

CASE REPORT

Unicystic ameloblastoma of the mandible

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ABSTRACT

Unicystic ameloblastoma refers to those cystic lesions that show clinical, radiographic or gross features of a jaw cyst but on histologic examination show a typical ameloblastomatous epithelium lining the cyst cavity, with or without luminal and/or mural tumor proliferation unicystic ameloblastoma is a less encountered variant of the ameloblastoma and believed to be less aggressive. As this tumor shows considerable similarities with dentigerous cysts, both clinically and radiographically the biologic behaviour of this tumor group was reviewed. Moreover, recurrence of unicystic ameloblastoma may be long delayed and a long-term post-operative follow up is essential for proper management of these patients. Here we are presenting a case of unicystic ameloblastoma in a 18 year old female patient.

Key words: Luminal proliferation, stellate reticulum, unicystic ameloblastoma

INTRODUCTION

Ameloblastomas are benign tumors whose importance lies in its potential to grow into enormous size with resulting bone deformity. They are typically classified as unicystic, multicystic, peripheral and malignant subtypes.^[1] The unicystic ameloblastoma is a less encountered variant of the ameloblastoma, referring to those cystic lesions that show clinical and radiographic characteristics of an odontogenic cyst but in histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor proliferation.^[2] This paper illustrates a case of unicystic ameloblastoma of the mandible in an 18-year old female.

CASE REPORT

An 18-year-old female reported to the Department of Oral Pathology and Microbiology Subharti Dental College, Meerut, with a complaint of swelling over the right side of face for past one month with associated pain while swallowing. On extraoral examination a diffuse swelling in the right lower posterior region of face was seen [Figure 1]. Intraoral examination revealed very slight diffuse swelling in the posterior region of right mandible showing no facial asymmetry.

CT scan revealed a large unilocular radiolucency associated with an impacted lower right second molar and showing thinning out and partial destruction of the lower border of mandible [Figures 2-4].

A clinical diagnosis of Ameloblastoma/Dentigerous cyst/Odontogenic keratocyst was made. Incisional biopsy was advised and specimen submitted for histopathological examination. Microscopically hematoxylin and eosin stained section showed thin cyst lining comprising of few cells in thickness and uniform in appearance with fibrocellular connective tissue capsule. Based on the above features, a diagnosis of dentigerous cyst was made [Figure 5]. Repeat incisional biopsy was undertaken and sections from this revealed characteristic follicles within the stroma and was diagnosed as follicular ameloblastoma [Figure 6a and b].

Treatment

Right hemimandibulectomy and microvascular reconstruction with free fibular flap was done. Peroneal artery anastomosed with right facial artery, peroneal vein anastomosed with right external jugular vein.

Microscopically hematoxylin and eosin stained sections revealed the presence of odontogenic epithelium lining cyst lumen of variable thickness ranging from few layers to multiple cell layers [Figure 7] showing typical cytomorphic features of ameloblastoma given by Vickers and Gorlin, including as basal cell layer composed of columnar cells displaying hyperchromatism, palisaded nuclei, with reverse polarity and subnuclear vacuole. A thin overlying layer of stellate reticulum like cells was also evident [Figure 8]. Parts

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of the lining showed luminal proliferation whereas some places showed proliferation into the capsule resembling ameloblastoma and the diagnosis of unicyclic ameloblastoma was made [Figure 9].

DISCUSSION

Unicyclic ameloblastoma, a variant of ameloblastoma was first described by Robinson and Martinez.^[3] A review of



Figure 1: Diffuse swelling in the right lower posterior region

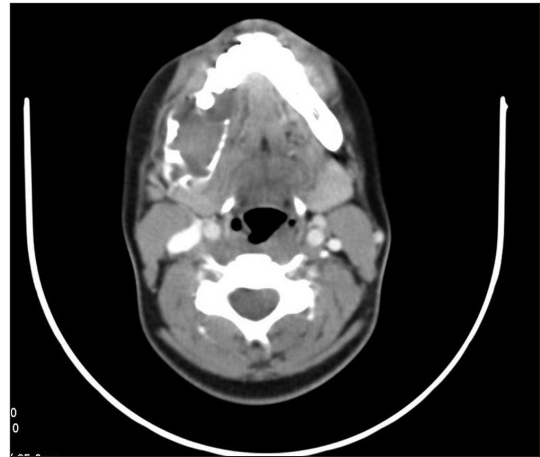


Figure 2: CT scan (Axial view) Body and ramus of mandible just above the level of occlusal plane

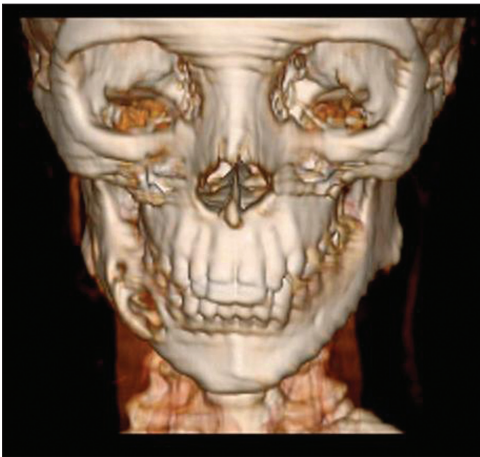


Figure 3: 3D reformatted CT image of lingual view showing unilocular radiolucency with an impacted molar in right mandibular region and thinning of lower border of mandible



Figure 4: 3D reformatted CT image of lingual view showing unilocular radiolucency with an impacted molar in right mandibular region and thinning of lower border of mandible

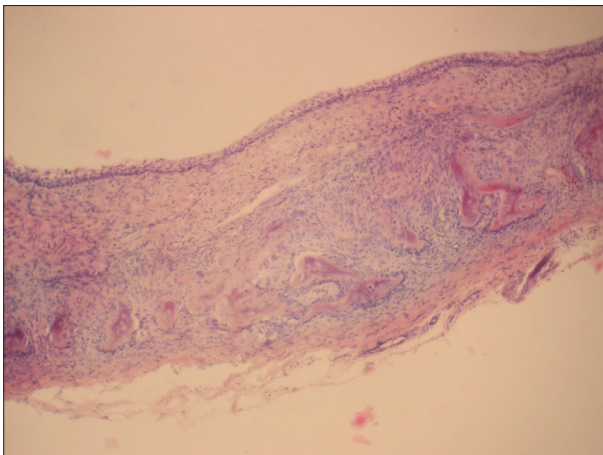


Figure 5: Thin cystic lining and fibrocellular capsule with numerous bony spicules (H and E, 4×)

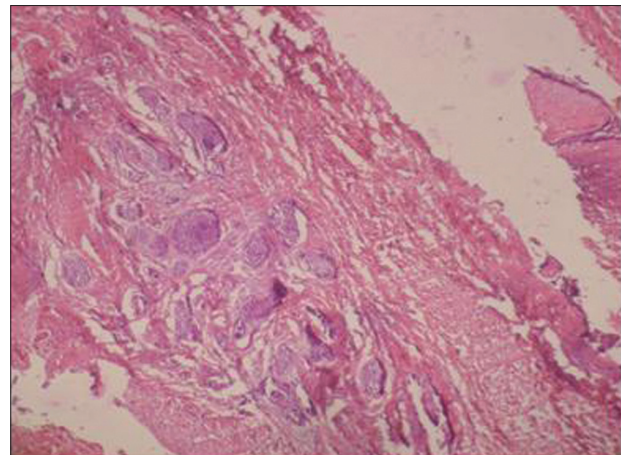


Figure 6a: Section showing ameloblastic follicles within the stroma (H and E, 4×)

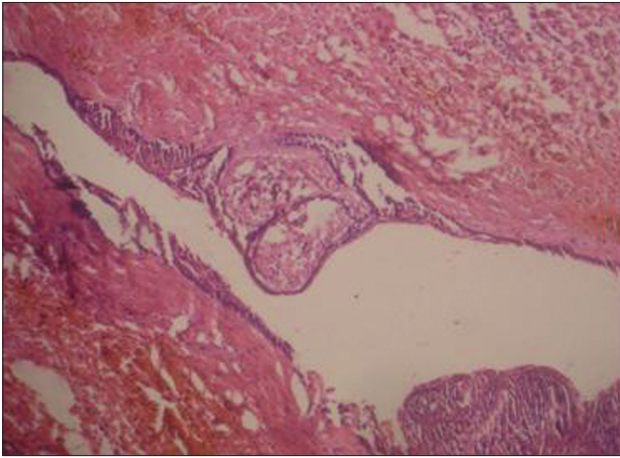


Figure 6b: Section showing ameloblastic follicles within the stroma (H and E, 10×)

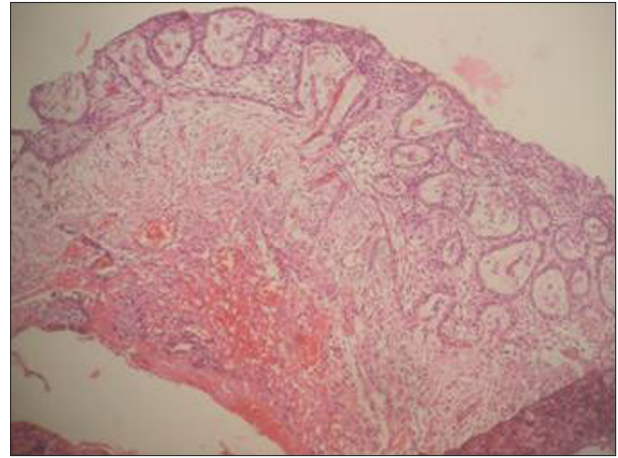


Figure 7: Section showing cystic lining proliferation within the surrounding capsule (H and E, 4×)

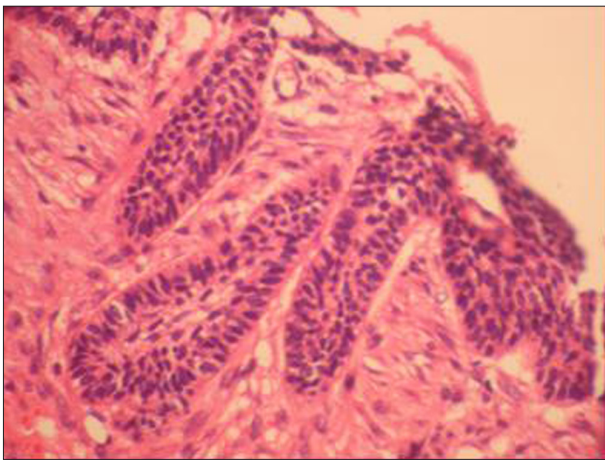


Figure 8: Basal cell layer composed of columnar cells displaying hyperchromatism, palisaded nuclei, with reverse polarity and thin overlying layer of stellate reticulum like cells was also seen (Vicker's & Gorlin criteria) (H and E, 10×)

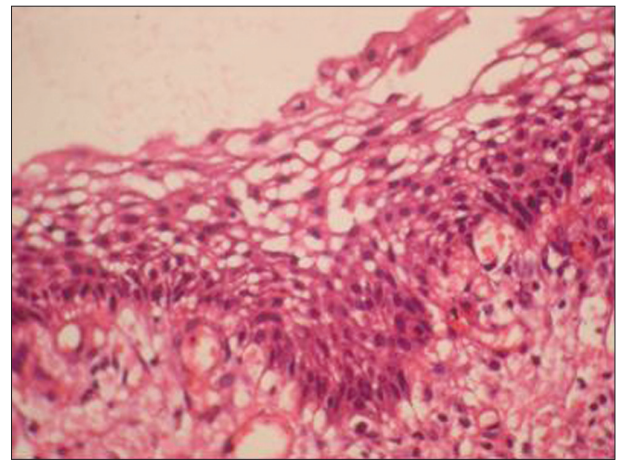


Figure 9: Section showing luminal proliferation (H and E, 10×)

English literature from case reports and minor reviews since 1977 disclosed total number of 152 cases of unicyclic ameloblastoma, with relevant information regarding age and gender. The age of the patients was 25.5 years with 46% of cases occurring in the second decade and they are most often seen in male patients.^[4] More than 90% are found in the mandible, usually in the posterior region. The lesion is often asymptomatic, although a large lesion may cause painless swelling of the jaws.^[5] Patient age and tumor location in our case was in general agreement with these reports. According to Reichart relative frequency has been reported as between 5% and 22% of all subtypes of ameloblastomas. No data is available concerning prevalence and incidence of unicyclic ameloblastoma. There have been many debates regarding whether unicyclic ameloblastoma develops de novo or arises in an existing cyst.^[6] Leider *et al*,^[7] proposed three pathogenic mechanisms for the evolution of Unicyclic ameloblastoma: reduced enamel epithelium, from dentigerous cyst and due to cystic degeneration of solid ameloblastoma.^[7] It often

involves an impacted tooth and the focal area of the cystic tumor lining is often composed of a nonspecific, thin epithelium that mimics the dentigerous cyst lining.^[2] In the present case the association with an impacted tooth and presence of non specific thin epithelium lining in focal areas of cystic tumor supporting the second hypothesis i.e. arising 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 as seen in the present case. Based on the character and extent of tumor cell proliferation within the cyst wall, several histologic subtypes of unicyclic ameloblastoma are recognized, which include simple cystic type, intraluminal and mural type. Gardner in 1984 has pointed out that there is a difference in biological behaviour between these lesions.^[8] Features of both luminal as well as mural type of ameloblastoma were evident in our case. Till now no recurrence has been seen and patient is still regularly being followed up.

Perhaps the most important consideration regarding unicyclic ameloblastoma is that of biologic behaviour. It has been widely stated that these lesions are less aggressive

than their solid or multicystic counterparts and should be treated by enucleation or curettage. However, Gardner has pointed out that there is a difference in biological behaviour between those lesions that are simply cystic or show intraluminal proliferation and those in which the epithelium penetrated and breaches the fibrous wall, therefore having the capacity to invade the cancellous bone.

It has been suggested that recurrence following conservative surgery is more likely to occur in the third group therefore these lesions should be treated in the same manner as solid ameloblastomas.^[9] Whatever surgical approach the surgeon decides to take, long-term follow-up is mandatory, as recurrence of unicyclic ameloblastoma may be long delayed.^[10]

CONCLUSION

The diagnosis of unicyclic ameloblastoma was based on clinical, histopathologic and CT features. Unicyclic ameloblastoma is a tumor with a strong propensity for recurrence, especially when the ameloblastic focus penetrates the adjacent tissue from the wall of the cyst. The ability to predict this potential occurrence prior to surgery would greatly enhance therapeutic strategies for reducing the incidence.

Hence, the Pathologist should examine the tissue sections carefully in an attempt to determine whether ameloblastoma has penetrated the wall of the cyst or not so that the complications can be minimized.

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