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Case Report

Unicystic Ameloblastoma of Maxilla: A Rare Case Report

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Abstract

Ameloblastoma is a rare neoplasm of the mandible and maxilla of odontogenic epithelial origin. It has multiple histologic variants. Most common subtype is the multicystic variant of ameloblastoma while its another variant unicystic ameloblastoma (UA) is relatively uncommon which usually occur in younger populations. In this article, we report a case of Unicystic Ameloblastoma in a 22 year old female which was provisionally diagnosed as dentigerous cyst based on clinico - radiographic features and was treated conservatively. Detailed microscopic examination revealed features of Unicystic ameloblastoma which requires long term follow-up to check for recurrence.

Keywords: Unicystic ameloblastoma, Dentigerous cyst.

INTRODUCTION

Ameloblastoma is the most common tumor which originates from the odontogenic epithelium [1], but it is still comparatively rare, comprising about 1% of tumors and cysts arising in the jaws. It may arise from the epithelial lining of a dentigerous cyst, from the remnants of the dental lamina and enamel organ, or from the basal layer of the oral mucosa, sometimes in a multicentric fashion [2-4]. Based on the tumor behavior, prognosis, clinical, radiographic and histopathological examination, Leon Barnes has categorized ameloblastoma into four types: Solid/ multicystic ameloblastoma (SMA), Unicystic ameloblastoma (UA), peripheral ameloblastoma (PA) and Desmoplastic Ameloblastoma (DA) [5, 6]. The term unicystic is derived from the macroscopic 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 [7]. Ameloblastoma occurs in all areas of jaws, but the mandible is the most commonly affected area. Within the mandible, the molar-angle-ramus area is commonly involved and they are occasionally associated with unerupted third molar teeth [8].

CASE REPORT

A 22 year female presented to the Department of Dentistry of our institute with a painless swelling in right maxillary area of size 3X2X1 cm externally for past 8 months. On intraoral examination, mouth opening was adequate, lateral jaw movement was normal and a swelling of size 3X2X1 cm was papable next to the right lateral incisor. The swelling was non tender, firm in consistency, overlying mucosa was unremarkable without any discharge. Radiographically, a well defined, unilocular radiolucent lesion containing an impacted tooth was seen. Any internal septation or sclerotic area was not seen (Figure-1). The provisional diagnosis of dentigerous cyst was made. A surgical anucleation of the cyst was done and specimen was sent to the Department of Pathology.

Grossly, a grey-brown cyst wall like soft tissue piece was received measuring 2.4X1.8X0.6 cm. On microscopic examination, cyst wall comprising of fibrous tissue lined by stratified squamous epithelium was seen. Luminal surface showed a foci of proliferating epithelium projecting into lumen. The cells were cuboidal to columnar exhibiting peripheral palisading. Areas of haemorrhage were also seen. Histological features were consistent with unicystic plexiform ameloblastoma (Figure 2 & 3).

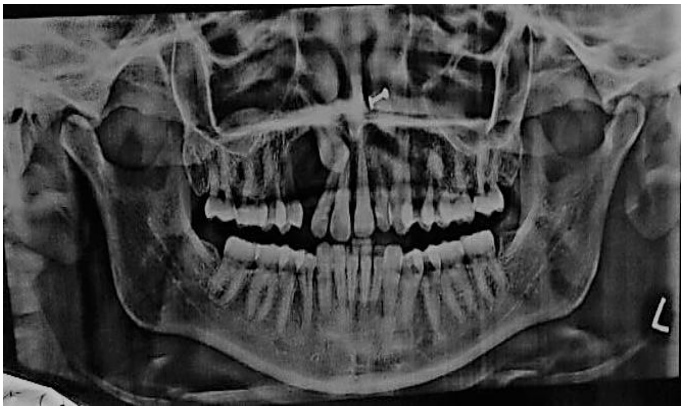

Fig-1: Panoramic radiograph showing impacted permanent maxillary right canine in a well-defined radiolucency, expansion and thinning of the inferior border of the maxilla, displacement of the anterior teeth

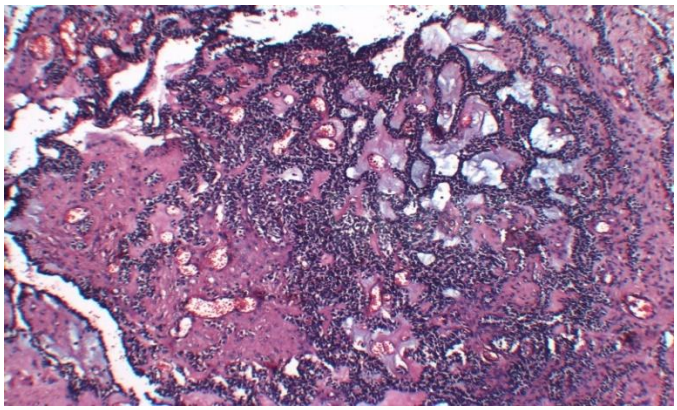

Fig-2: Unicystic plexiform ameloblastoma. Low-power view demonstrating a foci of proliferating epithelium projecting into lumen. (100X, H&E)

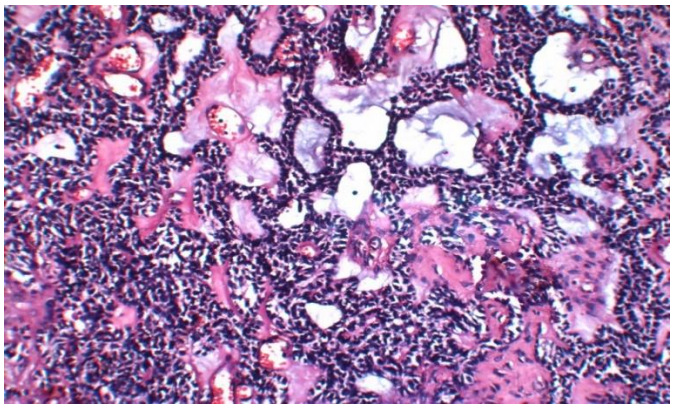

Fig-3: Unicystic plexiform ameloblastoma. Higher magnification showing cuboidal to columnar cells exhibiting peripheral palisading. (400X, H&E).

DISCUSSION

Unicystic ameloblastoma are relatively less common, accounting for about 6% of ameloblastomas. It usually occurs in younger age group, around 16–20 years with 50% of the cases occurring in the second decade of life. It often shows male predilection and commonly involves the posterior mandible and ascending ramus [9]. In our case, the lesion occurred in the maxilla with an impacted right canine tooth in a female patient of 22 years.

Unicystic ameloblastoma is usually asymptomatic, although a large tumor may cause painless swelling of the jaws with facial asymmetry. The clinical and radiographic findings in most cases of unicystic ameloblastoma suggest it to be an odontogenic cyst, particularly dentigerous cyst associated with impacted tooth and most commonly found in relation to mandibular third molar as seen in our present case. However, few cases are not associated with impacted teeth which are considered as non-dentigerous variant [10].

Unicystic ameloblastoma present as either unilocular or multilocular radiolucency radiographically, though there is a clear predominance of unilocular configuration in majority of studies. This predominance was exceptionally marked for the dentigerous variant where the unilocular: multilocular ratio was 4.3:1.2. For the non dentigerous type, this ratio was 1.1:1 [11]. In our case, well defined unilocular radiolucency was noted associated with an impacted tooth which is of dentigerous variant.

The pathogenesis of unicystic ameloblastoma remains obscure and it is not clear whether it develops de novo or from a preexisting cyst. The neoplasm originates within the jaws from epithelium that is involved in odontogenesis. Potential sources of epithelium include enamel organ, odontogenic rests (cell rest of Malassez, cell rest of Serres) reduced enamel epithelium and epithelial lining of odontogenic cyst especially dentigerous cyst [7]. Since the present case is associated with an impacted tooth along with the presence of non-specific thin epithelial lining in focal areas, it seems to arise from preexisting dentigerous cyst. Possibility of misdiagnosing such cases as dentigerous cyst poses a problem where repeat and deeper biopsies are advisable to reveal the underlying tumorous proliferation [12].

The minimum criteria to diagnose a lesion histopathologically as unicystic ameloblastoma is the demonstration of a single cystic sac lined by odontogenic ameloblastomatous epithelium which is seen only in focal areas [13].

CONCLUSION

Ameloblastoma is a locally aggressive tumor of odontogenic origin. Treatment decisions for ameloblastoma are based on the individual patient situation and the best judgment of the surgeon. Owing to the higher prevalence among odontogenic tumors cases of ameloblastoma should be studied carefully, correlating their histologic pattern with biologic behavior to detect subtle changes in histology that may predict aggressive behavior. Prognosis is good if an early diagnosis of the lesion is made with prompt surgical intervention. Long term follow-up is mandatory since recurrence may occur years after removal. Regular postoperative radiographic examination can play an important role in minimizing recurrences.

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