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Case Report

Dentistry Section

Ameloblastoma Arising from A Dentigerous Cyst-A Case Report

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ABSTRACT

We are reporting a case of an ameloblastoma which arose in the wall of a dentigerous cyst. The clinical, radiographic and histological characteristics were similar to those of dentigerous cysts, as were seen on doing an incisional biopsy. Enucleation was done intraorally under local anaesthesia. Post-operative excisional biopsy revealed strands and cords arising from the cystic lining, which are suggestive of ameloblastic changes.

Keywords: Unicystic ameloblastoma, Plexiform ameloblastoma, Enucleation

CASE REPORT

A 14-year-old boy was referred to the Oral and Maxillofacial Surgery Clinic with an asymptomatic swelling on his right cheek [Table/Fig-1], which extended from the lower border of his mandible to the tragus of his ear and antero-posteriorly, from the corner of his mouth to the posterior border of the ramus of the mandible. His past medical history was noncontributory. An intraoral examination revealed a right mandibular, buccal, cortical expansion extending from the anterior border of the ramus to the first molar and extending buccally into the cheek, with normal overlying mucosa [Table/Fig-2]. Egg shell crackling was observed on palpation of the swelling. A panoramic radiograph [Table/Fig-3] revealed a single, unilateral, well defined, radiolucent area in the right mandibular ramus, which extended 1 cm below the sigmoid notch to the lower border of the mandible, superioinferiorly involving the anterior border of the ramus and extending anteriorly along the alveolar crest, upto the distal surface of the first molar. Posteriorly, the lesion was found to extend to the posterior border of the ramus, leaving 1.5 cm of the posterior border of the ramus intact. Crown of the malposed, unerupted mandibular right third molar was displaced to the lower border of the mandible. Under local anaesthesia, an aspiration which was done, revealed a straw-coloured fluid, which suggested that lesion was a dentigerous cyst, which was confirmed by doing an incisional biopsy. Enucleation of the lesion, along with the unerupted third molar, was done under local anaesthesia [Table/Fig-4]. [Table/Fig-5] shows the enucleated cystic cavity and the inferior alveolar neurovascular bundle intact after enucleation [Table/Fig-6].

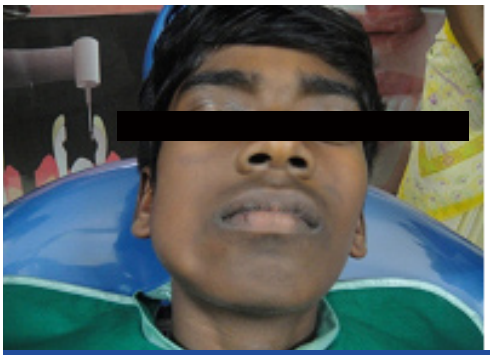
HISTOPATHOLOGICAL FINDINGS

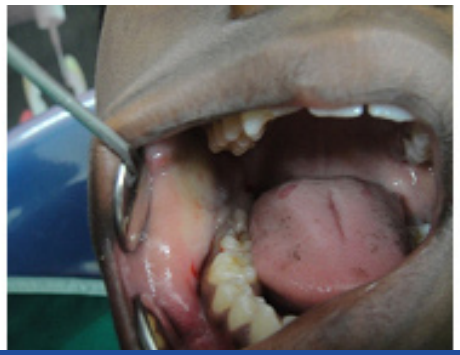
Incisional biopsy was suggestive of the lesion being a dentigerous cyst with 2-3 layers of epithelial lining and inflammatory component in the stroma.

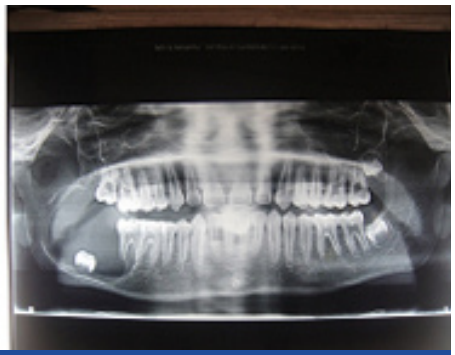
An excisional biopsy of the specimen, which was done, showed areas with 2 to 3 layers of thick epithelium, with some areas showing a proliferative lining also. The underlying stroma was condensed and it had areas which suggested that they had odontogenic epithelium. These were extensions from the intra capsular proliferation of the cyst wall, in the form of anatomizing strands and cords. The histological features suggested that the lesion was a dentigerous cyst with amelobastomatous changes [Table/Fig-7,8]. Due to the pattern of epithelial proliferations, we could suggest that the growth had formed into either an intraluminal/ plexiform unicystic ameloblastoma (a nodular proliferation into lumen without infiltration of tumour cells into connective tissue wall), as was classified by Ackerson 15. No recurrence was noticed clinically and radiographically [Table/Fig-9] during the Post-operative follow-up of six months [Table/Fig-9]. The H & E Stained soft tissue sections, under 40x resolution, showed odontogenic epithelium which was arranged in a bilayered plexiform pattern and each strand was peripherally bonded by the tall columnar cells, with reverse polarization and hyper chromatic nuclei. The columnar cells also demonstrated intra nuclear vacuolization. The connective tissue stroma tended to be loosely arranged and vascular, thus confirming it to be a plexiform ameloblastoma.

DISCUSSION

Dentigerous cysts (DCs) are one of the most common types of cysts which occur in the jaw. A typical DC clinically presents as an asymptomatic unilocular radiolucency which encloses the crown of an unerupted or an impacted tooth. In most of the cases, the diagnosis of a DC is straight forward; but even radiographically, a 'typical' DC can be diagnosed as something else, such as a dental follicle, a hyper plastic dental follicle, an odontogenic keratocyst [a keratocystic odontogenic tumour (KCOT)] or a

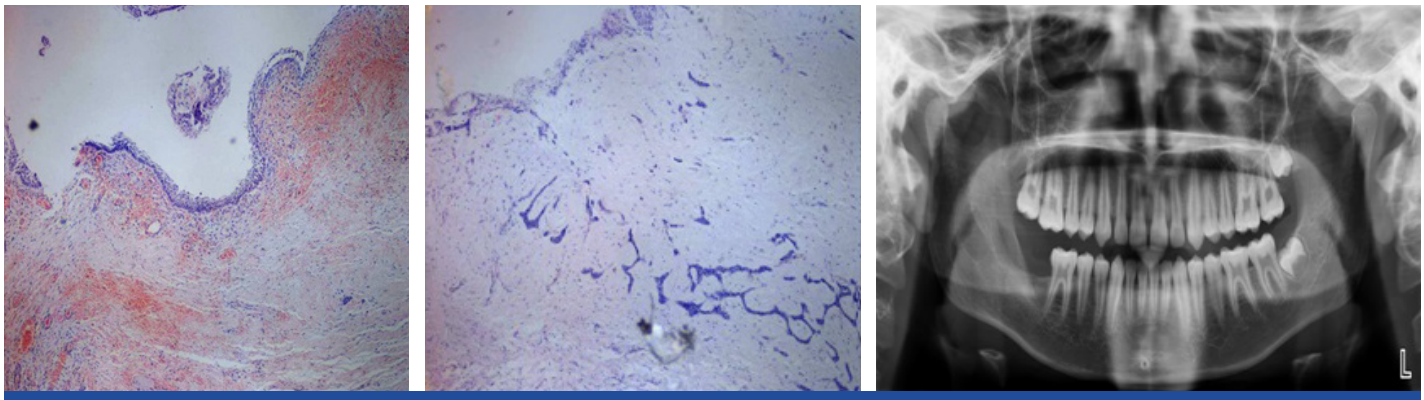






[Table/Fig-1]: Extra oral clinical photograph
[Table/Fig-2]: Intraoral view
[Table/Fig-3]: Radiograph showing the radiolucent lesion

[Table/Fig-4]: Intra operative view
[Table/Fig-5]: Cystic cavity after enucleation
[Table/Fig-6]: Inferior alveolar neurovascular bundle intact after enucleation



[Table/Fig-7]: Histological picture showing features of dentigerous cyst
[Table/Fig-8]: Histological picture showing features of ameloblastoma
[Table/Fig-9]: Six months follow up radiograph

unicystic ameloblastoma on histological analysis [1]. The histological diagnoses of these lesions are therefore critical [2].

A dentigerous cyst is the most common cause of pericoronal radiolucency which is associated with impacted teeth [3]. Because they are asymptomatic, dentigerous cysts are usually diagnosed on routine dental radiographs. The diagnosis of a dentigerous cyst is based on a combination of radiographic and histopathological features [4]. Dentigerous cysts form within the lining of the dental follicles when fluid accumulates within the follicular epithelium and the crown of developing or unerupted tooth [5].

Most of the dentigerous cysts manifest in the second and third decades of life, with peak incidences in teenages, often developing around the crowns of mandibular third molars [6] as it was seen in our case. An ameloblastoma is a benign and a locally aggressive tumour which arises from the mandible or less commonly, from the maxilla. Unicystic ameloblastomas are variants of ameloblastomas, which were first described by Robinson and Martinez, which refer to those cystic lesions that show clinical and radiological characteristics of odontogenic cysts, but which on histological examination, show typical ameloblastomatous epithelium which lines part of the cyst cavity, with or without a luminal or mural tumour proliferation [7]. Prior to this report, this variant had been referred to as a mural or an intra luminal ameloblastoma. In a clinico-pathological study done on 57 cases of unicystic ameloblastomas, Ackerson classified this entity into three histological groups: luminal unicystic ameloblastoma, intraluminal / plexiform unicystic ameloblastoma and mural unicystic ameloblastoma.

Fifteen percent to 20% of all unicystic ameloblastomas form in the wall of dentigerous cysts. Since 1925, many had reported the development of ameloblastomas within the walls of odontogenic cysts, among which the most commonly cited were dentigerous cysts [8]. The immuno-histochemical data on Ki-67 expression in ameloblastomas which arise from dentigerous cysts confirm the hypothesis that ameloblastomas which arise from dentigerous cysts have a biological behaviour which is similar to that of unicystic ameloblastomas [9]. The term, 'plexiform unicystic ameloblastoma' refers to a pattern of epithelial proliferation that has been described in cystic lesions of the jaws. It has been considered as a hyper plastic epithelium, rather than an ameloblastoma by some pathologists, because it does not exhibit histological criteria which were previously accepted for ameloblastomas. Gardner et al., in their article, provided histological evidence that plexiform unicystic ameloblastomas were in fact, variants of conventional unicystic ameloblastomas, by reporting ten cases of unicystic ameloblastomas that exhibited both patterns. Further evidence of the ameloblastomatous nature of plexiform unicystic ameloblastomas is that their biological behaviour, even when this pattern occurs alone, is similar to that of conventional unicystic ameloblastomas [2]. Occurrence of ameloblastomas in children and adolescents who are below 18 years of age is uncommon. Only 14.6% of cases ameloblastomas were seen in children and adolescents among 206 cases which were evaluated by Lucas [10].

In view of the reported ameloblastomatous potential of dentigerous cysts, it is thus important to be able to recognize true ameloblastomatous epithelium from ameloblastoma -like epithelium. In most cases of odontogenic cysts, the presence of an ameloblastomatous epithelial lining in inflamed odontogenic cysts is insufficient to diagnose unicystic ameloblastomas, unless other more diagnostic features of unicystic ameloblastomas are evident [11]. In such cases, other diagnostic criteria which are included to make a diagnosis of unicystic ameloblastomas, as were described by Vickers and Gorlin, are cysts which are lined by an ameloblastic epithelium, with a tall columnar basal layer , a sub nuclear vacuole, reverse polarity of hyper chromatic nucleus and a thin layer of oedematous, degenerating stellate reticulum like cells on surface [12].

CONCLUSION

In the present case, making a diagnosis was possible only because histopathological examination of the enucleated material was

performed. Thus, a histological examination is the most sensitive tool which can be used for differentiation of dentigerous cysts from unicystic ameloblastomas. It highlights the importance of enucleation as the first choice of treatment for large, cystic lesions, instead of conservative procedures like decompression and marsupialization which are used for children. Though marsupialization might help in the preservation of vital structures, keeping in view, the potential of more aggressive transformation of the cystic lining, complete removal of the lining by enucleation, with emphasis on possible preservation of vital structures, as in our case, clubbed with a thorough follow-up, is more appropriate.

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