

Unicystic Ameloblastoma of the Mandible: A Case Report and Review of Literature

Harsha Gorrela^{1*} Prashanth Jakkula² Aditya Mohan Alwala³ Ramesh K⁴ Rathod Prakash⁵

¹Professor and Head, Department of Oral and Maxillofacial Surgery, MNR Dental College and Hospital, Hyderabad, Telangana, India.

²PG Student, Department of Oral and Maxillofacial Surgery, MNR Dental College and Hospital, Hyderabad, Telangana, India.

³Associate Professor, Department of Oral and Maxillofacial Surgery, MNR Dental College and Hospital, Hyderabad, Telangana, India.

⁴Associate Professor, Department of Oral and Maxillofacial Surgery, MNR Dental College and Hospital, Hyderabad, Telangana, India.

⁵Assistant Professor, Department of Oral and Maxillofacial Surgery, MNR Dental College and Hospital, Hyderabad, Telangana, India.

ABSTRACT

Background: Ameloblastoma is a true neoplasm of odontogenic epithelial origin. It is the second most common odontogenic neoplasm, and only odontoma outnumbers it in reported frequency of occurrence, with strong predilection for the posterior region of the mandible. Unicystic ameloblastoma (UA) refers to those cystic lesions that show clinical, radiographic, or gross features of a cyst, but on histologic examination show a typical ameloblastomas epithelium lining part of the cyst cavity, with or without luminal and/ or mural tumor growth. It accounts for 5-15% of all intraosseous ameloblastomas. We report a case of unicystic ameloblastoma in a 52-year-old male.

Keywords: Odontogenic neoplasm, unicystic ameloblastoma, peripheral osteotomy.

INTRODUCTION

Many benign lesions cause jaw swellings, and these can be divided into those of odontogenic and non-odontogenic origin [1]. The most common tumor of odontogenic origin is ameloblastoma which develops from epithelial cellular elements and dental tissues in various phases of development [2]. The vast majority of ameloblastomas arise in the mandible, and the majority of these are found in the angle and ramus region. There are three forms of ameloblastomas, namely multicystic, peripheral, and unicystic tumors [3]. More than 80% of all ameloblastomas are solid or multicystic variants, with unicystic ameloblastoma being an important clinicopathologic form of ameloblastoma and occupying the remaining 20% of the cases along with peripheral ameloblastoma [4]. Unicystic ameloblastoma (UCA) is the most common term used to designate its different pathological entities [5]. Unicystic tumors include those that have been variously referred to as mural ameloblastomas, luminal ameloblastomas, and ameloblastomas

arising in dentigerous cysts. The goal of treatment of ameloblastoma is to achieve complete excision and appropriate reconstruction [6]. We present a case of unicystic ameloblastoma in a 52 year old male.

CASE REPORT

A 52 year old male reported to the department of oral and maxillofacial surgery, MNR dental college and hospital, with a chief complaint of pain and swelling in the lower left anterior region of mandible since two weeks. Extra oral examination revealed facial asymmetry and a diffuse swelling in submental region extending to the base of the mandible with mild obliteration of the of mentolabial sulcus which is measuring about 2.5cm×2cm in size (Fig-1). On palpation, the swelling was firm in consistency and tender, no local rise of temperature, it was non pulsatile and non-compressible, and no discharge was present. A

Received: Feb. 13, 2021; Accepted: Mar. 17, 2021

*Correspondence Dr. Harsha Gorrela.

Department of Oral and Maxillofacial Surgery, MNR Dental College and Hospital, Hyderabad, Telangana, India.

Email: drgharshaomfs@gmail.com

single left submandibular lymph node of size measuring about 0.5cm×0.5cm in size was palpable, which was firm in consistency, mobile and non-tender. On intra oral examination a diffused swelling was noted in the left mandibular labial vestibule in relation to 31,32,33,34 and 35 regions (Fig -2).



Fig 1: Extra oral swelling in the submental region



Fig 2: Intraoral diffuse labial swelling.



Fig 3: OPG – radiolucent lesion in anterior region of mandible with root resorption.

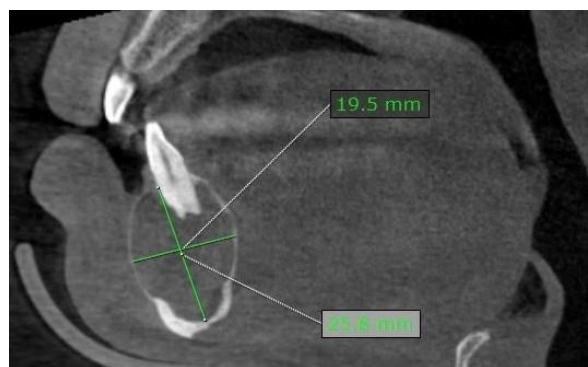


Fig 4: CT scan – Bucco lingual cortical plate expansion



Fig 5 : 3D CT – Buccal cortical plate perforation

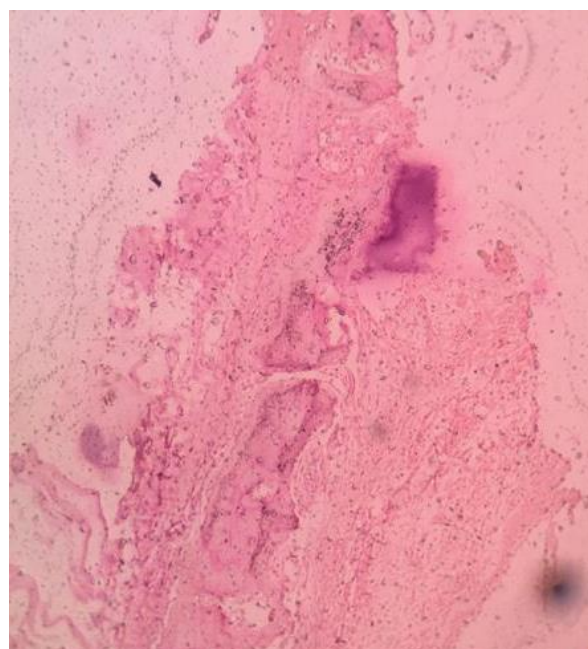


Fig 6 : HPE – features suggestive of cystic cavity with ameloblastomatous lining

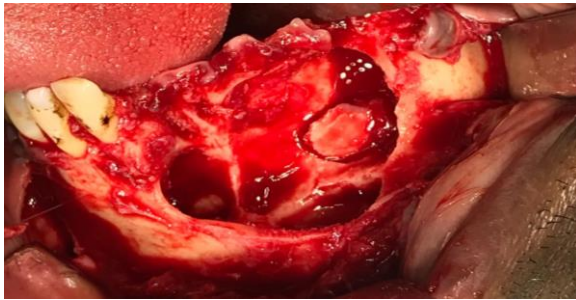


Fig 7: Cystic enucleation.



Fig 8: OPG 1 year follow up.

OPG revealed a well-circumscribed unilocular radiolucent lesion extending from the apical region of the right canine to left second premolar. Root resorption was observed in relation to 31 to 35 teeth (Fig- 3). CBCT scan revealed an expansile radiolucent lesion of size 2.6cm x2cm extending from 31tooth to 35 tooth region with perforation of the buccal cortical plate, the lingual cortical plate and the lower border of the mandible were intact (Fig- 4). 3D CT scan revealed buccal cortical plate perforation in relation to 31,32,41,42 teeth (Fig -5).

FNAC and incisional biopsy were done. Histopathological examination revealed a cyst lined by ameloblastic epithelium with a tall columnar basal layer, subnuclear vacuoles, reverse polarity of hyperchromatic nucleus, and a thin layer of edematous, degenerating stellate reticulum like cells on the surface, features which were suggestive of unicystic ameloblastoma (Fig - 6).

Enucleation of the cystic lesion along with peripheral osteotomy was performed under general anesthesia (Fig - 7). The entire cystic lining was excised and sent for histopathological examination which gave a confirmatory diagnosis of unicystic ameloblastoma. The patient was followed up for a

period of two years with no evidence of recurrence (Fig - 8).

DISCUSSION

Ameloblastoma is a benign, locally aggressive odontogenic neoplasm with variable clinical expression and accounts for 1% of all cysts/tumors of jaws and 18% of all odontogenic neoplasms. It is typically slow growing, locally aggressive and rarely metastasizes but has a high rate of recurrence (55–90%) if not removed adequately. As per the WHO system of 2003, ameloblastoma is classified based on differences in biologic behavior, treatment plan and recurrence rate as follows:

- (1) Classic solid/multicystic ameloblastoma,
- (2) Unicystic ameloblastoma,
- (3) Peripheral ameloblastoma,
- (4) Desmoplastic ameloblastoma, including the so-called hybrid lesions [3,7].

Unilocular ameloblastoma (UA) is a rare type of ameloblastoma, accounting for about 6% of ameloblastomas. It usually occurs in a younger age group of 16-20 years, with about 50% of the cases occurring in the second decade of life which correlates with the reported case. More than 90% are located in the mandible in the posterior region, followed by parasymphysis region [9].

There seems to be no significant sex predilection for unicystic ameloblastoma as documented in the available literature. Although el-abdin and ruprecht [10] found a higher female preponderance. Histologically, the minimum criteria for diagnosing a lesion as UA is the demonstration of a single cystic sac lined by odontogenic (ameloblastomatous) epithelium often seen only in focal areas. Ackermann et al in their landmark study classified the histological subtypes into three patterns, namely luminal (type 1), intraluminal (type 2) and mural patterns (type 3) [2,6].

The treatment of unicystic ameloblastoma can be radical or conservative [11-14]. The radical approaches can be achieved by resection of the lesion followed by insertion of the reconstructive plates [11,13,14]. Resection may cause possible deformities in the face and results in impairment of mastication, swallowing, speech and facial asymmetry [11,12,14]. Conservative treatment consists of enucleation, enucleation followed by

application of carnoy's solution and marsupialization followed by enucleation [11-14].

Nakamura, et al. compared the long-term results between different approaches to 78 patients with ameloblastomas [12]. They found that conservative treatments including marsupialization and enucleation followed by sufficient bone curettage were useful and reduced the need for jaw resection.

Lau and Samman reported that the recurrence rates for unicystic ameloblastomas were 3.6% after resection, 30.5% after enucleation alone, 16% after enucleation followed by application of carnoy's solution, and 18% after marsupialization with or without further treatment [11,14].

H.P. Philipsen, P.A. Reichart suggested that the definitive diagnosis of a cystic ameloblastoma is only possible after the whole tumor tissue is examined. (Isacson et al., 1986; Pogrel and Montes, (2009). It is thus difficult to decide preoperatively if enucleation is enough for a cystic ameloblastoma or not [3].

Stoelinga et al in their study on incidence, multiple presentation and recurrence of aggressive cysts of the jaws reported that resection was preferred earlier, but it is associated with anatomic and functional morbidity. The advantage of carnoy's solution and peripheral osteotomy is that it eliminates epithelial remnants and satellite cysts [15].

As our case was diagnosed as unicystic ameloblastoma, we decided to perform enucleation followed by peripheral osteotomy under general anesthesia (Fig- 9). Enucleation with peripheral osteotomy seems to have advantages over other conservative methods in treating large mandibular cystic ameloblastomas [16]. Advantage of enucleation with peripheral is that the inferior border of the mandible is conserved and integrity of inferior alveolar nerve is also preserved. This procedure also avoids an extraoral incision taking esthetics into consideration. In our case the patient was treated under general anesthesia via a transoral approach, teeth in continuity with the lesion were extracted. Enucleation of the lesion was done and Curettage of the grossly uninvolved bony

bed was performed with a large acrylic bur. Inferior mandibular border continuity was maintained.

Shi S et al, proposed that, by using Enucleation /Peripheral osteotomy, the contour and functions of the patients were well preserved. Firstly, the mandible continuity was maintained after enucleation followed by peripheral osteotomy. Removal of the infiltrative tumor islands in the residual bone walls is critical to reduce recurrence. The authors have thus used enucleation combined with peripheral osteotomy for the management of large mandibular cystic ameloblastomas in fourteen patients [16].

Ademola Abayomi Olaitan et al. reviewed 315 cases of ameloblastoma in which they suggested that after enucleation, resection with 1 cm of apparently normal bone will be adequate for mandibular lesions, inclusion of about 1.5 cm to 2 cm of apparently normal bone may probably be required in maxilla to eradicate recurrences [17].

CONCLUSION

The diagnosis of unicystic ameloblastoma was based on clinical, clinicopathologic and radiographic features. Unicystic ameloblastoma is a tumor with a strong propensity for recurrence, especially when the ameloblastic focus penetrates the adjacent tissue from the wall of the cyst. The ability to predict this potential occurrence prior to surgery would greatly enhance therapeutic strategies for reducing the incidence. Hence, the Pathologist should examine the tissue sections carefully in an attempt to determine whether ameloblastoma has penetrated the wall of the cyst or not so that the complications can be minimized. As the variant in the above case report is a mural type of ameloblastoma and more than 1cm of healthy inferior border of the mandible was present, conservative management has been planned. The enucleation combined with peripheral osteotomy method can preserve the function and esthetics well, and at the same time, thorough trimming off the residual tumor islands in the bone walls, keep the recurrence rate acceptably low. It also has advantages over other common conservative methods. This enucleation with peripheral osteotomy method is an effective and reliable option for the management of large mandibular cystic ameloblastoma, particularly the primary

tumor and in adolescent patients. However long term follow up is necessary to detect any recurrences.

CONFLICTS OF INTEREST

The authors declare they have no potential conflict of interests regarding this article.

REFERENCES

1. Kahairi A, Ahmad RL, Wan Islah L, Norra H: Management of large mandibular ameloblastoma - a case report and literature reviews. Archives of Orofacial Sciences. 2008; 3(2):52-55.
2. Handa H, Bailoor DN, Naidu G, Shrivastava K, Raghuvanshi V Unicystic Ameloblastoma of mandible . Aggressive treatment A myth or a need. Case report and Extensive review of literature Volume 12, Issue 2 (Nov.- Dec. 2013), PP 26-31.
3. Philipsen HP, Reichart PA: Classification of odontogenic tumors and allied lesions. Odontogenic tumors and allied lesions Quintessence Pub. Co. Ltd 2004. 21-3.]
4. Unicystic ameloblastoma a variant of ameloblastoma first described by robinson and Martinez in 1997 (quintessence publication.co.ltd.london , 20014:pp 77-86
5. H. P. Philipsen and P. A. Reichart, "Unicystic ameloblastoma. A review of 193 cases from the literature," *Oral Oncology*, vol. 34, no. 5, pp. 317-325, 1998)
- 6 . Chana, Jagdeep S, Yang-Ming Chang, Wei , Fu-Chan , Shen , Yu-Fen , Chan Chiu-Po, Lin Hsiu-Na, Tsai Chi-Ying, Jeng Seng-Feng: Segmental mandibulectomy and immediate free fibula osteoseptocutaneous flap reconstruction with endosteal implants: An ideal treatment method for mandibular ameloblastoma. Plast Reconstr Surg 2004; 113(1):80-87].
- 7.V. Nagalaxmi, Mithare Sangmesh, Kotya Naik Maloth, Srikanth Kodangal,Vani Chappidi, and Stuti Goyal Unicystic Mural Ameloblastoma: An Unusual Case Report. Volume 2013, Article ID 957418,
- 8.Pizer ME, Page DG,Svirsky JA: Thirteen-year follow-up of large recurrent unicystic ameloblastoma of the mandible in a 15-year old boy. J Oral Maxillofac Surg 2002; 60:211-5.),
- 9.. K.R.K.Kumar,G.B.George,S.Padiyath,and S.Rupak,"Mural unicystic ameloblastoma crossing the midline: a rare case report, "International Journal of Odontostomatology,vol.6,no. 1, pp. 97-103, 2012.
- 10.Unicystic ameloblastoma in the Sudan H. El-Abdin and A. Ruprecht: Unicystic ameloblastoma in the Sudan. Int. J. Oral Maxillofac. Surg. 1989; 18: 64-67
- 11.Enucleation of Ameloblastoma - Case Report of 16-Year-Old Male Anusha A Hennedige*, Dimitrios Doumptiotis and Syed Ahad
12. Lau SL, Samman N. Recurrence related to treatment modalities of unicystic ameloblastoma: a systematic review. Int J Oral Maxillofac Surg 2006;35:681-90.
13. Seintou A, Martinelli-Kläy CP, Lombardi T. Unicystic ameloblastoma in children: systematic review of clinicopathological features and treatment outcomes. Int J Oral Maxillofac Surg 2014;43:405-12.
14. Kim J, Nam E, Yoon S. Conservative management (marsupialization) of unicystic ameloblastoma: literature review and a case report. Maxillofac Plast Reconstr Surg. 2017;39(1):38. Published 2017 Dec 25. doi:10.1186/s40902-017-0134-0.
15. Stoelinga PJW, Bronkhorst FB. The incidence, multiple presentation and recurrence of aggressive cysts of the jaws. J Craniomaxillofac Surg 1988; 16: 184-95.
16. Shi S, Liu Y, Shan Y, Fu T, Zhao S, Enucleation combined with peripheral ostectomy: its role in the management of large cystic ameloblastomas of the mandible, Journal of Cranio-Maxillofacial Surgery (2014); 42 (8): 1659-63
17. Ademola Abayomi Olaitan, Davis Sunday Adeola, Emmanuel Oladepo Adakeye, Ameloblastoma : clinical features and management of 315 cases from Kaduna, Nigeria, Journal of Cranio Maxillofacial surgery 1993;21(8): 351- 355.