Case Report

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UNICYSTIC AMELOBLASTOMA: LUMINAL AND INTRALUMINAL TYPE

Pranav Parashar, * Arti Chauhan, ** Urvashi Tomar, ***
Dheeraj Sharma †

* Senior Lecturer, Department of Oral & Maxillofacial Surgery, Index Institute of Dental Sciences, Indore, Madhya Pradesh, India
** Senior Lecturer, Department of Oral & Maxillofacial Surgery, KD Dental College & Hospital, Mathura, Uttar Pradesh, India
*** Senior Lecturer, Department of Oral Pathology, Index Institute of Dental Sciences, Indore, Madhya Pradesh, India
† Senior Lecturer, Department of Oral Pathology, Index Institute of Dental Sciences, Indore, Madhya Pradesh, India

ABSTRACT

Ameloblastoma is a true neoplasm of odontogenic epithelial origin. It is the second most common odontogenic neoplasm, and only odontoma outnumbers it in reported frequency of occurrence. Unicystic ameloblastoma (UA) refers to those cystic lesions that show clinical, radiographic, or gross features of a mandibular cyst, but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumor growth. It accounts for 5-15% of all intraosseous ameloblastomas. We report a case of young female with a multilocular radiolucent lesion in left mandibular region. Histopathologic examination revealed Unicystic ameloblastoma intraluminal and mural type. The patient was treated conservatively and enucleation was done.

KEYWORDS: Unicystic; ameloblastoma; odontogenic

INTRODUCTION

The unicystic ameloblastoma (UA) is a less encountered variant of the ameloblastoma. It appears more frequently in the second or third decade with no sexual or racial predilection. It is almost exclusively encountered asymptomatically in the posterior mandible.[1] The clinical and radiological simulation of an UA with mandibular cyst often leads to simple enucleation of lesion and further its recurrence. This report highlights the importance of histopathologic analysis of any pathology in jaws even if it seems innocuous in clinical as well as in radiological examination.

CASE REPORT

A 14 year old female presented with a slowly growing swelling in the left cheek region since 3 years. She was apparently alright 7 years back then she came up with the complaint of swelling for which she was diagnosed with dentigerous cyst and enucleation and extraction of 36 was performed. Since a period of 3 years she developed swelling in the same region which increased in size in a span of last 6 months. On extraoral examination a diffuse swelling was seen extending superioinferiorly from tragus to 1cm below the lower border of mandible (Fig. 1). Anteroposteriorly from left side of ala of nose to the tragus region. Swelling was hard in consistency, non fluctuant and was not attached to overlying skin. There was no associated pain or discharge. Neck nodes were not palpable. On intraoral examination, dome shaped solitary swelling about 4 x 3cm seen on left side extending from 33 to 37 region (Fig. 2). Overlying mucosa was normal with no discharge. Vestibular obliteration was present.

INVESTIGATIONS

Routine investigations like complete blood count was done, which was normal. Orthopantomogram (OPG) (Fig. 3) revealed well defined multilocular radiolucency in left angle of mandible region extending from 34 to involve complete ramus upto the sigmoid notch. Internal content appeared to be consisting of locules and impacted 38 pushed to the inferior border at the angle of mandible. Roots of 32, 33, 34 and 35 were tipped mesially. Incisional biopsy was done and sent for histopathologic examination.

DIFFERENTIAL DIAGNOSIS

In differential diagnosis, ameloblastoma, odontogenic keratocyst (OKC), dentigerous cyst, central giant cell granuloma (CGCG), calcifying epithelial odontogenic tumor (CEOT), odontogenic myxoma were considered. In CEOT focal areas of calcifications are seen, but in our case, multilocular radiolucency without any radio opaque flecks was present. Linear expansion of

Fig. 1: Extraoral picture

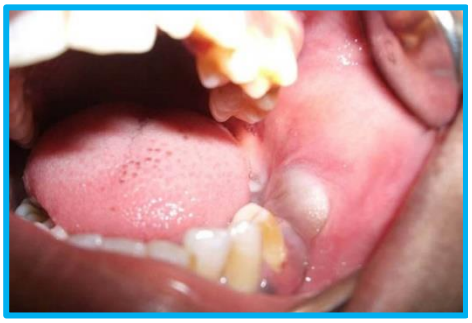

Fig. 2: Intraoral picture

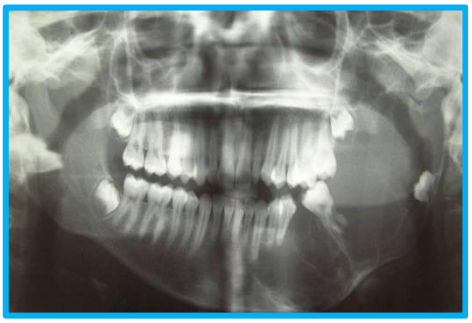

Fig. 3: Orthopantomogram

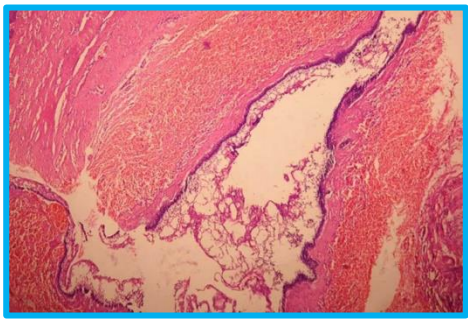

Fig. 4: Cystic lumen with wall

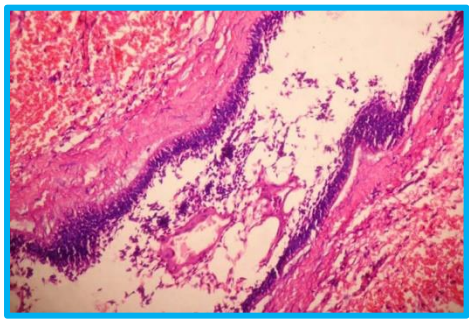

Fig. 5: Luminal and Intraluminal proliferation

OKC through medullary spaces without any buccolingual expansion ruled out OKC. Lesions like CGCG and odontogenic myxoma were also ruled out based on their clinical and radiological features.

HISTOPATHOLOGY

The histopathologic examination of the biopsy tissue revealed cystic cavity with connective tissue wall. The cystic lining consists of ameloblastic epithelium with hyperchromatic polarized basal cell layer (Fig. 4). Plexiform type of epithelial proliferations were seen within the lumen (Fig. 5). Fibrous connective tissue wall was seen with no evidence of invasion or islands of epithelium within the connective tissue suggestive of Unicystic ameloblastoma (luminal and intraluminal type).

TREATMENT

In the present case, the histopathologic examination of the biopsy sample showed ameloblastic changes confined to luminal and intraluminal area i.e., Subgroup 1.2. Hence, finally taking into account the age of patient, clinical and radiologic features, the enucleation procedure was chosen as the treatment of choice, for it has least patient morbidity and effect on the quality of life is minimal. The patient is being followed up at regular intervals to check for any recurrences

DISCUSSION

UA, first described by Robinson and Martinez in 1977, refers to those cystic lesions that show clinical, radiologic or gross features of a mandibular cyst, but on histologic examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with/without luminal and/or mural tumor growth. It accounts for 5-10% of all intraosseous ameloblastomas.[1-3] Between 50 and 80% of cases are associated with impacted tooth, the mandibular third molar being

most often involved. The 'dentigerous' type occurs 8 years earlier on average than the 'non-dentigerous' variant. Histologically, the minimum criterion for diagnosing a lesion as UA is the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas. UA should be differentiated from odontogenic cysts because the former has a higher rate of recurrence than the latter.[4] In a clinicopathologic study of 57 cases of unicystic ameloblastoma, Ackermann[5] classified this entity into the following three histologic groups:

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

Another histologic subgrouping by Philipsen and Reichart[6] has also been described:

- Subgroup 1: Luminal UA
- Subgroup 1.2: Luminal and intraluminal
- Subgroup 1.2.3: Luminal, intraluminal and intramural
- Subgroup 1.3: Luminal and intramural

The UA diagnosed as subgroups 1 and 1.2 can be treated conservatively (careful evaluation), whereas subgroups 1.2.3 and 1.3 showing intramural growths require radical resection.[1] Recurrence is also related to the histologic subtypes, among which those invading the fibrous wall have a rate of 35.7%, but others have a rate of only 6.7%.[7] UA's are considered to be less aggressive form of ameloblastomas and can be successfully removed by simple enucleation or less aggressive surgery. Recurrence rates are also related to the type of initial treatment. Lau *et al*., reported recurrence rates of 3.6% for resection, 30.5% for enucleation alone, 16% for enucleation followed by Carnoy's solution application, and 18% by marsupialization followed by enucleation (where the lesion reduced in size).[8]

CONCLUSION

Unicystic Ameloblastoma is the rare type of ameloblastoma. Around 50-80% cases are associated with impacted tooth mainly third molar. The minimal histologic diagnostic criteria for unicystic ameloblastoma is considered the presence of single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas. Conservative treatment with enucleation is required. Regular follow-up should be done .

CONFLICT OF INTEREST & SOURCE OF FUNDING

The author declares that there is no source of funding and there is no conflict of interest among all authors.

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