Unicystic Ameloblastoma: An Unusual Case Report

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Abstract

Ameloblastomas have many microscopic and clinical variations. Typically it has three biologic variants which include solid, cystic and peripheral, out of which 14% to 22% of all cases are unicystic. Rare variantslike granular cell subtype composed exclusively of eosinophilic granular cells present inboth peripheral and inner layers of tumoral islands. This is a unique case of cystic ameloblastoma affecting a 16-year-old male with an unusual radiographic and microscopic pattern. This rare variant will create awareness for oral and maxillofacial pathologists for a proper pathway of diagnosis.

Keywords: *Intraluminal, Mural, Plexiform, Ameloblastoma.*

Introduction

Ameloblastoma is a locally aggressive epithelial odontogenic benign neoplasm, which has several different microscopic variants.[1-3] It is divided into three groups solid, peripheral and cystic (unicystic). The most common location is the posterior mandible.[4,5] Unicystic ameloblastomas are seen in the second decade of lifemore often in younger patients. The large lesions may cause a painless swelling of the jaws which is often asymptomatic.[5] Unicystic ameloblastoma is classified into the mural, luminal andintraluminal (plexiform) based on microscopic features. The fibrous wall of the cyst is infiltrated by typical follicular or plexiform ameloblastoma in mural type. The tumor is confined to the luminal surface of the cystin luminal type when one or more nodules of ameloblastoma project from the cystic lining into the lumen is called as the intraluminal type.[4-6] Radical surgery is the treatment of choice for mural ameloblastoma andenucleation is the treatment of luminal and intraluminal ameloblastoma.[4] Granular cytoplasmic changes are seen in the stellate reticulum regions of the ameloblastomatous follicles and interlacing cords and nests and it's a rare variant. In peripheral ameloblastic cells,g ranular changes can also be seen[1,6] this case report aimed to present a rare challenging case of ameloblastomain a 16-year-old male in the posterior aspect of the mandible.

Case Report: A male of 16-year was referred for evaluation of the painless expansion on the left lower jaw region to the Department of Oral and Maxillofacial Pathology of Institute of Dental Sciences On intraoral examination there was a swelling in the posterior area of left mandible extending from 36 to 43 region. On palpation the swelling was, non-tender, bony hard to firm in consistency with eggshell crackling. The radiograph (Figure 1) revealed a well defined cortical unilocular radiolucent lesion with radiopacity extending from 36 to 43 region superiorly till the alveolar ridge and inferiorly to the lower border of mandible and teeth displacement concerning 31, 41,42,43,44,45 region. Given the radiographic feature,it was diagnosed as ameloblastoma. An incisional biopsy was performed under local anesthesia to make a final diagnosis. The routine hematoxylin and eosin (H & E)was done for

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histopathologic study. On microscopic examination it showed anepithelial lining with ameloblast like cells over which there was presence of stellate reticulum like cells proliferating into the underlying connective tissue wall in intraluminal and mural pattern many networks of interconnected strands of ameloblast like cells were also present in the connective tissue wall with areas of cystic degeneration at places (Figure 2, 3) and minimal degree of chronic inflammatory cell infiltration.

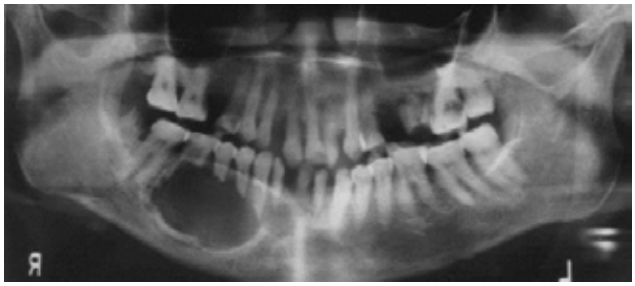

Figure 1. Welldefined corticated unilocular radiolucent lesion with radiopacity

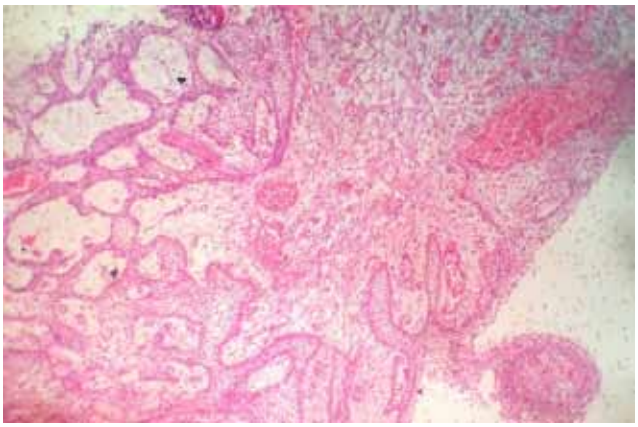

Figure 2. Low power reveals plexiform pattern with mural proliferation

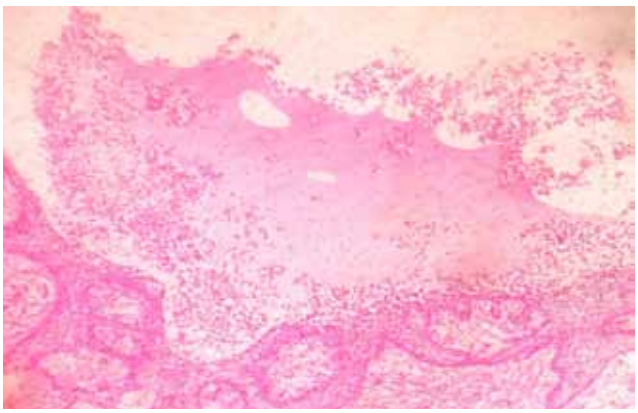

Figure 3. High power reveals plexiform pattern with mural proliferation and luminal variant of ameloblastoma

Differential Diagnosis: Ameloblastoma, Central giant cell granuloma (CGCG) Odontogenickeratocyst (OKC), Calcifying epithelial odontogenic tumor (CEOT).

Discussion

Ameloblastoma is a locally aggressive odontogenic benign, neoplasm which accounts for 1% of all cysts/ tumors of jaws with variable clinical expression and 18% of all odontogenic neoplasms.[6,7] Ameloblastoma is classified as per the WHO system of 2003, based on differences inbiologic behavior, treatment plan and recurrence rate as follows:

(1) Solid/multicysticameloblastoma,

(2) Unicysticameloblastoma,

(3) Peripheralameloblastoma,

(4) Desmoplasticameloblastoma.[7].

6% of ameloblastomas are unicysticameloblastoma which is a rare type of ameloblastoma. It usually occurs in the age group of 15–20 years, with about 50% of the cases occurring in the second decade of life as in our case[8,9]. The male to female ratio of 1.5:1. More than 90% are located in the posterior region of the mandible, the parasymphysis region, the anterior maxilla, and the posterior maxilla[9].

Three pathogenic mechanisms for the evolution of Unicystic Ameloblastoma was proposed by Leider et al.[10].

(1) During the development of the tooth, the epithelium of enamel is reduced which is associated with ameloblastic transformation with subsequent cystic development.

(2) Ameloblastoma arises in the neoplastic ameloblastic epithelium in which there is the temporary non-neoplastic stratified squamous epithelial lining.

(3) When a solid ameloblastoma undergoes cystic degeneration with subsequent fusion of multiple microcysts and develops into unicystic lesions.

Histologic subclassification by Phillipsen and Reichart are described by[7]

Subgroup 1—luminal;

Subgroup 1.2—luminaland intraluminal;

Subgroup 1.2.3—luminal, intraluminal and intramural;

Subgroup1.3—luminalandintramural

Here the present case reveals a swelling in the left lower jaw region of a 16-year-old male radiographically unilocular radiolucency with radiopacity with histopathologic compatibility with intraluminal and mural variant of plexiform pattern.

Conclusion

Unicystic ameloblastoma can be diagnosed based on clinical, radiological, histopathologic, and CT features Recurrence rate was high, especially when the ameloblastic focus penetrates the adjacent tissue from the wall of the cyst. Radiographically, unicystic ameloblastomas show a single large unilocular radiolucency whereas most of ameloblastomas show multilocularity, Unicystic variant of ameloblastoma with aggressive histologic features can be successfully treated with marsupialization with subsequentenucleation, and its an alternative to resection.

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Ethical Approval: Approved

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