

Central Odontogenic Fibroma of Maxilla: A Rare Case

P Balaji¹, Ashish Gupta², Poornima Govindraju³, Vinitra Vasan², Usha Jambunath²

¹Professor, Department of Oral Medicine and Radiology, Rajarajeswari Dental College and Hospital, Bengaluru, Karnataka, India,

²Post-graduate student, Department of Oral Medicine and Radiology, Rajarajeswari Dental College and Hospital, Bengaluru, Karnataka, India,

³Reader, Department of Oral Medicine and Radiology, Rajarajeswari Dental College and Hospital, Bengaluru, Karnataka, India

Abstract

Central odontogenic fibroma (COF) is a rare benign odontogenic tumor characterized by variable amounts of inactive-looking odontogenic epithelium embedded in a mature fibrous stroma corresponding to 0-5.5% of all odontogenic tumors. COF may be seen at any age, but it is diagnosed most frequently in patients between the second and fourth decades of life. The lesion is asymptomatic except the swelling of the jaw. It occurs slightly more often in the mandible, and the prevalent site in the mandible is the molar-premolar region whereas in maxilla it occurs anterior to the first molar. The periphery usually is well defined where smaller lesions are usually unilocular, and larger lesions show a multilocular pattern. The internal septa may be fine and straight, as in odontogenic myxomas, or it may be granular, resembling those seen in giant cell granulomas. COF may cause expansion with maintenance of a thin cortical boundary or on occasion can grow along the bone with minimum expansion, similar to an odontogenic myxoma. Tooth displacement is common, and root resorption has also been reported. Here, we present a rare case of COF in the left premolar area of the maxilla in a 14-year-old male.

Key words: Anterior maxilla, Central odontogenic fibroma, Maxilla

INTRODUCTION

Central odontogenic fibroma (COF) is a rare benign odontogenic tumor characterized by variable amounts of inactive-looking odontogenic epithelium embedded in a mature fibrous stroma¹ which corresponds to 0-5.5% of all odontogenic tumors recorded in various studies.²⁻⁴ COF may be seen at any age, but it is diagnosed most frequently in patients between the second and fourth decades of life.⁵ The lesion is asymptomatic except the swelling of the jaw.⁶ It occurs slightly more often in the mandible, and the prevalent site in the mandible is the molar-premolar region whereas in maxilla it occurs anterior to the first molar. The periphery usually is well-defined where smaller lesions are usually unilocular, and larger lesions show a multilocular pattern. The internal septa may be fine and straight, as in odontogenic

myxomas, or it may be granular, resembling those seen in giant cell granulomas. COF may cause expansion with maintenance of a thin cortical boundary or on occasion can grow along the bone with minimum expansion, similar to an odontogenic myxoma. Tooth displacement is common, and root resorption has also been reported. Here, we present a rare case of COF in the left premolar area of the maxilla in a 14-year-old male.

CASE REPORT

A 14-year-old male patient reported to the department of oral medicine and radiology with a chief complaint of swelling on the left side of the face since 6 months (Figure 1). The patient first noticed the swelling 6 months back, and it was increasing in size gradually. The swelling is not associated with pain. There was no history of trauma or any decayed teeth in that region. The patient's medical history and other histories were non-contributory.

On general physical examination, the patient was moderately built and nourished; all his vital signs were within normal limits. Extraoral examination revealed asymmetry of the face with a diffuse swelling on the left

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www.ijss-sn.com

Month of Submission : 01-2015
Month of Peer Review : 01-2015
Month of Acceptance : 02-2015
Month of Publishing : 03-2015

Corresponding Author: Dr. Ashish Gupta, Department of Oral Medicine and Radiology, Rajarajeswari Dental College and Hospital, #14 Ramohalli Cross, Mysore Road, Bengaluru, Karnataka, India. Phone: +91-9886527337. E-mail: ashes4987@gmail.com

middle third of the face measuring around $6\text{ cm} \times 6\text{ cm}$ in size, extending from the left ala of the nose till 5 cm anterior to the tragus of the left ear anteroposteriorly and from the left infraorbital margin till the left corner of the mouth. The overlying surface was normal. The swelling was firm to hard in consistency with diffuse borders and was non-tender on palpation (Figure 1).

On intraoral examination, swelling measured around $4\text{ cm} \times 3\text{ cm}$ in size, extending from left maxillary lateral incisor till the left maxillary second molar causing expansion of the cortical plate and vestibular obliteration. The swelling was non-tender and non-fluctuant on palpation. Clinically, left maxillary canine was missing (Figure 2).

Based on patient's history and clinical findings, working diagnosis of dentigerous cyst involving left maxillary canine was made. Clinical differential diagnosis of adenomatoid odontogenic tumor and fibrous dysplasia - monostotic type was considered.



Figure 1: Extra oral photograph with swelling on the left side of the face



Figure 2: Intra oral photograph showing buccal cortical expansion

Patient was subjected to chair side investigations like electric tooth vitality test which revealed positive response in all teeth of the second quadrant and all routine blood investigations were within normal limits. Aspiration could not be performed as there was no yielding point for needle insertion.

Patient was subjected to radiographic examination. Orthopantomograph revealed a multilocular radiolucency extending from left maxillary central incisor till the second premolar anteroposteriorly and from the crest of the maxillary arch from the region of missing left canine till the left infraorbital margin (Figure 3). The left canine was displaced superiorly till the infraorbital margin and the left maxillary sinus was obliterated.

Intraoral periapical radiograph showed an area of multilocular radiolucency with missing left maxillary canine (Figure 4). Thin, fine, straight bony septa intersecting at certain angles were also seen.

Occlusal radiograph showed an area of multilocular radiolucency extending from the left maxillary lateral incisor till the second premolar anteroposteriorly (mediolaterally) and impacted canine could be noted (Figure 4). Based



Figure 3: Orthopantomograph showing lesion in the left maxilla



Figure 4: Intraoral periapical radiograph and occlusal radiographs showing internal structure of the lesion

on all the radiographic findings, differential diagnosis of dentigerous cyst, keratocystic odontogenic tumor, ameloblastoma, odontogenic myxoma was considered.

Further to know the extent of the lesion computed tomography (CT) of the head was advised which showed a multilocular lesion (Figure 5). Expansion and thinning of buccal cortical plate with thin and straight septa were also noted.

Following CT examination, an incisional biopsy was performed which was evaluated histologically. The features were suggestive of odontogenic fibroma, following which partial maxillectomy was done, and the specimen was evaluated histologically again. The biopsy specimen revealed highly cellular connective tissue stroma made-up of mature collagen fibers interspersed with plump fibroblasts (Figure 6). Few odontogenic islands were evident. The feature was again suggestive of odontogenic fibroma.



Figure 5: Computed tomography showing lesion in sagittal and coronal sections

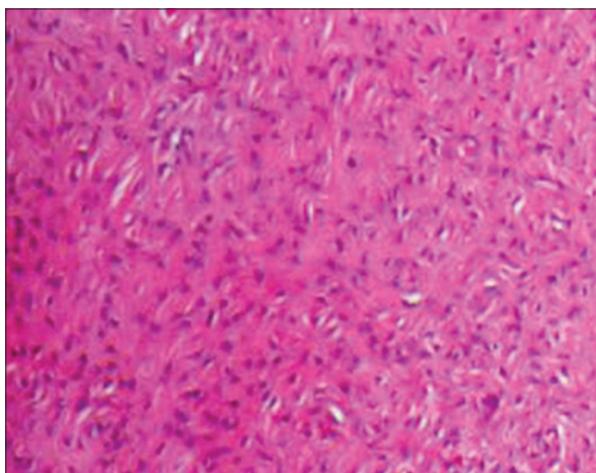


Figure 6: Photo micrographic picture (x40)

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

DISCUSSION

COF is defined as a fibroblastic neoplasm containing varying amounts of apparently inactive odontogenic epithelium.⁷ It can originate from the mesenchymal tissues of dental origin that includes dental follicle, dental papilla or periodontal ligament.^{5,8} COF is a benign odontogenic neoplasm which is not completely understood.⁹ Revised WHO histological typing of odontogenic tumors by Kramer (1992) included this entity under "odontogenic ectomesenchyme with or without included odontogenic epithelium."¹⁰ The WHO panel decided to consider the simple type of odontogenic fibroma under the heading of myxoma. It is the most collagenous variant of the histologic spectrum of odontogenic myxomas, myxofibromas, and odontogenic fibromas.¹¹ It is suggested that the terminology "odontogenic fibroma WHO type" can be renamed as "odontogenic fibroma complex type" or "fibroblastic odontogenic fibroma" which could be considered as a more appropriate name.¹¹

The literature review showed that COF is a very rare odontogenic neoplasm which accounts for only 0-5.5% of all odontogenic tumors.⁷ It is seen in wide age ranges and frequently diagnosed in patients between the second and fourth decades of life and in the current case the patient was in the second decade. Female predilection is observed in many reports, but equal distribution has also been reported between males and females by Kaffe *et al.*; Mosqueda-Taylor *et al.*; Veeravarmal *et al.*^{5,7,8} According to the literature available both the jaws have been affected equally, in the maxillary arch it involves anterior segment as seen in the present case, and mandibular lesions affect the premolar and molar areas. The present case manifested as a slow growing lesion in the anterior region of the maxilla. COF causes bony expansion and displacement of the adjacent teeth.^{10,12}

Radiographically, the lesions are associated with the crown of an unerupted molar, premolar, or incisor tooth. COF usually has well-defined borders, but may also present with scalloped margins as seen in the present case. The internal structure normally appears as a unilocular radiolucency which may exhibit a multilocular appearance as seen in the present case.^{5,7,8} The presence of calcifications in the form of flecks is interpreted as a mixed lesion with a characteristic "ground glass" appearance. The lesion and the surrounding normal bone interface may be well-demarcated with sclerotic borders. The appearance may suggest encapsulation, but the presence of a capsule has

not been reported. In spite of that, many cases appeared to be infiltrative.^{5,8,10-12}

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

CONCLUSION

As the occurrence of COF is very rare, the purpose of this report is to present an additional case of COF as well as to highlight the significance of clinical, radiological, and histological features which helps in differentiation of the lesion from other lesions and to understand the need of long-time post-operative follow-up and to develop a better understanding of COF.

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How to cite this article: Balaji P, Gupta A, Govindraju P, Vasan V, Jambunath U. Central Odontogenic Fibroma of Maxilla: A Rare Case. *Int J Sci Stud* 2015;2(12):189-192.

Source of Support: Nil, **Conflict of Interest:** None declared.