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Case Report

Dentistry Section

Case of Unicystic Ameloblastoma with Features of Ameloblastic Fibro-dentinoma

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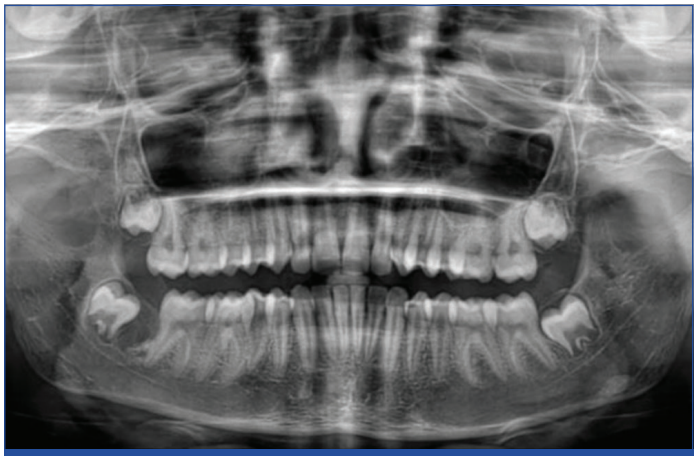
ABSTRACT

Hybrid odontogenic tumours occur rarely in the oral cavity. The basic histogenesis of these lesions is not clear. Hybrid odontogenic tumours characteristically exhibit two or more distinct morphological features that may lead to a diagnostic dilemma. Unicystic Ameloblastomas (UA) are the second most common odontogenic tumour. Ameloblastic Fibro-dentinoma (AFD) is a rare mixed odontogenic tumour. Both UA and AFD commonly occur in the second decade of life, may be asymptomatic with a slight female predilection, and predominantly occur in the mandibular posterior region. Herein, the authors report a rare case of a 15-year-old female patient complaining of swelling in the right lower posterior jaw region in the last two months. The lesion was surgically enucleated and sent for histopathological examination. On histopathological examination, the lesion exhibits areas of both UA and AFD, representing a rare occurrence of a hybrid odontogenic tumour. The present case report highlights the unique combination of two odontogenic tumours of different origins.

Keywords: Dysplatic dentin, Hybrid, Odontogenic tumour, Unilocular radiolucency

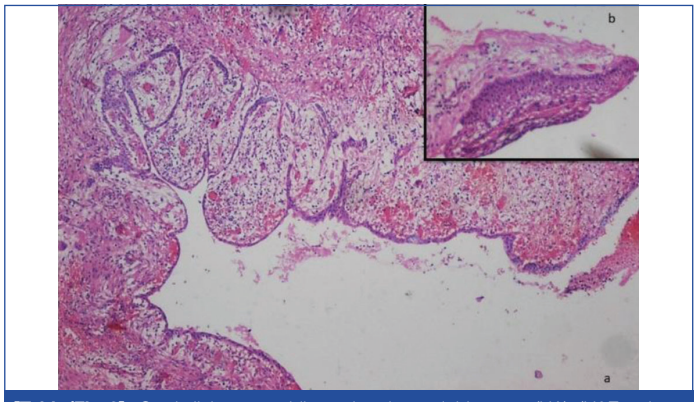
CASE REPORT

A 15-year-old female patient visited a private dental clinic with a chief complaint of pain and swelling over the right posterior lower jaw region in the last two months. The swelling was gradually increasing in size. Past medical and dental history were not significant. On general examination, the patient was normal. Intraoral examination revealed obliteration of the right lower posterior buccal vestibule. The swelling measured approximately 2×1 cm in an antero-posterior direction. No signs of paresthesia or discharge was observed. On palpation, the swelling was bony hard in consistency. An Orthopantomogram (OPG) revealed a unilocular radiolucency in relation to the impacted third molar on the right-side of the mandible. Mixed radio-density was also observed [Table/Fig-1].



[Table/Fig-1]: Orthopantomogram (OPG) showing unilocular radiolucency in-relation to impacted 48. Irregular border and mixed radio density is evident.

The lesion was provisionally diagnosed as an odontogenic cyst of the right posterior mandible, and cystic enucleation was performed with curettage. The impacted tooth 48 was also removed. Tissue was sent for histopathological examination for a confirmatory diagnosis. Macroscopically, small bits of soft tissue specimen showed a thin cystic lining, with a larger bit measuring approximately 1×1×0.5 cm in size and cream to brown in colour. On microscopic examination, the cystic lumen was lined by basal tall columnar to cuboidal cells with hyperchromatic basophilic nuclei placed away from the basement membrane. Superficially, loosely arranged star-shaped cells were observed resembling stellate reticulum. Just below the cystic lining, the connective tissue capsule was delicate with a moderate number of chronic inflammatory cells and areas of haemorrhage. These features were suggestive of UA [Table/Fig-2].

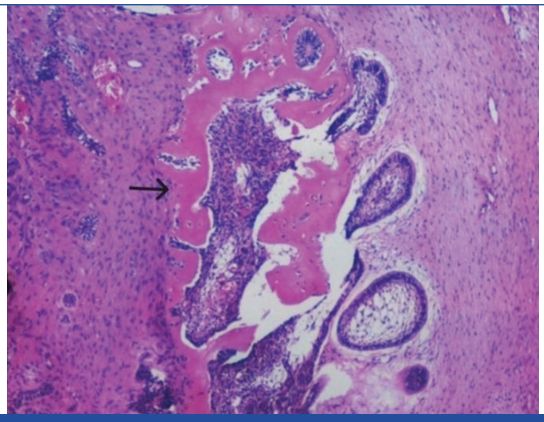


[Table/Fig-2]: Cystic lining resembling unicystic ameloblastoma (UA). (H&E stain, a-40x and b-100x).

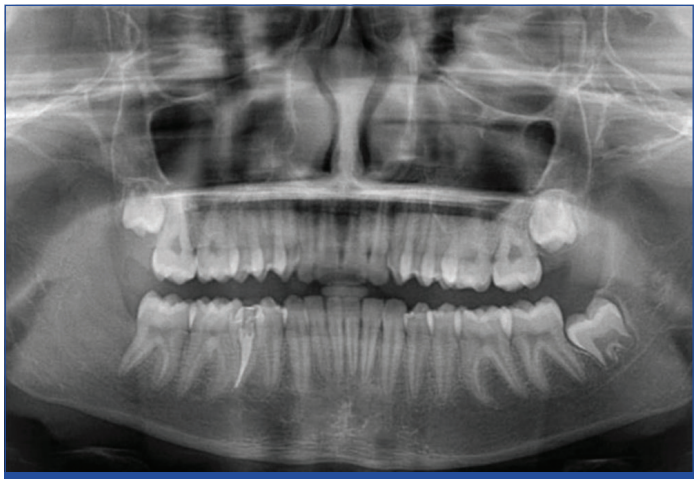
The peripheral connective tissue capsule was quite cellular, resembling primitive ectomesenchyme, i.e., dental papilla. The ectomesenchymal component exhibited a few odontogenic areas arranged in the form of strands, cords, and islands of varying sizes. Odontogenic islands showed peripheral tall columnar cells with hyperchromatic nuclei placed away from the basement membrane and central stellate reticulum-like cells resembling ameloblastic islands. A few ameloblastic islands showed hyalinised areas of dysplastic dentin-like material on the periphery suggestive of AFD [Table/Fig-3]. Histologically, the lesion was diagnosed as a hybrid odontogenic tumour exhibiting the combined features of UA with areas of AFD. After an eight months follow-up period, the lesion completely healed, and no recurrence was noted [Table/Fig-4].

DISCUSSION

Hybrid odontogenic lesions are described as lesions that show characteristic histological features of two or more odontogenic



[Table/Fig-3]: Stroma showed ameloblastic islands with hyalinised areas of dysplastic dentin like material (black arrow) on the periphery. (H&E stain, 40x).



[Table/Fig-4]: Postoperative OPG after eight months showing complete healing of the lesion without radiolucency.

tumours or cysts involving the same primary site [1]. Both UA and AFD commonly exhibit a unilocular radiolucency, associated with an impacted tooth, occur in the first two decades of life with male predominance, and involve the mandibular posterior region [2]. UA may arise: 1) de novo; 2) from the basal cells of the reduced enamel epithelium of an unerupted tooth; 3) from the epithelial lining of a pre-existing odontogenic cyst; 4) by cystic degeneration and fusion of ameloblastic islands of conventional ameloblastoma. Histologically, the epithelial lining shows cuboidal to tall columnar cells with hyperchromatic nuclei placed away from the basement membrane in a palisaded pattern [2,3].

AKA dentinoma or fibroameloblastic dentinoma (AFD) is a neoplasm of odontogenic origin showing both epithelial and ectomesenchymal tumour components. The epithelial component shows primitive ameloblasts or dental lamina-like cells arranged in strands, cords, or in islands. Central stellate reticulum-like cells may or may not be present. These epithelial components have an induction capacity to form dysplastic dentin [3].

The biological nature of AFD is still a subject of controversy between a true neoplasm and a hamartoma. According to Reichart PA and Phillipsen HP and the 2005, World Health Organisation (WHO) classification, AFD was classified as a separate entity under mixed odontogenic tumours [3]. AFD is excluded from the 2017 and 2022 (5th edition) WHO classification with the explanation that once the dental hard tissue is formed, it means the lesion has a tendency to develop into odontomas. So, these lesions can be called developing odontomas [4,5]. According to Sánchez-Romero C et al., no evidence exists that both AFD and Ameloblastic Fibro-Odontoma (AFO) are developing odontomas because each of these tumours has its clinicopathological features. AFD has no potential to produce enamel/enameloid; therefore, it cannot mature into an odontoma [6]. AFD, AFO, and ameloblastic fibromas exhibit Rapidly Accelerated Fibrosarcoma (B-RAF) p.V600E, unlike odontomas. These findings suggest that atleast a few cases of AFDs are neoplastic in nature rather than hamartomas [5,6]. So, in the present case report, the author represents a developing odontoma, in particular as AFD, depending on histopathological features.

This is a unique case of asymptomatic unilocular radiolucency around the crown of the 48 tooth in a 15-year-old female patient. On biopsy, the lesional tissue histologically exhibited the features of both UA and AFD. UA rarely associates with other odontogenic tumours like adenomatoid odontogenic tumour, etc., [7]. Similarly, only a few cases are reported previously which histologically show the features of both ameloblastoma and odontogenic tumour of mixed origin [Table/Fig-5] [8-11]. The present case is one among the rare case reports of a hybrid tumour which exhibits UA-like cystic lining and the capsule with focal areas of AFD.

Hybrid odontogenic tumours are defined as lesions showing combined histopathological characteristics of two or more previously recognised odontogenic tumours and/or cysts of different categories [1]. The histogenesis of hybrid tumours is always considered a topic of controversy. Atarbashi-Moghadam S et al., and Yeh TH et al., discussed that some hybrid odontogenic tumours are two distinct entities arising in the same location, or the hybrid tumours can occur due to the pluripotential nature of the odontogenic epithelium [12,13]. According to Thapa G et al., numerous theories were proposed in the literature regarding the histogenesis of hybrido-odontogenic tumours [14] like a) Collision theory [3]; b) Transformation theory; c) The combination theory; d) The conversion theory and; e) The composition theory. Hybrid odontogenic tumours were not included in the recent 2017 WHO classification, though a good number of cases of hybrid odontogenic tumours were reported in the literature. In the present case, tumour origin can occur by any one of the ways.

S. No.	Reference/Year	Age/Sex	Site	Sign/symptom	Radiographic feature	Microscopic findings	Treatment	Recurrence
1	Chen SH et al., [8]/ 1991	5/M	Mandibular molar, ramus area	Swelling	Multilocular radiolucency	Conventional ameloblastoma and ameloblastic fibroma	Enucleation	No
2	Economopoulou P and Sotiriadou S [9]/1998	26/F	Mandibular premolar-molar area.	Painful swelling	Unilocular radiolucency	Unicystic ameloblastoma and ameloblastic fibroma	Enucleation	No
3	Nafarzadeh SH [10]/ 2011	16/M	Mandibular premolar to second molar apical region	Painless Swelling	Unilocular radiolucency	Unicystic ameloblastoma and ameloblastic fibroma	Excisional biopsy with bone curettage	No
4	Alampally HS [11]/ 2014	28/m	Mandibular left premolar to right canine region.	Painful swelling	Well defined unilocular radiolucency with few tooth like radiopacities.	Unicystic ameloblastoma (UA) and compound odontoma.	Enucleation	Not mentioned
5	Present case, 2024	15/F	Right mandibular third molar area.	Painful Swelling	Unilocular radiolucency	Unicystic ameloblastoma and ameloblastic fibro-dentinoma	Enucleation with curettage.	No

[Table/Fig-5]: Comparison of demographic, clinical, radiographic and microscopic features of all the four cases histologically showing ameloblastoma and mixed odontogenic tumour (ameloblastic fibroma/ameloblastic fibro-odontoma/odontoma) [8-11].

Depending on the size of the lesion, conservative surgical excision or enucleation is the treatment of choice. None of the reported cases showed recurrence similar to the present case.

CONCLUSION(S)

Considering the literature, this is one of the very few reported cases histologically exhibiting the features of both UA and AFD. The histogenesis of these lesions is still an enigma. Enucleation and conservative surgical excision are the treatments of choice for localised hybrid odontogenic tumours. More cases should be reported with long-term follow-up to confirm the clinical significance of these lesions and the recurrence rate.

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