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A Case Report of Plexiform Unicystic Ameloblastoma

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Abstract:

Unicystic / Cystic ameloblastoma (UCA) is a single cystic cavity which shows ameloblastomatous differentiation in the lining. It is a rare, benign, locally invasive odontogenic neoplasm of young age that shows clinical, radiographic, or gross features of an odontogenic cyst, but it histologically shows typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth. It accounts for 5-15% of all intraosseous ameloblastomas. We report a case of unicystic plexiform ameloblastoma in a 18-year-old female

Key words: Unicystic, ameloblastoma, invasive, plexiform

1. Introduction

Ameloblastoma is the most common neoplasm arising from the primary odontogenic or tooth-forming tissue. Ameloblastomas are benign tumors whose importance lies in their potential to grow to enormous sizes, with resulting bone deformity.¹ Unicystic ameloblastoma (UA) is a variant of ameloblastoma. Robinson and Martinez first described this entity in 1977.² The term unicystic is derived from the macro- and microscopic appearance, the lesion being essentially a well-defined, often large, monocystic cavity with a lining, focally but rarely entirely composed of odontogenic (ameloblastomatous) epithelium.³ The relative frequency of occurrence of UA has been reported to be between 5% and 22% of all types of ameloblastomas. Ameloblastoma has been categorized broadly into three biologic variants: cystic (unicystic), solid, and peripheral.⁴ Various studies report that between 15 to 30% of all ameloblastomas form in the wall of a dentigerous cyst.⁵ Unicystic / Cystic ameloblastoma arises in cystic cavity which shows ameloblastomatous differentiation in the epithelial lining. We report a case of plexiform unicystic ameloblastoma in an 18-year-old asymptomatic female patient.

An 18-year-old female patient, residing in Bhoirwada, Panvel, came to the OPD of YMT dental college with the chief complaint of maligned upper front teeth. The patient's medical and dental history was non-contributory. General examination revealed that she was well oriented with time space and person. On extraoral examination, a slight swelling was noted over the left mandibular angle area. Lymph nodes were non-palpable, and the muscles of mastication and the temporomandibular joints (TMJ) were normal, with no evidence of clicking, crepitation or tenderness.



Figure 1: Profile Photo

Intraoral examination revealed slight buccal expansion and more pronounced lingual cortical expansion in the area of 37. Application of pressure on the lingual aspect of the swelling and on the mucosa distal to 37, elicited tenderness. Based on the clinical examination, she was advised with a routine intra-oral periapical radiograph, occlusal and panoramic radiograph .



Figure 2: Intraoral View

Radiographic examination revealed an oval shaped unilocular radiolucency with well corticated borders surrounding the crown of unerupted 38 extending from mesial of 37 to the anterior border of mandibular ramus in a circumferential manner. Root resorption was noted with mesial and distal roots of 37.

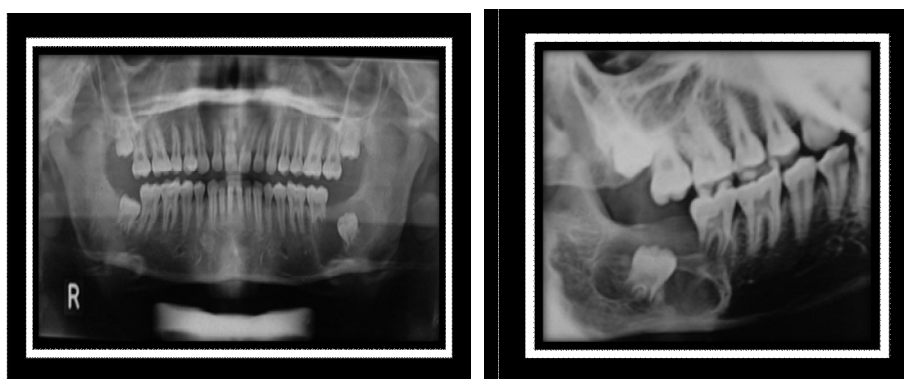


Figure 3: Radiographs Showing Cystic Lesion

Fine needle aspiration cytology revealed a hemorrhagic fluid aspirate which revealed acute inflammatory cells. Based on the above findings, a provisional diagnosis of dentigerous cyst was given. A differential diagnosis of unicystic ameloblastoma was also considered. Incisional biopsy revealed an ameloblastomatous change in the lining of odontogenic cyst. The lesion was treated by enucleation and curettage and submitted for histopathological examination. The histopathology revealed palisading basal cells with hyperchromatic nuclei and reversal of polarity in some areas resembling ameloblasts. Suprabasal layer showed stellate reticulum like appearance. Connective tissue capsule was made up of dense bundles of collagen fibers with moderate infiltration of chronic inflammatory cell infiltrate.

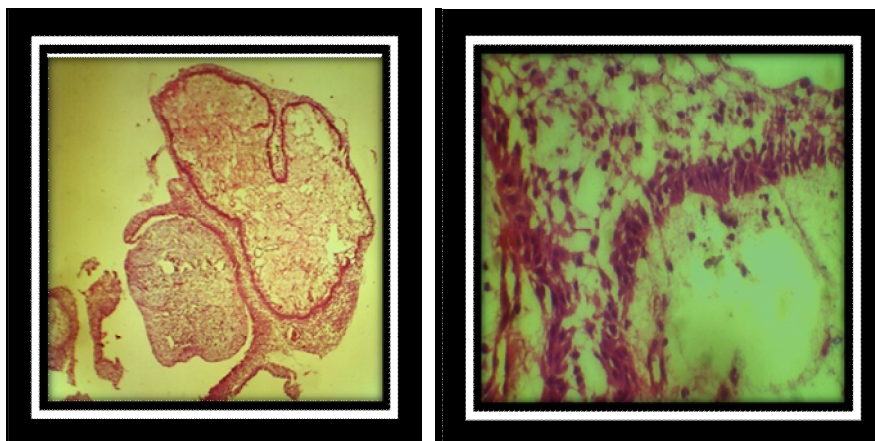


Figure 4: Histopathology

A final diagnosis of unicystic plexiform ameloblastoma was given based on the histopathology. Postsurgical healing was uneventful. A follow up was done after a week with no postoperative complications.

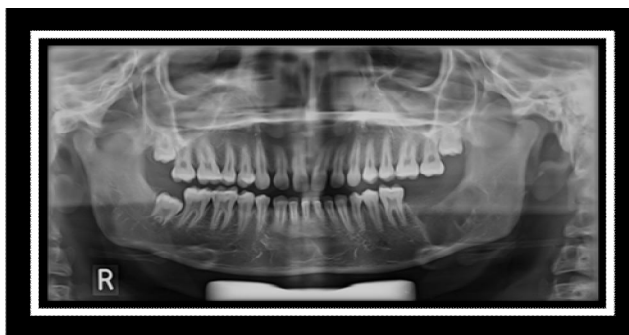


Figure 5: Follow up

2. Discussion

UCA occurs usually in the younger age group of 16-20 years. It has slight male predilection in a ratio of 1.6:1. The mandible is affected more often than the maxilla. These tumors are most commonly encountered in the posterior mandible followed by the parasymphysis region, anterior maxilla, and the posterior maxilla.⁶ Eversole et al. and Paikkatt et al. identified predominant radiographical patterns for UCA: unilocular, scalloped macromultilocular, pericoronal, interradicular, or periapical expansile radiolucencies.⁷

Ackermann et al. classified UA into three types based prognostic and therapeutic implications.

In type 1, the tumor is confined to the luminal surface of the cyst with a lining of ameloblastomatous epithelium. Type 2 is characterized by epithelial nodules arising from the cystic lining and projecting into the cyst lumen. There is no evidence of infiltration of the fibrous cystic wall in either type of lesion. In type 3, the fibrous wall of the cyst is infiltrated by a trabecular pattern that resembles the plexiform pattern seen in conventional ameloblastoma. In addition, it is characterized by a basal layer of columnar cells with hyperchromatic nuclei. These cells are loosely cohesive and resemble stellate reticulum epithelium.^{8,9}

Treatment of UA continues to be controversial. Influencing factors are age, general health, clinico-radiographic variant, anatomic locations and clinical behavior of the lesion. Available treatment options are enucleation, enucleation followed by use of Carnoy's solution, marsupialisation followed by enucleation, marginal resection and aggressive resection.¹⁰ Unicystic ameloblastomas and dentigerous cysts have an identical clinical and radiographic appearance. So in these cases, clinic and radiographic appearance is not reliable for diagnosis¹¹. Hence, we conclude that the surgical protocol must include the postoperative histopathologic examination for all lesions to rule out any ameloblastomatous changes, so that the patient can be followed up properly to take care of any recurrences happening.

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