Case Report

Ameloblastomatous Calcifying Odontogenic Cyst in the Mandible – A case report of a rare histological variant

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Abstract

Calcifying odontogenic cyst (COC) is an uncommon benign cystic neoplasm of odontogenic origin, which shows extensive diversity in its clinico-pathological appearances and biological behavior. Ameloblastoma is one of the well-known frequently occurring odontogenic tumours in head and neck region. It can be associated with calcifying odontogenic cysts (COCs), but only a few case reports have been reported in the literature. Here in this paper we report a rare case of calcifying odontogenic cyst with ameloblastomatous proliferation. This is a rare histological variant occurred in mandible of a 26 years old male.

Keywords: Calcifying odontogenic syst, Ameloblastomatous, Neoplasm

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Introduction

Calcifying odontogenic cyst (COC) is a well circumscribed, solid or cystic lesion derived from odontogenic epithelium which develops from reduced enamel epithelium or remnants of odontogenic epithelium in the follicle, gingival tissue or bone but contains ghost cells and spherical calcifications. It was first described by Gorlin and colleagues,[1] are now included in the group of odontogenic tumours. World Health Organization (WHO) international classification proposed in 1992, under the sections of tumors of odontogenic epithelium with odontogenic ectomesenchyme with or without dental hard tissue formation.[2] The COC represents a heterogeneous group of lesions that exhibit a variety of clinicopathologic and behavioral features. COC has been categorized under two basic groups namely, cystic and

neoplastic.[3] It has no sex predilection with equal distribution in maxilla and mandible and may occur at any age with peak incidence in second decade of life. Radiographically they appear unilocular radiolucency with well defined margins and diffused calcifications. They may also show ameloblastomatous proliferative activity intraluminally or intramurally (ameloblastomatous COC). Neoplastic variants of COC, which showed a solid growth pattern consisting of ameloblastoma-like strands and islands of odontogenic epithelium infiltrating into mature fibrous connective tissue is described in this case report.[4]

Case Report

A 26 years old male patient was referred to the department of Oral and Maxillofacial Surgery with the chief complaint of an insidious onset of slow growing swelling in the right side of mandible in premolar-molar region since 3 months. It was associated with mild pain.

On extraoral examination, a diffuse swelling with obvious asymmetry of face in the lower third area was noted involving the ramus, body of the mandible extending posteriorly till the condylar area. On intraoral examination, an ill-defined oval shaped swelling measuring about 6 cms x 7 cms with smooth surface was noticed. On bidigital palpation buccal and lingual cortical plate expansion was observed extending from distal of 46 till the anterior border of ramus. The overlying mucosa was normal with no sinus or scar formation. The swelling was non- tender and non–fluctuant in nature with no associated mobility of teeth. The patient did not reveal any relevant medical history.

The Orthopantomogram findings revealed unilocular radiolucency on the right side of mandible involving ramus, body of mandible, sigmoid notch, coronoid process and the neck of the condyle with smooth radiopaque borders. Impacted third molar was also noted associated with the radiolucency.

The history, clinical and radiographic features indicated a provisional diagnosis of unicystic ameloblastoma and segmental resection of right mandible with disarticulation of condyle was performed and the resected specimen was sent to the department of oral pathology for histopathological examination which showed expansion of the cortical plates with perforation of the cortex, and while opening a large cystic cavity was noted with numerous solid areas. (Figure 1)

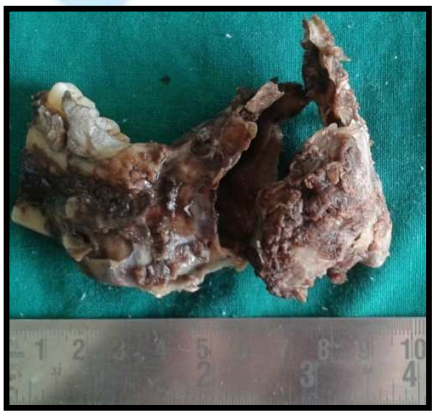

Figure 1: Resected gross specimen of the mandible showing expansion of the cortical plates with perforation of the cortex and a large cystic cavity with solid areas.

The tissue was routinely processed and thorough microscopical evaluation was done. The histopathological examination revealed cystic cavity lined by odontogenic epithelial lining with connective tissue capsule (Figure 2).

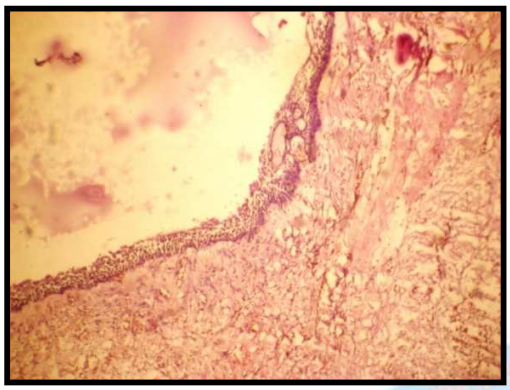

Figure 2: Showing cystic cavity lined by odontogenic epithelial lining with connective tissue capsule.(H and E, 4x)

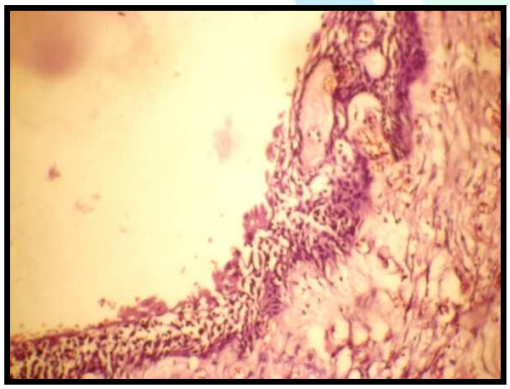

Figure 3: showing epithelium 3 to 5 layers thick with tall columnar basal, superficial stellate reticulum like cells and also showing numerous masses of eosinophilic ghost cells. (H and E; 10x)

The epithelium was 3 to 5 layers thick and showed tall columnar basal cells with hyperchromatic nucleus arranged in palisaded manner. (Figure 3). The superficial cells have much cell layer thickness and resembled stellate reticulum like cells and also showed numerous masses of eosinophilic ghost cells (Figure 4).

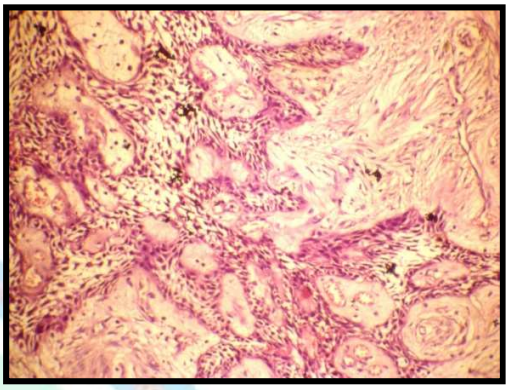

Figure 4: showing odontogenic epithelium showed areas of extensive ameloblastomatous proliferation projecting in to the lumen exhibiting plexiform pattern. (H and E , 10x)

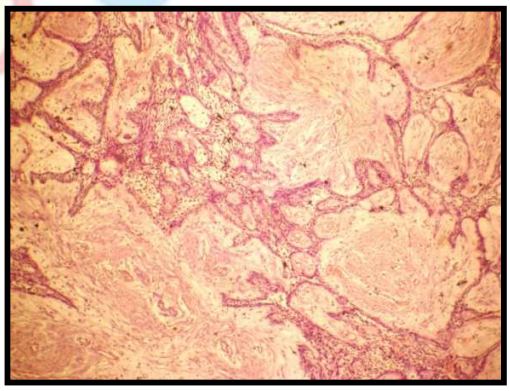

Figure 5: Showing intraluminal and intramural ameloblastomatous proliferation from the lining epithelium. (H and E, 10x)

The odontogenic epithelium showed areas of extensive ameloblastomatous proliferation projecting in to the lumen exhibiting plexiform pattern (Figure 4) were also observed along with few independent follicles in the stroma (Figure 5). Significant inductive changes were also noted surrounding the follicles.

Based on the above findings and correlation with the clinical features a histopathological diagnosis of ameloblastomatous calcifying odontogenic cyst was made. The postoperative course was uneventful. Follow up of the case was done and 8 months have passed since the surgery with no evidence of recurrence.

Discussion

Jaws are the most common site for the occurrence for the epithelial-lined cysts derived from remnants of the odontogenic apparatus. Odontogenic cysts can as either be of developmental or inflammatory origin. The calcifying odontogenic cyst (COC) is a rare example of a developmental odontogenic cyst, its occurrence constituting about 0.37% to 2.1% of all odontogenic tumors.[5,6]

The calcifying odontogenic cyst (CGCOC) was first described by Gorlin et al. who were impressed by the significant presence of the so-called ghost cells.[1] World Health Organization's publication Histological Typing of Odontogenic Tumors,[7] in 2005 has classified all COCs under the category of odontogenic tumours.[8]

As COC has a diverse histopathology, and there has always been confusion regarding its behavior as whether it is a cyst, neoplasm, or hamartoma. Although, there is a wide acceptance of it as a cyst, some investigators consider it to be a neoplasm. [9] Due to this dualistic concept, some subtypes of this cystic variant included the odontoma-associated type of COC and the rare ameloblastomatous proliferating type. On the other hand, it has been recognized that the lining epithelium of the odontogenic cysts can give rise to ameloblastoma. COCs may exhibit extensive intramural ameloblastic-like proliferation which makes the diagnosis to be challenging, due to the overlap of histological features.[10]

Ameloblastomatous COC, which is considered to be a rare histologic variety, microscopically resembles unicystic ameloblastoma except for the ghost cells and calcifications within the proliferative epithelium. It occurs only intraosseously and this variety of COC is different from that of a true ameloblastoma that arises in COC. In contrast to ameloblastoma ex COC, the ghost cells and dystrophic calcifications are within the proliferative epithelium, which lacks histopathologic criteria as suggested by Vickers and Gorlin, and is confined to the cyst lumen.[9]

According to the review of literature done so far on previously published articles around 30 cases of ameloblastomatous COC were reported. It was found that the occurrence of this hybrid tumor was noticed with a wide age group ranging from 11 yrs to 59yrs (mean=29yrs), with a slight female predilection. The most common sight

Table 1: Previously published cases of Ameloblastomatous Calcifying odontogenic cyst:

Reference	Year	No of cases	Mean age (in y)	Gender	Location	Follow up and prognosis
Hong et al[17]	1991	11	NS	NS	NS	NS
Yoshida[11]	2001	07	11-38	3-M 4- F	painless swelling i.r.t Maxilla-6, Mandible-1	no EOR in 6 cases, after 6 y of follow up
Aithal D et al[18]	2003	1	28	F	painless swelling i.r.t mandible	no EOR after 2 y of follow up
Lida et al[15]	2004	1	M	M	painless swelling i.r.t Mandibular body	no EOR after y of follow 13 y of follow up
Eshghyar[19]	2006	3	NS	NS	NS	NS
Kamboj et al[4]	2007	1	58	F	painless swelling i.r.t mandibular ramus	no EOR after follow up
Masshadi et al[20]	2008	1	13	M	painless swelling i.r.t mandibular body	no EOR after 15m of follow up
Kamran N et al[9]	2009	1	22	M	painless swelling i.r.t mandible	no EOR after 14m of follow up
Gupta N et al [21]	2011	1	65	M	painless swelling i.r.t mandible angle	no EOR after follow up
Yuvanati et al[23]	2012	1	63	F	painless swelling i.r.t mandible	no EOR after 10m of follow up
Yadavalli et al[3]	2-12	1	19	F	painless swelling i.r.t maxilla	no EOR after 1 year of follow up
Harkanaw al[5]	2013	1	24	F	painless swelling i.r.t buccal vestibule	no EOR after 2 y of follow up
pesent case	2014	1	26	M	painless swelling i.r.t mandibular body	no EOR after 5m of follow up

NS: Not stated, m-months, y-years, EOR- evidence of recurrence, irt- in relation to

of occurrence of tumor was on the mandibular body with a few cases reported in maxilla. (Yoshida et al).[11] There was no evidence of recurrence in most of the cases except few. (6 cases in the study done by Yoshida[11] recurrence was noticed)

Many authors (Buchner, pretorious)[12,13] have suggested that if COC is associated with an ameloblastoma, its behaviour and prognosis will be that of an ameloblastoma, not of a COC. Even the behaviour and prognosis of this hybrid tumor is more or less almost similar to ameloblastoma, and not like COC, so undoubtedly it should be treated accordingly. The present case did not show any evidence of recurrence after the treatment, but it is no doubt that careful post-operative observations are necessary for COCs which are associated with an ameloblastoma.[14] So based upon the above mentioned features the best treatment option of this cystic lesion includes enucleation with long term follow up. The recurrence of the lesion depends on the completeness of cyst removal. The prognosis is good for cystic COC and less certain for neoplastic COC.[15]

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