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Case Report

Dentistry Section

Ameloblastic Fibrodentinoma of Anterior Mandible: A Rare Case Report

NIKHAT MUKHTAR GAZGE[1], BALAJI PACHIPULUSU[2], POORNIMA GOVINDRAJU[3], YOGESH PAWAR[4]

ABSTRACT

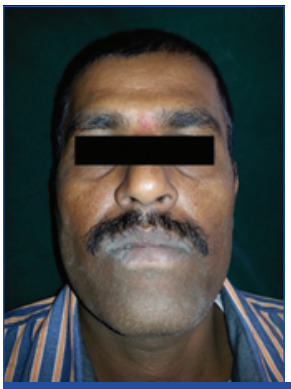
Ameloblastic Fibrodentinoma (AFD) is an extremely rare odontogenic tumour which is a variant of Ameloblastic Fibro-Odontoma (AFO) consisting of both epithelial and connective tissue components of the dental tissue. It was first reported by Field and Ackerman in 1942 and represents less than 1% of all odontogenic tumours in most of the published literature worldwide. It generally occurs during childhood as a slow growing asymptomatic swelling in the posterior mandible. Hereby a unique case of AFD in a 35-year-old male who presented with a gradually growing painless swelling in his anterior mandible region is reported.

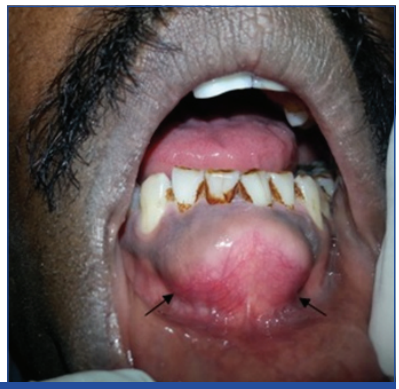
Keywords: Ameloblastic fibro-odontoma, Dentinoid, Mandible, Odontogenic tumours, Odontoma

CASE REPORT

A 35-year-old male reported to the Department of Oral Medicine and Radiology with a chief complaint of swelling in his lower front teeth region since six months [Table/Fig-1]. He noticed a swelling six months back, which was insidious in onset, initially small in size and had gradually progressed to the present size of approximately 2.5×2.5 cm. It was not associated with pain, paresthesia or discharge. There was no history of trauma to the front region of the face. No other associated symptoms were present. His personal history revealed that he was a gutkha chewer since five years (10 times per day). His medical, drug and family history was non-contributory. The patient was moderately built and nourished with all vitals within normal limits.

Extraoral examination revealed no gross facial asymmetry and no regional lymphadenopathy. Intraoral examination of the lesion proper revealed a solitary, well-circumscribed, sessile, dome-shaped swelling present on the labial aspect of the lower anterior region measuring about 2.5×2.5 cm extending mediolaterally from 32 to 42 regions and superoinferiorly from marginal attached gingiva to labial vestibule [Table/Fig-2]. There was no evident swelling on the lingual aspect of the lower anterior region. The overlying mucosa appeared smooth, stretched and pink in colour with no ulceration or visible pulsations. There was no evidence of blood or pus discharge. The surrounding mucosa appeared normal. On palpation, all inspectory findings were confirmed. The swelling was indurated, hard in consistency and non-tender with no yielding points or cortical expansion. The swelling was non-fluctuant, non-reducible and non-compressible. The swelling had well-defined margins and regular edges.

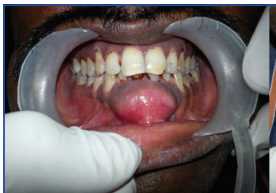


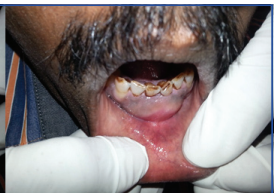


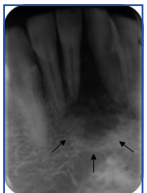
[Table/Fig-1]: Extraoral photograph of the patient. **[Table/Fig-2]:** Dome-shaped swelling present irt the buccal aspect of the lower anterior region.

The hard tissue findings included: anterior deep bite [Table/Fig-3a]; severe attrition irt lower anteriors [Table/Fig-3b]; gingival recession irt 31, 32, 41, 42, 15, 23; periodontal pocket and grade I mobility irt 31, 41. Other findings included missing tooth irt 26; proximal caries irt 45; restoration irt 46, 16, 37, 47. The patient was subjected to electric pulp test which revealed a normal response irt 32, delayed response irt 31, no response irt 41, and normal response irt 42. Based on the history and clinical findings, a provisional diagnosis of radicular cyst irt 41 secondary to trauma due to anterior deep bite was given. Clinical differential diagnosis of ameloblastoma, cemento-ossifying fibroma, ossifying fibroma, osteoma, torus mandibularis, chondromas, mature osteoblastoma and central odontogenic fibroma was considered.

Complete haemogram revealed all values within normal limits. The patient was subjected to radiological investigations. IOPAR revealed a diffuse radiolucency measuring about 1.5×2 cm extending mediolaterally from 42 to 31 region and superoinferiorly from alveolar crest till 0.7 cm below the periapical region of 41. The lesion was surrounded by a thin sclerotic rim and lateral displacement of the roots of 31 and 41 was also seen. The internal structure showed enlarged marrow spaces towards the lower half of the lesion. Interdental bone loss was evident between 31-41, 41-42 [Table/Fig-4]. A mandibular cross-sectional occlusal radiograph revealed no bony expansion or cortical plate thinning [Table/Fig-5]. OPG revealed a diffuse radiolucency at the periapical region of 41 surrounded by a sclerotic rim and missing tooth irt 26 [Table/Fig-6]. A 2D Radiological differential diagnosis of periodontitis/periodontal abscess, central giant cell granuloma, ossifying fibroma was considered.

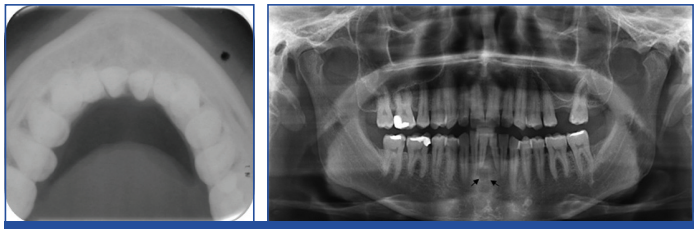






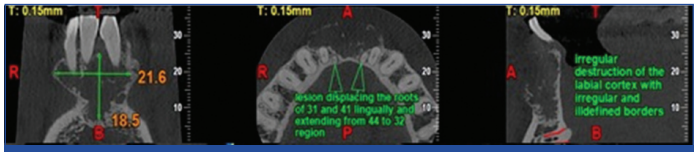
[Table/Fig-3a,b]: Anterior deep bite and severe attrition irt lower anteriors.
[Table/Fig-4]: IOPAR revealing diffuse radiolucency with enlarged marrow spaces surrounded by a sclerotic rim irt 31, 41, 42. Lateral displacement of roots of 31, 41 can be seen.

Due to the inability to view the lesion clearly in two-dimensional radiographs, the patient was advised to undergo CBCT (NewTom, kV: 90, Exposure time: 9.0s, mAs: 54.83, FOV: (5×5) HiRes, Axial thickness: 0.150 mm). CBCT revealed a poorly defined multilocular hypodense lesion with irregular borders, measuring

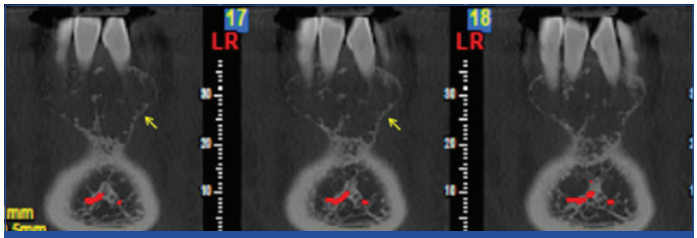


[Table/Fig-5]: Mandibular cross-sectional occlusal radiograph revealing no cortical expansion. **[Table/Fig-6]:** OPG revealed diffuse radiolucency present at the periapical region of 41 surrounded by a sclerotic rim.

about 21×18×12 mm in between 31 and 41. Labial expansion with cortical plate destruction was noted with roots of 31 and 41 laterally and lingually displaced [Table/Fig-7a-c]. In the coronal sections, the inferior border of the lesion showed tiny locules which were uniform and in close proximity towards each other [Table/Fig-8a-c]. Along with this, the presence of few fine curved septations gave a honey comb appearance. The CBCT findings were suggestive of a mildly aggressive lesion possibly secondary to chronic irritation due to anterior deep bite. Hence, ameloblastoma, central giant cell granuloma and odontogenic myxoma were considered as 3D radiological differential diagnosis.



[Table/Fig-7a-c]: CBCT (Coronal, Axial & Sagittal sections) revealing poorly defined hypodense lesion with few fine opaque septations giving a honey comb appearance.

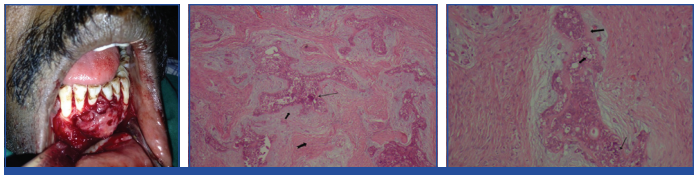


[Table/Fig-8a-c]: CBCT (Coronal sections) revealing small uniform locules in close proximity at the periphery of the lesion giving a honey comb appearance (Line arrow).

FNAC could not be performed as the lesion was hard in consistency with no yielding points. The lesion was surgically excised under local anaesthesia and sent for histopathological assessment [Table/Fig-9]. The H&E stained sections showed strands and islands of odontogenic epithelium in cell rich primitive ectomesenchyme. The epithelial islands were peripherally made up of tall columnar cells with reversal of polarity and hyperchromatic nucleus. The centre of islands showed stellate reticulum-like cells similar to ameloblastic follicles [Table/Fig-10a,b]. The follicles of odontogenic epithelium were seen surrounded by a halo of pale eosinophilic hyalinised dysplastic dentin resembling dentinoid-like material. Areas of basophilic inductive changes showing coarse fibre bundles with elongated cells were seen in the vicinity of the epithelial islands [Table/Fig-10b]. There was no evidence of enamel matrix or enamel formation. Nuclear pleomorphism and mitotic figures could not be found. The presence of characteristic perifollicular dentinoid like material was suggestive of a histopathological diagnosis of ameloblastic fibrodentinoma giving a final diagnosis of AFD. No recurrence was noted within eight months of follow-up.

DISCUSSION

Ameloblastic fibrodentinoma is a rare mixed odontogenic tumour similar to ameloblastic fibroma with inductive changes leading to dentin formation [1-3]. To begin with, it was earlier known as “Dentinoma”, first described in 1936 by Straith and was categorised into mature and immature types depending on its progress. Since 1992, AFD was synonymously recognised



[Table/Fig-9]: Lesion surgically excised under local anaesthesia.
[Table/Fig-10a]: Low power (10X) photomicrograph reveals islands of odontogenic epithelium (Line Arrow) in cell rich primitive ectomesenchyme (Notched Arrow). Areas of inductive changes seen as basophilic areas surrounding the epithelial islands (Block Arrow). **[Table/Fig-10b]:** High power (40X) photomicrograph showing epithelial island with tall columnar cells (Line Arrow) and stellate reticulum like cells (Block Arrow) in the centre surrounded by pale eosinophilic dentinoid like material (Notched Arrow).

as an immature dentinoma [2]. The two main subtypes of AFD are peripheral and central [2]. A rare subtype is the pigmented variety as reported by Takeda Y et al., [4]. Azaz B et al., reported only nine cases of AFD between 1942 and 1967 [5] and only 15 cases had been reported in English language literature upto 2009 [1]. A recent systematic review of AFD cases has shown about 60 cases of central variety and 4 cases of peripheral variety have been reported so far [3,6].

Clinically and pathologically AFD, AFO, and odontoma are almost the same [7] but were listed as distinct entities in the 2005 third edition of WHO classification of odontogenic tumours [8]. However, some authors believe that mixed odontogenic tumours like AF, AFD and odontoma are developmental stages of the same lesion with AF at one extreme, odontomas at the other extreme and AFD in an intermediate stage [7,9,10]. Hence, it was concluded that there was little evidence to justify classifying AFD and AFO as independent entities and the decision was made to group them under odontomas as developing odontomas [7, 11]. AFO and AFD can be distinguished based on the presence of tooth germ elements such as enamel and dentin in combination or only dentin in isolation [1]. This case was considered as a true AFD due to the sole presence of dentin.

AFD commonly affects young males (male to female ratio of 3:1) as a slow-growing painless swelling of the jaws [12]. The average age of occurrence reported by Azaz B et al., was 18 years [5]. There is a greater incidence of posterior location of the lesion with increasing age [12] and predominantly occurs in the mandibular posterior region [1]. Rare sites such as nasal floor and gingiva have also been reported [2,13]. AFD of the posterior mandible was recently reported in a 27-year-old male patient by Shwetha V et al., [14]. Our case was quite unique as in contrast to the literature, our patient was a 35-year-old male with the lesion at an anterior location.

Radiologically, it presents as a well-defined radiolucent lesion with radiopaque flecks surrounded by a thin sclerotic rim. Multilocular lesions are also possible [12]. The lesion is often associated with single or multiple unerupted teeth especially primary incisors or permanent molars [12]. Hence, it is observed as a pericoronal radiolucency with varying degrees of opacity making it difficult to differentiate from a developing odontoma [15]. It should be differentiated from cemento-ossifying fibroma, calcifying epithelial odontogenic tumour, odonto-ameloblastoma, ameloblastic fibro-odontoma or a large odontoma. Interestingly, this case was not associated with an unerupted tooth. A 2D image revealed a radiolucent lesion extending from the alveolar crest to the periapical region surrounded by a sclerotic rim whereas 3D images revealed a honey comb pattern.

Histopathologically, it is comprised of an odontogenic ectomesenchyme that resembles the dental papilla and epithelial strands and nests that resemble the dental lamina and enamel organ with the presence of dentin formation [10]. Few odontogenic epithelial islands or follicles lined by tall cuboidal to columnar cells with central stellate reticulum-like cells similar to ameloblastic follicles are seen. Cystic degeneration in the follicles can also be seen [3]. Dentin deposition is seen in the form of osteodentin,

dentinoid or tubular dentin. It may also be seen entrapping the odontogenic epithelium and ectomesenchymal cells. Some areas of hyalinisation which may or may not be dentin are also evident [1]. It is imperative to note that no enamel or enamel matrix is seen in cases of AFD [3]. This case also showed odontogenic epithelium resembling dental lamina arranged in cell rich primitive ectomesenchyme. Dentinoid is a dysplastic form of dentin which neither contains tubules nor fulfils the criteria for atubular dentin, and which is located in a close anatomical relationship to odontogenic epithelium. It may or may not exhibit cellular inclusions [16]. All the characteristic features of dentinoid material as described by Gardner and Farquhar [16] were present. The pale eosinophilic dentinoid material was identified due to its intimate association to the odontogenic epithelium. No special stains were used. Interestingly, inductive changes were seen as basophilic coarse fibre bundles around the epithelial islands. The presence of ameloblast like cells with dentinoid like material at the epithelial-mesenchymal tissue interface confirmed our diagnosis of AFD.

As the tumour is well-encapsulated, it is treated by enucleation or excision with a very low recurrence rate. It rarely progresses to malignancy as Ameloblastic Fibrodentino Sarcoma [12]. Presence of irregular borders, expansion, and perforation of the cortices should be interpreted with caution and possibility of malignant odontogenic tumour should be suspected. In the malignant counterpart, only the mesenchymal component undergoes a malignant transformation while the epithelial component does not show any chance of cancer [4]. Giraddi G et al., suggested a radical resection especially if the lesion is aggressive showing erosion and perforation of the cortical plate [1]. This case was treated by surgical excision with no recurrence in eight months follow-up period.

CONCLUSION

This is a rare case as AFDs are more common in younger individuals and are associated with impacted teeth. It was diagnostically challenging as there was absence of an unerupted tooth associated with the asymptomatic gradually growing swelling. The clinical findings and vitality test pointed towards radicular cyst. Moreover, there was a radiographic diagnostic dilemma due to variation in the findings of 2D and 3D imaging. This case also highlights the importance of considering rare lesions such as AFD in differential diagnosis.

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PARTICULARS OF CONTRIBUTORS:
1. Editorial Coordinator, Department of Oral Medicine and Maxillofacial Radiology, Indian Dentist Research and Review, Bengaluru, Karnataka, India.
2. Professor and Head, Department of Oral Medicine and Maxillofacial Radiology, Rajarajeswari Dental College and Hospital, Bengaluru, Karnataka, India.
3. Professor, Department of Oral Medicine and Maxillofacial Radiology, Rajarajeswari Dental College and Hospital, Bengaluru, Karnataka, India.
4. Founder and Director, Department of Oral Medicine and Maxillofacial Radiology, Dr. Pawar's Multispeciality Dental Clinic, Satara, Maharashtra, India.

NAME, ADDRESS, E-MAIL ID OF THE CORRESPONDING AUTHOR:
Dr. Nikhat Mukhtar Gazge,
#39, 2nd Cross, 1st Main, Brindavan Nagar, Tavarekere Road, Bengaluru-29, Karnataka, India.
E-mail: dr.nikhatgazge@gmail.com

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