

Unicystic ameloblastoma: A case series

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Abstract

Ameloblastoma is locally intrusive and is envisioned to be a neoplasms of benign type originated from odontogenic epithelium type, which generally affects the mandible. Ameloblastoma comprises different types with precise clinical and histological features. Apart from all these usually encountered varieties there are not so generally occurring types of the ameloblastoma, unicystic type of ameloblastoma being one among those signifies to that cystic lesions that display features clinically, radiographically or gives gross descriptions of jaw cyst but on histological inspection displays an epithelium which is seen lining the cystic cavity typically ameloblastomatous cells type, with or without luminal and or mural tumor proliferation. Albeit unicystic ameloblastoma being an infrequently encountered type of ameloblastoma and is well-thought-out to be less aggressive, this tumor displays significant similarities with Dentigerous cyst equally when seen clinically and on radiographs too, it befalls difficult to conclude the correct diagnosis and suitable form of treatment. We are bestowing a case report of 15 histologically diagnosed unicystic ameloblastoma cases, among which 4 comprised intraluminal type and 11 mural type.

Keywords: Ameloblastoma, Unicystic, Mural, Tumor, Cyst.

Introduction

Dated back to year 1977 Robinson and Martinez was the first one to define unicystic ameloblastoma, a type of ameloblastoma.¹ Unicystic ameloblastoma being one among those cystic lesions that display features clinically and on radiograph or gives gross descriptions of a mandibular cyst.² Principally a well-defined lesion is seen, monocystic cavity with a lining frequently sizable is seen with lining, focally but comprised of odontogenic (ameloblastomatous) epithelium.³ Some investigators believed that the unicystic ameloblastoma develops from pre-existing odontogenic cysts, it develops mostly from a pre-existing cyst generally a dentigerous cyst, while some other authors support that the cyst arises de novo. Author Robinson and Martinez debated that as the epithelium of odontogenic cysts and ameloblastoma are supposed to have a communal ancestry, Feasibility of a switch from a non-neoplastic cyst to a neoplastic cyst is more, albeit it occurs infrequently.⁴

The development of unicystic ameloblastoma can be described by three pathogenic mechanism: First is reduced enamel epithelium, second is from dentigerous cyst and third is due to cystic degeneration of solid ameloblastoma.⁵ In approximately 50% incidence the unicystic ameloblastoma is seen to appear in second decade of life.^{6,7} In patients with unicystic ameloblastoma cases where tumour is not related with an unerupted tooth, the incidence ratio of male to female is completely upturned i.e 1:1.8.⁸ Unicystic ameloblastoma commonly presents with local swelling, intermittent pain, and lip numbness, as well as discharge or drainage in cases of secondary infection⁹ radiographically, unicystic ameloblastomas have been seen to be presented into two main patterns: one is unilocular and second one multilocular. In most the cases the dominance of unicystic ameloblastomas have been clearly seen for the unilocular pattern.¹⁰ The radiological descriptions of

unicystic ameloblastoma are classically unilocular and there is a round area of radiolucency. Hence, this lesion is frequently misdiagnosed as an odontogenic keratocyst or dentigerous cyst.¹¹

Histological classification given by Ackermann, Ackermann classified unicystic ameloblastoma into three histologic groups as tabulated² in Table 1:

Table 1: Ackermann classified this entity into three histologic groups

1) Luminal unicystic ameloblastoma, in this presentation tumour confined to the luminal surface of the cyst.
2) Intraluminal/plexiform unicystic ameloblastoma, in this presentation nodular proliferation into lumen without infiltration of tumour cells into connective tissue wall is seen.
3) Mural unicystic ameloblastoma, in this invasive islands of ameloblastomatous epithelium in the connective tissue wall not involving the entire epithelium.

Classification given by Philipsen and Reichart^{8,13} in Table 2 has also been explained.

Table 2

Subgrouping by Philipsen and Reichart: Subgroup
1: luminal UA; Subgroup
1.2: luminal and intraluminal; Subgroup
1.2.3: luminal, intraluminal and intramural; and Subgroup
1.3: luminal and intramural.

Accordingly, a definitive final diagnosis of unicystic ameloblastoma can only be made by thorough clinical, radiographical and histological examination of the lesion.

Table 3

	Total no. of cases	Mural type	Intraluminal type	Multilocular	Unilocular
Maxilla	6.6%	6.6%	0%	0%	6.6%
Mandible	93.33%	66.66%	26.66%	26.66%	66.66%

As reported by different authors unicystic ameloblastoma is reported to be less aggressive so, the response of unicystic ameloblastoma to enucleation or curettage is extra favourable than the classic solid or multicystic ameloblastoma. Better prognosis of ameloblastoma as explained by many examples is that the involvement of only the epithelial lining of the cyst or projecting into its lumen is seen in ameloblastoma.¹⁴

In the present case presentation of a sequence of unicystic ameloblastoma of distinguishing series of 15 cases with Unicystic ameloblastoma (out of which 11 are mural type and 4 are intraluminal proliferation type) which were reported in the year 2016 -2018 in the OPD of HIDS Paonta Sahib.

Case Report

In our present case report we are reporting a series of 15 cases, histologically diagnosed unicystic ameloblastoma among which 11 out of 15 diagnosed with unicystic ameloblastoma with mural proliferation and 4 cases out of 15 were diagnosed with unicystic ameloblastoma intraluminal proliferation type. To establish the clinicopathologic and biologic conduct of this tumour group, the clinic pathological characters of 15 unicystic ameloblastoma were reviewed. The sequence shows approximately 73.33% cases with mural type proliferation unicystic ameloblastoma and rest with intraluminal type. The age finding ranged from 20-40 years 93.33% occurred in the mandible and rest in maxilla. (Table 3)

Patients narrated with complaint of puffiness on Right side in 11 cases and on the left side crossing midline in 4 cases since 5-6 months in all the reported cases. (Fig. 1, 2) In all the incidents patient had undertaken treatment of cyst marsupialisation around 5-6 years back for the same. In the reported cases on intraoral examination, bony extension with hard painless swelling was seen extending from lower right to third molar region to canine region without affecting oral mucosa i.e with intact mucosa in 93.33% cases is seen. All the teeth in the lesional area were vital and the patients experienced no correlated pain and difficulty in opening or closing the mouth, nor while chewing and articulating.

The OPG revealed (Fig. 3), an spread-out unilocular radiolucency in 7 cases and was multilocular in 8 cases. Microscopically (Fig. 4-7) under scanner view given H and E stained soft tissue section showed cystic lining with underlying fibrous connective tissue wall. Under lower magnification all tumours showed a generally monocystic growth pattern, epithelial lining the cyst wall with typical ameloblastomatous cells in the basal layer with hyper chromatic nuclei, showing reversal polarity with basilar cytoplasmic vacuolization. Generally monocystic growth pattern was seen in all the reported tumors, 4 out of total 15 were compromised intraluminal tumour nodules and the

remaining 11 out of total 15 mural type. The cystic lining seen is consistently showed, at least in part, a typical ameloblastomatous pattern that is often accompanied by epithelial areas of various histological appearance. On the basis of above mentioned conclusions the final diagnosis of unicystic ameloblastoma type intra luminal type and intra mural type was made.



Fig. 1: Extraoral pre operative photograph



Fig. 2: Intra oral photograph



Fig. 3: OPG of patient showing well defined unilocular radiolucency on right ramus and body region extending till the mandibular canine region

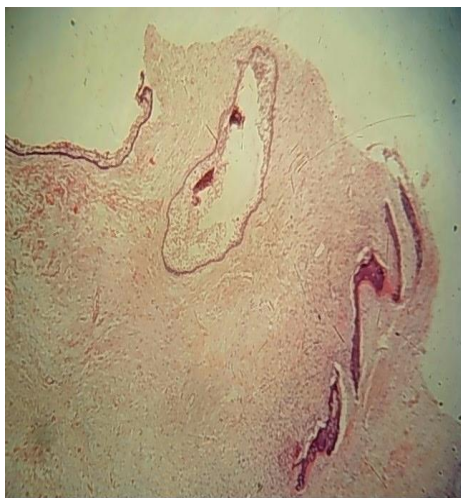


Fig. 4: Microphotograph under scanner (4X) view showing cystic lining with underlying fibrous connective tissue wall



Fig. 5: Microphotograph under lower magnification showing epithelial cells lining the cyst wall with typical ameloblastomatous cells in the basal layer. (10X)

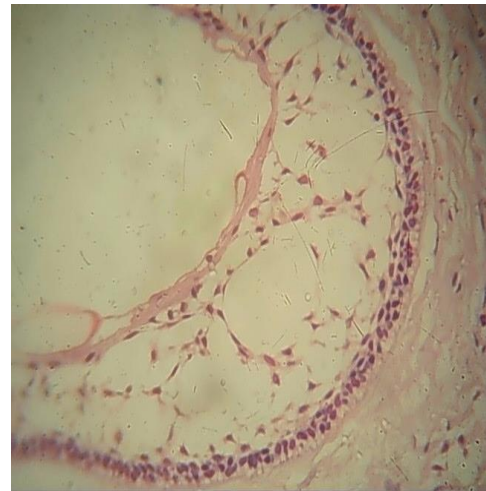


Fig. 6: Microphotograph showing typical ameloblastomatous cells in the basal layer with hyperchromatic nuclei, showing reversal of polarity with basilar cytoplasmic vacuolization and overlying epithelial cells are seen loosely cohesive and resembling stellate reticulum.(40X)

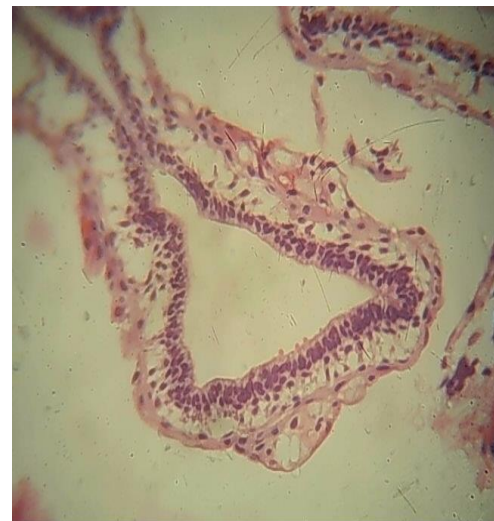


Fig. 7: Cystic fibrous wall is seen with follicular ameloblastoma islands.(40X)

Discussion

In the reported cases, the provisional opinion of dentigerous cyst was made bestowing to radiographic and clinical conclusions but histopathological investigation revealed ameloblastic variations. Notwithstanding the descriptions about unicycstic ameloblastoma may in typical, compare approvingly with its solid or multicystic equivalent in rapports of clinical presentation and response to treatment, the subsections of maxillary lesions or tumors comprehending invading islands in the fibrous wall could have high risk of recurrence. Additionally, recurrence of unicycstic ameloblastoma may be long deferred and long term post-operative follow up is needed for the proper management of these patients. Reappearances is seen to be emerged to relate to histologic subtypes of unicycstic

ameloblastoma with those that are seen invading the fibrous wall having a rate of 35.7% but other types having rate of 6.7%. The follow up of patients at regular intervals are to be taken up, to check for any reappearance. In the reported sequences of series of 15 cases in this case report, feasibility of final diagnosis was only made because of histopathological examination which was implemented in the enucleated material. The standard duration recommended for follow up for the patents diagnosed with ameloblastoma in previous report was 5 years after the surgical treatment. In the present cases no recurrence has been reported from past two years.

Conclusion

It is mostly challenging to diagnose unicyclic ameloblastoma completely relying on the clinical and radiographically descriptions due to the resemblance between both, same way in the present reported series of 15 cases, the diagnosis was feasibly achieved only because of histopathological examination which was implemented in the enucleated material. In the regular practice concluding that surgical protocol must include the post-operative histopathological examination for all the lesions to rule out any ameloblastomatous changes is mandatory, so that the patient can be followed up properly to take care of any recurrences happening in the course of time.

Conflict of Interest: None.

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