Marsupialization of unicystic ameloblastoma: A conservative approach for aggressive odontogenic tumors

Dogan Dolanmaz, Osman A Etoz[1], Alper Pampu[2], Abdullah Kalayci, Omer Gunhan[3]

Department of Oral and Maxillofacial Surgery, Selcuk University, Konya, [1]Erciyes University, Kayseri, [2]Department of Oral and Maxillofacial Surgery, Karadeniz Techniqual University, Trabzon, [3]Department of Pathology, Ankara, Gülhane Military Medical Academy and Medical Faculty, Turkey

ABSTRACT

Unicystic ameloblastoma (UA) is known as a distinct entity which has a less aggressive behavior when compared with conventional ameloblastoma. In this report, we have presented two cases of UAs, (of which one case showed a more aggressive behavior with mural invasion into the adjacent tissues and granular cell differentiation), both of which were successfully managed with enucleation following marsupialization. We aim to highlight how this method can be used for the successful management of such cases, rather than following more aggressive approaches. In both the cases, marsupialization was done for the UA lesions initially and follow-ups were maintained. When the tumor size had regressed on radiographic follow up, an enucleation procedure with ostectomy of the margins was carried out. Special importance was also given to the endodontic treatment of the teeth involved in the area of the lesion. The patients were free of the condition and did not show any signs of recurrence on radiographic follow-ups even after 30 months of the final procedure. Granular variant of UA is quite rare and had been considered to be more aggressive. Marsupialization of UA is an alternative treatment option of resection even for more aggressive variants, as long as the histological behavior of the lesion was carefully evaluated and strict radiographic follow-up is maintained.

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Ameloblastoma is the most common odontogenic tumor of the jaws.[1] Although it has been accounted as a benign neoplasm, aggressive behavior, local invasion potential and tendency to recur make it difficult to manage.[1] There are several clinical variants of intra-osseous ameloblastomas, such as: solid, multicystic and unicystic. Unicystic ameloblastoma (UA) is a distinct entity which was first described by Robinson and Martinez in 1977.[2] Although it has a less aggressive behavior when compared with conventional ameloblastoma, it has a considerable recurrence rate of 3.6 to 30.5%.[3] However, prognosis of UA mostly depends on the treatment option as well as the histopathologic variant of UA. Ackermann classified UA into 3 subtypes, and briefly mentioning them: subtype 1- unilocular cystic luminal; subtype 2- intraluminal with a solid growth inside lumen of the cystic lesion,; and, subtype 3- intraluminal growth with mural invasion within adjacent tissues.[4]

Although various treatment modalities have been identified, the primary choice for multicystic or solid types' of ameloblastoma is the total removal of the lesion; while the unicystic variant can be successfully managed with either enucleation or marsupialization as long as a close follow-up is maintained.[5] We present 2 cases UAs, one of which had a solid granular growth inside the lesion with mural invasion, which were successfully treated with conservative enucleation following marsupialization.

Address for correspondence:
Dr. Osman A. Etoz,
E-mail: osmanetoz@yahoo.com

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CASE REPORTS

Case 1

A 17 year-old male patient was referred to the clinic with the chief complaint of a painless swelling in the right mandibular premolar region without any sign of sensory impairment. Panoramic view of the patient revealed a well defined radiolucent area extending from the right lateral

incisor to the distal root of the first molar tooth [Figure 1]. Under local anesthesia, an incisional biopsy was performed and the lesion was decompressed between two premolar teeth and left uncovered with the aid of an acrylic stent. Histopathological evaluation of the lesion revealed luminal type UA without any tumor cells within the cyst wall [Figure 2]. The patient was scheduled for radiographic follow-up after an interval of three months. Post 18 months of marsupialization, the diminished lesion was completely enucleated with peripheral ostectomy to ensure complete removal of the margins. The apical portions of the aforementioned teeth were resected and allogenic bone graft material was placed in the cavity. There were no signs of recurrence even at 30 months of follow-up [Figure 3].

Case 2

A 52 year old otherwise healthy edentulous woman came to our clinic with an asymptomatic swelling on her left mandible. The patient complained of a slow growing swelling in the region of the ramus of the left mandible, without any signs of sensory disturbance. A well-defined unilocular radiolucent area, occupying almost whole of the left mandibular ramus with an unerupted third molar was seen on the panoramic radiograph [Figure 4]. Simultaneous decompression of the lesion with incisional biopsy was carried out and an acrylic obturator was made to keep the lesion uncovered. A solid growth with a diameter of 1 cm which developed through the lumen of the cystic cavity was detected and removed. Histopathologic findings of the lesion revealed granular UA with mural invasion [Figure 5]. Post 18 months of marsupialization, the impacted tooth and the regressed lesion was enucleated, and peripheral ostectomy was performed. At 30 month follow-up, the lesion was completely healed without any sign of recurrence [Figure 6].

DISCUSSION

The rationale for marsupialization is reducing the size of the lesion to ease total removal. Lau and Samman reviewed treatment modalities for UA and reported that the highest recurrence rate (30.5%) was observed with single enucleation, while the lowest (3.6%) was observed with resection.[3] Accordingly, they also found that recurrence rate was decreased (18%) when marsupialization was applied prior to curettage. These findings were related to the results of Sampson and Pogrel,[1] in which smaller lesions express better response to conservative management. Both lesions in our report were first marsupialized, and after they regressed in size, they were enucleated.

In general, unilocular lesions of the jaws are treated conservatively, by following enucleation and curettage procedures, regardless of the microscopic features. However, the histological characteristic of UA should be considered while making the treatment choice.[3,4] Lesions with subtype 1 and subtype 2 histological patterns give better response to conservative treatments like marsupialization and enucleation, while more aggressive treatment options could be considered for subtype 3 lesions for UA.[5] Hence, it has been suggested that an incisional biopsy should be done to determine the histopathologic subtype, for a thorough management of UA.[3] On the other hand, it has been reported that definitive diagnosis cannot be made unless histologic examination of the entire specimen is performed, since unique epithelial characteristics of UA may or may not be seen in the different parts of the lesion.[5] Furuki *et al.*[5] reported three cases of UA recurrence after marsupialization. They mentioned that scalloping of sclerotic margin of the lesion is an early radiographic sign of recurrence; and concluded that the first and second histological variants of UA could be managed via conservative manner as marsupialization or enucleation, while the third variant of UA would need more radical approaches. However they did not mention about peripheral ostectomy during enucleation after marsupialization which might be a possible reason for recurrence. The first case in this report was luminal type UA, which might be considered as subtype 1 and it was successfully managed with enucleation following marsupialization. Nevertheless, according to Sampson and Pogrel, marsupialization is associated with a high tumour recurrence rate, since the tumor cells may be left within the adjacent cancellous bone.[1] In contrast with literature, an acceptable outcome was obtained in our second case which had mural invasion through adjacent bone and had intraluminal granular growth within the cyst lumen that was consistent with subtype 3. Granular type of ameloblastoma- in particular with unicystic variant, is rarely documented, and the existence of granular cells within ameloblastoma might be an indicator of aggressive behavior.[6] However, granular structures within UA seems to be a histological variant and does not affect prognosis.

Though the success rate of marsupialization mostly depends on the histological behavior of UA, outcome of marsupialization is affected by various factors, such as: age, technique of marsupialization, removal of solid growths during incisional biopsy, close radiographic follow-up and effectiveness of enucleation after marsupialization. In the present report, the first case was a good candidate for marsupialization because of the histological type of the lesion and younger age of the patient. In addition, one of the possible reasons of uneventful healing in the first case might be preoperative endodontic management and intraoperative apical resection of the teeth which had been associated with the lesion before enucleation. However, in the second case the outcome of the treatment was surprisingly favorable. This may be due to some factors as: (i) the solid granular growth inside the lumen of the lesion had been able to removed during incisional biopsy; (ii) histologic feature of the lesion was exactly determined and considered during enucleation procedure, and the lesion

Figure 1: Panoramic radiograph of the first patient before marsupialization, revealing a well defined radiolucent area extending from the right lateral incisor to the distal root of the first molar tooth

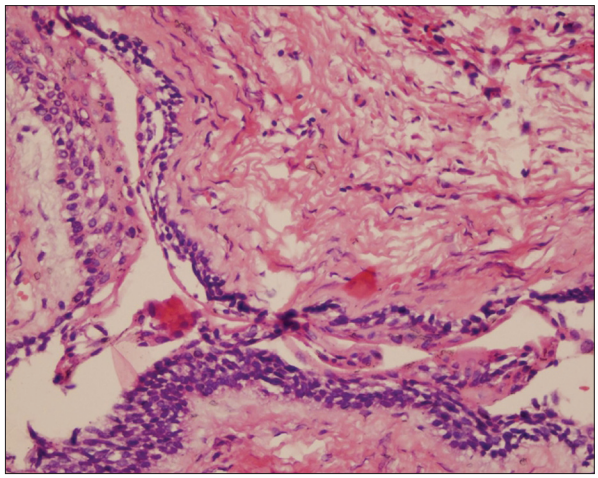

Figure 2: Loose squamoid (*stellate reticulum* like) cells above ameloblast-like cells within the cystic tumor of the first patient. No tumor cells were detected within the cyst wall upon histopathologic examination. (Hematoxylin and eosin, original magnification 100×)

Figure 3: Panoramic radiograph of the first patient 2 years post the complete enucleation procedure with peripheral ostectomy, with no signs of recurrence

Figure 4: Panoramic radiograph of the second patient before marsupialization, showing a well-defined unilocular radiolucent area, occupying almost whole of the left mandibular ramus with an unerupted third molar

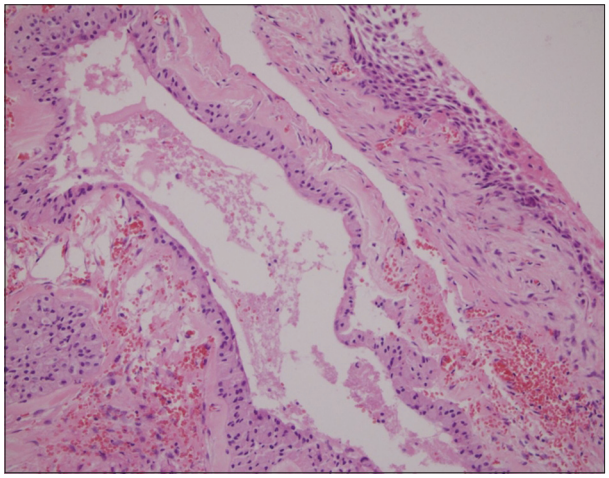

Figure 5: Unicystic ameloblastoma in the second patient that involved granular cell differentiation upon histopathologic examination (Hematoxylin and eosin, original magnification 100×)

Figure 6: Panoramic radiograph of the first patient 2 years post the complete enucleation procedure with peripheral ostectomy, with no signs of recurrence

was more aggressively curetted; (iii) a strict radiographic follow-up was maintained.

CONCLUSION

Unicystic variant of ameloblastoma with aggressive histologic behavior also might be successfully treated with marsupialization with subsequent enucleation, and this approach can be considered as an alternative to resection. However, shrinkage of the lesion may not be always symmetric, and the extent of the lesion should be carefully evaluated during the enucleation phase of the treatment. Clinicians should also perform a close radiographic follow-up and consider radical treatment options in case of suspicious radiographic changes during the marsupialization follow-up period. Removal of solid structures within the lesion during incisional biopsy, more aggressive enucleation with peripheral ostectomy, with accurate endodontic management of teeth in the area of the pre-existing lesion, help in improving the treatment outcome.

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