Clinical Report

Three-Dimensional Volumetric Analysis of Unicystic Ameloblastoma before and after Marsupialization Using OsiriX Software

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Abstract: The aim of this study was to evaluate the effect of marsupialization therapy for unicystic ameloblastoma by using three-dimensional (3-D) volumetric analysis with OsiriX software. This retrospective study assessed patients with unicystic ameloblastoma treated using marsupialization. Four patients (age, 29.00 ± 21.65 years) were included in this study, all of whom had mandibular ameloblastoma. The period between computed tomography before and after marsupialization was 5.75 ± 1.71 (4–8) months. In 3-D volumetric analysis using OsiriX software, the mean volume of the lesions before marsupialization was 22.91 ± 28.74 (6.23–65.86) cm3, and that after marsupialization was 11.28 ± 15.60 (2.19–34.62) cm3. The mean reduction in volume was 11.63 ± 13.16 (4.03–31.24) cm3, and the volumetric reduction ratio was 56.42 ± 7.60 (47.43–64.76)%.

Key words: Unicystic ameloblastoma, Three-dimensional reconstruction image, Volumetric analysis, OsiriX software

Introduction

Ameloblastoma is a benign odontogenic tumor with an invasive and destructive growth pattern1–5. The World Health Organization classifies ameloblastomas into four types: the solid/multicystic type, the unicystic type, the extraosseous-peripheral type, and the desmoplastic type6. The solid/multicystic type accounts for 90% of all ameloblastomas, whereas the other types are relatively rare5. Ameloblastoma is treated by radical resection or conservative treatment such as enucleation, curettage, decompression, and marsupialization1,2. Previous articles have reported that conservative treatment is associated with a high recurrence rate1,2. The unicystic type has a lower recurrence rate compared with the other types and is the only type that responds to conservative treatment6. Although progress in reconstructive surgery, extensive jaw resection influences quality of life and causes numerous complications2,3. Therefore, particularly for young patients with unicystic ameloblastoma, conservative treatment should be favored over other treatment methods.

OsiriX is an image-processing software dedicated to Digital Imaging and Communications in Medicine (DICOM) images, for Apple Macintosh7–9. It supports multiple medical imaging modalities, including computed tomography (CT), magnetic resonance imaging (MRI), positron emission tomography, and ultrasonography5,9. Clinicians can use OsiriX to build three-dimensional (3-D) images from DICOM files obtained from CT or MRI. Furthermore, OsiriX enables the extraction of a region of interest and the measurement of the area and volume10.

Over half a century has passed since the usefulness of marsupialization of unicystic ameloblastoma was first reported10. Nevertheless, few studies have conducted 3-D volumetric analysis to determine the effectiveness of marsupialization. In this study, we performed 3-D volumetric analysis of unicystic ameloblastoma before and after marsupialization using OsiriX software to evaluate the effects of marsupialization therapy.

Materials and Methods

Patients

We designed a retrospective study assessing patients diagnosed with ameloblastoma at the University of Fukui Hospital between May 2007 and April 2015. Only patients with unicystic ameloblastoma treated with marsupialization were included in this study.

Evaluation

We analyzed the location, form, and histological type of ameloblastoma, as well as which teeth were extracted. Furthermore, we performed 3-D volumetric analysis using OsiriX software to evaluate the effects of marsupialization on unicystic ameloblastoma. We recorded the period between CT examinations before and after marsupialization, reduction in volume, and volumetric reduction ratio. This study was approved by the Institutional Research Board and Ethics Committee of the Faculty of...
Fifteen patients were diagnosed with ameloblastoma, four of whom (two men and two women) underwent marsupialization for unicystic ameloblastoma and were included in this study (Table 1). The youngest and oldest patients were 11 and 58 years old, respectively, and the mean age ± standard deviation was 29.00 ± 21.65 years. In all cases, the mandible was involved, and the plexiform ameloblastoma was detected; the lesions were located in the mandibular molar region in three cases. At the time of marsupialization, the mandibular third molar was extracted in Case 1 because it was impacted in the lesion, and the mandibular second molar was extracted in Case 3 because of root resorption associated with the lesion.

The mean and standard deviation of the period between the CT examinations before and after marsupialization was 5.75 ± 1.71 (4–8) months (Table 2). In 3-D volumetric analysis using OsiriX software, the mean and standard deviation of the volume of ameloblastoma before marsupialization was 22.91 ± 28.74 (6.23–65.86) cm$^3$, whereas that after marsupialization was 11.28 ± 15.60 (2.19–34.62) cm$^3$. The mean and standard deviation of the reduction in volume was 11.63 ± 13.16 (4.03–31.24) cm$^3$, and the volumetric reduction ratio was 56.42 ± 7.60 (47.43–64.76)% (Figs. 1 and 2).

In all cases, remarsupialization or removal surgery was performed after marsupialization, and no recurrence occurred during the follow-up period.

**Case presentation (Case No. 1)**

An 11-year-old boy was referred to our department complaining of swelling and pain in the left cheek. Intraoral examination revealed buccal gingival swelling from the left lower first molar to the second molar; however, no percussion pain or mobility of the left lower first and second molars was observed. Panoramic radiography revealed a radiolucent lesion from the left mandibular body to the ascending ramus, impacted left lower third molar, shortened distal root of the left lower first molar, and shortened mesial and distal roots of the left lower second molar (Fig. 3). CT examination showed an osteolytic change from the left mandibular body to the ascending ramus and left lower impacted third molar within the lesion. MRI examination showed a relatively well-defined unicystic lesion; low and uniform signals were detected on T1- and T2-weighted images, respectively, indicating mural projection and

![Image](image_url)
substantially uniform contrast enhancement. Based on a clinical diagnosis of an odontogenic tumor, biopsy, extraction of the impacted third molar, and marsupialization were performed on the buccal aspect of the lower first and second molars under general anesthesia. The lesion was cystic, and a yellow transparent serous fluid was observed. Intraoperative frozen diagnosis showed ameloblastoma. After an impression of the fenestration window was taken, the area was packed with antibiotic gauze. Seven days after the operation, the antibiotic gauze was removed and a silicon obturator fabricated on the cast and designed to avoid occlusal interference was used to maintain the fenestration window. Marginal adaptation of the obturator was performed with a secondary silicon impression material. The obturator negated the requirement for frequent gauze changes. CT examination 8 months after the first marsupialization revealed that the lesion was diminished, and new bone formation was observed in the lingual and buccal regions, including the alveolar bone beneath the first and second molars and on the mandibular canal (Fig. 3). Eight months after the first marsupialization, enucleation of the tumor under general anesthesia was also performed with ease and the enucleated specimen had a smooth outer surface. Subsequently, bone scraping using a round diamond bur was performed with an endoscope. A piezosurgery system was used to scrape the bone near the mandibular canal. After enucleation of the tumor, intraoperative frozen diagnosis revealed no tumor cells in the surgical margins. Histopathological examination of the enucleated specimen revealed ameloblastoma cells from the outer to luminal surfaces of the thick fibrous tissue (Fig. 4). Marsupialization was performed again to open the bony cavity after tumor enucleation. One year and 2 months later, scar tissue was removed from the bone surface of the markedly diminished fenestration. The removed scar tissue was histologically proven to contain a small nest of odontogenic epithelium surrounded by granulation tissue, indicating residual tumor cells. Panoramic radiography and CT revealed new bone formation and no tumor recurrence. The patient’s facial appearance had become symmetric, no neuropathy was observed, and the first and second molars were successfully preserved. Panoramic radiography performed 2 years after the first marsupialization revealed that the roots of the first and second molars were slightly extended (Fig. 5). The left lower first and second molars responded positively to an electric pulp test. No recurrence was observed during the 2-year follow-up period.
Discussion

In this study, the average volume reduction ratio was 56.42% at 4 to 8 months after marsupialization. Additional conservative treatments were performed after marsupialization; however, radical resection with surgical margins was avoided because marsupialization reduced the volume of ameloblastomas in all cases. Furthermore, no recurrence was observed during the follow-up period. In particular, despite the relatively large ameloblastoma and involvement of the tooth roots in the lesion in Case No. 1, marsupialization made it possible to avoid tooth extraction and deactivation of the dental pulp of the mandibular first and second molars. This result showed that marsupialization can prevent a major decline in a patient’s quality of life after treatment for ameloblastoma.

Although marsupialization and decompression are different procedures, they share the principle aim of reducing the size of cystic lesions by lowering the pressure of cystic fluid and inducing bony apposition to the cystic walls. Marsupialization and decompression were suggested by Partsch and are referred to as the Partsch I procedure. The Partsch II procedure comprises enucleation and primary closure. Decompression involves making a small opening in the cyst and keeping it open by using a rigid drainage tube that facilitates the irrigation of the cavity and prevents food impaction or microorganism accumulation. In contrast, marsupialization, which is considered the more definitive treatment, involves converting the cyst into a pouch by creating a large window in the bone, and then connecting the inner cystic wall to the oral mucosa, with exposure of the cyst lining to the oral environment. The disadvantages of these techniques are secondary infection and deformation caused by opening the cyst without primary closure.

Nakamura et al. analyzed cases of cystic ameloblastoma treated with marsupialization. They reported that marsupialization was more effective in teenaged patients and that the potential for new bone formation associated with the effect of marsupialization was influenced mainly by the age of patients. Furthermore, four out of the five cases in which marsupialization was extremely effective showed unicocular appearance on radiographs.

Previous studies on the 3-D volumetric analysis of the effect of decompression surgery for odontogenic cystic lesions including ameloblastoma have reported that the volume reduction was related to the duration of decompression and the original size of the lesion. In particular, Jeong et al. reported that decompression was more effective when performed for more than 6 months. Asutay et al. also performed 3-D volumetric analysis of the effect of decompression and reported that the percentage of volume decrease of ameloblastoma was 44.50 ± 14.41% from the preoperative volume to the volume 6 months postoperation. Shudou et al. performed 3-D volumetric analysis of the effect of marsupialization surgery for odontogenic keratocyst lesions and reported that the volume of the lesions was negatively correlated with the duration after marsupialization. These studies suggested that despite the longer treatment period and the need for additional treatments, marsupialization and decompression are useful surgical interventions for large odontogenic cystic lesions.

The progress of medical technology has contributed to innovations in surgical procedures and led to superior treatment outcomes compared with conventional procedures. Image processing software is useful in pre- and postoperative evaluation. Real-time 3-D navigation of images enables operators to carefully evaluate the surgical field and target lesion, and facilitate minimally invasive and accurate surgery. In addition, robot-assisted surgery has been developed, enabling highly accurate and minimally invasive surgical procedures. Development of technology such as devices and software should continue in medical and dental fields in the future.

In conclusion, marsupialization contributed to reducing the volume of unicystic ameloblastoma and to avoiding radical resection, which reduces quality of life. The results of this study suggest that marsupialization should be favored over other methods in the treatment of unicystic ameloblastoma.

Conflict of Interest

The authors declare no conflict of interest.

References