IOSR Journal of Dental and Medical Sciences (IOSR-JDMS)
e-ISSN: 2279-0853, p-ISSN: 2279-0861.Volume 13, Issue 4 Ver. IV. (Apr. 2014), PP 80-82
www.iosrjournals.org

Mural Variant of UnicysticAmeloblastoma

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Abstract: *As per our knowledge of recent literature, Ameloblastoma is a true neoplasm of odontogenic epithelial origin is known to be the second most common odontogenic neoplasm after odontoma which is said to have the highest rate of occurrence.The incidence rate of Ameloblastoma, combined with its clinical behaviour, makes ameloblastomaone of the most significant odontogenicneoplasm. Unicysticameloblastoma (UA), a variant of Ameloblastoma is known to comprise of those cystic lesions that show clinical, radiographic, or gross features of a cyst, but on histological examination it shows a typical ameloblastomatous epithelium lining which is part of the cyst cavity, with or without luminal and/or mural tumour growth.*
Key Words: *Ameloblastoma, Neoplasm, Odontogenic Tumor*

I. Introduction

In our knowledge there are many lesions comprising either of hard bony or soft tissue in origin that are known topotentiate mandibular swellings, and they can be grouped under lesion having an odontogenic and nonodontogenic origin. The commonly associated tumor of odontogenic origin is Ameloblastoma which is known to develop from epithelial cellular elements and dental tissues in their various phases of development.In majority ,more than 80% of all ameloblastomas are solid or multicystic variants, while unicysticameloblastoma being another clinicopathologic form of ameloblastoma occupies the remaining 20% of the cases along with peripheral Ameloblastoma[1]. Thus, basedupon the patterns of the lesion observed clinically and its prognosis, three subgroups of ameloblastomas can be presently distinguished: (1) the 'conventional or classical', intraosseous, solid or multicysticameloblastoma; (2) the unicysticameloblastoma; and (3) the peripheral Ameloblastoma[2]. Recent studies on Ameloblastoma do indicate another entity known as thedesmoplasticameloblastomawhich might be qualified as a fourth subgroup of ameloblastomas because of its unique biological behaviour, radiographic appearance, and variedhistological features [1].

Robinson and Martinez[3] in 1977 described unicysticameloblastoma as a subvariant of ameloblastomaknown to associated with a large cystic cavity showing either mural or luminal proliferation of ameloblastictumor cells. In 1984,Eversole et al[4] described desmoplasticameloblastoma, a new subvariantof ameloblastoma which presents a varied histopathological and clinical patterns findings. Histologically, it is characterized by extensively collagenizedstroma along with the presence of small nests and strands of odontogenic epithelium[5].

Unicysticameloblastoma,a less encountered variant of the Ameloblastoma, is known to have clinical and radiographic characteristicssimilarto that of an odontogenic cyst but histologically it shows a typical ameloblastomatous epithelium lining the part of the cyst cavity, with or without the presence of luminal and/ormural tumor proliferation[6].

Ameloblastoma, a benign, locally aggressive odontogenic neoplasm with varied clinicalfeature accounts for nearly 1% of all cysts/tumors of jaws and around 18% of all odontogenic neoplasms. It is a slow growing, locally aggressive which is known to metastasizes rarely but is known to have a high rate of recurrence (55–90%) if not removed adequately[7].UnicysticAmeloblastoma (UA) accounting for around 6% of Ameloblastomas is known to usually occur in a younger age group of 16–20 years, with about 50% of the cases occurring in the second decade of life [8,9]. The gender distribution shows a slight male predilection with a male to female ratio of 1.6 :1, however the ratio is changed when the tumor is not associated with an unerupted tooth, and ratio of male to female ratio being 1:1.8[2]. Mucosal ulceration is generally rare but may be caused by continuous growth of the tumor[10].

The UnicysticAmeloblastoma, a distinctive type of ameloblastoma,can befurther sub-divided into four different groups as follows : luminal (1), luminal and intra luminal (1.2), luminal, intra luminal, and intra mural (1.2.3), luminal and intramural (1.3) types[11]. Amongst the following types the unicysticameloblastomas associated with muralproliferation is considered to be aggressive and therefore should be treated in the same manner as solid multicysticameloblastoma, while the other subvariants can be treated in a conservative fashion [11,12].

Leider et al has proposed three pathologic mechanisms for development of UnicysticAmeloblastoma[13].

a. The reduced enamel epithelium associated with a developing tooth undergoes ameloblastic transformation with subsequent cystic development.
b. Ameloblastomas arise in dentigerous or other types of odontogenic cysts in which the neoplastic ameloblastic epithelium is preceded temporarily by a non-neoplastic stratified squamous epithelial lining.
c. A Solid ameloblastoma undergoes cystic degeneration of ameloblastic islands with subsequent fusion of multiple micro cysts and develops into a unicystic lesion.

The radiographic appearance of UCAs has been divided broadly into 2 main types :unilocular and multilocular, with the unilocular patternbeing more dominant. This preponderance is predominantly seen for the dentigerous variant, where the unilocular to multilocular ratio is 4.3:1, and for the nondentigerous type, this ratio is 1.1:1[14] . The involved teeth show varying degrees of root resorption[13].

Histologically, Ackermann et al[11] classified unicysticameloblastoma into 3 histological subvariants

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

The above mentioned classification was further modified by Philipsen and Reichart[13] as already stated above as:
Subgroup 1 – luminal unicysticameloblastoma.
Subgroup 1.2 – luminal and intraluminal
Subgroup 1.2.3 – luminal, intraluminal and intramural
Subgroup 1.3 – luminal and intramural.

A final confirmatory diagnosis of unicysticameloblastomashould only be done by histological examination of the entire lesion and cannot be made solely on clinical or radiographic grounds.The epithelial lining of a UCA is not always uniformly characteristically distinct and is often lined partly by a nonspecific thin epithelium that may mimics the dentigerous cyst lining[7]. Thus, true nature of the lesion becomes evident only after complete enucleation when the entire specimen is available for microscopy[10]

Thus histologically, the minimum criteria for diagnosing a lesion as UCA are the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas[15]

II. Conclusion

The final diagnosis of unicysticameloblastoma should not be based on clinical, radiological findings alone but histopathology should be adviced for a final diagnosis . It is a tumor with a strong inclination for recurrence, especially when the ameloblasticfoci perforates the adjacent tissuefrom the wall of the cyst. Unicystic variant of ameloblastoma with aggressive histologic behaviour also might be successfully treated with marsupialisation with subsequent enucleation, and this approach can be considered as an alternative to resection. Oral health care providers should be aware of the unilocularradiolucencies of the jaws as this lesion could be unicysticameloblastoma having strong propensity for recurrences and timely intervention and conservative surgical treatment may improve treatment outcome and potential complications associated with larger resection.

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