

Case study

Long standing ameloblastoma mimicking residual cyst – a unique case report.

ABSTRACT :- Unicystic ameloblastoma is a rare type of ameloblastoma, accounts for about 6 % of all ameloblastomas. Great difficulty exists in differentiating dentigerous cyst from UA. The unicystic ameloblastoma deserves special consideration on the basis of its clinical and radiologic appearance, its histopathology, and its response to treatment. In the present case complete enucleation was performed followed by excisional biopsy revealing ackermans type three variant. The purpose of this case report is to present a case of unicystic ameloblastoma that was long standing and appeared after tooth extraction and was misdiagnosed as residual cyst.

KEYWORDS :- UA , tumour , long standing , cyst.

Introduction :-

Unicystic ameloblastoma, described by Robinson & Martinez in 1977, is one of three clinical variants of ameloblastoma, the other two being the more common intraosseous solid or multicystic (conventional).¹⁻⁴ VICKERS & GORLIN in 1970⁵ described 3 distinct histopathological features for unicystic ameloblastoma and these were slightly modified by LEIDER et al. In 1985⁶. ACKERMAN et al. in 1988 reported a series of 57 unicystic ameloblastomas and studied their histological features in detail.⁷ Type 1 – A unilocular cystic lesion lined by epithelium which in areas shows the criteria defined by VICKERS & GORLIN⁵. Type 2 – A nodule arising from the cyst lining, projecting into the lumen of the cyst, and comprising odontogenic epithelium with a plexiform pattern which closely resembles that seen in the plexiform ameloblastoma. Type 3 – The presence in the connective tissue wall of the cyst, of invasive islands of ameloblastomatous epithelium.⁸ The reported recurrence rate after treatment of unicystic ameloblastoma ranges from 10 to 25%^{7,9,10,6,3,11}.

The purpose of this case report is to present a case of unicystic ameloblastoma that was long standing and appeared after tooth extraction and was misdiagnosed as residual cyst.

Case report :-

A female patient aged 28 years reported to the department with a well-defined swelling in the right back region of lower jaw (figure 1), since past 1 month, patient had a history of extraction wrt 46 from a local clinician, following which the swelling appeared and gradually increased in size, the swelling was non reducible and did not responded to any medication ,the swelling was 3*2 cm in dimension, swelling was tender and fluctuant in consistency and the overlying mucosa was smooth and normal, patient was advised orthopantamogram, the report clearly showed the presence of a well-defined radiolucency, with corticated smooth margin and radiolucent cavity, fine needle aspiration biopsy was performed under local anaesthesia which revealed a yellowish brown colour fluid (figure 2) . Based on the history, clinical and radiological examination the provisional diagnosis of residual cyst was made.

Patient underwent enucleation of the cyst under local anaesthesia, the complete cyst was removed in total and was sent for biopsy (figure 3, figure 4)

The histopathological report showed sections of cystic architecture with evidence of an area of ameloblatomatous lining and moderately fibrous capsule with evidence of few ameloblastomatous follicle, giving it an impression of unicystic ameloblatoma group-3(disconnected follicles by ackerman 1988).(Figure 5)

Patient was recalled back for further management patient didn't reported back.

DISCUSSION :-

UA is a rare type of ameloblastoma, accounts for about 6 % of all ameloblastomas. Great difficulty exists in differentiating dentigerous cyst from UA. However, following manifestations favors UA. Defect in the wall of a cyst, unilocular cystic lesion extending into the ramus, expansion of both buccal and lingual cortex (tumor usually grows buccally and lingually, whereas the cyst grows toward most dependent part, i.e. buccally), presence of erythematous and granulomatous tissue at the marginal gingival (mucosal ulceration) with the absence of the bony cortex, and associated healthy primary dentition.¹²

The unicystic ameloblastoma deserves special consideration on the basis of its clinical and radiologic appearance, its histopathology, and its response to treatment³. It has been suggested that for all unilocular lesions, an excisional biopsy by enucleation should be carried out. If the histopathological diagnosis shows Ackerman type 1 or type 2 unicystic ameloblastoma, then follow-up and a wait and see policy is advocated till recurrence is noted. However, for a pathological diagnosis of Ackerman type 3 resection is recommended. The rationale for treatment without an incisional biopsy is that a small tissue may not reflect all types of Ackerman unicystic ameloblastoma; thus, the chance of under diagnosis is high.³

Enucleation alone yielded the highest recurrence rate among all treatment (30.5%). Two possible explanations: firstly, cystic lining of the tumor is inadequately removed; secondly, ameloblastic tumor cells can invade the cancellous bone to a certain extent.¹² Enucleation followed by application of Carnoy's solution has resulted in a recurrence rate of 16.0% which is the best except for resection.¹² The recurrence rate could even lower than reported, if the closely related teeth with tumor are extracted.¹² Because in an attempt to preserve the tooth without damage, tumor remnants may be left around the tooth apex or root and these may lead to recurrence.^{11,12} Carnoy's solutions a powerful fixative penetrates the cancellous spaces and thus fixes the remaining tumor cells.

In the present case complete enucleation was performed followed by excisional biopsy revealing ackermans type three variant. Patient was recalled for marginal resection but didn't turned up. We also support the idea of being minimally aggressive for type 1 and 2 variants and resections for type 3 variant.

References

1. Piscevic A, Gavric M, Sjerobabin I. Maksilofacialna Hirurgija, Izdavacka Agencija "Draganic", Beograd, 1995. pp. 344–6. 2.
2. Li TJ, Kitano M. Reviewing the unicystic ameloblastoma: a clinicopathologically distinct entity. *Oral Med Pathol.* 1997;2:61–8
3. Robinson L, Martinez MG. Unicystic ameloblastoma: A prognostically distinct entity. *Cancer* 1977; 40: 2278–2285.
4. P. K. Lee¹, N. Samman¹, I. O. Ng², Unicystic ameloblastoma—use of Carnoy's solution after enucleation, *Int. J. Oral Maxillofac. Surg.* 2004; 33: 263–267.
5. Vickers RA, Gorlin RJ. Ameloblastoma: Delineation of early histopathologic features of neoplasia. *Cancer* 1970; 26: 699–710
6. Leider AS, Eversole LR, Barkin ME. Cystic ameloblastoma. A clinicopathologic analysis. *Oral Surg Oral Med Oral Pathol* 1985; 60: 624–630.
7. Ackermann GL, Altini M, Shear M. The unicystic ameloblastoma: A clinicopathological study of 57 cases. *J Oral Pathol* 1988; 17: 541–546
8. S. L. Lau, N. Samman, Recurrence related to treatment modalities of unicystic ameloblastoma: a systematic review, *Int. J. Oral Maxillofac. Surg.* 2006; 35: 681–690.
9. Gardner DG, Corio RL. The relationship of plexiform unicystic ameloblastoma to conventional ameloblastoma. *Oral Surg Oral Med Oral Pathol* 1983; 56: 54–60.
10. Gardner DG, Corio RL. Plexiform unicystic ameloblastoma. A variant of ameloblastoma with a low-recurrence rate after enucleation. *Cancer* 1984; 53: 1730–1735.
11. Shteyer A, Lustmann J, LewinEpstein J. The mural ameloblastoma: A review of the literature. *J Oral Surg* 1978; 36: 866–872.
12. Ritesh Kalaskar, Ashok S. Unawane,¹ Ashita R. Kalaskar, Conservative management of unicystic ameloblastoma in a young child: Report of two cases, *Contemp Clin Dent.* 2011 Oct-Dec; 2(4): 359–363. 12

Legends

FIGURE 1:-	Patient reported with facial asymmetry
FIGURE 2 :-	Aspiration positive showing yellowish brown fluid
FIGURE 3:-	Enucleation of cyst under anesthesia
FIGURE 4:-	Cyst removed in toto
FIGURE 5:-	Histopathological section revealing area of ameloblatomatous lining and moderately fibrous capsule

IMAGES

FIGURE 1 :-



FIGURE 2:-



FIGURE 3:-



FIGURE 4 :-



FIGURE 5 :-

