Central Odontogenic Fibroma: A Case Report and Review of Literature
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Abstract
Central odontogenic fibroma is an extremely rare benign tumor, accounting for less than 0.1% of all odontogenic tumors. Clinically, the lesion is slow growing and produces cortical expansion. Radiologically, the most common finding is multilocular radiolucency. Histologically, the lesion is characterized by mature collagen fibers and numerous fibroblasts. COF responds well to conservative surgical excision with no tendency for recurrence. Here, a case of central odontogenic fibroma of the maxilla in a 15-year-old boy is described. The lesion was an asymptomatic swelling with well-defined borders covered by normal mucosa. The lesion presented as a unilocular radiolucency with foci of radio opacity in relation to the root of the maxillary anterior teeth. The lesion was surgically removed and analyzed histopathologically. There were no postoperative complications.

Keywords: Odontogenic Fibroma; Benign Neoplasm; Fibrous Tissue; Desmoplastic; Fibroblasts.

Introduction
Central odontogenic fibroma (COF) is an uncommon lesion almost always associated with the crown of an unerupted tooth. It has been reported in patients of age ranging from 4 to 80 years and shows strong female predilection. Smaller odontogenic fibromas are asymptomatic and on radiographic examination tend to be well-defined, unilocular, radiolucent lesions. Larger lesions tend to be multilocular, probably showing sclerotic borders.1 Central odontogenic fibroma (COF) is an uncommon benign neoplasm composed by varying amounts of inactive-looking odontogenic epithelium embedded in a neoplastic mature and fibrous stroma.2 The lesion may evolve from a dental germ (dental papilla or follicle) or from the periodontal membrane, and therefore is invariably be related to the coronal or radicular portion of teeth. Due to its non-exclusive histological features, this lesion may be confused with other entities, such as hyperplastic dental follicles, odontogenic myxomas, and desmoplastic fibromas, which highlight the importance of clinico pathological correlation in the diagnosis of odontogenic fibromas.3

Controversy exists as to concept and definition. At present the term COF is applied to two histological types of lesions: the epithelium-poor type (formerly termed simple type) and the epithelium-rich type (formerly termed complex or WHO-type). Sub variants of the two types (among which is the granular cell variant) have been described.4 Since then, other authors have reviewed and reclassified available material using these criteria. Regezi et al., found no case of odontogenic fibroma in a series of 706 odontogenic tumors. Gardner in 1980 attempted further clarification of lesions previously described as odontogenic fibroma and classified them into 3 different, yet probably related lesions.5 The third type, a more complicated lesion with features of dysplastic dentine or cementum-like tissue and varying amount of odontogenic epithelium WHO type was similar to the calcifying odontogenic tumor described by Pindborg in WHO publication in 1971, Gardner designated it as odontogenic fibroma (WHO type). The distinguishing features between the 2 lesions are that in the calcifying odontogenic tumor but not in the odontogenic fibroma (WHO type) this lesion stains positive with amyloid stains.

Case Report
A 15 years old male patient reported to the department of oral and maxillofacial surgery with a chief complaint of painless, hard swelling, gradually increasing in size since last six months in right upper jaw. There was no history of trauma or any decayed teeth in that region. The patient had no relevant medical history. On examination, there was slight swelling in right maxillary anterior region causing slight facial asymmetry. The swelling was non tender (Fig 1a and 1b). Intra orally hard, non tender swelling about
3 × 3 cm size was present on the buccal aspect, extending from right maxillary central incisor to maxillary first premolar. The involved teeth were mobile. The teeth were non tender to vertical and lateral percussion. Oral hygiene status was fair. The patient’s blood reports were within normal limits. Intraoral periapical and orthopantomograph revealed well circumscribed radiolucency of about 3 × 3 cm size with well-defined borders and some radio opaque flecks within the radiolucency. The roots of tooth #12, #13 and #14 were displaced (Fig 1c). Aspiration was negative and there was absence of any fluid, blood or pus.

Figure 1: The extraoral clinical appearance of the patient (a and b) with the orthopantomograph showing a mixed lesion.

The differential diagnosis of odontogenic fibroma, ossifying fibroma, desmoplastic fibroma, adenomatoid odontogenic tumor, odontogenic myxoma, calcifying Odontogenic cyst, unicystic ameloblastoma, and central giant cell granuloma was made. The lesion was surgically excised and the specimen was sent for histopathological examination for confirmatory diagnosis. The excised mass had glistening smooth surface (Fig 2a). Healing was uneventful. Histopathological examination revealed whorled pattern of fibroblastic activity with interlacing collagen bundles and uniform distribution of fibroblasts. Numerous amorphous calcifications were seen in supporting collagen, which were suggestive of simple Odontogenic fibroma (Fig 2b, c and d). Lesion was surgically removed. Post operatively the outcome was satisfactory on follow up.

Discussion
Gardner in 1980 classified lesions described as COF into three categories: 1) the hyperplastic dental follicle; 2) a fibrous neoplasm with collagenous fibrous connective tissue containing odontogenic epithelium – (simple type); and 3) a lesion with dysplastic dentine or tissue like cementum and odontogenic epithelium (WHO type). The ill-defined nature of central odontogenic fibroma is illustrated by the plethora of alternative terms which include odontogenic fibroma, nonosteogenic fibroma of the jaw, osteogenic fibroma, odontogenic fibroma with calcification and central fibroma of the mandible (Appel and Verbin, 1985). As the histological features of some of these tumours are identical to appearances seen in hyperplastic dental follicles or the walls of dentigerous cysts it is probable that some of the reported cases represent these entities rather than distinctive neoplasms.

In the review by Handlers et al, the 39 cases found revealed an incidence of 22 cases in the maxilla to 17 cases in the mandible with a female: male ratio of 3:1 and an age range of 11-80 years. The COF is a benign neoplasm reported in the literature that is
usually diagnosed in the second and third decade of life.\textsuperscript{8} The most usual sign is swelling of the mandible or maxilla and less frequently pain and paresthesia are observed. Wesley et al in 1975 suggested a set of criteria for diagnosing odontogenic fibroma as follows:\textsuperscript{9} Clinically, the lesion is central in the jaws and has a slow persistent growth that results in painless cortical expansion. Radiologically, its appearance varies, but, like the ameloblastoma and odontogenic myxoma, most examples are multilocular 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. 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. The lesion is benign and responds well to surgical enucleation with no tendency to undergo malignant transformation.

Figure 2: The gross soft tissue specimen (a). The hematoxylin and eosin stained photomicrograph at low power view (b), at high power view (c and d) showing collagenous stroma with fibroblasts presenting in whorled pattern and eosinophilic calcifications.

According to the latest classification of odontogenic tumors reported by Gardner (1996),\textsuperscript{3} the odontogenic fibroma (OF) is classified as a benign lesion derived from “odontogenic ectomesenchyme with or without odontogenic epithelium”. This classification has been applied to various types of lesions, characterized by the presence of connective tissue containing islands and strands of odontogenic epithelium, and sometimes associated with the presence of dystrophic calcification. Gardner reviewed the information concerning the central odontogenic fibroma identifying lesions with two different histologic patterns. The first type, classified as simple, contained fibrous tissue with various amounts of collagen and the second type, which has been referred to as the WHO type or complex type, contained fibrous tissue with myxoid area associated to odontogenic epithelium. Moreover, dysplastic dentin and cementum-like material could be found.\textsuperscript{5}

The simple type COF is the most collagenous variant on the histologic spectrum of myxoma, myxofibroma and odontogenic fibroma. The simple type COF is an expansile lesion that does not infiltrate the surrounding bone, whereas the myxoma does. Our present case showed simple type of COF. The simple type COF is defined as, an expansile, non-infiltrating connective tissue lesion resembling dental follicle. It is relatively acellular, the fibres being quite...
delicate, and there is considerable amount of ground substance yielding a fibromyxoid quality. It may show inactive-looking rests of odontogenic epithelium but seldom are they numerous. Occasionally, nondescript calcifications are found.\textsuperscript{10}

Importance of making a correct differential diagnosis of COF with endodontic lesions showing the radiolucent image has been argued. Radiologically, COF manifests as a uni or multilocular image with well defined margins and surrounded by a sclerotic halo.\textsuperscript{11} Ameloblastic fibromas are distinguished from COF by the fact that both the epithelial and mesenchymal components are neoplastic, while in COF, is only the mesenchymal.\textsuperscript{12} Distinguishing COF from desmoplastic fibroma of the jaws remains an insoluble problem. Microscopically COF exhibit relatively abundant ground substance (which gives the paraffin sections a bluish tinge when stained by hematoxylin and eosin) and not being particularly collagenous. By definition, desmoplastic fibromas are collagenous. Theoretically, distinguishing between these two lesions is important because desmoplastic fibromas infiltrate surrounding tissues while odontogenic fibromas do not. Wider surgical margins would be indicated if the lesion was considered to be a desmoplastic fibroma. Recurrences of COF are not common.

In conclusion, it is essential that oral and maxillofacial surgeons, radiologists and pathologists integrate all relevant and available information to come up with a correct diagnosis of central odontogenic fibroma and appropriate disease management.

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