

Conservative Management of a Huge Multilocular Unicystic Ameloblastoma: A Case Report

Mithileswer Kadiyala^{1*} Swetha Karipineni² Srikanth Gunturu³ Vijayalakshmi Uppuluru⁴
Koteswara Rao Nadella⁵ Neelima Kadiyala⁶

¹MDS, Department of Oral and Maxillofacial surgery, Drs.S & NR Siddhartha Institute of Dental Sciences, Vijayawada, India.

²MDS, Department of Oral and Maxillofacial surgery, Drs.S & NR Siddhartha Institute of Dental Sciences, Vijayawada, India.

³MDS, Department of Oral and Maxillofacial surgery, Drs.S & NR Siddhartha Institute of Dental Sciences, Vijayawada, India.

⁴MDS, Department of Oral and Maxillofacial surgery, Drs.S & NR Siddhartha Institute of Dental Sciences, Vijayawada, India.

⁵MDS, Department of Oral and Maxillofacial surgery, Drs.S & NR Siddhartha Institute of Dental Sciences, Vijayawada, India.

⁶MDS, Consultant Orthodontist, SS Dental and Implant Centre, Visakhapatnam, India.

ABSTRACT

Background: Unicystic ameloblastoma is a rare subtype of Ameloblastoma commonly affecting younger age group. It affects mostly mandible than maxilla. Usually it is unilocular without any bony septa in the lesion. There was considerable debate in the management of huge ameloblastomas. Some authors support resection of tumor while others considered managing it in conservative ways. In this article we present a case report of huge multilocular unicystic ameloblastoma of a young boy who was managed with enucleation with no evidence of recurrence.

Keywords: Multilocular, Unicystic, Ameloblastoma, conservative management, enucleation.

INTRODUCTION

Among all the odontogenic tumours classified, Ameloblastoma is the most common one encountered in Clinical scenario. It accounts for almost 11% of all the odontogenic tumors and 1% of all tumors of the maxilla and mandible [1, 2, 3]. Clinical variants of ameloblastoma are divided into Solid [or multicystic], Unicystic, and Peripheral types. The first two varieties are intraosseous while the third one is a soft tissue counterpart [4]. Ameloblastomas are well known for their local aggressive nature and tendency to recur. Prime treatment of any ameloblastoma is surgical whether to remove conservatively or aggressively [5]. Literature shows there is considerable debate regarding the most appropriate treatment method for surgical removal of the tumour. These include conservative approaches like enucleation, curettage and cryosurgery [6, 7, 8] or radical approaches like marginal resection, segmental resection or composite resection. [9,10,11]. Solid variants of

ameloblastoma are locally invasive and tend to recur if not adequately excised. Whereas the unicystic variant which was first described by Robinson and Martinez is a less aggressive one and many authors reported that it can be conservatively managed. [12,13]. We report a case of large multilocular unicystic ameloblastoma of mandible treated conservatively.

CASE REPORT

A 16 year old male patient had reported to the dental clinic with a chief complaint of swelling in relation to left side of the lower jaw since 8 months. History of presenting illness revealed swelling had started 8 months back as a small one which increased in size slowly and reached the current size. On examination the swelling was large and bony hard, non-tender extending from corner of the mouth to the posterior mandible. Swelling was seen lingually also causing bicortical expansion. An orthopantomogram (OPG) was advised. OPG (Fig.1)

Received: Oct. 17, 2018; Accepted: Nov. 2, 2018

*Correspondence Dr. Mithileswer Kadiyala.

Department of Oral and Maxillofacial surgery, Drs.S & NR Siddhartha Institute of Dental Sciences, Vijayawada, India.

Email: mithileswerkadiyala@gmail.com

revealed a large multilocular lesion extending from left canine to second molar region. An impacted third molar was present distal to the second molar. Radiolucency was observed just mesial to the third molar. There was resorption of roots of premolar and molar teeth. The swelling was aspirated and clear yellow straw colored liquid observed. The lesion was suspected as a cystic lesion with tendency to expand the cortex suggestive of Dentigerous cyst. But since the roots are also resorbed, we again had a dilemma of an odontogenic tumor like Ameloblastoma. Since it is a cystic lesion we then included Unicystic ameloblastoma in our list of differential diagnosis. An incisional biopsy was taken in most prominent area of swelling. The Histopathological examination report confirmed it as Unicystic ameloblastoma-luminal variety. A CT scan was advised to reveal the extent of lesion three dimensionally and it showed a massive lesion causing erosion of cortical bone but maintaining adequate lower border thickness (Fig 2,3). Review of literature regarding unicystic ameloblastoma suggested enucleation for young and adolescent patients with minimum recurrence rate, thus causing minimum aesthetic defects caused by resection and reconstruction.

Patient consent for the enucleation of the lesion under general anesthesia was taken after explaining the patient that a second surgery might require if lesion recurs. Patient was more willing to take a chance for thorough enucleation.

Under general anesthesia, mucoperiosteal flap was elevated carefully exposing the expanded cortices on both buccal and lingual side from left canine to distal to second molar tooth. All the affected teeth from canine to second molar were extracted. Later the lining of the lesion was slowly separated from the bone using a curette. Thin weakened bone was removed along with the lining. The lining was not fragile and was thick and easily separated from the healthy bone. Impacted third molar tooth was not found attached with the lining but still was removed to maintain the surgical margin of enucleation. Finally the entire lesion was separated from the soft tissue attachment and defect was irrigated with antibiotic solution and saline. Adequate amount of mandibular lower border was present which did not require to place a reconstruction plate (Fig.4).

Both buccal and lingual flaps were sutured with 3-0 vicryl.

Patient was instructed not to have hard food and was prescribed calcium supplements along with antibiotics. Patient had mild paresthesia in the lower lip and chin which subsided gradually. He was reviewed after one month, six months and twelve months postoperatively and bone formation was observed in the OPG taken (Fig 5 & 6).



Fig 1: Preoperative OPG



Fig 2: CT scan image 1



Fig 3: CT scan image 2



Fig 4: CT scan image 3



Fig 5: Immediate Postoperative OPG



Fig 6: Twelve months Postoperative OPG

DISCUSSION

Surgical management of ameloblastoma depends on several factors like location, size and extent of the lesion, age of the patient, histological variant of the lesion. For example, a small sized mandibular unicystic lesions in young patient might be treated with an enucleation whereas a large solid ameloblastoma affecting half of the mandible in an adult patient might be opted for resection. Many studies reported that there is considerable rate of recurrence in the lesions treated by conservative approach [5, 14, and 15].

A unicystic intraosseous ameloblastoma has distinct clinical, radiographic and histopathologic characteristics [16, 17].

There are three histopathologic variants of unicystic ameloblastoma (UA). The first type is the luminal

UA, where the tumour is confined to luminal surface of the cyst with fibrous wall. The second type is the intraluminal UA, with nodular extension of tumor into cystic cavity. Both these types don't tend to recur because of their confinement with in the cystic wall. But in the third variant, called mural UA, the fibrous wall of the cyst is breached by the tumor leading to recurrence if treated with the conservative means[18].

So a special caution is required while treating Solid multicystic ameloblastoma and mural unicystic ameloblastoma because both these variants show the tendency to recur. This doesn't mean that they should be radically resected. J. Haq et. al argued that it is not necessary to resect each and every multicystic variants and a simple conservative enucleation followed by sterilization of cystic cavity with Carnoy's solution promised reduced recurrence rate [19]. They also proposed that under proper surveillance even recurrent lesions can be eliminated by a second enucleation rather than resecting the jaw and causing facial deformity and morbidity to the patient who is relatively young [19].

In our case the Histopathological report was luminal variety of unicysticameloblastoma. Hence enucleation of the lesion was done even though the pathology seems to be aggressive in the CT scans. Adequate thickness of lower border of mandible was intact and hence no reconstruction plate was required. The patient was followed up for 1 year and did not show any recurrence. Dental rehabilitation on the involved side is to be planned using strategic implants on a later date.

CONCLUSIONS

A long term follow up of the patient is required to rule out any chance of recurrence of the lesion. Since the patient is young, we tried to maintain the natural contour of the jaw by removing only lesion in the conservative approach thus trying to eliminate the complications associated with reconstruction followed by resection of the jaw.

CONFLICT OF INTEREST

No potential conflict of interest relevant to this article was reported.

REFERENCES

1. Gorlin RJ, Chaudhry AP, Pindborg JJ: Odontogenic tumors: Classification, histopathology clinical behaviour in man and domesticated animals. *Cancer* 14:73, 1961
2. Adekeye EO: Ameloblastoma of jaws: A survey of 109 Nigerian patients. *J Oral Surg* 38:36, 1980
3. Adekeye EO, Lavery KM: Recurrent ameloblastoma of the maxillofacial region. Clinical features and treatment. *J MaxillofacSurg* 14:153, 1986
4. Gardner DG: A pathologist's approach to the treatment of ameloblastoma. *J Oral MaxillofacSurg* 42:161, 1984
5. Sehdev MK, Huvos AG, Strong EW, et al: Ameloblastoma of maxilla and mandible. *Cancer* 33:324, 1974.
6. Takahashi K, Miyauchi K, Sato K: Treatment of ameloblastoma in children. *Br J Oral MaxillofacSurg* 36:453, 1998
7. Nakamura N, Higuchi Y, Tashiro H, et al: Marsupialization of cystic ameloblastoma: A clinical and histopathologic study of the growth characteristics before and after marsupialization. *J MaxillofacSurg* 53:748, 1995
8. Shigeru U, Mushimoto K, Shirasu Re: Prognostic evaluation of ameloblastoma based on histologic and radiographic typing. *J MaxillofacSurg* 47:11, 1989
9. Olaitan AA, Adeola DS, Adekeye EO: Ameloblastoma: Clinical features and management of 315 cases from Kaduna, Nigeria. *J Cranio maxillofac Surg* 21:351, 1993
10. Gardner DG, Pecak AMJ: The treatment of ameloblastoma based on pathologic and anatomic principles. *Cancer* 46:2514, 1980
11. Molla MR, Shaheed I, Shrestha P: Ameloblastoma a clinical study of 13 cases. *Bangladesh Med Res Counc Bull* 17:29, 1991
12. Robinson L, Martinez MG: Unicysticameloblastoma: A prognostically distinct entity. *Cancer* 40:2278, 1977
13. Shteyer A, Lustman J, Lewis-Epstein J: The mural ameloblastoma: A review of literature. *J Oral Surg* 36:866, 1978
14. Shatkin S, Hoffmeister FS: Ameloblastoma: A rational approach to therapy. *Oral Surg* 20:421, 1965
15. Waldron CA: Ameloblastoma in perspective. *J Oral Surg* 24: 331, 1966
16. Economopoulou P, Sotiriadou S. An unusual tumor of the mandible with features of unicysticameloblastoma and ameloblastic fibroma. *J Oral MaxillofacSurg* 1998;56:1196-200.
17. Philipsen HP, Reichart PA. Unicysticameloblastoma. A review of 193 cases from the literature. *Oral Oncol* 1998;34:317-25.
18. Leider AS, Eversole LR, Barrin ME. Cystic ameloblastoma: a clinicopathological analysis. *Oral Surg Oral Med Oral Pathol* 1985;60:624-30.
19. J. Haq et al, Argument for the conservative management of mandibular ameloblastomas. *Br J Oral Maxillofac Surg* 2016;54:1001-05.