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Ameloblastoma: A case report

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Abstract

Ameloblastoma is an odontogenic neoplasm whose significance lies in their impending nature to grow to huge size resulting in bone distortion. Unicystic ameloblastoma (UA) is a variation of the solid or multicystic ameloblastoma. It comprises for 5-22% of all intraosseous ameloblastomas. Biologically it is less aggressive variant which mimics an odontogenic cyst clinically as well as radiographically. Hence the histophathological examination is obligatory to diagnose such cases. This article presents a case which was diagnosed initially as an odontogenic cyst which further on histopathological examination was diagnosed as Unicystic ameloblastoma of mural variety.

Keywords: Ameloblastoma, Odontogenicneoplasm, Unicystic Ameloblastoma, Unicystic, Intraosseous

Introduction

Ameloblastoma is a common odontogenic tumour accounting for around 18% of all odontogenic jaw tumors.(1) In 1827it was first recognized by Cusack and was named as adamantinoma by the French physician Louis-Charles Malassezin in 1885, additionally it was renamed as ameloblastoma in 1930 by Ivey and Churchill.(2) It is defined as unicentric, non-functional, intermittent in growth anatomically benign and clinically persistent by Robinson. The ameloblastoma is divided into three clinicopathological groups. These are solid or multicystic, unicystic and peripheral (extraosseous). Multicystic variety is the most common. It accounts for 86% of cases. Unicystic is the less common variant of ameloblastoma with 6% prevalance.(3) It is commonly seen in young patients and rarely seen over 40 years of age and commonly involves most posterior part of mandible. This paper presents a rare case of unicystic ameloblastoma in an aged patient involving anterior part of mandible.

Case Report

A 75 yr old male reported with the complaint of missing teeth. On examination maxillary and mandibular arches were completely edentulous with the presence of root remnant in 34 region. Clinically a bluish discolouration was noticed over the mandibular incisor region(Fig. 1). On palpation buccal cortical plate appeared to be expanded in the same region. Patient did not give any history of associated pain, difficulty in mouth opening, tenderness with mastication and no pus discharge. Personal history and medical history was non-contributory. A provisional diagnosis of residual cyst was given.

An orthopantamogram (OPG) was done, which revealed a large unilocular radiolucency extending from 34 to 44 region, measuring approximately 6x3cm in dimensions having scalloped and corticated margins and a radiopaque shadow of a root piece remnant in 34 region.(Fig. 2) Occlusal radiograph revealed buccal as well as lingual cortical plate expansion and thinning (Fig. 3). A radiographic diagnosis of keratocystic odontogenic tumour was given.

Enucleation of the lesion was performed and sample sent for histopathological examination (Fig. 4). After histopathological examination it was diagnosed as unicystic ameloblastoma of mural variety (Fig. 5).

Table 1: Ackerman histological classification

Group	Explanation
I	Luminal UA (tumour confined to the luminal surface of cyst)
II	Intraluminal / plexiform UA(nodular proliferation into the lumen without infiltration of tumour cells into the connective tissue wall)
III	Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium)

Table 2: Philipsen and Reichart histological classification

Sub groups	Explanation
1	Luminal UA
1.2	Luminal and intraluminal UA
1.2.3	Luminal, intraluminal and intramural UA
1.3	Luminal and intramural UA

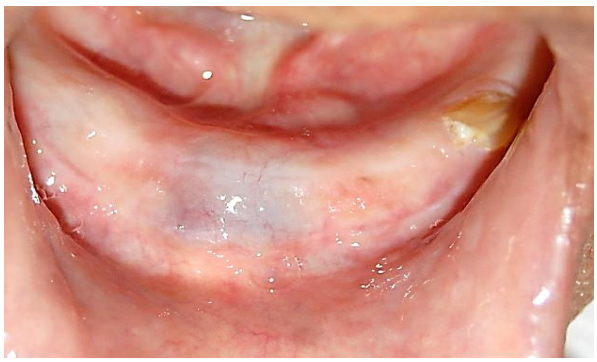

Fig. 1: Bluish discolouration over the mandibular incisor region

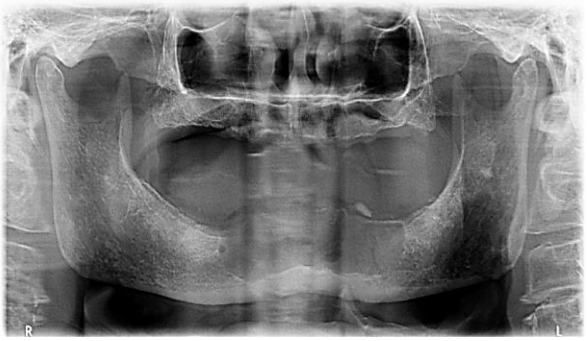

Fig. 2: Panoramic Radiograph

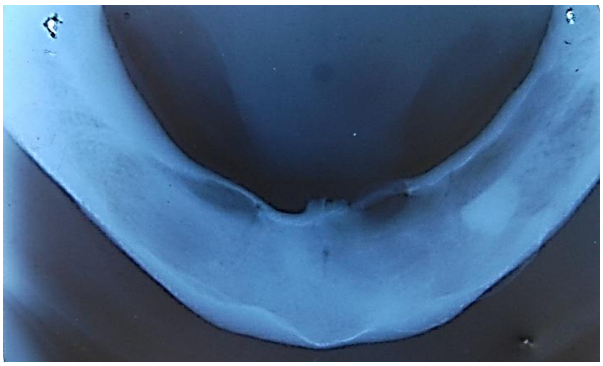

Fig. 3: Occlusal radiograph

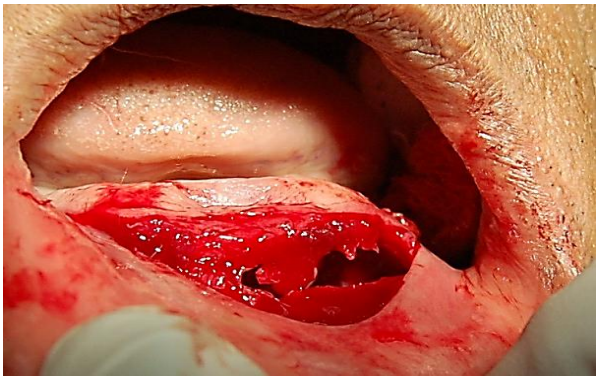

Fig. 4: Surgical Enucleation

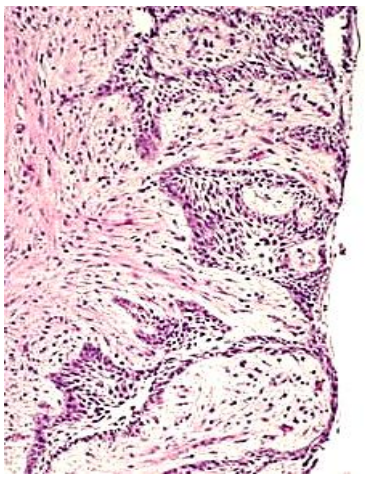

Fig. 5: Histopathological depiction

Discussion

Unicystic ameloblastoma sometimes present diagnostic difficulty when it mimics other clinical entities as in present case. Unicystic ameloblastoma which is less aggressive variant of ameloblastoma was first described by Robinson and Martinenz in 1977. This basically refers to the lesions that appear clinically and radiographically as odontogenic cyst, but reveal in histological examination as a typical ameloblastomatous epithelium lining the cavity with or without luminal and/or mural tumour proliferation.

It is less aggressive variety of ameloblastoma which is commonly seen in younger age group. Most commonly involves third molar region of the mandible and 50% to 80% of the cases are associated with impacted third molars.[4]

Three various pathogenic mechanisms are proposed by Leider et al for the progression of unicystic ameloblastoma. a) The reduced enamel epithelium which is associated with a developing tooth undergo ameloblastic transformation with cystic development. b) Ameloblastomas begin from dentigerous cyst or other odontogenic cysts in which the neoplastic ameloblasticepithlium is created temporarily by a non-neoplastic stratified squamous epithelial lining c) A solid ameloblastoma undergoes cystic degeneration of ameloblastic islands with consequent fusion of multiple microcysts and forms into an unicystic lesion.[5]

Ackerman has given a histological classification[4] (Table 1) Another histological classification is given by Philipsen and Reichart[4] (Table 2).

As far as the treatment of UA is concern, the UA which are diagnosed as subgroups 1,1.2 can be treated by simple enucleation and those which are diagnosed as 1.2.3,1.3 require radical resection.[4] Diagnosis of UA plays a key role in treatment planning as this variety of ameloblastoma has a lesser recurrent rate as compared to characteristic ameloblastoma. According to literature various methods have been studied to differentiate UA from odontogenic cysts like expression of blood cell

carbohydrates, imaging studies like contrast enhanced MRI but proved to be inefficient. The variations in levels of activities of oxidative enzymes, diaphorases, acid phosphatases and naphthol esters have been studied, in ameloblastoma there were uniformly low oxidative, enzymatic activities in the epithelium and widespread, activity of alkaline phosphatase in the stroma. Calretinin is a calcium binding protein found in normal human tissues and tumours like ameloblastoma. Studies have shown that calretinin is expressed only by UA indicating that it may be a specific marker for ameloblastoma.[6] A definitive diagnosis of UA can be made only after examining the whole specimen. Hence, incision biopsy may not always be correct as the epithelium shows variation. Thus, multiple sections from the whole specimen should be examined for final diagnosis.

The overall recurrence rate of UA is reported less than 25%.[6] Hence it requires a regular follow up. In the present case, according to protocol we kept the patient under systematic follow up for a period of one year during which there was no complication or any sign of recurrence.

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