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Case report

Two-Stage Decompression by Delayed Enucleation in the Management of an Odontogenic Keratocyst in a Resource Limited Setting: A Case Report

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Abstract

Introduction: Odontogenic keratocyst (OKC) is a rare type of developmental cyst known for its aggressive behavior and high recurrence rate. It arises from remnants of the dental lamina within the mandible and maxilla. Diagnosis is typically based on clinical, radiographic and histopathological findings.

Case Presentation: An 8-year-old male child was diagnosed with odontogenic keratocyst after a thorough clinical and radiological evaluation. The pertinent clinical findings included swelling in the anterior mandible, obliteration of the vestibule, and destruction of the buccal cortical bone. The panoramic X-ray revealed a well-defined radiolucent area in the anterior mandible.

Clinical Discussion: Treatment consisted of decompression with a tube in situ followed by delayed enucleation as definitive management. This resulted in significant clinical and radiographic improvement of the affected mandible.

Conclusion: Cystic decompression with a tube in situ for continuous irrigation followed by conservative enucleation represents an effective treatment approach for odontogenic keratocyst particularly in younger patients.

Key words: Cyst, Odontogenic, Decompression, Enucleation

Introduction

An odontogenic cyst is defined as a cystic lesion in which the epithelial lining of the lumen is derived from epithelium involved in tooth development. The three main epithelial structures that are believed to cause odontogenic cysts include; reduced enamel epithelium (REE), Rests of Malassez remnants of the Hertwig's epithelial root sheath and remnants of the dental lamina (1, 2). There are diverse group of odontogenic cysts ranging from a small lesions detected incidentally during routine examinations to highly aggressive and destructive lesions that may, in rare case, undergo malignant transformation. The radiographic features may be well defined solitary lesion with smooth or scalloped margins, or it can be multilocular radiolucency (3). Recurrence is common after removal of the cyst. The mechanism of recurrence was proposed by Brannon in 1976, suggesting it was due to three different mechanisms: incomplete removal of the cyst, growth of new keratocysts from satellite cysts, and development of a new keratocyst in the area adjacent to the site of the primary keratocyst (4).

The management options for OKC include enucleation, surgical resection, and marsupialization. Other treatment options include adjuvant methods such as peripheral ostectomy, cryotherapy (liquid nitrogen) and Carnoy's solution (5). Nevertheless, there is still considerable uncertainty regarding which methods yield the lowest recurrence rates while minimizing morbidity and determining the optimal treatment option (6). Here we discuss decompression with delayed enucleation as a primary and effective treatment option for pediatric patients with mixed dentition as means of conservative treatment.

Case Presentation `

An 8-year-old male patient presented to the Department of Dentistry, Oral and Maxillofacial Surgery with a complaint of discomfort and noticeable swelling over anterior surface of the mandible which had lasted for 2 months. The patient did not report any associated pain and denied history of any medical illness. On physical examination there was swelling on the anterior region of the mandible from left primary canine region to right primary second molar region. The swelling continued to expand, causing further distension of the labial mucosa with obliteration of the lower vestibule. On palpation, the swelling was painless, fluctuant, and the overlying mucosa did not show any inflammatory signs. A mixed dentition (primary and permanent teeth) was present. Outward flaring of the lower permanent incisors was observed. Extraoral examination was unremarkable.

Diagnostic assessment: The differential diagnosis included Odontogenic keratocyst, Dentigerous cyst, ameloblastoma. On aspiration of the lesion there was thick cheesy, creamy material. Orthopantomograph (OPG) revealed a well-defined radiolucent lesion, extending from the mesial aspect of the erupting permanent mandibular right second premolar, to the left erupting permanent canine. There was an inferiorly displaced right permanent canine which was within the cyst and the lower permanent anterior teeth were drifted to the righted side (Fig.1). OKC was considered the most likely diagnosis as OPG revealed antero-posterior expansion of the lesion with thin well-defined borders and perforation of the buccal cortical plate, which are the characteristic radiographic features. Based on clinical as well as radiological evaluation, the final diagnosis of OKC was established.

Therapeutic intervention: Under general anesthesia**,** an incisional biopsy and decompression of the lesion were performed. A decompression tube was inserted into the cystic cavity and secured to the adjacent mucosa. The

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patient's family was advised concerning the need for continuous irrigation through the tube.

Follow-up and outcomes: Irrigation was initially performed at the hospital until the patient's family became familiar with the procedure and the patient was discharged. The patient was seen after one week, the inserted decompression tube was in situ and patent, with good healing of sutured mucosa. The initial incisional biopsy report revealed a benign odontogenic keratocyst. The patient was scheduled for a regular follow-up appointment. After 9 months, the patient underwent a control X-ray which revealed that the lesion had shrunk significantly in size. Additionally, bone deposition was confirmed by an OPG (Fig. 2**).**

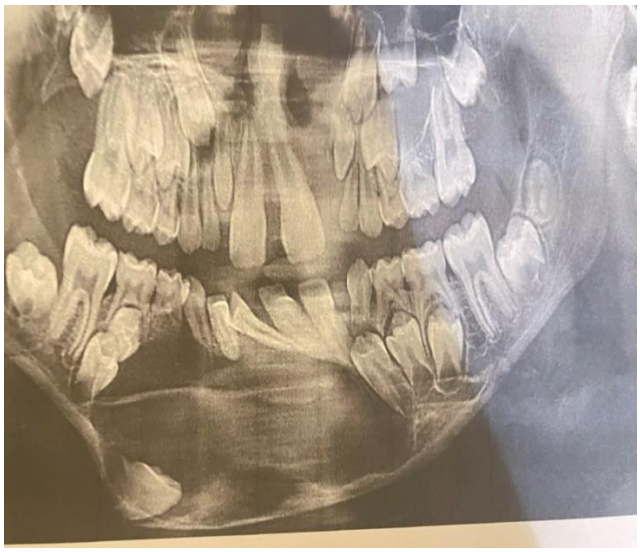

Figure 1: Preoperative OPG showing well defined radiolucent lesion in the mandible

After clinical and radiographic evaluation and confirming the size of the cyst was dramatically reduced, the patient was scheduled for complete enucleation of the cystic lining. Complete enucleation of the thickened cystic lining and cauterization were performed. The associated inferiorly displaced tooth was removed; otherwise other permanent teeth were preserved. The final histolopathological report also revealed an odontogenic keratocyst. Again six months after complete enucleation, the patient had control OPG revealing progressive bone deposition (Fig. 3). After the first year following complete enucleation, the patient came with a

Figure 2**:** Follow - up OPG after 9 months with decompression tube in situ.

Figure 3: Follow - up OPG after six months of complete enucleation with progressive bone deposition

follow-up X-ray that revealed adequate bone deposition and good remodeling of the mandible. Additionally, the alignment of permanent teeth, which had been pushed out of position due to mass effect, came to normal eruption position (Fig. 4).

Discussion

The management of odontogenic keratocysts (OKC) remains a hotly debated topic in oral and maxillofacial surgery. Despite numerous studies and systematic reviews on treatment options, there is a lack of consensus and no accepted protocol on the management of OKC (7). There are different treatment options available for this condition, including conservative approaches such as marsupialization, decompression, and enucleation, as well as aggressive bone resections and adjuvants methods.

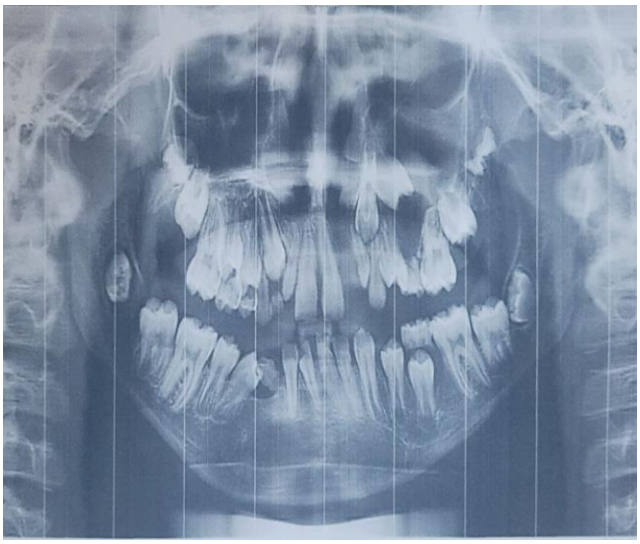

Figure 4: Follow - up OPG after 1 year of complete enucleation with sufficient bone formation and remodeling

Adjuvant methods such as peripheral ostectomy, cryotherapy (liquid nitrogen) and Carnoy's solution are considered aggressive forms of treatment which have shown more promising outcomes (7).A new adjuvant method has emerged recently, involving the topical application of 5-fluorouracil (5-FU) 5% cream following enucleation and peripheral ostectomy. This methods has a lower recurrence rate and less chance of alveolar nerve paresthesia compared to application Carnoy's solution (9,10). Radical methods involve mainly resection which yields the lowest rate of recurrence however causes significant morbidity. The increase in morbidity associated with the resection and the compromise in the quality of life in dealing with large lesions has led surgeons to consider decompression followed by enucleation as an alternative treatment option (11).

Decompression is a simple method in which a hole that connects the cystic cavity with the oral cavity is created by placing decompression tube. Cystic fluid is evacuated and the cystic pressure is reduced. This will stimulate new bone formation. It is considered to be the least invasive and most effective treatment of choice for OKC. This approach results in a reduction in lesion size which has a positive impact in reducing aggressive behavior commonly associated with these lesion (11). The decreased osmotic pressure caused by cystic fluid in the surrounding bony walls enables the cyst to shrink, promotes the deposition of new bone, reduces the risk of pathological fractures, lowers the rate of recurrence, preserves adjacent vital structures, and allows for the eruption of affected teeth (13). Furthermore, the decrease in osmotic pressure has a significant impact on the biological functions of cells, including proliferation, migration, and apoptosis (134)

At our institution, we have successfully utilized decompression of large OKC as a pre-treatment measure to reduce the size prior to complete enucleation. Because the cyst was large and the patient was a child in the with mixed dentition stage and preservation of the permanent teeth is mandatory which is difficult in primary surgical enucleation. Other treatment options like cryosurgery and Carnoy's solution are not readily available in our setup. In previous studies, decompression was considered an effective management option for treating large OKC. A tube was surgically inserted and left in place untill new bone formation and thickening of the cyst wall. Definitive management was then performed after six months which aligns with the approach taken in our current case report (14,15). Regarding recurrence, studies have suggested that no recurrence occurs

following decomposition and definitive management for large odontogenic keratocysts (16, 17)

Conclusion

Conservative decompression followed by enucleation, instead of aggressive resection, in younger patients with large odontogenic keratocysts. This surgical approach is particularly useful in situations where other adjuvant treatment options are not readily available, as in our setting. Long-term follow-ups are essential, including periodic radiographic examinations to monitor progress.

Consent for Publication

Written informed consent was obtained from the patient for the publication of this manuscript

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Ethical considerations

The ethical approval was waived by the Department of Dentistry and Maxillofacial Surgery, Addis Ababa University Research Ethics and Review Committee. Written informed consent was obtained from the parents for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Data availability statement

All relevant data are contained within the manuscript.

Conflicts of interest

The authors declare no competing interest.

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