

## Case Report

# Conservative treatment of a unicystic ameloblastoma by marsupialization with a favorable response: A case report and review of the literature

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## ABSTRACT

Ameloblastoma is a benign tumor of odontogenic epithelium which is slow growing, locally invasive, expansive that may result with asymmetries of the face. Conservative treatment of odontogenic tumors with marsupialization is not common but can be done successfully in those with cystic pattern. The aim of this case and literature review is to propose marsupialization as an initial procedure for its treatment. We have presented a case of an ameloblastoma involving 56-year-old male patient. Orthopantomography and computed tomography showed the lesion extended between the right molar region and left molar region of mandible with both buccal and lingual expansion and root resorptions. On clinical examination, expansions of mandible and slight tooth mobilities were seen. An incisional biopsy was carried out and histopathologic examination revealed ameloblastoma although clinical features of lesion were more suggestive of cyst. Our case was treated by obturator marsupialization with a good healing. This case and review of the literature discusses the available treatment option according to macroscopic features of jaw lesions and emphasizes on the importance of the most conservative treatment option. Conservative treatment preserves integrity of bones while radical treatment can leave major cosmetic and functional sequelae such as pathologic fractures, complication with reconstruction plates.

**Key Words:** Ameloblastoma, jaw cysts, jaw neoplasms, odontogenic tumors

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## INTRODUCTION

Ameloblastoma is a benign tumor of odontogenic epithelium which is slow growing, locally invasive, expansive. The major clinical forms of intrabony ameloblastoma are unicystic, multicystic, and solid types. The peripheral ameloblastoma is the visibly extraosseous soft-tissue form of the lesion found in the gingiva and mucosa of the alveolar process.<sup>[1]</sup> Numerous methods of treating this tumor have been

proposed in the literature. Sampson and Pogrel published an algorithm for the surgical management of ameloblastoma which reported favorable results with resection and unacceptable recurrence rates following curettage.<sup>[2]</sup>

Unicystic ameloblastoma (UA) was first described by Robinson and Martinez in 1977 as a special type of ameloblastoma.<sup>[3]</sup> Although currently, UA is classified

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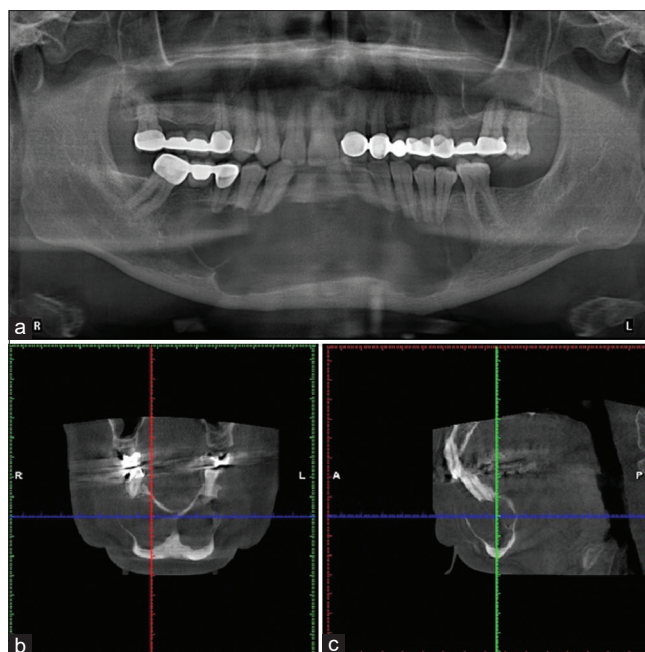
as a type of ameloblastoma by the World Health Organization, it is a distinct entity since it has less aggressive behavior compared with conventional ameloblastoma.

Various treatment modalities for UA have been used, such as segmental resection as normally used for conventional ameloblastoma; however, enucleation and marsupialization are other alternatives for treatment. Histopathologic variant is prognostic factor for UA beside the treatment option. According to literature resection of UA provides the lowest recurrence rate (3.6%) comparing with enucleation and marsupialization. Despite a high success rate for resection, conservative treatments to optimize quality of life are generally favored. Enucleation yielded the highest recurrence rate (30.5%), while marsupialization results with 18% recurrence rate.<sup>[4]</sup> We have presented a case of an UA which was successfully treated with marsupialization with a favorable response and reviewed the cases who were treated with marsupialization or decompression.

Review of articles published since 1995 using Medline and MeSH term “unicystic ameloblastoma” in combination with “decompression” and “marsupialization” were searched. Articles presenting cases and populations of patients with UAB treated by marsupialization or decompression were identified and included. Only articles in English language were included.

## CASE REPORT

A 56-year-old otherwise healthy male patient came to clinic with a swelling on his chin. The patient complained of a slow growing swelling in the region of corpus and symphysis mandible with a slight sensory disturbance. On clinical examination expansions on buccal mandible and slight tooth mobilities especially on left incisors were seen. Panoramic view of the patient revealed a well-defined radiolucent area extending from right molar region to the left molar region of mandible with root resorbtions [Figure 1a]. Computed tomography showed both buccal and lingual expansions and root resorbtions [Figure 1b and c]. The involved teeth gave late positive vitality test result, as evaluated by the electrometric vitality test. Patient consent form has been obtained. Before insicional biopsy, aspiration from the cystic cavity was carried on. Cholesterin crystals were seen in cavity. The lesion was suspected



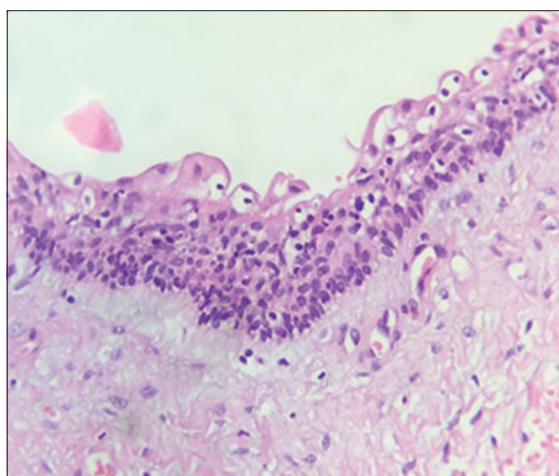
**Figure 1:** Panoramic radiograph of patient before marsupialization, showing a well-defined radiolucent area (a) coronal computed tomography (b) and sagittal tomography (c) showing weakened mandible and buccal and lingual expansions before treatment.

to be keratocyst since the teeth were vital and content of lesion was cystic fluid. Following advised consent from the patient about the surgical plan; left central and lateral teeth which have advanced mobility were extracted and through the extraction socket incisional biopsy was carried out under local anesthesia. It was noted that lesion had a cystic cavity in mandible with a thin cystic wall. Cystic lesion opening was sutured to obtain a marsupialization hole. Furthermore, an acrylic obturator was planned to keep the lesion uncovered and to avoid food impaction. Dental impression was taken with silicone and a casts were fabricated. Customized Hawley like acrylic plate was fabricated and applied to patient mouth on seventh day. Until the acrylic obturator was applied to mouth, marsupialization hole was preserved with daily exchanged gauze covered with topical antibiotic pomade. After application of acrylic obturator to mouth, the patient was instructed to maintain overall proper hygiene of the oral cavity through self-irrigation of cavity every day. The patient was scheduled for hygiene of cyst cavity follow-up weekly management. Histopathologic evaluation of lesion revealed luminal type unicystic ameloblastoma. Reverse polarization in basal layer and neoplastic epithelium overlaid with squamous epithelium

without intraluminal or extraluminal infiltration indicates luminal type UA [Figure 2]. The patient was scheduled for radiographic follow-up after an interval of 3 months. The lesion gave a good response and shrank with marsupialization [Figure 3]. Post 24 months of marsupialization, the diminished lesion was completely enucleated with peripheral osteotomy to ensure complete removal of margins. Lesion wall was thickened following 2 years' decompression comparing with thin wall at the beginning [Figure 4]. Postoperative healing was uneventful. There was no sign of recurrence with a follow-up of 42 months bone healing has been obtained in defect of lesion. In present case, stafne bone defect was diagnosed in the left angulus region with the aid of panoramic radiograph and three-dimensional computed tomography at the beginning. The lesion has appeared similar during the follow-up period [Figures 1 and 3].

## DISCUSSION

UA has been considered a special type with unilocular radiographic appearance, macroscopic cystic nature and the better response to conservative treatment.<sup>[5]</sup> Based on the character and extent of tumor cell proliferation within the cyst wall, three histologic subtypes of UA are recognized, which include those of simple cystic nature, those with intraluminal proliferative nodules and those containing infiltrative tumor islands in cyst wall. While the first two groups of lesions named with luminal and intraluminal subtypes may be treated more conservatively, third group named with mural

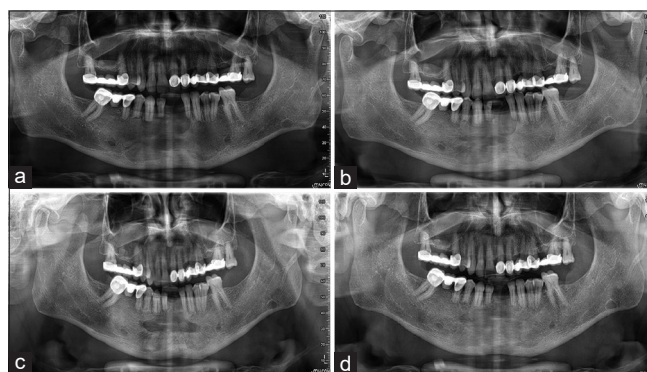


**Figure 2:** Reverse polarization in basal layer and neoplastic epithelium overlaid with squamous epithelium indicate unicystic ameloblastoma.

type is more prone to recurrence.<sup>[4]</sup> In our case, histologic examination revealed luminal type UA with no intraluminal proliferation or mural invasion.

Lee *et al.* analyzed 29 UA cases retrospectively and reported asymptomatic bony swelling of the jaws and unilocular radiolucency are the most common signs of lesion as found in our case.<sup>[6]</sup> A report published in 2004 by Abdelsayed *et al.* reported that a parathyroid hormone-related protein (PTHrP) was expressed by all lesions in a study of 30 unicystic and multicystic ameloblastomas. This local expression of PTHrP by tumor cells may readily provide a humoral cause of bone resorption and aggression by the tumor. The dense cortical bone of the lateral and medial borders, as well as the inferior border and symphysis of the mandible can be invaded and destroyed by the tumor following infiltration and resorption of cancellous bone.<sup>[7]</sup>

The treatment must be guided by consideration of the behavior and potential of the tumor, the growth characteristics of the various physical forms, the anatomic site of occurrence, the clinical extent and size of the tumor, and the histologic assessment of the specific lesion.<sup>[8]</sup> While solid and multicystic ameloblastomas are recommended to treat with resections even sacrificing inferior alveolar nerve and periosteum if they are involved, unicystic ameloblastomas could be treated with enucleation, resection or marsupialization. Extensive unicystic lesions should be biopsied in several sites in an attempt to obviate selection of nondiagnostic areas.<sup>[1]</sup> In our case, we have obtained biopsy twice to plan treatment accurately. Following the diagnosis of unicystic ameloblastoma with luminal type, lesion was decided to decompress to protect bone and teeth.



**Figure 3:** Panoramic view following 10 months' marsupialization (a) 24 months after marsupialization (b) after enucleation (c) no recurrence in 16 months after enucleation (d).



**Figure 4:** After marsupialization for removal of diminished lesion an incision was made along the marsupialization hole (a and b) lesion was enucleated and peripheral osteotomy was performed (c)

Nakamura *et al.* discussed the effectiveness of marsupialization for cystic ameloblastoma to reduce the tumor volume and minimize the extent of surgery. They suggested the possibility of conservative management of ameloblastomas, avoiding radical surgeries that are often associated with various complications such as removal of teeth, masticatory dysfunction, facial deformity and abnormal jaw movement, even after successful reconstruction.<sup>[9]</sup> Marsupialization is supported as a presurgical treatment for UA by many researchers.<sup>[10-19]</sup> Some studies that mainly investigate efficacy of marsupialization on cystic lesions supported marsupialization for UA.<sup>[20-22]</sup> Prasad *et al.* treated 5 cases with UAs with decompression in which decompression period range between 3 months and 24 months.<sup>[23]</sup> Even mural type ameloblastoma was successfully treated with decompression without recurrence up to three years.<sup>[24]</sup> Predominantly, acrylic devices were used for marsupialization while Liu *et al.* designed a vacuumed system to yield faster bone gain.<sup>[25]</sup> We have reviewed the literature to understand effectiveness of marsupialization on UA patients. There are 16 published papers that evaluated marsupialization treatment in UA patients [Table 1]. The follow-up period for UAs which were treated with marsupialization ranged between 8 months and 10 years. Nine mural type, 6 intraluminal type, and 4 luminal type UA were investigated (subtype of remaining cases was not mentioned) and 14 out of 72 cases (19%) showed recurrences up to about 5 years. Marsupialization of UA results with eruption of accompanying tooth while the lesions had been shrinking.<sup>[9,15,26]</sup> Some lesions resolve with marsupialization without need of any further surgery (Xavier, Kim). Furthermore, Gülsen *et al.* used decompression in a case with calcifying epithelial odontogenic tumor with a successful result.<sup>[27]</sup> Decompression

brings some benefits that are maintenance of pulp vitality, preservation of the inferior alveolar nerve, preservation of the mandibular contour and growth, prevention of fracture of the jaw, and low risk for recurrence.<sup>[28]</sup> In our case, we avoid further tooth loss and mandibular discontinuity. Remaining teeth whose roots were resorbed due to lesion, gave vital response after marsupialization. Since the lesion shrank and moved away from roots, we preserved the vitalities of remaining teeth.

Although there are many articles that support conservative treatments for UA, some researchers confront this idea.<sup>[29,30]</sup> Sampson and Pogrel reviewed the records of 26 patients referred for the management of mandibular ameloblastoma and reported that all patients treated with curettage alone developed recurrence while no recurrence was observed after resection.<sup>[2]</sup> Meshram *et al.* and Seintou *et al.* reviewed children with UA since UA have younger age predilection. Majority of their cases were treated with enucleation. Recurrences are reported to be related with enucleation treatment.<sup>[5,31]</sup> Li *et al.* reported clinicopathologic features of 33 UA cases who were predominantly treated with enucleation and showed recurrences in 4 years to 11 years period following enucleation. Second and third decades are peak time for diagnosis of UA.<sup>[32]</sup> Lee *et al.* supported enucleation of lesion and application of Carnoy solution which results with lower recurrence rate than enucleation alone. Furthermore, all recurrences involved type 3 lesions with mural invasion by ameloblastoma and presented between 1 and 4 years after initial treatment.<sup>[6]</sup>

Some studies claimed that the following marsupialization the tumor becomes more invasive and has a potential to infiltrate into the surrounding tissues.<sup>[9,33]</sup> Cuboidal cell type is an indicator for expansive pattern while columnar cell type shows

**Table 1: Review of published cases of unicystic ameloblastoma**

	Number of patients	Histologic type	Treatment modality	Follow up	Recurrence	Recurrence time
Nakamura <i>et al.</i> <sup>[9]</sup>	16	1 UA	1 marsupialization 4 marsupialization+enucleation	NM	9	NM
Furuki <i>et al.</i> <sup>[12]</sup>	3	1 UA (luminal) 1 UA (intraluminal) 1 UA (mural)	Marsupialization	NM	3	64 25 and 11 months
Matsumoto <i>et al.</i> <sup>[16]</sup>	1	1 UA	Marsupialization+enucleation	2.5 years	No	-
Liu <i>et al.</i> (2006) <sup>[25]</sup>	1	1 UA (mural)	Marsupialization (with a vacuumed intraoral device) + enucleation+curettage	22 months	No	-
Sharma <i>et al.</i> <sup>[18]</sup>	1	1 UA (luminal)	Marsupialization+enucleation	1 years	No	-
Dolanmaz <i>et al.</i> <sup>[10]</sup>	2	1 UA (luminal) 1 UA (mural)	Marsupialization+enucleation + curettage	30 months	No	-
Carneiro <i>et al.</i> <sup>[11]</sup>	1	1 UA with pathologic fracture	Marsupialization+enucleation + cryosurgery	3 years	No	-
Sano <i>et al.</i> <sup>[17]</sup>	1	1 UA (intraluminal)	Marsupialization+enucleation	5 years	No	-
Tomita <i>et al.</i> <sup>[15]</sup>	1	1 UA	Marsupialization+enucleation	10 years	No	-
Prasad <i>et al.</i> <sup>[23]</sup>	5	4 UA (mural) 1 UA (intraluminal)	1 decompression 4 decompression + enucleation	6.5 years-4.5 years-19 months-2 years-1.5 years	1	5.2 years
Saravanakumar <i>et al.</i> <sup>[14]</sup>	2	1 UA (Intraluminal) 1 UA (Mural)	decompression+enucleation + iliac graft	2 years	No	-
Xavier <i>et al.</i> <sup>[24]</sup>	1	1 UA (mural)	Decompression	3 years	No	-
de Paulo <i>et al.</i> <sup>[13]</sup>	1	1 UA (intraluminal)	Marsupialization+enucleation+ curettage	3 years	No	-
Meshram <i>et al.</i> (2017) <sup>[5]</sup>	2	1 UA (luminal) 1 UA (intraluminal)	Marsupialization + enucleation + curettage	4 years 3 years	No	-
Kim <i>et al.</i> <sup>[26]</sup>	1	1 UA	Decompression	49 months	No	-
Yang <i>et al.</i> <sup>[19]</sup>	33	33 UA	Marsupialization + enucleation	8 months to 7 years	1	In 2 years

UA: Unicystic ameloblastoma; NM: Not Mentioned

invasive pattern.<sup>[9]</sup> Nakamura *et al.* evaluated outcomes of marsupialization for 24 UA cases. Cuboidal cells transformed to columnar or basal cell types after marsupialization which may be related with change in the pattern of lesion after marsupialization.<sup>[9]</sup> Zhang *et al.* investigated success of marsupialization in 48 patients with UA. Patients associated with high TP53 and interleukin 1 $\alpha$  (IL-1 $\alpha$ ) expression predicts a better response to marsupialization than TP53- and IL-1 $\alpha$ -negative UA.<sup>[34]</sup>

Recurrence after marsupialization treatment has been reported in some cases.<sup>[5]</sup> Marsupialization was extremely effective in our case since the lesion almost disappeared. Recurrence was reported mainly in cases that respond less ideal to marsupialization. Recurrent cases were treated successfully by enucleation and bone curettage.<sup>[9]</sup> Recurrences were reported to be mostly related with mural type among UA subtypes.<sup>[6]</sup> Our case was diagnosed as luminal type UA which has the best prognosis. However, it is impossible to rule out mural invasion with one

incisional biopsy of the lining of a UA because of potential for taking a nonrepresentative tissue sample. Therefore, there is a dilemma whether the patient should receive further treatment to eliminate possible residual ameloblastoma tissue in surrounding cancellous bone or regularly observed with radiographs for early detection of possible recurrence.<sup>[6]</sup> Although our case was diagnosed as luminal type with the best prognosis, we have performed peripheral ostectomy after enucleation and the most important point is the patient is under our close follow-up period of marsupialization was suggested minimum 21 months in cases respond well to marsupialization.<sup>[9]</sup> In our case, we had run the marsupialization period of 24 months till the lesion nearly disappeared. Meshram *et al.*, after 14 and 20 months marsupialization, lesions were enucleated in young patients.<sup>[5]</sup>

Although younger patients have been reported to respond well to marsupialization while older ones not,<sup>[9,26]</sup> cystic ameloblastoma shrunked fastly in our 56-years-old case. Zheng *et al.* evaluated factors

affect the outcome of marsupialization in patients with UA. Resorption of root, histopathologic subtype, and perforation of cortical bone were the main factors that predicted the outcome.<sup>[34]</sup> Moreover, a long-term follow-up is important for conservative treatment of UA because more than half of recurrences occur within 5 years of the treatment.<sup>[35]</sup>

## CONCLUSION

Marsupialization is a useful procedure for UAs providing improved quality of life with maintenance of jaw continuity. It is emphasized that even if the tumor resembles an odontogenic cyst, it has a potential to proliferate after marsupialization. Consequently, clinical, radiographic follow-up should be performed periodically.

### Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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### Conflicts of interest

The authors of this manuscript declare that they have no conflicts of interest, real or perceived, financial or nonfinancial in this article.

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