WORLD JOURNAL OF PHARMACEUTICAL RESEARCH
SJIF Impact Factor 7.523
Volume 6, Issue 15, 547-555. Case Report ISSN 2277– 7105

UNICYSTIC AMELOBLASTOMA

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Article Received on
23 Sept. 2017,
Revised on 14 Oct. 2017,
Accepted on 05 Nov. 2017
DOI: 10.20959/wjpr201715-9840

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ABSTRACT

Unicystic ameloblastoma is a unique type accounting for 5% to 22% of all ameloblastomas. It is usually seen in females during the second – third decade, with a predilection for posterior mandible. Unusual occurrence of ameloblastoma during pregnancy has been discussed in the literature, with only 3 reported cases till date. The first case was reported in 1957. Here we present a case report of a 31 year old female who was diagnosed with unicystic ameloblastoma on right mandibular anterior region, within 1 month post partum, along with a review of the related literature.

KEYWORDS: unicystic ameloblastoma, pregnancy, odontogenic tumors.

INTRODUCTION

Cysts in the maxillofacial region are fairly common entities. Development of a tumor similar to a cyst in the maxillofacial region is uncommon. Also, such occurrences associated with pregnancy is considered to be rare and poses a challenge to the clinician in the management. Case reports of ameloblastoma occurring during pregnancy are still scarce in the literature. We report a case of mural variant of unicystic ameloblastoma that occurred in the right anterior mandible of a 31-year-old female 55 days post-partum.

CASE REPORT

A 31 year old female patient presented to the Department of Oral Medicine and Maxillofacial Radiology with a swelling in the lower right front tooth region since 1 month. The swelling was gradual in onset and attained the present size with associated pain. H/o mild mobility and displacement of the lower right front teeth was elicited. There was no h/o trauma, bleeding or pus discharge.

Medical history revealed that she was hypothyroid and had h/o caesarean delivery 1 month back.

On examination, a solitary 3*4cm swelling was present in the right lower face region extending supero-inferiorly from lower lip to the lower border of the mandible and antero-posteriorly from 1.5 cm posterior to the mental region to a line drawn from the commissure of the lip to the base of the mandible. Overlying skin was normal in colour. Swelling was tender and firm on palpation.

Intraoral examination revealed a 5*4 cm swelling in relation to right mandibular anterior region, extending antero-posteriorly from 41 to 44 region and supero-inferiorly from 1mm below the gingival margin to the lower vestibular region, with evident vestibular obliteration. The overlying mucosa was erythematous and tender. Buccally, the swelling was firm in consistency while, lingually, the area was soft in consistency.(figure 1(a) and (b)) On electric pulp testing, 41 and 42 were non-vital and 43 showed a delayed response. Aspiration revealed a straw coloured fluid (figure 2). After aspiration, subsequent reduction in the size of the swelling was noted. Based on the above findings, a provisional diagnosis of radicular cyst was given.

OPG showed a well-defined pear-shaped radiolucency on right mandibular anterior region extending anteroposteriorly from 31 to 43 and superoinferiorly from alveolar crest to 1 cm below the apices of teeth in its greatest dimension with smooth curved margins with displacement of 42 and 43. (figure 3) CBCT examination (figure 4A) showed perforation of buccal and lingual cortical plates and teeth were displaced distally and mesially, respectively. Considering the size of the lesion and the perforation of both the cortical plates, an incisional biopsy was performed and sent for histopathological examination, which was conclusive of unicystic ameloblastoma with mural proliferation.(Figure 5). The lesion was marsupialized. Marked reduction in size of the lesion was noted after 8 months and the area was resected along with placement of a bone graft harvested from the symphysis region. (Figure 4 B,C,D).

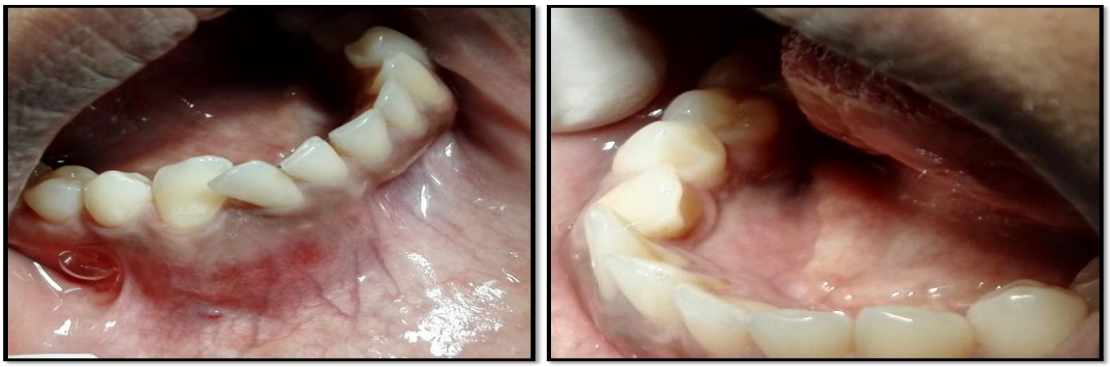


Fig. 1: Intraoral swelling at the right anterior mandibular region between 42 and 43, with obliteration of buccal and lingual vestibule and displacement of the teeth.

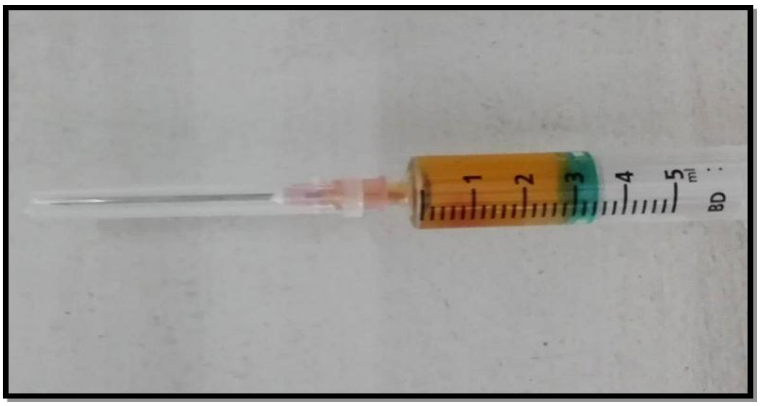


Fig. 2: Fine needle aspiration showing straw-coloured aspirate.

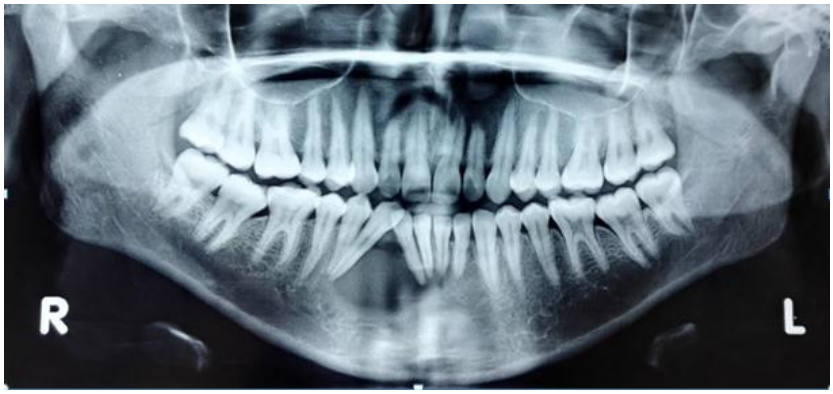
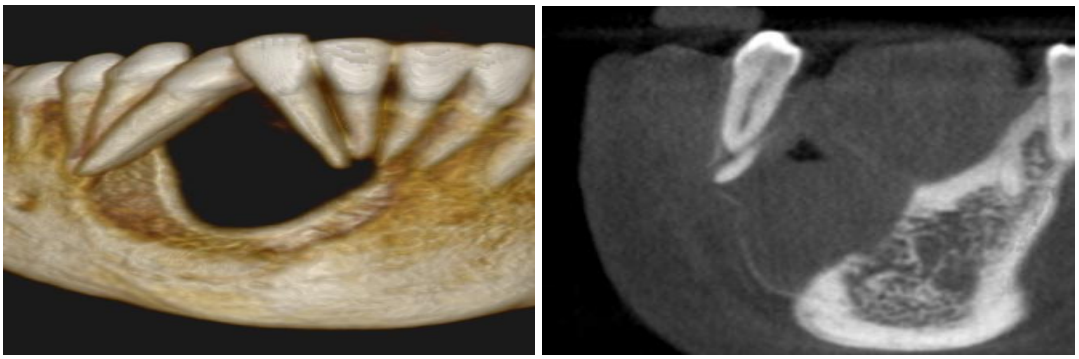


Fig. 3: OPG showing a well-defined unilocular radiolucency between 42 and 43 region with displacement of adjacent teeth.

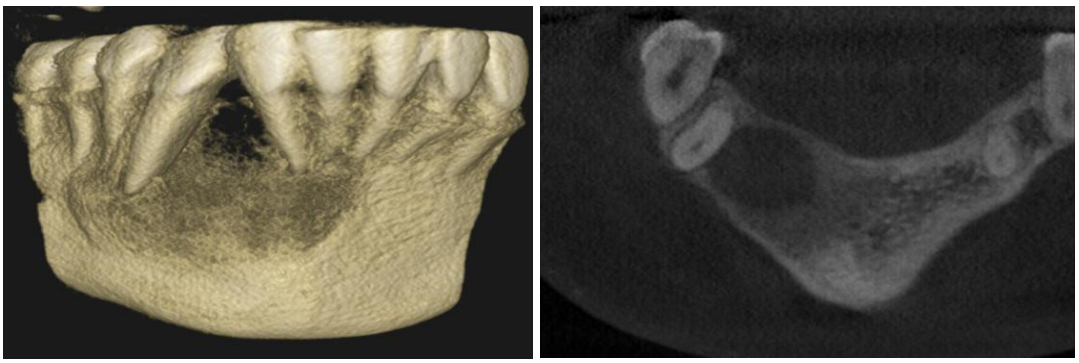
4. A



(a) (b)

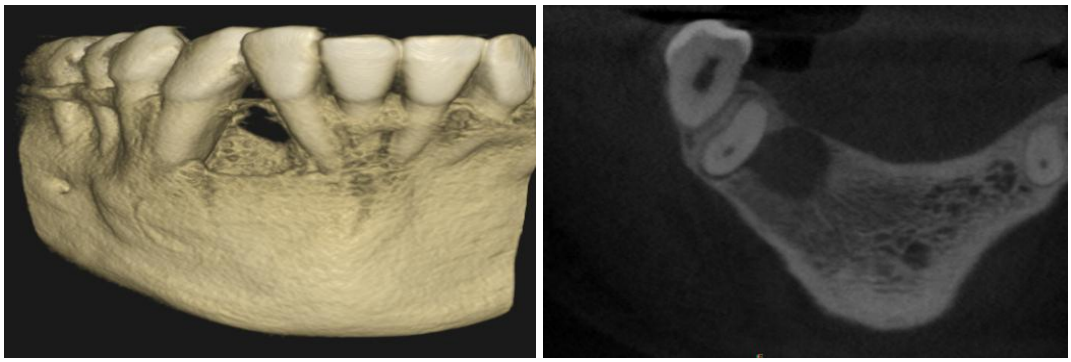
Fig. 4 A: Pre-operative CBCT : (a) 3D reconstruction showing perforation of buccal and lingual cortical plates; (b) Coronal section showing thinning of buccal and lingual cortical plates.

4. B.



(a) (b)

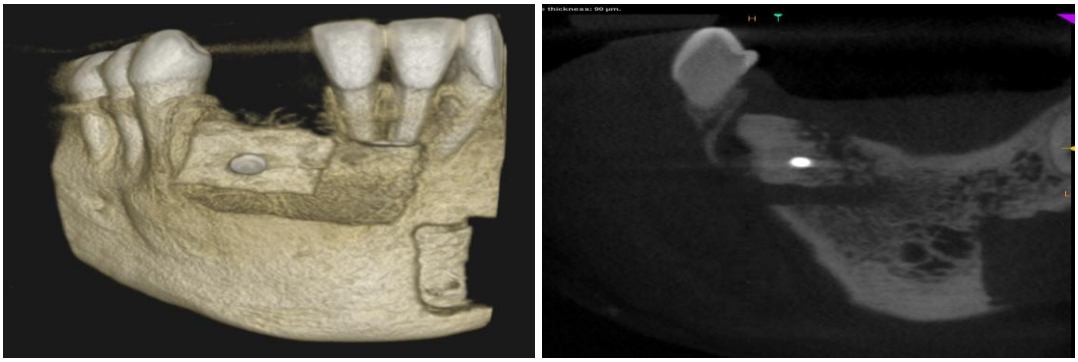
4. C.



(a) (b)

Fig. 4 B, C: (a) 3D reconstruction and (b) coronal section showing bone formation in relation to buccal, lingual and inferior portion of the cystic cavity leading to subsequent reduction in the size of the lesion.

4. D.



(a) (b)

Fig. 4 D: Post-operative CBCT – (a) 3D reconstruction showing bone graft fixed with screw in relation to 42, 43 region.

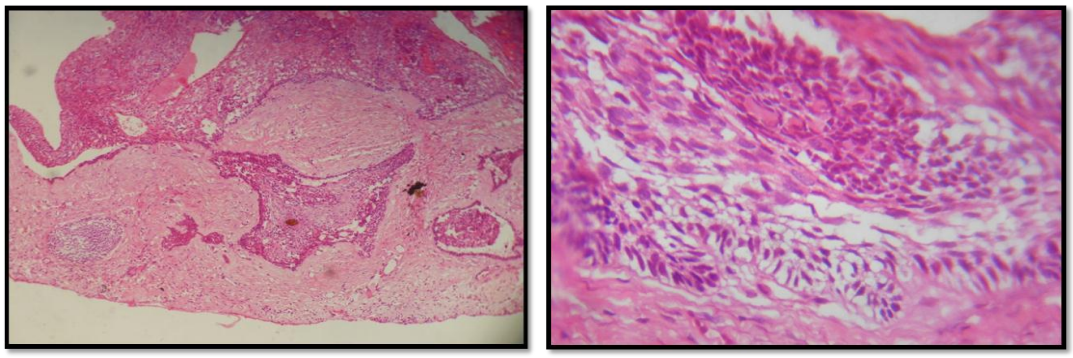


Fig. 5: Histopathological specimen showing Odontogenic islands in the underlying connective tissue capsule.

DISCUSSION

Unicystic ameloblastoma was first described by Robinson and Martinez in 1977. It refers to those cystic lesions that show clinical, radiographic, or gross features of a mandibular cyst, but on histological examination show a typical ameloblastomatous epithelium lining part of the cyst cavity, with or without luminal and/or mural tumour growth. It is characterized by slow growth and is relatively a localized aggressive cystic lesion.[1] It was considered as a distinct subtype of ameloblastoma about 4 decades ago, based on clinical and radiological features as well as distinct histopathological features.[2]

Leider et al.[3], proposed three pathogenic mechanisms for this cystic entity

- Ameloblastic transformation of the reduced enamel epithelium, associated with developing tooth, which later undergoes cystic changes.
- Ameloblastomas may arise in dentigerous or other types of odontogenic cysts in which neoplastic ameloblastic epithelium is preceded temporarily by non-neoplastic stratified squamous epithelial lining.
- Cystic degeneration occurs in a solid ameloblastoma, with subsequent fusion of multiple microcysts and develops into a unicystic lesion.

The reason why some ameloblastomas become completely cystic may be related to epithelial dysadhesion (e.g. defective desmosomes) or the intrinsic production of proteinases (e.g. metalloproteinases, serine proteinases).

Eversole et al.[4] in their series of 31 cases reported the typical demographics of unicystic ameloblastoma. There are basically 2 variants of UCA: associated with an unerupted tooth (dentigerous variant) and the one associated with an erupted tooth (non-dentigerous variant). The present case is of non- dentigerous variant as it is not associated with an unerupted tooth. The characteristics and their similarity to the present scenario are tabulated below.

NON-DENTIGEROUS VARIANT[4]

Characteristics	Usual presentation	Present case
Age distribution	35.2 years	31 years
Gender Distribution	Male: female = 1:1.8	Female
Aspirate	Straw-coloured	Straw-coloured
Location	Maxilla:mandible relationship: 1:4.7 Molar-ramus region- 77% Premolar region- 10%	Symphysis-parasymphysis region

	Symphysis – 13%	
Radiographic features	U/L: M/L = 1.1:1	Unilocular appearance
	Pear shaped radiolucency interposed between contiguous teeth, causing divergence of the roots.	Similar presentation

Patients usually present with swelling and facial asymmetry, pain being an occasional presenting symptom.[5] Cases with similar presentation were previously reported in the literature.[6]

What makes this case report unique is the development of unicystic ameloblastoma within 1 month of post partum.

Earlier three cases of ameloblastoma during pregnancy have been reported in the literature.[7],[8],[9] Studies show conflicting results on the development of odontogenic tumors during pregnancy. Pregnancy hormones promote development, growth, and birth of the newborn. Herberts and Sandstrom et al[7] showed that these hormones can influence the growth and development of tumors. This concept was supported by Gordy et al.[8] who suggested that hormonal action modulates the lesions during pregnancy, promoting rapid growth of the ameloblastoma.

A case of keratocystic odontogenic tumor (KCOT) in a pregnant woman was reported by Kornafel O.et al[10]. Similarly Sekiya et al.[10] reported a case of adenomatoid odontogenic tumor (AOT) in which the tumor cell survival was accomplished by Bcl-2 upon continuous stimulation of estrogen during pregnancy. This favoured the fact that hormones released during pregnancy are probably able to influence growth and development of odontogenic tumors directly or indirectly. This was contradicted by Bhandari and Kothan et al[11] who revealed no such dependence of estrogen or progesterone for the growth of AOT.

An inflammatory mechanism has been proposed by Omoregie et al.[12] in the aetiology of unicystic ameloblastoma arising from neoplastic transformation of non-neoplastic epithelial lining of odontogenic cyst such as radicular cyst, dentigerous cyst and odontogenic keratocyst. They observed that such transformations were common in females and reinforces the hypothesis that hormonal factors may play a significant role in their occurrence.

The less aggressive behaviour, better prognosis and reduced recurrence rates of unicystic ameloblastoma compared to solid and multicystic ameloblastomas, which is locally invasive

and highly destructive, tend to favour a conservative management such as enucleation, curettage, and marsupialization.

Treatment modalities are dictated by the size, anatomical location, histologic variant, and anatomical involvement. The present case was managed with marsupialization. The rationale for marsupialization is to reduce the size of the lesion, to enable ease of total removal and avoid morbidity of the patient.[13] Conventionally, choosing the treatment is based on the exact histology of the UCA. In cases of the intraluminal or plexiform patterns with no penetration of the fibrous tissue capsule by the ameloblastic cells, enucleation generally suffices. But if there is a mural component that extends into the wall to the level of the interface with the bone, bony resection is necessary to ensure adequate removal. Follow up for atleast 10 years is recommended as the recurrence interval is approximately 7 years.[14] The recurrence rate is decreased (18%) when marsupialization is applied prior to curettage.[15]

CONCLUSION

This case report emphasizes the need for the strict maintenance of oral health and monitoring of patients during pregnancy. The concept regarding the possible influence of pregnancy hormones on the growth and development of tumors, particularly ameloblastoma, is still scarce in the literature. Hence, further studies should be conducted to explore and unveil the truth regarding the incidental appearance of odontogenic tumors in pregnancy.

KEY POINTS

- Currently, according to update from the 4th edition of the 2017 WHO classification of Head and Neck Tumours, the mural variant of unicystic ameloblastoma is recognized as having the same biological aggressiveness as conventional ameloblastoma.
- Clinically, UA is characterized by perforation of buccal and lingual cortical plates unlike radicular cyst (which was considered as a provisional diagnosis), where lingual cortical plate perforation occurs rarely.
- Radiographically, it appears as a pear shaped radiolucency interposed between contiguous teeth, causing divergence of the roots.
- Care must be taken in pregnancy to rule out etiological factors which could predispose towards inflammation within the oral cavity and future studies are required to elucidate the possible role of hormones in the development of odontogenic tumours.

REFERENCES

1. H. P. Philipsen and P. A. Reichart. Unicystic ameloblastoma. A review of 193 cases from the literature. Oral Oncology, 1998; 34(5): 317–325.
2. Wright J M, Vered M. Update from the 4th edition of the World Health Organization Classification of Head and Neck Tumors: Odontogenic and Maxillofacial Bone Tumors. Head and Neck Pathol. February 2017.
3. Leider AS, Eversole LR, Barkin ME. Cystic Ameloblastoma: A clinicopathological analysis. Oral Surg Oral Med Oral Pathol, 1985; 60: 624-30.
4. Langland.O.E, Langlais.R.P.Diagnostic Imaging of the jaws. Williams & Wilkins. 1995.
5. Handa H, Bailoor DN, Naidu G, Shrivastava K, Raghuvanshi V. Unicystic Ameloblastoma of mandible. Aggresive treatment A myth or a need. Case report and Extensive review of literature. IOSR-JDMS, 2013; 12(2): 26-31.
6. Savithri V, Janardhanan M, Rakesh S. Unicystic Ameloblastoma as a Differential Diagnosis for Odontogenic Cysts. Oral Maxillofac Pathol J., 2014; 5(1): 466-469.
7. Herberts G, Sandstrom J. Ameloblastoma occurring and recurring during pregnancies. Acta Otolaryngol, 1957; 48: 327–32.
8. Gordy FM, Holder R, O'Carroll MK, Krolls SO. Growth of an ameloblastoma during pregnancy: opportunity lost? Spec Care Dentist, 1996; 16: 199–203.
9. da Silva.H, Costa. E.S.R. Ameloblastoma during pregnancy: a case report. Journal of Medical Case Reports, 2016; 10(1): 244.
10. Kornafel.O, Jaźwiec.P, Pakulski.K. Giant Keratocystic Odontogenic Tumor of the Mandible – A Case Report. Pol J Radiol, 2014; 79: 498-501.
11. Bhandari N, Kothan M. Adenomatoid odontogenic tumour mimicking a periapical cyst in pregnant woman. Singapore Dent J., 2010; 31(1): 26-29.
12. Omoregie.F.O, Sede.A, Ojo.A.M. Ameloblastomatous change in radicular cyst of the jaw in a nigerian population. Ghana medical journal. June 2015; 49(2): 107-111.
13. Dolanmaz. D, Etoz. O. A, Pampu. A, Kalayci. A, Gunhan. O. Marsupialization of unicystic ameloblastoma: A conservative approach for aggressive odontogenic tumors. Indian Journal of Dental Research, 2011; 22(5): 709-712.
14. Philipsen HP, Reichart PA et al: Odontogenic tumors and allied lesions.London, Quintessence publishing, 2004.
15. Lau SL, Samman N. Recurrence related to treatment modalities of unicsytic ameloblastoma: a systematic review. Int J Oral Maxillofac Surg, 2006; 35(8): 681-90.