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CASE REPORT

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Unicystic ameloblastoma in a 9-year-old child treated with a combination of conservative surgery and orthodontic treatment: A case report

Tingting Guo[1] | **Ci Zhang**[2] | **Jian Zhou**[1]

[1]Department of General Dentistry and Emergency Dental Care, Beijing Stomatological Hospital, Capital Medical University, Beijing, China

[2]Department of Orthodontics, Beijing Stomatological Hospital, Capital Medical University, Beijing, China

Correspondence
Jian Zhou, Department of General Dentistry and Emergency Dental Care, Beijing Stomatological Hospital, Capital Medical University, Tian Tan Xi Li No.4, Beijing 100050, China.
Email: zhoujian@ccmu.edu.cn

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Abstract

Unicystic ameloblastoma (UAM) in the pediatric population is a rare clinical entity that has not been well addressed in the literature. Radical approaches affect a growing young patient's physical and psychological development, so conservative approaches are widely used for management in children. This report describes the case of a 9-year-old girl with UAM of the mandible, which also involved the impaction of the first and second premolars. Marsupialization with orthodontic treatment was performed to shrink the lesion and upright the first premolar involved in the tumor. Ten months after marsupialization, the lesion had healed entirely. The combination of conservative surgery and orthodontic treatment effectively shrank the lesion, preserving mandibular growth, preserving the involved first and second premolars, and promoting the eruption of the teeth.

KEYWORDS

marsupialization, orthodontic treatment, tooth eruption, unicystic ameloblastoma

1 | INTRODUCTION

Unicystic ameloblastoma (UAM) is defined as a variant of intraosseous ameloblastoma that occurs as a single cystic cavity. UAM is believed to be less aggressive and to respond more favorably to conservative treatments than the multicystic or solid type.[1] Various treatment modalities, such as segmental or marginal resection for UAM, have been used, similar to those generally used for conventional ameloblastoma, and more conservative treatments have also been frequently reported.[2] The traditional complete resection of the lesion site could impact a growing young patient's physical and psychological development. Therefore, conservative treatments are widely applied for treating UAM in children.

This report presents the case of mandibular UAM in a 9-year-old girl who was successfully treated by marsupialization and orthodontic treatment without the need for tooth removal.

2 | CASE REPORT

A 9-year-and-2-month-old girl with a chief report of swelling in the left mandibular premolar region was referred to our medical center. The patient had no systemic health

FIGURE 1 (A) Panoramic radiograph obtained at the first visit revealing a unilocular, well-defined, radiolucent lesion surrounding the unerupted left lower first and second premolars. (B, C) Intraoral photographs. Arrow indicates mucosal swelling. Images obtained at 9 years and 2 months of age

conditions. Extraoral examination revealed facial symmetry, and intraoral examination revealed mucosal swelling extending from the left lower first deciduous molar to the left second deciduous molar region, mixed dentition, and unerupted left lower first and second premolar. (Figure 1B,C).

A panoramic radiograph revealed a well-circumscribed, unilocular radiolucency in the region of the left lower premolars, extending from the distal canine area to the mesial root of the first molar area, with unerupted first and second premolars located lower than their healthy right counterparts. The roots of both the left lower first and second deciduous molars were absorbed to the neck of the teeth. (Figure 1A).

Cone-beam computed tomography (CBCT) was performed to confirm the location of the lesion and the relationship between the lesion and the adjacent anatomical structures. (Figure 4A–C).

The cystic lesion was treated by marsupialization together with the extraction of the left lower first and second deciduous molars. Biopsy was conducted (age: 9 years and 2 months) to shrink the lesion. Ibuprofen was used to relieve pain after surgery. After histopathologic evaluation, the lesion was diagnosed as UAM. (Figure 2).

Radiographs taken 5 months (age: 9 years and 7 months) after marsupialization showed that the lesion margin had lost clarity and that regenerated bone had replaced the low-density area. The first and second premolars had begun to erupt. However, the direction of the eruption of the first premolar was mesioangular, and no space was present between the left canine and the first premolar. (Figure 3A,D).

Thus, a segment arch appliance was bonded to the left canine and first molar of the mandible to uphold the leeway space with a coil spring. Then, an orthodontic button was bonded to the left first premolar, and distal traction was applied to the tooth using an elastic power chain. (Figure 3B,E).

Five months after orthodontic appliance placement (age: 10 years), we observed that the first premolar had erupted in place. (Figure 3C,F).

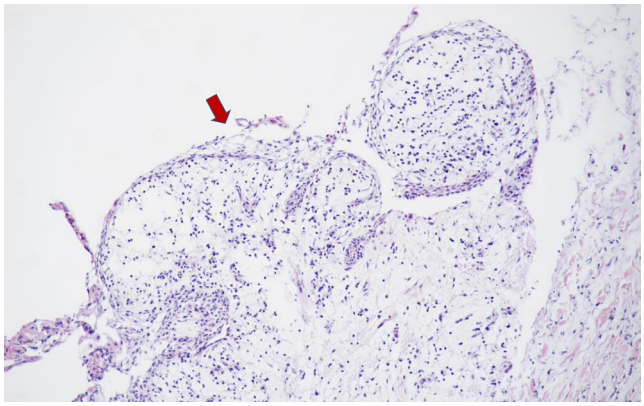

FIGURE 2 Histological findings (10×) of the specimen obtained at marsupialization revealed a simple cystic-type (type I) ameloblastoma. Arrow indicates loosely arranged suprabasal cells. Specimen obtained at 9 years and 2 months of age

Cone-beam computed tomography 10 months (age: 10 years) after marsupialization showed that the radiolucent area had disappeared entirely. (Figure 4D–F).

3 | DISCUSSION

The treatment of UAM has been controversial and can be conventional, radical, or conservative. Conventional approaches consist of segmental or marginal resectioning with subsequent reconstructive procedures often being required. Conversely, conservative treatments comprise marsupialization plus second-stage curettage. No substantial evidence proves one treatment modality as the most effective, and many reasons exist for this uncertainty.

There is a consensus among various authors that ameloblastoma has to be treated aggressively to avoid recurrences. However, an aggressive surgical approach in pediatric patients could result in numerous complications, such as functional and masticatory changes, mutilations, and loss of permanent teeth involved in the tumor and facial deformities. Therefore, there is a dilemma regarding the applicability of an initial radical extensive surgery in children. In children, the treatment of unicystic ameloblastoma is

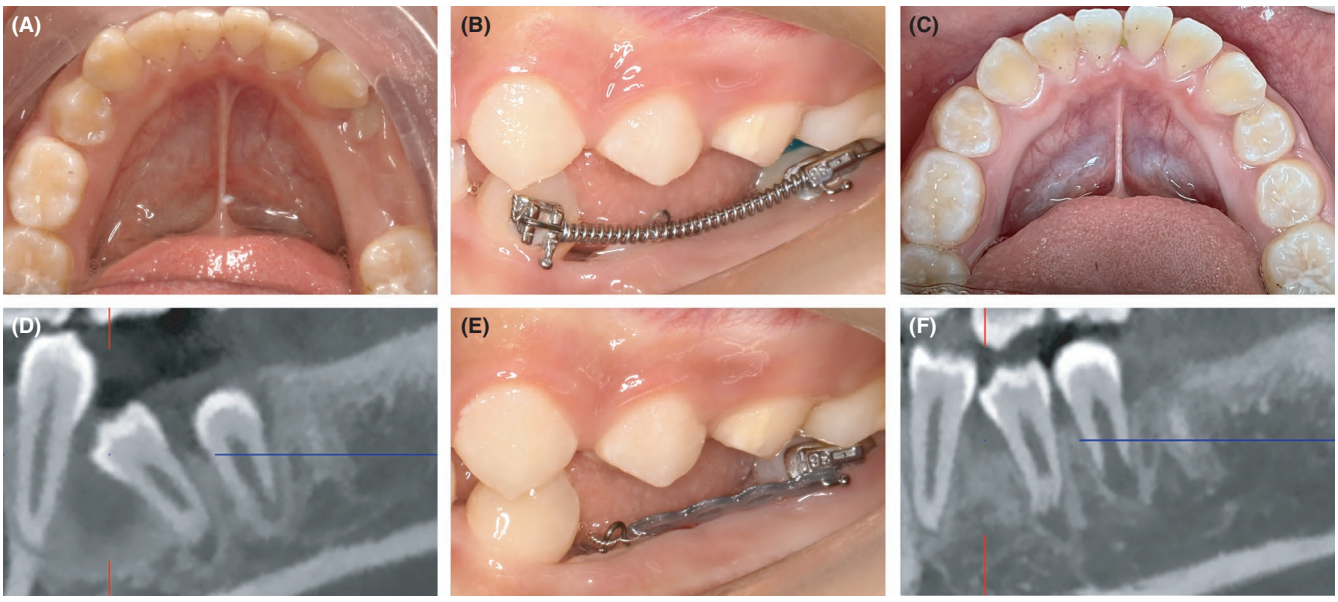

FIGURE 3 (A, D) Postoperative intraoral photographs and radiographs taken after 5 months show that the direction of the eruption of the first premolar was mesioangular, and no space was present between the left canine and the first premolar. The lesion margin had lost clarity, and regenerated bone had replaced the low-density area. Images obtained at 9 years and 7 months of age. (B, E) Segment arch appliance was bonded to the left canine and first molar of the mandible to uphold the leeway space with a coil spring, and distal traction was applied to the tooth using an elastic power chain. (C, F) Intraoral photographs and radiographs taken 5 months after orthodontic appliance placement, and the first premolar erupted in place. Images obtained at 10 years of age

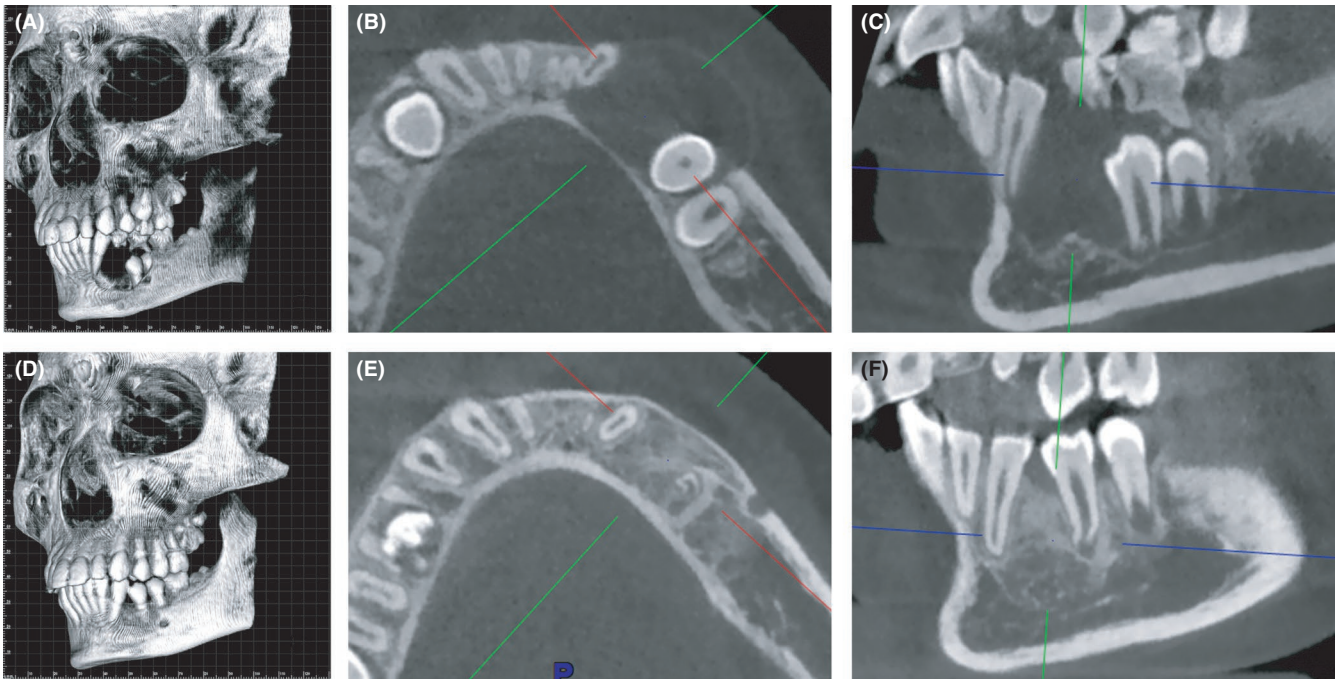

FIGURE 4 (A–C) Cone-beam computed tomography images taken at the first visit. (A) CBCT 3D reconstruction shows a low-density area surrounding the unerupted left lower first and second premolars. (B) Axial CBCT view shows an expansion of the buccal and lingual cortices in the mandible. (C) Coronal CBCT view at the level of the mandible. Images obtained at 9 years and 2 months of age. (D–F) Postoperative CBCT taken 10 months after marsupialization. (D) CBCT 3D reconstruction shows regenerated bone surrounding the left lower first and second premolar. (E) Axial CBCT view shows normal trabeculae replacing the low-density area in the mandible. (F) Coronal CBCT view at the level of the mandible. Images obtained at 10 years of age

complicated by the following key factors: 1) continued facial growth, different bone physiology (greater percentage of cancellous bone, higher bone turnover, and reactive periosteum), and the presence of unerupted teeth; 2) difficulty in making the initial diagnosis; and 3) predominance of the unicystic type of ameloblastoma.

Given that mandible resection in pediatric patients may lead to complications, such as dysfunction and deformity, our patient was submitted to conservative treatment to preserve the permanent teeth and as much bone as possible, to avoid esthetic deformities.

Marsupialization is considered a good way to exteriorize or decompress a cyst. Marsupialization followed by enucleation is one therapeutic approach for ameloblastoma,[3] which was our original treatment plan. Surprisingly, after 10 months, we observed that the lesion had completely healed. There are several reports of cystic ameloblastoma cases in which the tumor completely disappeared after marsupialization alone, suggesting that marsupialization helps avoid wide resection of the mandible in patients with UAM.[4] For our patient, we assumed that the lesion fully recovered because the pathological classification was simply cystic (type 1), which favored a successful outcome of conservative management.

In the present case, the lesion was histologically diagnosed as ameloblastoma on biopsy, and we assumed that the tumor was not entirely enucleated by marsupialization but remained around the crown of the first and second premolars. Accordingly, we performed careful follow-up to monitor the tumor size. Orthodontic treatment was applied in addition to marsupialization to allow the teeth to erupt. In similar cases, orthodontic treatment has been successfully applied for tumors involving the crown of the unerupted teeth.[5] After 10 months, the lesion in our patient had disappeared entirely, and the first and second premolars had erupted to a normal level. Long-term follow-up with radiography is mandatory for the early detection of recurrences. We are still monitoring our patient annually using radiography.

The treatment we chose avoided the resection of the mandible and induced eruption of the teeth involved in the tumor. Thus, the combination of conservative surgery and orthodontic treatment can be advantageous for managing UAM in children.

ACKNOWLEDGEMENTS

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CONFLICT OF INTEREST

The authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS

Tingting Guo examined and treated the patient, conceiving the idea for the case report, leading the case report writing, with Ci Zhang did the orthodontic part. Jian Zhou supervised the entire case report and revised the manuscript. All authors read and approved the final manuscript.

CONSENT

Written informed consent was obtained from the legal guardian of the patient (the father of the child) for publication of this case report and any accompanying images.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon reasonable request.

ORCID

Tingting Guo https://orcid.org/0000-0002-0570-968X

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