Appearances are a glimpse of the unseen: Odontogenic Keratocyst

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ABSTRACT

Introduction: Odontogenic Keratocyst (OKC) is an enigmatic developmental cyst that needs special attention due to its aggressive behavior, autonomous growth, high recurrence rate and sometimes, silent clinical manifestation of lesions to large size. These peculiarities, combined with their frequencies, delayed diagnosis, prognosis makes the treatment often challenging.

Case discussion: This case report demonstrates the management of an extensive infected OKC with surgical endodontics. A 52-year-old patient reported with swelling in the palate. After thorough examination clinically and radiographically with the help of periapical, occlusal radiographs, orthopantomogram and CT scan the character, extent and the size of the lesion was assessed and a treatment protocol was formulated. The protocol included a conventional endodontic therapy followed by cystic enucleation, apicectomy, root end filling with biodentine and open packing.

Conclusion: Successful treatment of OKC depends on accurate diagnosis, appropriate non-surgical and surgical intervention and periodic follow up in order to prevent the appearance of new lesions in the area. Enucleation followed by open packing can be suggested as a choice of conservative treatment with low recurrence rate for large OKC. However, due to the clinical behaviour of OKC, a periodic follow up is mandatory.

Key words: odontogenic keratocyst, surgical endodontics, enucleation,
INTRODUCTION

Jaw cysts are very common due to the presence of odontogenic epithelium remnants. Cysts constitute about 17 percent of the tissue specimens submitted to oral pathology biopsy services.\(^1\) The odontogenic keratocyst (OKC) is classified as a developmental cyst derived from the enamel organ or from the dental lamina.\(^2\) The definition “odontogenic keratocyst” first was proposed by Philipsen in 1956, when he separated seven jaw cysts from cholesteatomas occurring in other cranial areas. OKC constitutes 5.4-17.4 percent of all odontogenic cysts.\(^3,4\) OKC is one of the rare odontogenic cysts, which attracts many researchers due to its unique characteristics as it reflects aggressive clinical behavior, histologically high mitotic rate and association with genetic and chromosomal abnormalities.\(^5\)

OKCs appear as well-defined radiolucencies, which can be either unilocular or multilocular. Unilocular OKCs can be located periapically, simulating periapical cysts; surrounding the crown of unerupted teeth, simulating dentigerous cysts; between the roots of teeth, simulating lateral periodontal cysts or lateral radicular cysts; or in the maxillary midline, simulating nasopalatine duct cysts. Large unilocular OKCs can be indistinguishable from cystic ameloblastomas. Multiple OKCs are also associated with Nevoid basal cell carcinoma syndrome (NBCCS).\(^1,6,7\)

Differential diagnosis in endodontics may raise concerns when destructive lesions resorb significant amounts of alveolar bone. Generally, a sequence of clinical examinations is recommended for evaluating pulp health and a possible endodontic etiology of the bony lesion (Pitt Ford & Patel 2004).

CASE REPORT

A 52 year old patient reported to Department of Conservative Dentistry & Endodontics, KVG Dental College with a complaint of swelling in the palatal region since 2 years. There was no history of any recent trauma or any associated pain in the affected region. He gave history of rapid progression of the lesion to current size in past 4-5 months. On intraoral examination a large swelling in the palatal region was clinically visualised extending roughly from the canine region to the first molar region. There are multiple missing teeth in the maxillary arch and a root stump seen in relation to tooth number 25 (figure 1). On further evaluation with an Electric pulp tester tooth number 23 and 24 showed negative response hence concluded to be non vital.

The intraoral periapical radiograph, occlusal radiograph and an orthopantamograph revealed a large periapical radiolucency in relation to upper left canine, 1\(^{\text{st}}\) premolar and extending upto 2\(^{\text{nd}}\) premolar region, even including the palatal bone (figure 2,3,4). Based on the history, clinical and radiographic diagnosis, a provisional diagnosis of extensive radicular cyst was made with a differential diagnosis of odontogenic keratocyst or ameloblastoma.
It was decided to perform the root canal treatment for the nonvital teeth. After complete oral prophylaxis, rubber dam was applied. Following access cavity preparation, necrotic tissue was removed, working length determined, and cleaning and shaping of the canals were done. Obturation was done with lateral condensation of gutta-percha (figure 5,6,7).

Periapical endodontic surgery was planned after the conventional endodontic treatment and a Computed Tomography scan was done in order to assess exactly the extent of the lesion. As from the CT scan, the cystic radiolucency was involving parts of the palatal bone, even parts nasal bone and extending till the first molar region posteriorly (figure 8).
The surgery was performed under General Anaesthesia, where complete enucleation of the cyst (figure 9,10,11) was performed followed by root resection, retrograde preparation and root end filling using Biodentine™ (Septodont, Saint-Maur des Fosses, France) (figure 12,13).
The resulting cavity was packed with iodoform gauze impregnated with bacitracin ointment. The packing was replaced during the recall visits biweekly for three months following the initial surgery (figure 14).

Histopathology revealed, cystic cavity lined by stratified squamous epithelium of 8-10 cell layer thick and pallisading and polarized basal cells with few areas of parakeratinisation. The underlying connective tissue consisted of infiltrate of chronic inflammatory cells suggestive of infected Odontogenic Keratocyst (figure 15).

DISCUSSION

The OKC was first described by Phillipsen (1956). This lesion is classified by the World Health Organization as a developmental, noninflammatory odontogenic cyst (Kramer et al. 1992) arising from rests of dental lamina cells (Tsukamoto et al. 2001). Histologically, it is characterized by a uniform epithelial layer with a corrugated para-keratinized luminal layer and a prominent basal-cell layer (Ali & Baughman 2003).

OKC is one of the most commonly encountered odontogenic entities and requires special consideration because of its known aggressive behavior and tendency to recur. Clinical evidence of its known aggressive behavior is supported by reported cases penetrating the cortical bone and involving adjacent soft tissues, as well as extending to the skull base from the mandible or to the orbit and infratemporal fossa from the maxilla.

The common radiographic features are unilocular or multilocular well-circumscribed radiolucent lesions surrounded by a thin sclerotic border. When unilocular radiolucent OKC is encountered, it is difficult to distinguish it from other odontogenic or nonodontogenic cysts; when the multilocular variant is present, it is difficult to differentiate it from other odontogenic or nonodontogenic neoplasms (e.g., ameloblastoma, myxoma). OKCs can be located at the periodontal region of teeth, thus resembling periradicular cysts; or they may envelope the crowns of unerupted teeth, mimicking dentigerous cysts; or they can be sited between the roots of the teeth, simulating lateral periodontal cysts or lateral radicular cysts; or they can be located at the maxillary midline, suggestive of a nasopalatine duct cyst. Radiographically, large OKCs in the mandible can be indistinguishable from cystic ameloblastomas. One radiographic feature that may suggest the diagnosis of OKC is that OKCs tend to grow in an anterior-posterior direction within the medullary cavity of the bone without causing obvious bone expansion. Conventional radiographic imaging, such as panoramic and intraoral periapical films, is usually adequate to determine the location and estimate the size of a OKC. With larger lesions, CT scans are required. CT scans of OKCs show 3D extension, sharply defined borders, and contents of water.
In a systematic review, Blanas et al. (2000) analysed treatment methods and the associated prognoses of OKC. The authors reported that the literature concerning OKC was limited to retrospective consecutive case series. Surgical approaches based on complete resection showed the lowest recurrence rate (0%) but the highest morbidity rate (including discomfort, pain and paraesthesia). Simple enucleation was reported to have a recurrence rate of 17–56%. Simple enucleation combined with adjunctive therapy, such as the application of Carney’s solution or decompression before enucleation, was reported to have recurrence rates of 1–8.7%.16,17

In the current case, an extensive lesion was treated with surgical endodontics. After completion of root canal therapy, enucleation of the cyst was carried out followed by root end resection and retrograde filling with Biodentine. A critical step in the endodontic surgery is to identify any areas of possible leakage, such as an accessory canal, an isthmus, dentinal cracks, a gap between the existing root- canal filling and the pulp canal wall, and areas of the root canal that have not been negotiated or filled by the orthograde approach. Biodentine™ was developed by Septodont’s Research Group as a new class of dental material which could conciliate high mechanical properties with excellent biocompatibility, as well as a bioactive behavior.8,18,19

Laurent et al. were the first to show the promising biological properties of Biodentine on human fibroblast cultures. The main benefits of Biodentine over other products are the bioactive behavior, reduced setting time (compared to MTA), better mechanical properties, sealing ability and higher push out bond strength.20

Enucleation of the OKCs followed by open packing has been suggested as another conservative method of surgical treatment. The current patient was treated with enucleation followed by open packing. The resulting cavity was irrigated with mixture of normal saline and chlorhexidine gluconate for a full-of glass and also packed with iodoform gauze impregnated with bacitracin ointment to minimize the risk of recurrence in each recall visits. Regular recall visits are required to ensure cyst involution and the opportunity for appropriate treatment should be an evidence of recurrence. The benefit of this protocol lies in the minimal surgical morbidity. In addition, associated structures such as the inferior alveolar nerve and developing teeth are less vulnerable to the damage.2,10,11

CONCLUSION

The radiographic and clinical characteristics of OKCs are not pathognomonic signs and may lead to a difficult diagnosis especially when this lesion is adjacent to teeth with nonvital pulps or inadequate root fillings. When an orthograde treatment is not feasible, or is ineffective, a retrograde treatment with a biopsy is recommended because an OKC in an unusual location may simulate a periapical lesion.

REFERENCES