Case Report

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Unicystic Plexiform Ameloblastoma in a Rare Location

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Abstract

Unicysticameloblastomas (UA) are a rare variant of ameloblastomas and refers to those cystic lesions that show clinical, radiographic and 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. Unicysticameloblastoma is less encountered variant of the ameloblastoma and believed to be less aggressive in nature. They are characterized by slow growth and locally aggressiveand most commonly seen in posterior portion of the mandible. Here we report a case of unicysticplexiformameloblastoma in symphyseal region in 53 years old male.

Keywords: Mural Growth; Plexiform; Unicystic.

Introduction

A unicysticameloblastoma (UA) was first described by Robinsonand Martinez in 1977 [1]. Although it is a rare variant of ameloblastomas, it has a relatively benign biologic behavior and better response to conservative treatment [2]. It accounts for 15% of all intraosseousameloblastomas and often affects the younger population, most of the cases occurring in the second decade of life.

Unicysticameloblastomas have a slight male predilection and frequently occurs in the posterior mandible [3]. Radiographically, the lesions commonly show expansive unilocular radiolucencies with a well-demarcated border. Approximately 50-80% of cases are associated with an impacted or unerupted tooth [4]. Three histological types are recognized according to the degree of ameloblastomatous epithelial extension, namely luminal, intraluminal and mural types [5].

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Case Report

A 53 years old male patient visited us with the swelling in the symhyseal region of the mandible since 5 months. Swelling was sudden in onset and gradually progressed to the present size. Patient had no pain and paresthesia in the swelling region and patient gives history of extraction of lower front teeth two years back. On inspection a swelling noticed, measuring about 4 x 3 cm over the symphyseal region extending from right angle of mouth to left side. Superiorly just below the lower lip region to lower border of the mandible inferiorly. The overlying skin appears to be normal. On palpation all inspectory findings were confirmed and swelling was soft to firm in consistency and it was fluctuant. Intra orally, vestibular obliteration was noticed in the labial vestibule extending from 35 to 45 region and mucosa over the swelling seems to be normal. Based on history and clinical examination provisional diagnosis of residual cyst was made. Occlusal radiograph [Figure 1] showed radiolucent lesion extending from 35 to 45 in the periapical region. Expansion of labial cortical plate noted extending from 35-43 region. Posterior extent of cortical expansion is not defined. After radiological examination, differential diagnosis ofodontogenickeratocyst and ameloblastoma was made. Incision biopsy was made at the representative area of the swelling. On histopathological examination, H and E stained [Figure 2] section revealed cystic lesion lined by epithelium of

odontogenic in origin, 2-3 cell layered thick showing budding into underlying connective tissue. In many areas, epithelium shows intraluminal proliferation which is proliferated in the form of follicles and follicles lined [Figure 3] by ameloblast like cells and connective tissue stroma in the centre of the follicle and follicles were surrounded by stellate reticulum like cells which represents plexiform ameloblastomatous in nature. In few areas the connective tissue capsule surrounding the cyst is extremely thin. All these features were suggested of unicystic plexiform ameloblastoma. After histopathological diagnosis swelling was removed by means of enucleation with bony resection. Patient is followd up till date.

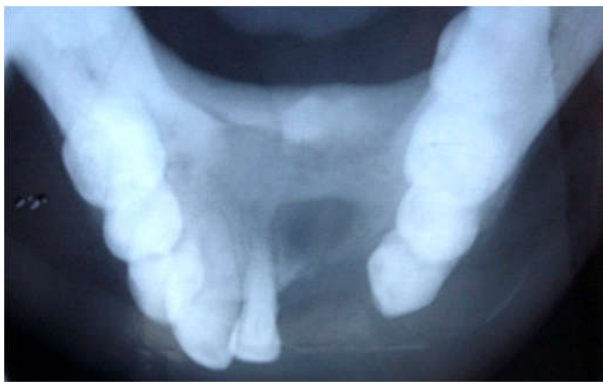

Fig. 1: Occlusal radiograph showing showing radiolucent lesion extending from 35 to 45 in the periapical region

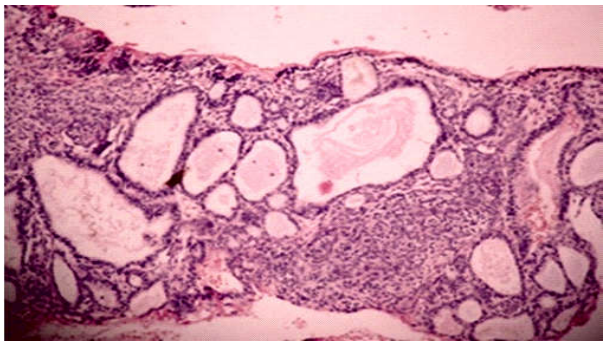

Fig. 2: Photomicrograph [10X] showing cystic lesion lined by epithelium of odontogenic in origin, 2-3 cell layered thick showing budding into underlying connective tissue

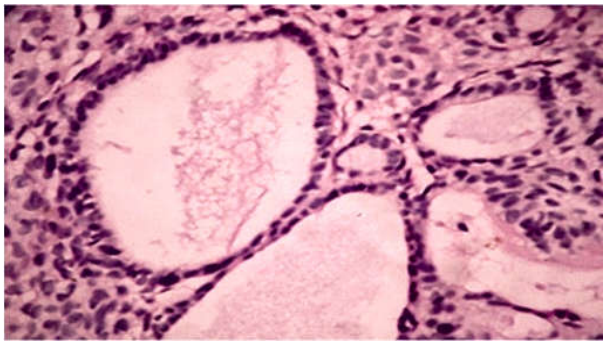

Fig. 3: Photomicrograph [40 X] showing and follicles lined by ameloblast like cells and connective tissue stroma in the center and follicles were surrounded by stellate reticulum like cells

Discussion

Unicysticameloblastoma is a rare varient of ameloblastoma, accounting for about 6% of ameloblastomas[1,2]. About 50% of the cases occur in the second and third decades of life. The mandible is affected more often than the maxilla. These tumors are most commonly encountered in the posterior mandible followed by the parasymphysis region, anterior maxilla, and the posterior maxilla, but in our case UA was seen in the symhyseal region which is extreamly rare[1,4]. Clinically and radiographically, the unicystic ameloblastoma often has the appearance of a dentigerouscyst.this feature was not in consistant with our case. A confirmatory diagnosis of unicystic ameloblastoma was made by histopathological evaluation of biopsy specimens [4] .

Ackermann et al Classified this Entity into 3 Histologic Groups: [5]

Group 1 – Luminal unicysticameloblastoma lesions consist of a unilocular cyst lined by epithelium that in some areas shows ameloblastic transformation without infiltration into the connective tissue wall.

Group 2- Intraluminal/plexiform unicystic ameloblastoma lesions consist of a unilocular cyst with the lining epithelium showing a nodular proliferation of plexiform ameloblastoma into the lumen without infiltration of tumor cells into the connective tissue wall.

Group 3- Mural unicystic ameloblastoma lesions have invasive islands of ameloblastomatous epithelium in the connective tissue wall that may or may not be connected to the cyst lining epithelium.

Adenomatoid odontogenic tumor, odontogenic keratocyst, residual cysts, giant cell lesions and sometimes solid ameloblastoma can be the differential diagnoses [6]. The treatment depends on the histological pattern of the ameloblastoma. In cases of the luminal, intraluminal or plexiform patterns, enucleation generally suffices but if there is a mural component, bony resection is necessary to ensure adequate removal [7]. In our case it was of plexiform variant which was treated by enucleation with bony resection.

Conclusion

This article tries to highlight some about unicystic

plexiform ameloblastoma, based on a case report, and also tries to emphasize the fact that recent insights into the biologic nature of these lesions seem to suggest that they could be more aggressive than previously thought.

Referrences

1. Paikkatt VJ, Sreedharan S, Kanna VP. Unicysticameloblastoma of the maxilla: A case report. J Indian SocPedodPrev Dent. 2007; 25: 106–10
2. Gardner DG, Corio RL. plexiform unicystic ameloblastoma: A variant of ameloblastoma with a low recurrence rate after enucleation. Cancer. 1984; 53: 1730–5.
3. Li TJ, Wu YT, Yu SF, Yu GY. Unicysticameloblastoma: a clinicopathologic study of 33 Chinese patients.Am J Surg Pathol. 2000; 24: 1385-92.
4. Ramesh RS, Manjunath S, Ustad TH, Pais S, Shivakumar K. Unicysticameloblastoma of the mandible – an unusual case report and review of literature. Head Neck Oncol. 2010; 14; 2: 1
5. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: A clinicopathological study of 57 cases. J Oral Pathol. 1988; 17: 541–6
6. Gardner DG, Corio RL. Plexiform unicystic ameloblastoma. A variant of ameloblastoma with a low-recurrence rate after enucleation.Cancer. 1984 Apr 15; 53(8): 1730-5.
7. Bisinelli JC, Ioshii S, Retamoso LB, Moyses ST, MoysesSJ,Tanaka OM. Conservative treatment of unicysticameloblastoma.Am J OrthodDentofacial Ortho. 2010; 137: 396-400.