limited growth potential and invasiveness.

Till now, i.e. inclusive of our case a total of 73 cases of COF comprising of simple and WHO type, have been identified. Out of these 73 cases, 17 cases of WHO type have been proven histologically (Table 1).

### CASE REPORT

A male patient aged 15 years, reported to our department with chief complaint of painless swelling in the right lower face region since 2 years. The history of presenting illness revealed that the patient had undergone extraction of a tooth in the area of chief complaint 2 years back. Since then a swelling arose which gradually increased in size to its present size. The swelling was asymptomatic without pain or paresthesia or any other associated symptoms. All his vital signs were within normal limits.

Extraoral examination revealed asymmetry of face with a swelling on the right lower side of face measuring around 3 × 3 cm in size, ovoid in shape, extending from 1 cm away from midline and 1 cm medial to angle of mandible anteroposteriorly and from angle of mouth to 1 cm below the lower border of mandible superioinferiorly. The overlying surface was normal. Swelling was firm in consistency and the lower border of mandible was irregular with a hard structure present on palpation. Single right submandibular lymph node was palpable, solitary, firm, mobile and nontender.

Intraoral swelling was present measuring around 3 × 3 cm in size, extending from right mandibular canine to right mandibular first molar causing expansion of buccal cortical plate and vestibular obliteration (Fig. 1). The swelling was nontender and nonfluctuant on palpation. Clinically, right mandibular premolar was missing. There was grade I mobility in right mandibular canine.

A working diagnosis ‘dentigerous cyst’ involving right mandibular second premolar was made. Clinical differential diagnosis of adenomatoid odontogenic tumor and odontogenic myxoma was made.

Electric tooth vitality test demonstrated positive response. Hematology investigations were found to be normal. Fine needle aspiration biopsy using topical anesthetic was performed. No aspirate was obtained.

IOPA showed an area of mixed radiodensity with radiolucency and radiopacit. Absence of lamina dura of right mandibular canine was seen along with displacement of tooth and horizontal pattern of root resorption. Occlusal radiograph showed an expansion of buccal cortical plate (Fig. 2). Orthopantomogram revealed well-defined radiopaque borders

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Mandi: mandibular, Maxi: maxillary, Rt: right, M: male, F: female, NS: nothing specified, Age in years

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**Table 1:** Review of histologically proven COF (WHO type) updated as on January 1st, 2009
of a mixed radiodensity extending from right mandibular canine to mesial root of right mandibular first molar. Thin, fine, straight bony septa were seen due to entrapment of residual bone within the tumor. Mandibular right second premolar was impacted and displaced below the lesion near the inferior border of mandible. Root resorption and tooth displacement were also seen. CT axial sections showed homogeneous soft tissue mass measuring approximately 3.3 × 1.2 cm. Expansion and thinning of buccal cortical plate with thin and straight septa were also noted (Fig. 3).

Under general anesthesia, the lesion was enucleated and curettage was done and sent for histologic investigation. The biopsy specimen revealed mature fibrous proliferation with pronounced fibroblastic hypercellularity. Epithelial cell rests were scattered throughout the lesion. Focal areas showed calcified material resembling dentin or cementum in relation to epithelial islands (Fig. 4).

Based on clinical, radiological and histological findings, a final diagnosis of COF (WHO type) was made. The prognosis of the lesion was good and the patient is still under follow-up.

**DISCUSSION**

Shafer et al\(^1\) (1983) stated that of all odontogenic tumors, this lesion has the most poorly defined parameters. One must be careful when assessing the literature because some authors have lumped the central and peripheral variants into one group.

COF has been classically divided into two histologically distinct variants. The first has been called the simple type. The second type is designated the WHO type, contains mineralized material that has been interpreted as osteoid, cementum-like or dysplastic dentin. Dysplastic dentin is usually found in close to the odontogenic epithelium. The WHO type has more islands of odontogenic epithelium. Gardner (1980)\(^5\) discussed the histologic characteristics of these two types in detail. Most believe that WHO type occurs much less commonly.

Most lesions are seen as painless swelling and appear as an asymptomatic expansion of buccal and lingual cortical plates. It is a slowly enlarging hard swelling which is sometimes recognized by presence of slow growing diastema, due to the dislocation of the adjacent teeth. The affected patients may have mobility of teeth.\(^17\) According to Neville,\(^18\) there is a slight female preponderance with a ratio of 2.2 to 1. Ratio of women to men and the distribution of odontogenic fibroma in the jaws is as follows:

- **Maxilla:** Anterior region 29%, premolar region 6%, molar region 10%.
- **Mandible:** Anterior region 8%, premolar region 18%, molar region 29%.

Radiographically, the most common location was molar region in the mandible.\(^19\) The anterior mandible was affected in four cases; however, there was usually extension posteriorly to the premolar or molar region. The case being reported here in the right molar region in a 15-year-old boy is consistent with the clinical and morphological characteristics of COF stated before. A multilocular pattern was observed in at least half of the cases.\(^19\) In some reports, it was found that there are more unilocular than multilocular radiolucencies.\(^12\) The smaller lesions are unilocular while the larger ones tend to have...
scalloped margins or are multilocular. Expansion of the cortex occurs in large lesion, as in our case and perforation of cortex is not a feature. The margins are well-defined, circumscribed or well-demarcated in most cases, but there is usually no sclerotic margin. The septa of bone forming the locules may not appear very radiopaque.\textsuperscript{20} It is possible that some lesions contain radiopaque flecks, in rare instances the lesion may appear as a homogeneous radiopaque mass. Several cases were associated with an impacted tooth. Teeth may be displaced and root resorption may be seen. This feature coincides with ours as our patient had an impacted and displaced mandibular second premolar.

The mode of treatment of COF is enucleation and curettage. Recurrence is uncommon. But on the evidence of cases of recurrence, it is suggested that these patients must be followed up postoperatively. There has not been any recurrence in present case 3 months after surgery and the patient is still under follow-up.

**CONCLUSION**

The purpose of this report is to present an additional case of COF as well as to compare its clinical, radiological and histological features with the cases reported till date in the review and to understand the need of long-time postoperative follow-up and to develop a better understanding of COF.

**REFERENCES**