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Case Report

Central Odontogenic Fibroma: A Diagnostic Dilemma with Literature Review

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Abstract

Central odontogenic fibroma is a rare benign tumor accounting for only 0.1% of all odontogenic tumors. A slow growing tumor, usually asymptomatic and is found in routine radiographic examinations. It has been stated that of all odontogenic tumors, this lesion has the most poorly defined parameters. The present case is of a 27 year old female patient having central odontogenic fibroma which was incidentally found on a radiograph. The clinical features and radiographic appearances lead to a diagnostic dilemma between an inflammatory and a tumorous lesion. The diagnosis of the lesion was confirmed on histopathological evaluation after surgical enucleation.

Keywords: Central Odontogenic Fibroma; WHO Type; Odontogenic; Tumor; Depression; Dentinoid.

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Introduction

Central odontogenic fibroma (COF) is an extremely rare benign tumor accounting for only 0.1% of all odontogenic tumors[1] and accounts for 6.1% of all central odontogenic tumors.[2] This lesion is considered to be derived from ectomesenchymal tissue of dental origin such as periodontal ligament, dental papilla, or dental follicle. Clinically, it presents as a slow growing asymptomatic mass which, in most cases, can remain unknown until the appearance of a swelling. In more severe cases root resorption and displacement of adjacent teeth have been reported.[3] It is composed of varying amounts of inactive-looking odontogenic epithelium embedded in a neoplastic mature and fibrous stroma. Due to its non-exclusive histological features, this lesion may be confused with other entities, such as hyperplastic dental follicles, odontogenic myxoma and desmoplastic fibromas, which highlight the importance of clinicopathological correlation in the diagnosis of odontogenic fibromas.[4] The present case is discussed with clinical findings, radiographic and histopathological features of a central odontogenic fibroma in the left canine-premolar area of the maxilla in an adult female patient.

Case Report

A 27 year old female patient visited the department of Oral Medicine and Radiology with a chief complaint of pain in the upper left back region since two months. The pain was sudden in onset, mild, dull type and intermittent in nature which was experienced only while eating food. Patient gave a past dental history of a silver filling done five months back and an uneventful extraction three years back in the mandibular right posterior region. Intra orally a small circular depression measuring approximately 3 mm in diameter with a smooth base could be palpated on the palatal mucosa approximately 1.5 cm medial to the marginal gingiva of 24 and 25 (Fig 1a). There was secondary caries with amalgam restoration in 26 which was tender on vertical percussion. Tenderness on percussion was absent with 22, 23, 24 and 25. A single small reddish round swelling measuring approximately 1x1 mm in diameter was seen on the attached gingiva in relation to 26 which was non tender, soft in consistency, non-compressible, non-reducible and without any suppuration on palpation suggesting it to be a parulis (Fig 1b). Also there was a missing tooth i.e. 46. Following this a pulp vitality test was done for 22, 23, 24, 25 and 26 which revealed 23, 24, 25 and 26 to be non-vital teeth. A provisional diagnosis of dento-alveolar abscess secondary to secondary caries with 26 was made. An intra-oral periapical (IOPA) radiograph was advised for that region which showed a single ill-defined diffuse radiolucency at the apical 1/3rd of the palatal root with thinning of trabeculae in the region of 26 suggesting it to be rarefying osteitis and also a well-defined radiolucency around 25 which was extending further more anteriorly. So another IOPA of 23, 24, & 25

region was made, which showed a well-defined oval radiolucency extending antero-posteriorly from distal aspect of 22 to mesial aspect of 26 and superiorly from the floor of the nasal fossa and maxillary sinus to inferiorly to the middle 1/3rd of roots of 23, 24, 25, & 26 measuring approximately 2.7x1.5 cm (Fig 1c).

In order to visualize the entire radiolucency an occlusal radiograph and an orthopantamogram were also made (Fig 1d & e). The radiographs revealed a well-defined unilocular radiolucency measuring approximately 2.5x1.8x1.5 cm which had irregularly corticated superior margin and non-corticated inferior margin which extended between the teeth. The surrounding structures appeared to be normal. Radiographic differential diagnosis of periapical cyst, adenomatoid odontogenic tumor (extra follicular), Globulomaxillary cyst, nasopalatine cyst, odontogenic Keratocyst and central odontogenic fibroma was made. Fine needle aspiration cytology was done which gave a negative aspiration suggesting it to be more likely a tumor mass.

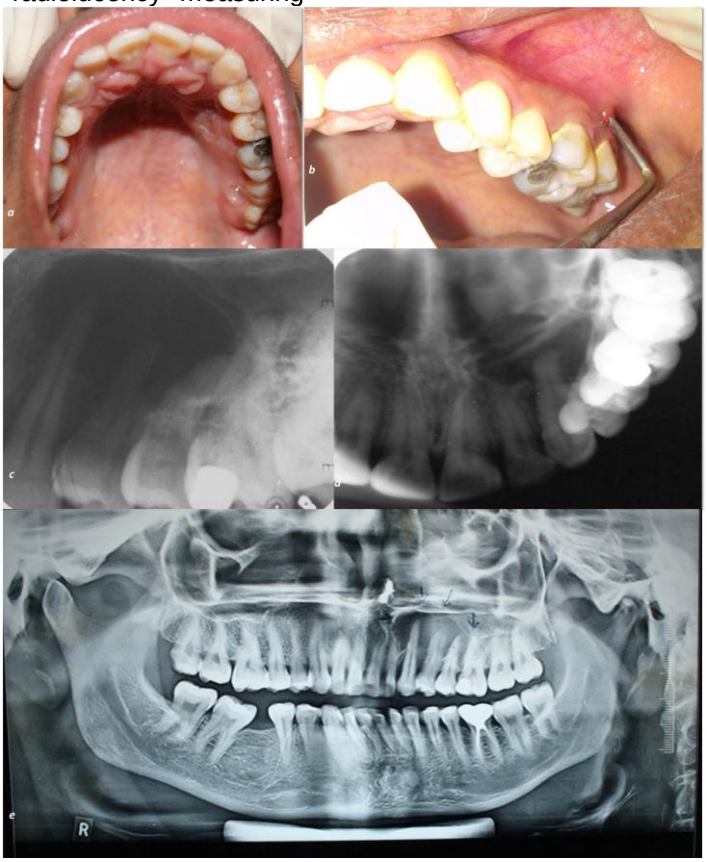

Figure 1: The Intra-oral photograph showing palatal depression between 24 and 25 (a) and parulis in relation to 26 (b). The Intraoral periapical radiograph showing a well-defined radiolucency in relation to 23, 24 and 25 and rarefying osteitis with 26 (c), maxillary occlusal view showing the medio-lateral extension of the lesion (d) and Orthopantamogram showing the superio-inferior and antero-posterior extension of the lesion (e).

This was preceded by root canal treatment of 23, 24, 25 and 26. Following this an enucleation was performed with apicectomy of 23, 24 and 25 under local anesthesia. A buccal muco-periosteal flap was raised and the lesion with the cortical bone that surrounded it was exposed. After removing the cortical plate, the soft lesion was enucleated from the bone (Fig 2a). The surgical specimen was fixed in 10% neutral formalin and submitted for histopathological examination (Fig 2b). The hematoxylin and eosin stained slide showed connective tissue showed devoid of epithelium. The connective tissue shows abundant uniformly arranged mature collagen fibers in the story form pattern with few inflammatory component mainly lymphocytes at places. In few areas dentin like material (dentinoid) in the form of spicules were evident. Odontogenic islands are widely distributed within the connective tissue. At places, bundles of wavy fibers with spindle shaped wavy nuclei indicative of nerve fibers are

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present (Fig 2c, d & e). By correlating clinical, radiographic and histopathological features a definitive diagnosis of central odontogenic fibroma, WHO type was made.

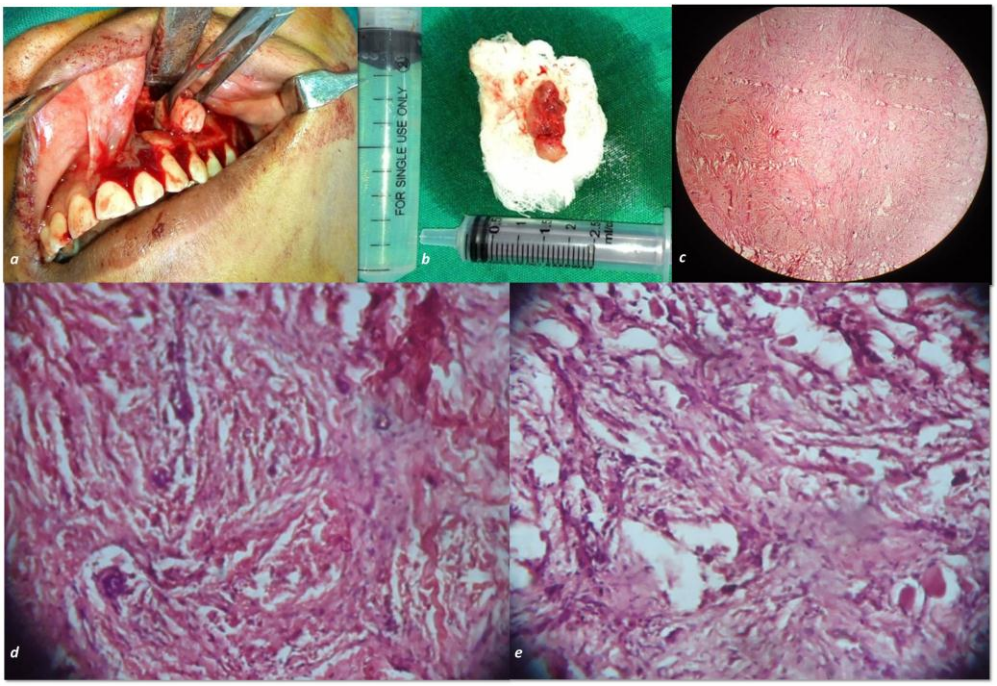

Figure 2: The Intraoperative clinical picture showing the enucleation of the lesion (a) with excised specimen (b). The photomicrograph of hematoxilin-eosin stained section at x10 showing dense fibrillar connective tissue with odontogenic epithelial islands (c), at x40 showing odontogenic epithelial islands in fibro-cellular stroma (d) and dentin like material (e).

Discussion

According to the World Health Organization (WHO) Classification, central odontogenic fibroma 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.[5] It was included in WHO classification of benign tumors in 1971 as a benign lesion derived from 'odontogenic ectomesenchyme with or without odontogenic epithelium.

Wesley et al.,[6] suggested a set of criteria for diagnosing odontogenic fibroma as follows:

a. Clinically, the lesion is central in the jaws and has a slow persistent growth that results in painless cortical expansion.
b. 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.
c. 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 are variable feature.
d. The lesion is benign and responds well to surgical enucleation with no tendency to undergo malignant transformation.

Sepheriadou Mavropoulou et al., (1985) believed it to be the counterpart to the peripheral odontogenic fibroma that arises from the periodontal membrane. 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.[7] Kaffe et al.,[8] in the meta-analysis of the literature with 51 lesions showed that the patients diagnosed with COF ranged from four to 80 years with a mean age of 34.4 years and a male to female ratio of 1:2.2. Of these 55% were located in the mandible and 45% in the maxilla. Of the mandibular lesions, 54% involved the molar while 32% involved the premolar region, of the maxillary lesions, the anterior area was mostly involved (65%).[8] 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.[9] In 2006, Buchner et al, studied the odontogenic tumors from northern California and reported 15 cases of COF. According to this study the age of patients ranged from 11 to 50 years with a mean age of 36 years, a male to female ratio of 15:1 and highest frequency of occurrence in the fifth decade of life. There was no significant predilection for the location either in the maxilla or in the mandible.[2] However, on a retrospective study of 8 clinical cases of COF, Hrichi et al.,[10] found a predilection for male sex (1.67:1) and the most common location of the tumor was on the mandible. The average age was 19.9 years with an age range of 11–38 years.[10] In the present case it was seen in a 27 year old female patient in the maxillary anterior region which is in accordance with the literature.

Clinically, the most frequently observed sign is swelling and moreover several clinical cases showed the presence of slowly growing diastema, due to the dislocation of the adjacent teeth. Clinical symptoms such as pain and paresthesia were uncommon.[11] The present case was asymptomatic and without any swelling and cortical expansion. Central odontogenic fibroma often presents with a cortical bony depression of the palatal contour which was seen in our patient.[12]

Radiologically, the COF has been described both as a well-defined radiolucent area that stimulates a unilocular ameloblastoma or odontogenic cyst and a multilocular radiolucent lesion with well-defined borders. Kaffe et al.,[8] in their study discussed that the majority of small COFs are unilocular radiolucent lesions (55%), whereas large lesions tend to be multilocular (29%) with most cases having well defined borders. They also found that in some instances it may exhibit a mixed radiolucent/radiopaque (12%) appearance with poorly defined or diffused borders. They concluded that the great variability in radiologic appearance of COF emphasizes that despite its rarity it should be considered in the differential diagnosis of all abnormal radiolucencies of the jaws.[8] In some cases due to its location near the root apices COF can mimic a lesion of endodontic origin. Periapical radiolucencies of non-endodontic origin occur infrequently so a proper diagnosis must be made keeping in mind the other etiologies. Though COF is rarely reported it must be considered as one of the differential diagnosis for periapical radiolucency associated with vital and non-vital teeth. In the present case the lesion was large, well defined, unilocular, associated with the roots of non-vital teeth and hence a provisional diagnosis of a periapical lesion was given.

Gardner made an attempt to classify COF into two variants namely: 1) Simple type and 2) WHO type. The basic difference between the simple and WHO type is that the stroma of the simple fibroma mimics that of a dental follicle from which it is probably derived. Histologically, the simple type exhibits relatively acellular delicate fibers which are interspersed with considerable amount of ground substance. On the other hand the stroma of the WHO type exhibits high cellularity. It occurs as fibroblastic strands which may be interwoven with less cellular areas. The epithelial rests are dispersed sparsely in the simple type. In contrast, the odontogenic epithelium is an integral component of the WHO type. The other difference between these two variants is the presence of foci of calcifications of the collagenous materials in the WHO type which are described as cementoid, osteoid, and dysplastic dentin by several other authors. The probable reason for dissimilar histological presentation of odontogenic fibromas is attributed to the tissue of origin.[13]

The current classification of odontogenic fibroma by WHO (2005) is: 1) The WHO variant, and 2) The non-WHO variant. The WHO variant is considered as a mesenchymal odontogenic tumor and is comprised of two distinct cell types, a fibrous element, and an epithelial component that resembles dental lamina or its remnants. In contrast, the non-WHO variant lacks an epithelial component and is said to be a monomorphic fibroblastic tumor, purported to be of odontogenic mesenchymal origin and ostensibly derived from pulpal or follicular fibroblasts.[14]

As COF is considered a benign odontogenic tumor the treatment of choice is enucleation with careful follow up for a few years, although few cases of recurrence have been reported. Dunlap and Barker[15] presented two cases of maxillary odontogenic fibroma treated by curettage with a follow-up of nine and ten years with no evidence of recurrence.[15] Also none of the eight cases reported by Hrichi et al.,[10] showed recurrence on a follow up of two years after

surgery. However, Ramer et al., reported a 12.8% (five out of 39 cases) rate of recurrence.[16] Although the tendency toward recurrence is relatively low, postoperative patient follow-up for five years is advisable.[17]

Conclusion

The purpose of this report is to present an additional case of this rare lesion, emphasize on the diagnostic difficulty of this lesion and put together the important findings known in the literature about COF.

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