Unicystic Ameloblastoma: A Case Report

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Date of Receiving : 23/July/2013
Date of Acceptance : 25/Aug/2013

Abstract: Unicystic Ameloblastoma includes several clinically-radiographical and histological types. Apart from commonly encountered clinico-pathological features, few variants show distinct biological behavior. In the present case, authors report a case of unicystic ameloblastoma with luminal type in 18 year old male. Radiographically, the lesion show features similar that of dentigerous cyst as the cyst was associated with impacted third molar tooth. Histopathologically, the cyst showed features of ameloblastoma lining the cyst cavity. Emphasis was given for the comprehensive discussion of various clinically-radiographical, histopathological and behavioral aspects of Unicystic Ameloblastoma.

Key words: Ameloblastoma, Cyst, Mandible, Unicystic

INTRODUCTION

Ameloblastoma is an odontogenic tumor which accounts for 1% of all oral tumors. It occurs due to the impairment of histodifferentiation and morphodifferentiation in the process of odontogenesis. Term "Ameloblastoma" includes several clinically-radiographical and histological types. Based on the clinical behavior and prognosis ameloblastoma can be distinguished in to: (1) Solid/ Multicystic ameloblastoma; (2) Unicystic ameloblastoma; (3) Peripheral ameloblastoma; (4) Desmoplastic ameloblastoma, including the so called hybrid lesions.

In 1977, Robinson and Martinez described unicystic ameloblastoma (UA) as a distinct variant of ameloblastoma with large cystic cavity showing either mural or luminal proliferation of ameloblastic tumor cells. UA accounts for 10% to 15% of all odontogenic tumors. The term UA refers to those cystic lesions that show clinical, radiographic or gross features of jaw cyst, but on histologic examination show a typical ameloblastomatous epithelial lining the cystic cavity. The present article reports a case of UA in 20 year old male. Also attempts have been made to discuss the clinical, radiographic and histopathological aspects of UA.

CASE REPORT

An 18 year-old male patient was referred to an oral and maxillofacial surgeon complaining of a 'little lump' in his mouth of about 6-month duration. On clinical examination, asymmetry was noted in the mandibular right quadrant. Examination revealed an asymptomatic bony hard swelling in the right posterior ramus mandible covered by normal mucosa. The patient had no paresthesia or limitation of functional mandibular movements. Mucosal ulceration and cervical lymphadenopathy was not detected. The patient complained only of low level discomfort to pressure and palpation. The entire teeth in the examined area responded within normal limits to thermal pulp testing (hot and cold), suggesting their vitality. Radiographic examination revealed a well defined radiolucent area surrounding the impacted third molar. The lesion was biopsied and sent for histopathological examination. It showed cystic lesion lined by ameloblastic epithelium with hyperchromatic nucleus showing reverse polarity. The overlying epithelial cells were loosely cohesive and resembled stellate reticulum. The subepithelial hyalinization was seen at many places, which is a characteristic feature of unicystic ameloblastoma [Figure 1 & 2].

The patient was scheduled for radiographic follow-up after an interval of three months. Post six months of marsupialization, the diminished lesion was completely enucleated with peripheral osteotomy to ensure complete removal of the margins. The apical portions of the aforementioned teeth were resected and allogenic bone graft material was placed in the cavity. There were no signs of recurrence even after 15 months of follow-up.

DISCUSSION

Unicystic ameloblastoma implies not a unilocular radiographic pattern but refers to the monocystic appearance grossly and microscopically. It usually occurs in younger age group of 16-20 years, with about 50% of cases occurring in the second decade of life as in our case. The distribution shows a slight male predilection with male to female ratio of 1.6:1. However, when the tumor is not associated with an unerupted tooth, the gender ratio is reversed to a male, female ratio of 1:1.8. More than 90% are located in the mandible in the posterior region. The clinical and radiographic findings of UA suggest that the lesion is an odontogenic cyst, particularly dentigerous cyst. However, few are not associated with impacted teeth which are called nondentigerous variant.

In the present case, cyst was associated with impacted third molar depicting as a dentigerous cyst in mandibular-ramus area. Predominant radiographic patterns of UA include unilocular, scalloped, macromultilocular, pericoronal, inter-radicular or periapical expansible radiolucencies. Present case showed unilocular radiolucency associated with impacted tooth appearing as a dentigerous cyst. Vickers and Gorlin proposed the diagnostic histopathological criteria, which include tall columnar basal layer, subnuclear cytoplasmic vacuoles, reverse polarity of hyperchromatic nucleus and a thin layer of edematous,
degenerating stellate reticulum over the tall columnar cells. The mural extension in to the cystic wall is the frequent seen feature, and the term mural UA is used when the thickened lining penetrates the adjacent capsular tissue. Present case showed above mentioned similar histopathological features without mural proliferation. Various classifications have been proposed for unicystic ameloblastoma, among them two important classification have been mentioned in Table 1. Present case was classified under group III of Ackermann classification.

Confirmation of histopathological diagnosis of UA is only achieved by excisional biopsy. As the incisional biopsy is not representative of entire lesion, part of the cystic cavity may mimic dentigerous lining or part of the stroma may consists of plexiform ameloblastomatous arrangement. Leider et al. proposed three pathognomonic mechanisms for the evolution of UA. (1) The reduced enamel epithelium which is associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development; (2) Ameloblastomas arise in dentigerous cysts or in others in which the neoplastic ameloblastic epithelium is preceded temporarily by a nonneoplastic stratified squamous epithelial lining; (3) A solid ameloblastoma undergoes cystic degeneration of the ameloblastic islands, with subsequent fusion of multiple microcysts and develops into unicystic lesions.

Treatment planning depends on the histological type of UA (Table 1). Cysts diagnosed under the subgroup 1 and 1.2 may be treated conservatively, where as the subgroup 1.2.3 and 1.3 should be treated aggressively. The histopathological typing of current case was under group 1 and hence the lesion wad treated conservatively. The recurrence rate of UA after conservative surgical treatment is reported to be 10% to 20%, irrespective of any surgical modality and hence mandates long follow up. Present case was followed for six months without recurrence.

CONCLUSION

The diagnosis of UA has to be achieved by the correlation of clinical, radiographic, and histopathological diagnosis. Proper excisional biopsy is mandatory to sign out the histopathological diagnosis of UA. Proper surgical intervention with long follow up is required.

REFERENCES


Table 1: Proposed Classifications of Unicystic Ameloblastomas

<table>
<thead>
<tr>
<th>Ackermann⁸,⁹</th>
<th>Philipson and Reichart⁸</th>
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<tbody>
<tr>
<td><strong>Group I</strong></td>
<td>Luminal UA (tumor confined to the luminal surface of the cyst)</td>
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<tr>
<td><strong>Group II</strong></td>
<td>Intraluminal/Plexiform UA (nodular proliferation into the lumen without infiltration of tumor cells into the connective tissue wall)</td>
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<tr>
<td><strong>Group III</strong></td>
<td>Mural UA (invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium)</td>
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<tr>
<td>Subgroup 1</td>
<td>luminal UA</td>
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<tr>
<td>Subgroup 1.2</td>
<td>luminal and intraluminal</td>
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<tr>
<td>Subgroup 1.2.3</td>
<td>luminal, intraluminal and intramural</td>
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<tr>
<td>Subgroup 1.3</td>
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Fig. 1- Photomicrograph showing ameloblastomatous epithelium lining the cystic cavity (H/E - X100)

Fig. 2- Photomicrograph showing ameloblast cells with reverse polarity of the nucleus