

CONSERVATIVE TREATMENT OF A MURAL-UNICYSTIC AMELOBLASTOMA IN CHILD PATIENT

Didem Sakaryalı, DDS, PhD

Asistant Professor, Department of Pediatric Dentistry,
Faculty of Dentistry, Baskent University,
Ankara, Turkey
ORCID: 0000-0001-7850-2375

Alev Alacam, DDS, PhD

Professor, Department of Pediatric Dentistry,
Faculty of Dentistry, Gazi University,
Ankara, Turkey
ORCID: 0000-0001-5328-8173

Gizem Dimililer, DDS, PhD

Specialist, Department of Oral and Maxillofacial Surgen,
Dentroyal Dentistry Clinic,
Çatalkoy, Kyrenia
ORCID: 0000-0001-5773-7917

ABSTRACT

Ameloblastoma is a benign tumor of odontogenic epithelium which could rarely be seen in childhood. This report describes a mural unicystic ameloblastoma case which was treated with marsupialization in a 12-years- old male patient. During six-years follow-up period there was no recurrence and the second molar eruption was completed. Conservative treatment options such as marsupialization might be preferred in children due to the continuing facial growth and development as a treatment option for ameloblastoma cases.

Correspondence**Didem Sakaryalı DDS, PhD**

Department of Pediatric Dentistry, Faculty of Dentistry,
Baskent University,
82th street, No:26, Bahcelievler, Cankaya, 06490, Ankara, Turkey
ORCID: 0000-0001-7850-2375
Phone: +90 312 203 0000 -1676
Fax: +90 312 246 6615
E-mail: dsakaryali@baskent.edu.tr

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ÇOCUK HASTADA MURAL-UNİKİSTİK AMELOBLASTOMANIN KONSERVATİF TEDAVİSİ

Didem Sakaryalı

Dr. Öğr. Üyesi, Baskent Üniversitesi, Diş Hekimliği Fakültesi,
Pedodonti Ana Bilim Dalı,
Ankara, Türkiye
ORCID: 0000-0001-7850-2375

Alev Alaçam

Prof. Dr., Gazi Üniversitesi, Diş Hekimliği Fakültesi,
Pedodonti Ana Bilim Dalı,
Ankara, Türkiye
ORCID: 0000-0001-5328-8173

Gizem Dimililer

Dr., Ağız Diş Çene Hastalıkları Cerrahisi Anabilim Dalı,
Dentroyal Diş Hekimliği Kliniği Çatalkoy,
Girne , Kıbrıs
ORCID: 0000-0001-5773-7917

ÖZ

Ameloblastoma, çocukluk çağında nadiren görülebilen iyi huylu odontojenik bir tümördür. Bu vaka raporunda, 12 yaşında bir erkek hastada marsupializasyon ile tedavi edilen bir mural-unikistik tip ameloblastoma olgusu sunulmuştur. Altı yıllık takip süresi boyunca ameloblastomanın tekrarlamadığı ve daimi ikinci molar dişin erüpsiyonunun tamamlandığı gözlemlenmiştir. Sonuç olarak, çocuklarda halen devam eden yüz büyümesi ve gelişmesi nedeniyle ameloblastoma gibi durumlarda marsupializasyon gibi konservatif tedavi seçenekleri tercih edilebilmektedir.

Sorumlu Yazar

Didem Sakaryalı

Baskent Üniversitesi, Diş Hekimliği Fakültesi,
Pedodonti Ana Bilim Dalı,
82. cadde, no: 26, Bahçelievler, Çankaya, 06490, Ankara, Türkiye
ORCID: 0000-0001-7850-2375
Telefon: +90 312 203 0000 -1676
Faks: +90 312 246 6615
E-mail: dsakaryali@baskent.edu.tr

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INTRODUCTION

Ameloblastoma is a benign oral tumor of odontogenic origin that has histological similarities to developing enamel.¹ The clinicopathologic types of ameloblastomas are multicystic, unicystic (with luminal, intraluminal and mural subtypes), peripheral and desmoplastic. Multicystic ameloblastomas are the most common (86%), followed by unicystic ameloblastomas (13%).²⁻⁵ There are three known pathogenic mechanisms related to the evolution of unicystic ameloblastomas: reduced enamel epithelium, dentigerous cysts and the cystic degeneration of a solid ameloblastoma. Ameloblastomas can also be classified into five histopathological subtypes: follicular, plexiform, granular, acanthomatous and cellular desmoplastic.^{2,6}

Ameloblastomas are the most frequently occurring type of odontogenic tumor, representing 1% of all oral tumors and 18% of all odontogenic tumors.⁷ They are usually diagnosed between the fourth and fifth decades of life; however, unicystic ameloblastomas have been diagnosed between the ages of 20 and 30 years, with no gender predilection.² In a review of 3,677 ameloblastoma cases, the average patient age was 36 years old, and the cases were uncommon in children.^{8,9} Keszler et al.¹⁰ and Saxena et al.¹¹ have also demonstrated that ameloblastomas are rare in young people. Ord et al.⁸ reported that the percentage of unicystic ameloblastomas in children under 10 years of age was only 2.2%. Saxena et al.¹¹ found that the reported number of pediatric jaw tumors over a six-year period was much lower than the total number of adult jaw tumors. However, in their survey, more ameloblastomas were reported in the odontogenic tumor samples.

The ratio of ameloblastoma incidence for the mandible compared to the maxilla is five-to-one. Unicystic ameloblastomas are frequently seen in the mandible, but there are a limited number of maxilla reports.^{8,9} Radiographically, 50% of ameloblastomas appear as multilocular radiolucent lesions with sharp delineations. Frequently, ameloblastomas are localized in the anterior region of the maxilla, especially in the canine and molar regions or in the mandible's ascending ramus.³ It is important to emphasize that ameloblastomas have an invasive nature and may lead to local malignancies.² About 10–15% of these tumors are associated with an unerupted tooth, and the cases are primarily diagnosed incidentally after radiographic examination following the detection of asymptomatic swelling during a routine clinical examination.

As symptoms persist, ameloblastoma patients often suffer from a painless, slow-growing swelling. However, pain, paresthesia, tooth loss and malocclusion can be symptoms of the growing pathology.^{1,11} Radiographical examination can reveal a round, unilocular or multilocular cyst. Ameloblastomas typically have a radiolucent appearance, and their shape resembles a focused honeycomb or a soap bubble. In occlusal radiographs, the swelling of both the lingual and the vestibular bone walls can be observed.¹

Central jaw tumors in children occur infrequently. They are often initially misdiagnosed as infections and improperly treated with antibiotics. In these cases, it is important to obtain a differential diagnosis that considers the various aspects of tumor and tumor-like lesions of the jaws.

Ameloblastomas can be treated by conservative or radical treatment modalities. Conservative techniques include curettage, enucleation, a combination of both enucleation and curettage, a combination of enucleation and Carnoy's solution, marsupialization, marsupialization followed by enucleation or curettage, and cryosurgery. Radical treatment options include enucleation and peripheral ostectomy, enucleation and resection, marginal resection, continuous or segmental resection, resection and cryosurgery, full-thickness resection of the mandible, hemimandibulectomy and segmental resection.^{1,4,12,13} Radical surgical procedures generally have been preferred for large lesions, but these procedures may result in the destruction of crucial anatomical structures, often leading to high morbidity and difficult rehabilitation.⁴ Chemotherapy and radiation are generally contraindicated.⁹ Due to the high recurrence rate, the current treatment modalities include extensive surgical excision.

Unicystic ameloblastomas may recur after a long delay, and long-term postoperative follow-up is essential for proper management of these patients.^{1,8,14} This case report presents a mural unicystic ameloblastoma that occurred in a 12-year-old boy who was treated with marsupialization, a conservative surgical approach. There was no recurrence during the six-year follow-up.

CASE REPORT

A 12-year-old boy was referred to the Pediatric Dentistry Department, Gazi University, Ankara, Turkey for dental treatment. During the clinical examination, an asymptomatic intraoral swelling was observed in the left mandibular posterior region (Figure 1). Radiographical evaluation revealed an unerupted permanent second molar that was



Figure 1. Intraoral asymptomatic swelling in the left mandibular posterior region

surrounded by a multilocular radiolucent area shaped like a soap bubble (Figure 2). Following an evaluation and consultation with the Department of Oral and Maxillofacial Surgery, Gazi University, he was prediagnosed with an ameloblastoma, and surgery to marsupialize the cystic lesion was recommended.

Informed consent was obtained from the patient's parent before the surgery. During the surgical procedure, the cystic lesion was marsupialized, an obturator was positioned for drainage and the tissue that was removed from the lesion was sent to a laboratory for histopathological



Figure 2. Preoperative panoramic radiograph showing multilocular radiolucent area surrounded the second permanent molar tooth

evaluation. The obturator was positioned at the posterior retromolar section of the mandible, and the region was irrigated with saline solution every day for one week (Figure 3). A panoramic radiograph was taken after the surgical intervention for comparison (Figure 4). The histopathological report identified ameloblastoma cells in the cystic lesion, leading to a definitive diagnosis of a mural unicystic ameloblastoma and confirming the prediagnosis. After the soft tissue healed, the first permanent molar tooth received a root canal treatment. The patient was monitored for ameloblastoma recurrence and eruption of the second molar with annual panoramic radiographs. Figure 5 shows the healing of the lesion in a panoramic radiograph that was taken one year after the surgery. After six years of follow-up, there was no recurrence, and the second permanent molar erupted completely without complications (Figure 6).

DISCUSSION

Ameloblastomas are benign odontogenic tumors of epithelial origin. They are the most common aggressive benign tumor of the jaws. Assessments of the clinical type and size of the tumor, as well as the age of the patient, are important for designing treatments for this type of tumor.^{2,8} The WHO defines an ameloblastoma as a local invasive polymorphic neoplasia that often has a follicular or plexiform pattern in a fibrous stroma. Its behavior has been described as benign but locally aggressive.^{2,3,15,16}

Differential diagnoses of ameloblastomas and dentigerous cysts should be conducted by observing radiographic differences and obtaining biopsies, as dentigerous cysts can also occur with an unerupted tooth and painless, asymptomatic swelling.¹⁷ In a previous study, approximately 70% of unicystic ameloblastomas were associated with



Figure 3. Intraoral photograph of the obturator after the surgery



Figure 4. Postoperative panoramic radiograph taken after the surgery



Figure 5. Panoramic radiograph of one-year follow-up after the surgery showing the healing of lesion



Figure 6. Panoramic radiograph of six-years follow-up after the surgery showing no recurrence and eruption of the second molar tooth

an impacted tooth, and 1.9% were related to an unerupted tooth.¹⁸ The radiographic appearance of an ameloblastoma is frequently similar to that of a dentigerous cyst, but the presence of root resorption or tooth displacement should alert the clinician to the possibility of an ameloblastoma.⁸ In a study by Ord et al.⁸, six out of eight children who had ameloblastomas were prediagnosed with dentigerous cysts. Ameloblastomas are characterized by three distinct patterns with a mixture of radiolucent and radiopaque areas. The most common form is multilocular with various cysts that are separated by soap-bubble-shaped osseous septas. The second type of ameloblastoma is recognized by a honeycomb pattern, and the third type, which is critical for differential diagnosis, is unilocular.² In the present case, a soap-bubble appearance was radiographically observed in the left retromolar portion of the mandible that surrounded the unerupted second molar. The differential diagnosis included a clinical and radiographical evaluation. The case was prediagnosed as an ameloblastoma, a finding that was supported by the postoperative histopathological report. The treatment of ameloblastoma in children is complicated by several factors, including continued facial growth, different bone physiology (a greater percentage of cancellous bone, an increased bone turnover, and a reactive periosteum), the presence of unerupted teeth, the difficulties of the initial diagnosis and the predominance of unicystic ameloblastomas.⁸ To choose the right treatment plan for an ameloblastoma, the clinical type, localization, tumor size, and the patient age should be assessed.² While some researchers advise using only enucleation or minimal treatment for children, others suggest applying wide resection since recurrence is common with this disease, and a cancellous bone with a lesion may grow rapidly, causing extensive destruction.^{8,16,19,20}

Researchers have also suggested that enucleation, rather than partial or complete jaw resection, is an appropriate therapy for unicystic ameloblastomas.^{1,16} Huang et al.²¹ reported a retrospective study of ameloblastoma cases. According to that study, enucleation with a peripheral osteotomy was performed from an intraoral approach, and an obturator was constructed for the postoperative irrigation of the cavity with boiled water or saline. Paulo et al.²² reported an extensive unicystic ameloblastoma case of an eight-year-old girl who was treated with marsupialization. After 36 months, her facial asymmetry decreased, and mandibular growth was observed with no recurrence. In the present case report, the lesion was located at the retromolar region of the mandible, which might have resulted in postoperative fractures. Accordingly, after the initial diagnosis, marsupialization, curettage and enucleation were discussed as potential treatments. The conservative surgical approach of marsupialization was chosen due to the child's continued growth and development. An obturator was placed in the operated region to irrigate the marsupialized cavity during secondary healing.

On the other hand, other diagnosis of unicystic ameloblastoma at histopathological levels may require a different approach. Some unicystic ameloblastomas may be histopathologically classified as mural and luminal. Luminal ameloblastomas originate from infiltrative tissues, so their treatment should be more aggressive.^{9,13,18,20,23} Due to the high recurrence risk, regular clinical and radiological evaluations should be conducted. The relative recurrence rates observed in the literature suggest that unicystic ameloblastomas are less aggressive than solid or multicystic ameloblastomas.^{1,13} The recurrence period is approximately five years; however, numerous reports have documented significantly longer recurrence times. Sengüven et al.⁵ reported a mural unicystic ameloblastoma located in the posterior mandible of a child that recurred after eight years as a solid ameloblastoma. They found that a mural unicystic ameloblastoma can infiltrate the surrounding bone, necessitating further treatment. They also pointed out the need for long-term follow-up, especially in pediatric groups. In the present mural unicystic ameloblastoma case, there was no recurrence during an extended follow-up period of six years.

Functional and aesthetic damage might result in the development of craniofacial anomalies for young patients. Conservative treatments are generally associated with a

minimal healing area and positive functional and aesthetic outcomes. However, care should be taken with unicystic ameloblastomas due to their invasive character and local aggressiveness. Conservative treatments could be preferred for benign oral tumors in young patients, depending on the patient's age, the development stage, the anatomical structures surrounding the tumor and the wideness of the tumor, but these cases must receive careful long-term follow-up.

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