Open Access Library Journal
2024, Volume 11, e12601
ISSN Online: 2333-9721
ISSN Print: 2333-9705

Conservative Management of Odontogenic Keratocyst: Two Case Reports of Marsupialization Followed by Enucleation

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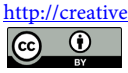
How to cite this paper: Moujoud, C., Bouzoubaa, S.M. and Yahya, I.B. (2024) Conservative Management of Odontogenic Keratocyst: Two Case Reports of Marsupialization Followed by Enucleation. *Open Access Library Journal*, **11**: e12601.
https://doi.org/10.4236/oalib.1112601

Received: November 8, 2024
Accepted: December 15, 2024
Published: December 18, 2024



Abstract

Odontogenic keratocysts (OKCs), first described by Philipsen in 1956, are benign but aggressive cysts of odontogenic origin with high recurrence rates [1]. The choice of treatment for keratocysts, depends on factors such as size, location, patient age, cortical perforation, and proximity to critical structures like teeth, the inferior alveolar nerve, the pterygomaxillary fossa, and the maxillary sinus. Current treatment modalities include curettage, marsupialization, enucleation, and surgical resection, frequently followed by reconstruction. For large cysts or those near vital structures, conservative treatments are recommended to preserve oral tissues and limit surgical damage to important anatomical structures [2] [3]. This paper presents two cases of odontogenic keratocyst treated with a conservative approach, combining marsupialization and subsequent enucleation. Marsupialization was initially performed to decrease the size of the cysts, allowing the preservation of critical anatomical structures and reducing the need for more extensive surgery. Once the cysts had sufficiently regressed, enucleation was carried out to fully remove the remaining lesion. This conservative method proved effective in managing the lesions while minimizing surgical risks. These cases highlight the potential advantages of a conservative management strategy for large OKCs.

Subject Areas

Dentistry

Keywords

Odontogenic Keratocyst, Marsupialization, Enucleation, Conservative Management

1. Introduction

Keratocystic odontogenic tumor (KCOT), also known as odontogenic keratocyst (OKC), is a benign intraosseous tumor of odontogenic origin that is relatively frequent in the oral cavity. It has a locally aggressive behaviour and exhibits a high rate of recurrence after treatment [1]. Histologically, KOT is characterized by a thin parakeratinized stratified epithelium [1] [2]. Bone resorption in the jaws is one of the most severe complications of KCOT and can easily give rise to large jaw defects or even pathologic fractures, especially when the KCOTs are large. Generally, KCOT can be treated by either enucleation or marsupialization, depending on its location, size, and proximity to vital structures such as the teeth, maxillary sinus, and mandibular canal [3]. Enucleation is the treatment of choice for small KCOTs and can be performed without damage to adjacent tissue, whereas marsupialization is usually performed for large ones, in order to minimize the cyst size and limit the extent of surgery.

The aim of these case reports is to describe the results obtained with a conservative surgical approach in two patients diagnosed with odontogenic keratocyst.

2. Case Reports

Case 1:

A 44-year-old female patient, in good general health, was referred to our service for the diagnosis and treatment of a mild swelling in the ramus region that had been evolving for one year.

On extraoral examination, inspection revealed facial asymmetry due to a slight swelling in the ramus area. Palpation identified a firm, painless mass with no associated lymphadenopathy. The intraoral examination was unremarkable.

The patient brought with her a cone beam computed tomography (CBCT), that demonstrated a well-defined, homogeneous radiolucent area occupying nearly the entire ascending ramus, while preserving the coronoid and condyloid processes. There was cortical perforation on both the internal and external bone tables, and the lesion had a very close relationship with the inferior alveolar nerve. (**Figure 1**)

We performed an aspiration, and the results were positive, leading us to suspect a cystic lesion.

Given the patient's motivation and her request for the surgery to be performed under local anesthesia, as well as the lesion's location and extent, which made the area fragile and placed in close proximity to the inferior alveolar nerve, marsupialization was chosen to avoid neuro-sensory damage, promote bone healing, and prevent pathological fractures.

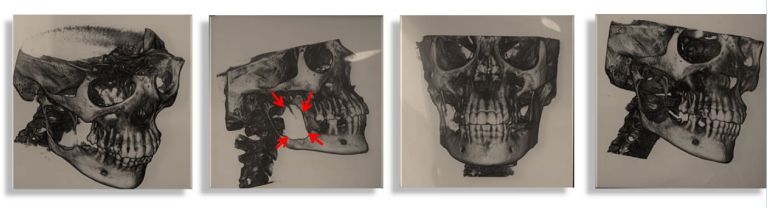
We carried out marsupialization and collected a surgical specimen, which confirmed the diagnosis of a keratocyst.

Only six months after marsupialization, the results were very promising, demonstrating spontaneous bone regeneration in the ramus, as well as a reduction in the volume of the lesion with peripheral regeneration. There was also an increase in the thickness of the internal and external bone tables, along with spontaneous bone

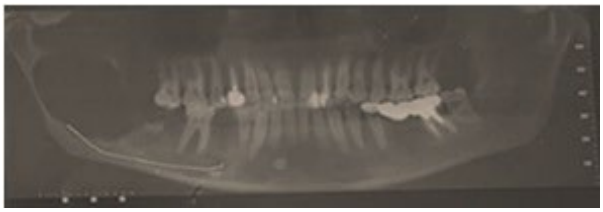
regeneration at the apex, which protected the inferior alveolar canal. (Figure 2)

At this stage, we chose to proceed with enucleation due to the encouraging results observed from the marsupialization. Enucleation would allow for the complete removal of the keratocyst while minimizing the risk of recurrence and further complications. (Figure 3)

Clinical follow-up showed complete clinical healing after three months. (Figure 4) Radiological follow-up after one year revealed complete bone healing in the ramus with no signs of recurrence. (Figure 5)



(A)



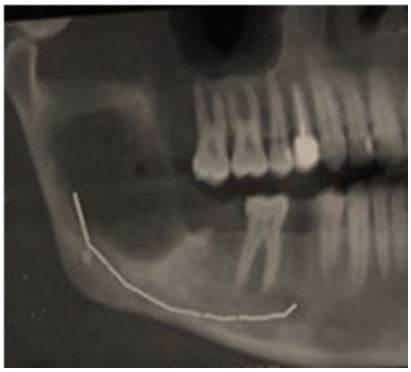
(B)

(C)

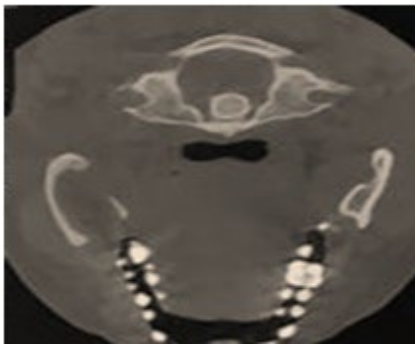
(D)

Figure 1. The initial cone beam. **(A):** 3D reconstruction revealed a radiolucent image at nearly the entire ramus. (B): The panoramic reconstruction revealed a well-defined, homogeneous radiolucent lesion, preserving the coronoid and condyloid processes. (C): The axial sections revealed the interruption of both the internal and external bone tables. (D): The oblique coronal sections revealed the close relationship of the lesion with the inferior alveolar nerve.

(A)



(B)



(C)

Figure 2. The control cone beam after six months of marsupialization. (A): The 3D reconstruction revealed spontaneous regeneration in the ramus. (B): The panoramic cut shows a reduction in the volume of the lesion after marsupialization. (C): The axial sections revealed an increase in the thickness of the bone tables after marsupialization.

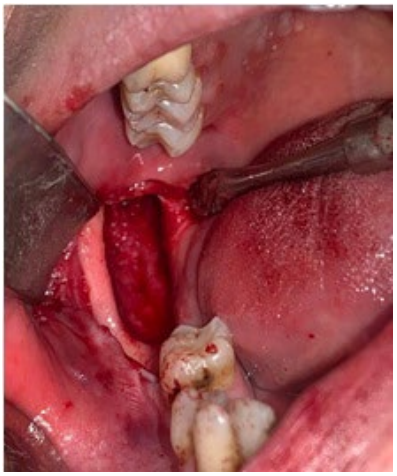


Figure 3. Per operative image illustrating the enucleation.

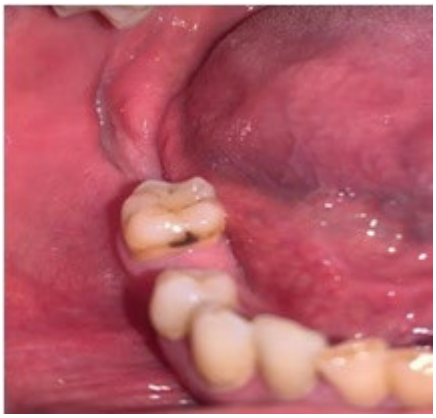


Figure 4. Clinical follow-up after 3 months showing complete healing of the enucleation site.

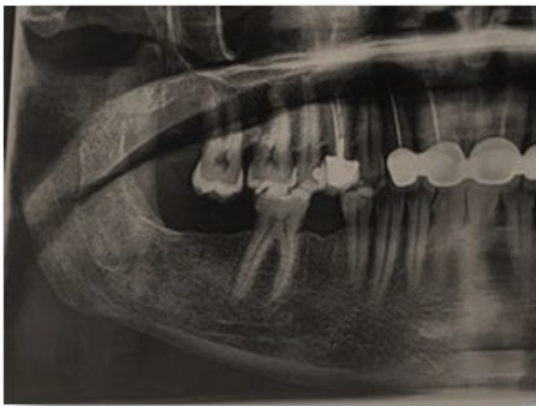


Figure 5. Radiological follow-up one year after enucleation showing complete bone healing.

Case 2:

A 36-year-old patient presented to our consultation at the Surgical Odontology Department of Ibn Rochd University Hospital of Casablanca following the incidental discovery of a bi-locular lesion in the posterior region of the left side of his lower jaw. The patient had no relevant medical history.

The extraoral clinical examination revealed no notable signs. The intraoral clinical examination revealed a slight filling adjacent to left mandibular molars. Palpation was painless, firm in consistency, and there were no lymphadenopathies.

The panoramic radiograph revealed a bilocular lesion associated with tooth 37, extending to the mandibular angle, with the inferior alveolar nerve obscured within the lesion. (Figure 6)

The 3D imaging revealed thinning of the bone tables and a close relationship with the inferior alveolar nerve. (Figure 7)

Given the extent and location of the lesion, along with its close relationship with the inferior alveolar nerve, marsupialization was chosen.

After obtaining a positive aspiration, we performed marsupialization and collected a surgical specimen that confirmed the diagnosis of a keratocyst. (Figure 8)

Radiographic Follow-up after 12 months of marsupialization shows a reduction in the volume of the lesion, an increase in the thickness of the bone tables, and bone healing apically that protects the inferior alveolar nerve. (Figure 9) (Figure 10)

An enucleation was then performed to eliminate the entire lesion. (Figure 11)

The clinical follow-up after one month shows complete mucosal healing. (Figure 12)

Radiological follow up at 12 months indicated marked new bone formation without any sign of recurrence. (Figure 13)

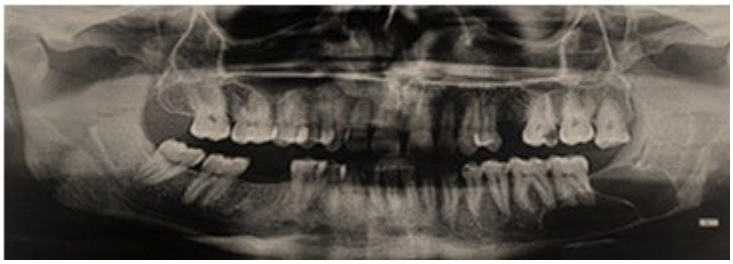


Figure 6. The panoramic radiograph revealed a bilocular lesion in the posterior region of the mandible.

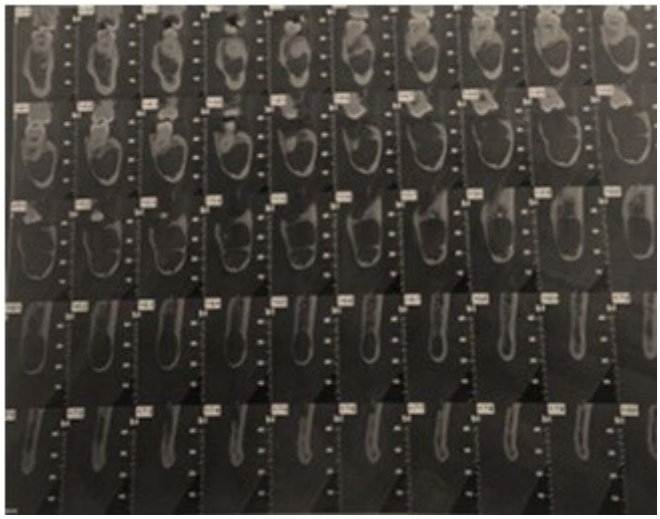


Figure 7. The initial CBCT.

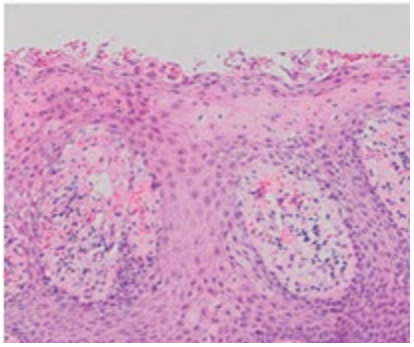


Figure 8. Histological section of a keratocyst. H&E stain.

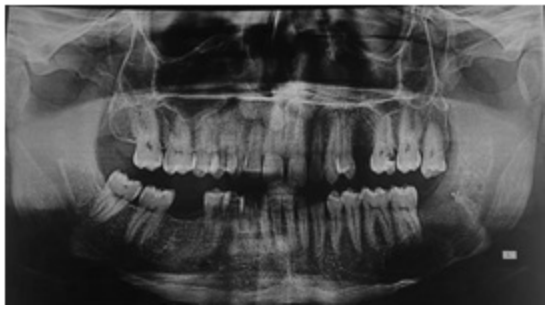


Figure 9. Radiograph after 12 months of marsupialization.

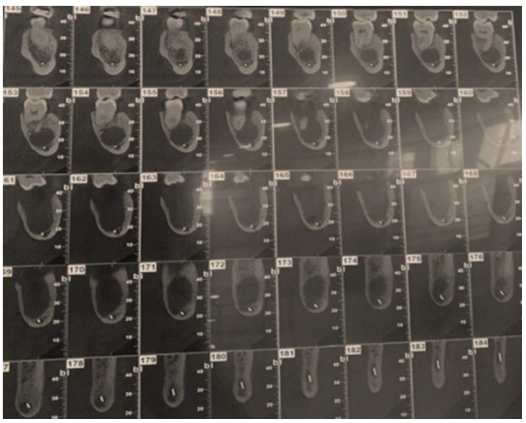


Figure 10. CBCT after 12 months of marsupialization.

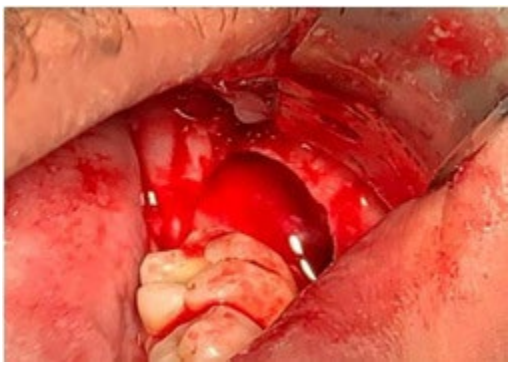


Figure 11. Per-operative photo of the enucleation.

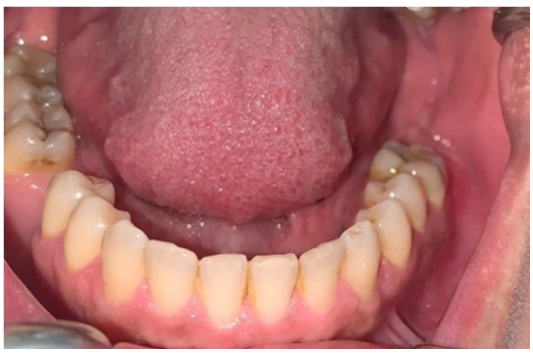


Figure 12. Clinical follow-up one month after enucleation shows complete healing.

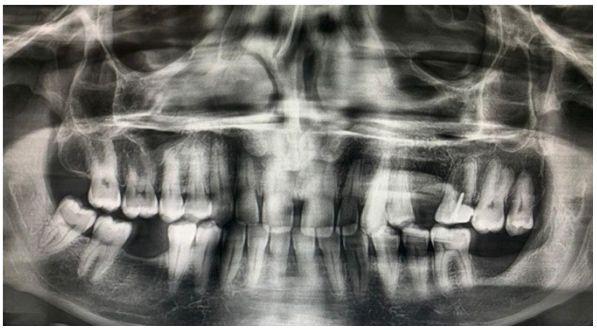


Figure 13. Radiological follow up after one year of enucleation shows complete bone regeneration at the site of the enucleation.

3. Discussion

Odontogenic keratocyst (OKC) is a type of developmental cyst that forms in the jaw due to the proliferation of remnants from the dental lamina or the basal layer of the oral epithelium within the mandible or maxilla [4]. It constitutes approximately 11.7% of all jaw cysts worldwide, making it the third most common type, following radicular and dentigerous cysts [5].

As classified by the WHO, OKC is a benign intraosseous tumor that can be either unilocular or multilocular, characterized by a lining of stratified squamous epithelium. It has the potential for aggressive and infiltrative behavior and is associated with a high recurrence rate ranging from 25% to 62.5% [6].

OKCs are most frequently observed in individuals during their second and third decades of life, with a higher prevalence in males. These cysts predominantly affect the mandible rather than the maxilla, with a common location at the angle of the mandible [4] [5]. These data are consistent with our case reports, where the keratocyst was presented in our two patients during the third decade of life at the mandibular angle.

Despite being classified as a benign lesion, the biological characteristics of the odontogenic keratocyst (OKC) lead to its consideration as an "aggressive" entity. This cyst has a propensity to recur and may grow to significant dimensions before detection, making it essential to carry out a thorough treatment involving complete removal of the cystic lesion to minimize or eliminate the risk of recurrence [7].

The high recurrence rate of odontogenic keratocyst can be attributed to the thin, delicate wall of the OKC, which makes it challenging to enucleate the cyst intact from the surrounding bone, along with the presence of small satellite cysts within the fibrous wall [8]-[10].

Currently, the treatment of choice for OKCs is still widely debated on the most effective treatment for OKC. Treatment modalities are generally divided into conservative and aggressive categories. The decision regarding treatment should consider various factors, including the patient's age, the cyst's size and location, any soft tissue involvement, prior treatment history, and the histological variant of the lesion [11] [12].

Aggressive treatments typically involve procedures such as osteotomy, lesion resection, the application of chemical agents like Carnoy's solution, cryotherapy with liquid nitrogen, or peripheral osteotomy. In contrast, conservative approaches usually include simple enucleation (with or without curettage) or marsupialization [7] [12] [13]. Notably, for patients in the first or second decade of life with unerupted teeth involved in KCOTs, aggressive surgery may not be the most suitable option compared to conservative treatment.

The advantages of marsupialization and decompression include:

- Preservation of oral tissues;
- Avoidance of surgical damage to critical anatomical structures, such as the inferior alveolar nerve;
- Maintenance of pulp vitality;
- Prevention of dental extractions and protection of developing teeth;
- Gradual reduction of the cystic cavity, which lowers the incidence of mandibular fractures;
- Low risk of recurrence;
- Minimal surgical morbidity.

However, there are also some disadvantages associated with this procedure, including the need to keep the cyst cavity clean to prevent infection, requiring the patient to irrigate the cavity several times a day; and the presence of pathologic tissue left in situ without a thorough histological examination [13].

Several studies investigating the effectiveness of marsupialization for keratocyst lesions through volumetric analysis with three-dimensional computed tomography (CBCT) have demonstrated its efficacy in reducing the size of odontogenic cysts and promoting bone healing [14] [15].

Additionally, other researchers have assessed the effectiveness of marsupialization using immunohistochemistry on odontogenic keratocysts. Their findings indicated that bone formation was significantly enhanced in the KCOT capsule wall adjacent to the bone following marsupialization, as evidenced by alkaline phosphatase activity assays and immunostaining for bone morphogenetic protein and an increase in fibrosis and qualitative changes in inflammation type [3] [16]. Overall, marsupialization appears to reduce the aggressive nature of OKCs in terms of growth characteristics [9].

Marsupialization has demonstrated its effectiveness as a preliminary treatment for large odontogenic keratocysts (OKCs) prior to enucleation. This treatment protocol was an effective and conservative approach to the management of the KCOT, enabling the reduction of the initial lesion, the preservation of anatomical structures and teeth, allowing a quicker return to function [9].

Research by Fadi Titinchi indicates that combining decompression with enucleation can lower recurrence rates by up to 15.2%. This decline in recurrence may be attributed to the metaplasia of the cystic lining, leading to the transformation of the odontogenic keratocyst into less aggressive lesions [17].

Marsupialization followed by enucleation performed after 12 to 18 months has been shown to lower the recurrence rate of odontogenic keratocysts (OKCs). In

the case of our patients, a conservative approach was chosen after considering factors such as the size, location, and radiological characteristics of the tumor, as well as the patient's preferences. The combination of marsupialization followed by enucleation demonstrated a positive outcome, with no signs of recurrence observed at the 12-month follow-up.

4. Conclusion

In conclusion, the conservative approach to treating keratocysts through marsupialization, followed by enucleation, has proven to be an effective strategy. This treatment not only facilitates the reduction of lesion size and promotes spontaneous bone regeneration but also minimizes surgical risks associated with extensive resections. Our clinical experience has demonstrated that this sequential management approach leads to favorable outcomes, including complete healing. Furthermore, the preservation of surrounding anatomical structures, such as the inferior alveolar nerve, enhances the patient's quality of life post-treatment. Therefore, marsupialization followed by enucleation represents a reliable and beneficial option for the management of keratocysts in clinical practice.

Conflicts of Interest

The authors declare no conflicts of interest.

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