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Case Report

Conservative Management [Combination of Marsupialization and Enucleation] of Unicytic Ameloblastoma in the Posterior Mandible of a Young Patient: A Case Report 2-Year Follow Up with No Recurrent

Maryam Johari[1], Hamed Gharuee[2], Sahar Latifi[3]* and Seyedeh Fatemeh Masudi[4]

[1]*Assistant Professor, Department of Oral and Maxillofacial Radiology, Babol University of Medical Science, Babol, Iran*

[2]*Oral and Maxillofacial Surgeon, Babol, Iran*

[3]*Postgraduate Student, Department of Oral and Maxillofacial Radiology, Babol University of Medical Science, Babol, Iran*

[4]*Medical Pathologist, Amol, Iran*

***Corresponding Author:** Sahar Latifi, Postgraduate Student, Department of Oral and Maxillofacial Radiology, Babol University of Medical Science, Babol, Iran.

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Abstract

Background: During this study, we present a case of unicystic ameloblastoma (UA) treated by marsupialization taken after surgical enucleation as a conservative treatment. UA could be a locally invasive benign odontogenic tumor with an epithelial origin. UA could be a common variation of ameloblastoma that occurs frequently in young adults. The primary treatment modality of UA is usually radical surgery. Our treatment priority is conservative treatments to reduce the consequences of aggressive treatments such as loss of structures and functional disorders, and psychological complications. Due to the proximity of the tumor to the tooth buds in developing young patients, aggressive treatment shouldn't be considered an elective treatment modality. In this case, we consider a combination approach of marsupialization and conservative enucleation to reduce these side effects. This report described a 13-year-old boy's case of UA within the left mandibular molar–ramus region. The lesion was marsupialized, and two months after marsupialization, surgical enucleation was performed that show no recurrent after 2 years of follow-up.

Conclusions: Conservative treatments for UA in young patients have more advantages. The results of our study provide that enucleation protects mandibular development, keeping up more involved teeth instead of aggressive treatments.

Keywords: Unicystic Ameloblastoma; Enucleation; Marsupialization; Conservative Treatment

Introduction

There are many lesions of odontogenic and non-odontogenic origin that cause swelling of the mandible. One of the most common lesions with an odontogenic origin is ameloblastoma. [1,2] Ameloblastoma is a neoplasm that develops from the remnants of the dental lamina and odontogenic epithelium. It is the second most frequent odontogenic tumor after odontoma. [2] It approximately accounts for 1% of all oral cancers. [3] Ameloblastoma is most common in people in their third and fourth decades of life, and it affects both men and women equally. [3] Ameloblastomas are classified as multicystic, peripheral, desmoplastic, or unicystic ameloblastomas (UAs) by the World Health Organization (WHO) Classification of Head and Neck tumors (2005).[4] According to 2017 WHO classification, Ameloblastomas are three types: conventional ameloblastoma, Unicytic ameloblastoma, and extraosseous/peripheral types. [5,6] Unicystic ameloblastoma (UCA) is a term used to classify its different pathological forms. Sometimes, unicystic ameloblastoma appears as a multi-cavity radiolucency, and the term "cystic ameloblastoma" is used. [7,8] Unicystic ameloblastoma is less common, refers to cystic lesions that show clinical and radiographic features of an odontogenic cyst, but on histological examination is a typical ameloblastomatous epithelium lining the cyst cavity. No ductal and/or mural tumor proliferation [5]. When Ameloblastomas are small in size, these may be discovered during a routine radiography examination. The mucosa over the mass is normal. Most cases of ameloblastoma don't report pain or paresthesia. Teeth displacement and mobility may occur [9-12].

In most cases, radical surgical excision with a 0,2 cm safety margin is the maintenance treatment modality for solid ameloblastomas. Masticatory dysfunction, the disorder in mandibular movements and facial deformity are just a few of the side effects of radical surgery [13]. The goal of this study is to present a case of unicystic ameloblastoma that was treated by mandibular marsupialization, followed by surgical enucleation after 2 months, as an alternative treatment plan for UA.

Case Presentation

The ethical approval number of this case report is IR.MUBABOL.REC.1401.160. It was confirmed by the Research Ethics Committee of Babol University of Medical Sciences, Babol, IRAN.

On October 2, 2020, a 13-year-old child was admitted to our medical faculty with a primary complaint of swelling in his left jaw. According to the patient, the swelling of jaw was small at first and reached its current size within four months.

Antibiotics was prescribed for the patient to reduce swelling and pain two months ago by another dentist. He had no history of systemic diseases.

Extraoral examination revealed widespread swelling on the left side of the mandibular angle, extending mesiodistally from the nasolabial fold to the posterior border of the ramus, and extending superoinferiorly from the ala tragus line to the lower border of the mandible. The skin color was normal. The swelling was non-tender, non-fluctuating, firm, and bony hard on palpation. The intraoral examination revealed mixed dentition with mucosal edema extending from the 36 tooth to the retromolar region. The retromolar area was inflammated without paresthesia. On 3 October 2020, an orthopantomogram (OPG) was performed. It showed a well-defined multilocular radiolucent lesion with a scalloped corticated border on the left side of mandible that extends from the 35 teeth to the coronoid process and condylar neck (76.8 mm), expansion and thinning in the anterior border of ascending ramus and the lower border of the mandible, elevation of 35 and 36 teeth and root Resorption of 36 tooth was noted (Figure 1).

Figure 1: A panoramic view revealed a well-defined corticated multilocular radiolucent lesion with the size f (76,8mm length) in the posterior left mandible and ramus and severe expansion of the buccal and lingual cortices. Note mesial displacement and root resorptions of the first molar (36) and inferior displacement of the mandibular canal. A small and incomplete internal septum due to the endosteal scalloping of the cortical plate is also shown.

Then Cone-beam computed tomography (CBCT) scan was obtained and revealed a radiolucent bilocular well-defined borders lesion with incomplete coarse septa, severe centric expansion measuring about (54.90 x 32.76mm) that extended from the alveolar crest to the inferior border of mandible and mesiodistally extending from 35 tooth to posterior border of the ramus, thinning of the inferior mandibular cortical border and root resorptions of 36Tooth was seen (Figure 2).

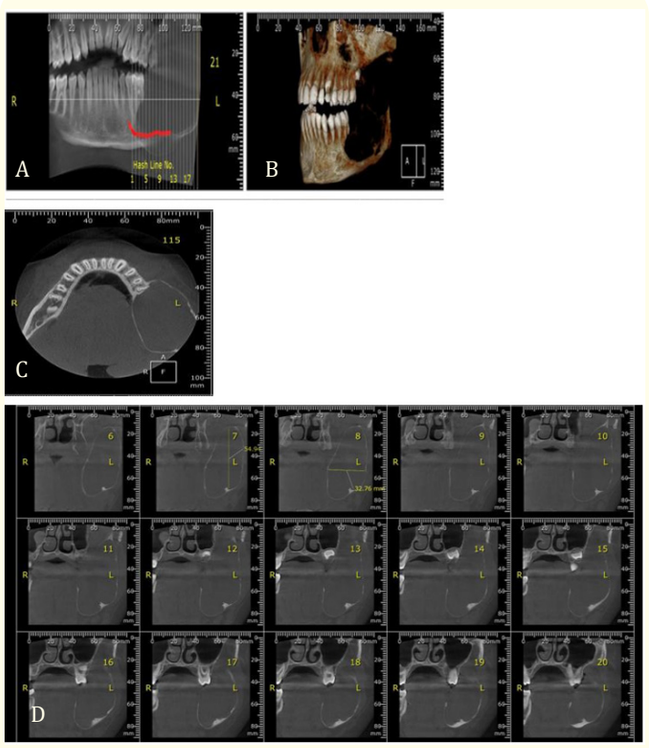


Figure 2:CBCT images A: panorama view with 10-mm slice thickness, B: 3D reconstruction showing destruction part of the mandible, C: Axial view, D: cross sectional view with 2-mm slice thickness revealed a bilocular radiolucent lesion that extended from the premolar region to the posterior border of ramus. Severe expansion and Thinning of the buccal and lingual cortical plates, root resorptions of 36Tooth was seen, Note an incomplete internal septum within the lesion, Perforation of the buccal and lingual cortex of the lesion is also shown.

Based on the clinical and radiography assessment ameloblastoma and Odontogenic keratocyst was considered differential diagnosis. Incisional biopsy Of the Lesion was performed under local anesthesia to confirm the Conclusive diagnosis. Histopathology examination showed neoplastic proliferation of ameloblastic epithelium including columnar basal cell in palisading arrangement with Small cytoplasm, hyperchromatic nuclei, polarized away from basement membrane with reverse polarization. Suprabasal cells show loosely textured and noncohesive with edema (Figure 3).

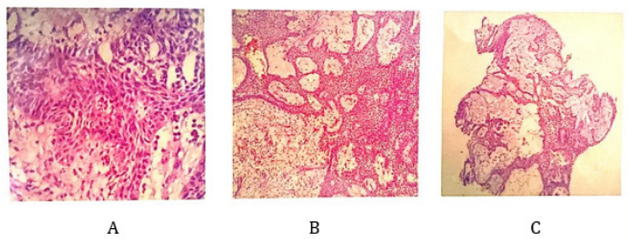


Figure 3: A: Micrograph of ameloblastoma showing the characteristic palisading and stellate reticulum. H and E stain H and E 40x B: Photomicrograph displaying features of the plexiform pattern of ameloblastoma, H and E 10x. C: Micrograph of ameloblastoma H and E 4x.

According to histopathologic examination, Ameloblastoma was considered the final diagnosis of the lesion. The parents were informed of the lesion, treatment plan, benefits and recurrent. After obtaining informed consent, the treatment procedure was started. The surgeon performed a Silastic drain to retain continuity between the lesion and oral cavity to reduce the lesion size by Marsupialization. In the first month of Marsupialization, weekly visits were planned for the patient and a panoramic radiograph was taken for follow-up that show the size of lesion didn't increase (Figure 4).

Figure 4: A panoramic radiograph 1 month after marsupialization displayed that the size of the lesion didn't increase.

2 months after Marsupialization, a panoramic radiograph was obtained that showed a slightly decreased in the lesion size (76.0 mm) (Figure 5,6).

A panoramic radiograph 3 months after marsupialization didn't show any change in the size of the lesion. So that 3 months after marsupialization, the drain was removed and Enucleation was performed under general anesthesia to completely remove the lesion with the 35 and 36 teeth. During the surgery, 5 ccs of autograft bone and 5 ccs of allograft bone were placed in equal proportions at the site of the lesion. The allograft bone contained spongy bone with a density of 1000-2000 μm and the autograft bone was obtained from the pelvic bone (Figure 7).

Figure 5: A panoramic radiograph 2 months after marsupialization with a Silastic drain showed a slight decrease in the lesion size.

Figure 6: A panoramic radiograph 3 months after marsupialization.

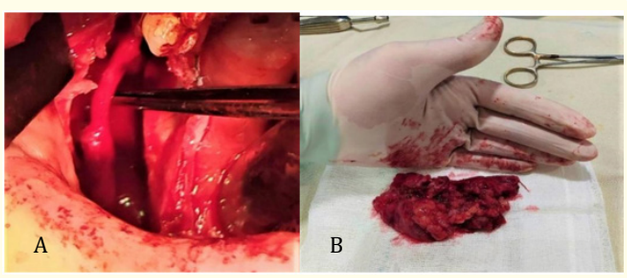


Figure 7: (A) The intraoral photography shows the inferior alveolar nerve in the peripheral osteotomy. (B). Macroscopic appearence of the tumor

On panoramic radiographs taken 3 months after Enucleation with Reconstruction of the mandible with the allograft combined with autograft (Figure 8).

3 months later, a panoramic radiograph was taken to follow the progress of the healing of the lesion. The panoramic 6-month Postoperative show the extent of the surgical scar cannot be detected, and the bone graft is replaced by a normal trabeculation pattern, the condylar and coronoid process have been reconstructed at well (Figure 9).

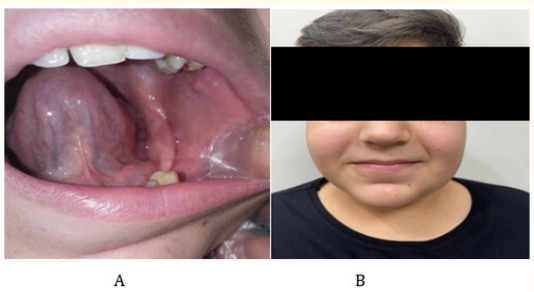
Figure 8: Postoperative panoramic radiograph 3 months after enucleation.

Figure 9: A panoramic radiograph 6-month Postoperative.

For follow, a panoramic radiograph was prepared 9 months after enucleation. the mandibular foramen and infra alveolar nerve canal can be seen clearly, condylar and coronoid processes have been reconstructed completely, cortical border of the alveolar crest and anterior border of the ramus is partially formed (Figure 10).

Figure 10: A panoramic radiograph 9 months after enucleation of the left side of mandible revealed a reconstructed at well.

Then a longer follow-up every 6 months was considered for the patient. 15 months Postoperative panoramic radiographs depicted: in the left side of mandible normal bone trabecular appearance, formation of the cortical border of alveolar crest, anterior wall of ramus and inferior Cortical border of mandible. The inferior alveolar nerve canal can be detected clearly (Figure 11-12).



A B

Figure 11: Postoperative photograph. (A) Intraoral photography, (B) Extraoral photography 15 months after enucleation.

Figure 12: Postoperative panoramic radiograph 15 months after enucleation.

The current follow-up radiography of the patient was obtained 2years after enucleation that showed the reconstructed bone with a normal bone trabecular appearance and No signs of recurrence (Figure 13).

Figure 13: Postoperative panoramic radiograph 2 years after enucleation.

Discussion

Ameloblastoma and odontogenic keratocyst, two different tumoral lesions of the mandible, have similar radiological and clinical appearances that are indistinguishable on initial evaluation. Although ameloblastoma is characterized by an expansion and larger size than the other compared lesions. It is fundamental to perform complementary examinations such as aspiration puncture and incisional biopsy to reach the final diagnosis [14].

Radical Surgery is the most common treatment approach for ameloblastoma, although whether to use conservative or radical procedures depends on the tumor type and Clinical presentation. The recurrence rate, mortality, morbidity, functional recovery, aesthetics of the patient, and the quality of life following treatment, are all factors to consider in deciding on the best treatment approach [15].

Qiao X., *et al.* 2021 performed a Systematic Review and Meta-Analysis to evaluate the recurrence rate of intraosseous ameloblastoma in patients treated with conservative and aggressive treatment approaches. The results revealed the recurrence rate for aggressive treatment (0.12) was significantly lower than for conservative treatment (0.30) [16].

Hendra., *et al.* 2019 performed a systematic review and meta-analysis to assess the recurrence rates of conservative treatment and radical surgery of solid or multicystic and unicystic ameloblastoma that revealed solid/multicystic ameloblastoma has more recurrences rate than unicystic type in both conservative treatment and radical surgery. The risk of recurrence in both types of ameloblastoma following radical Treatment was less than in conservative Treatment [17].

c. c. yew., *et al*. 2017 in a cross-sectional study of mandibular ameloblastomas treated by modified conservative management comprising surgical enucleation, peripheral osteotomy, and chemical cauterization and their outcomes Evaluated from 2006 until 2019, 23.2% recurrences were reported for the SMA group and no recurrences were observed for the UA cases [18].

Mitsu Meshram., *et al*. in 2017 in A case series of fifteen patients with unicystic ameloblastoma that were treated by various conservative methods such as marsupialization, enucleation with bone curettage over the last 6 years (2010-2015) and a methodical literature review of unicystic ameloblastoma treatment in young patients (<20 years) over the past 15 years (2001-2015).

The study revealed faster osseous regeneration after conservative treatment and owing growth potential, no clinical or radiographic evidence of recurrence and apparent deformity was reported in any of the cases in 4 years of follow-up [19].

Simon., *et al*. 2013 compared 32 patients who were treated radically for ameloblastoma and had immediate reconstruction during that surgery operation. The 32 patients were treated for the same tumor but did not have any subsequent reconstruction with an average follow-up time of 27.9 months. Patients who underwent reconstruction showed a higher quality of life compared to the group that did not get reconstructive surgery. In respect of solid food intake, aesthetics, and speaking, the results were significant [20].

S. Kalaiselvan., *et al*. in an evidence-based study in 2016, Evaluated the amount of safety margin necessary around the ameloblastic lesion by surgical, radiological, and histopathological methods in 25 cases of mandibular ameloblastoma. The results revealed the monocytic type had an infiltration rate of up to 0.2 cm, Acanthomatous ameloblastoma with 0.5 cm infiltration, and follicular and plexiform variants 0.75 cm. A regular monthly follow-up was performed for a period of at least 6 months up to a maximum of 12 months and then every 6 months for a period of 10 years. Therefore, that solid ameloblastoma is recommended and should be treated by segmental resection with a distance of 1.5 cm on both sides of the lesion [21].

Radical surgery reduces the chances of recurrence. However, it is almost certain to result in a significant bone defect, as well as severe facial deformities, malocclusion, lip paralysis, and impaired mastication. It may also have a negative influence on teenage patients' face development and psychological health [11-22].

Ameloblastoma is essentially a slow-growing benign tumor that is not life-threatening, minimizing recurrence at the expense of aesthetics and function does not seem reasonable. The treatment plan for large mandibular cystic ameloblastoma should be noted in postoperative quality of life, especially in young patients. Simple enucleation fails to the reduction of infiltrative islands of tumors in the bone borders, resulting in a high recurrence rate of up to 60% in unicystic cases and 60-80% in multicystic cases [11].

A better strategy for treating a large mandibular unicystic ameloblastoma should be considered to improve postoperative quality of life while also reducing the chance of recurrence, especially in young patients. The combination of conservative treatment and surgical modality was used in a teenage boy with large mandibular ameloblastoma. A follow-up regime for recurrent evaluation, according to the treatment plan employed, should be considered for at least a period of 15 years According to previous studies.

Conclusion

Unicystic ameloblastoma usually occurs in the young age group. It is very important to treat conservatively.

This case report presents successful treatment of pediatric unicystic ameloblastoma in a conservative manner involving marsupialization and enucleation to preserve of tooth buds and growing structures and function of structures while reducing loss of structures and psychological effects with no recurrent.

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