CASE REPORT

Multiple Odontogenic Keratocyst, Surgical Management: 14-Years Follow-up

Soundrapandian Karthikeyan[1], S Avinash Reddy[2], Sidhartha S P Behera[3]

ABSTRACT

Odontogenic cysts are considered as non-neoplastic benign lesions. Among the cysts, keratocyst odontogenic tumor (KCOT) is an intraosseous tumor characterized by parakeratinized stratified squamous epithelium and a potential for aggressive, infiltrative behavior, and for the possibility to develop carcinomas in the lesion wall. Thus, the aim of this study was to describe a clinical case of KCOT in a young patient and discuss the treatment alternatives to solve this case. A 13-year-old female was referred for the treatment of a giant lesion in her left side of the mandible. After the biopsy, a diagnostic of KCOT was made, and the following procedures were planned for KCOT treatment. After 14 years, no signs of recurrence were observed. In conclusion, this treatment protocol was an effective and conservative approach for the management of the KCOT, enabling the reduction of the initial lesion, the preservation of anatomical structures and teeth, and allowing quicker return to function.

Keywords: Keratocystic tumor, Odontogenic cyst, Odontogenic tumor, OKC.

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INTRODUCTION

Keratocyst odontogenic tumor (KCOT) is a benign neoplasm defined by the World Health Organization as a benign unicystic or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and a potential for aggressive, infiltrative behavior, and high recurrence rate.[1] Radiographically, is the most often unilocular or multilocular well-circumscribed radiolucent lesion, surrounded by smooth or scalloped margins with sclerotic borders. KCOT has presumably arisen from cell rests of the dental lamina or from offshoots of the basal cell layer of the oral epithelium. The differential diagnosis involves odontogenic cyst, dentigerous cyst, and ameloblastoma.[2-4] A perceptible number of cases is diagnosed incidentally during regular dental inspections.[5] It accounts for approximately 12–14% of all odontogenic cysts of the jaws. Some studies report recurrence rates for intraosseous odontogenic keratocysts ranging from 5% to 62%, although several studies examining a large number of cases indicate a recurrence rate of approximately 20–30% up to 10 years after treatment, though it is more common during the first 5–7 years.[5-7] The potential high risk of recurrence and the long intervals described in the literature explain the necessity for long-term follow-up. The conservative treatment for this pathology includes marsupialization, decompression, enucleation, and curettage. More aggressive approach is based on osteotomy, lesion resection, use of chemical agents like Carnoy's solution, and cryotherapy with liquid nitrogen or peripheral osteotomy.[8] Here, we report a clinical case in which KCOT was diagnosed and successfully treated with conservative marsupialization and the subsequent enucleation, with 14 years of follow-up.

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CASE REPORT

A 13-year-old female accompanied by his parents came with chief complaints of multiple swelling of the face. No systemic alterations and pain were reported. The patient denied use of alcohol and smoke. Clinical examination revealed intra- and extra-oral swelling, good plaque control, and no periodontal disease. A panoramic radiography and a computed tomography (CT) scan were requested to the patient. The panoramic image showed multiple radiolucent areas surrounded by a radiopaque halo around the unerupted teeth, blocking their eruption (Figure 1). Based on clinical and radiographic features, the diagnosis of a KCOT was suggested, and the treatment possibilities were discussed with the patient and parents. Initially, an incisional biopsy was performed to confirm the diagnosis. The

pathological findings were associated with KCOT and showed fragmented connective tissue, intraosseous cavity composed mainly by stratified squamous lining with acanthosis, and discontinuity of the subjacent connective tissue. In the alveolar bone was observed resorption areas with apposition areas characterized by reversion and/or incremental lines. No signs of malignancy were detected. The patient was informed about the diagnosis, and a minimally invasive surgery to KCOT removal was proposed and accepted by the patient. Written informed consent was obtained before initial treatment. The treatment of the KCOT started with the marsupialization of the lesion by excision of the overlying mucosa and opening of the window of 1.5 cm size into the cystic cavity and suturing the cyst lining to the oral mucosa. The cavities in the anterior part of the mandible were kept open using a syringe by the patient and suturing in place a short piece of drain. This approach allowed freely cyst drain. During the period of lesion decompression, the patient and family were instructed to maintain good oral hygiene and to clean the area with saline solution. At 3 months, after the marsupialization for decompression, the second approach was performed to remove the KCOT. Enucleation followed by the KCOT removal was performed. After 14 years, CT scan showed no evidence of lesion recurrence and great bone healing, and the second molar showed normal function with pulp vitality (Figure 2).

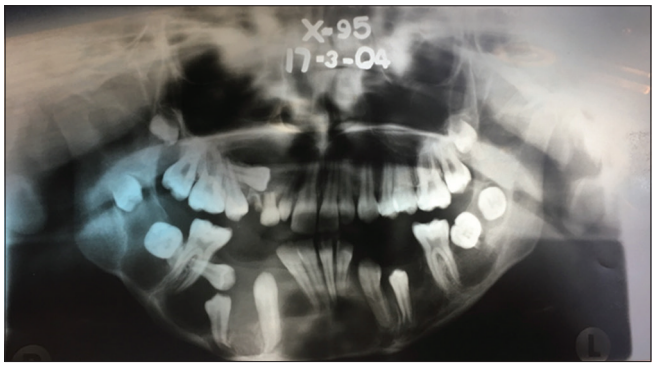

Figure 1: Pre-surgical panoramic radiographic image

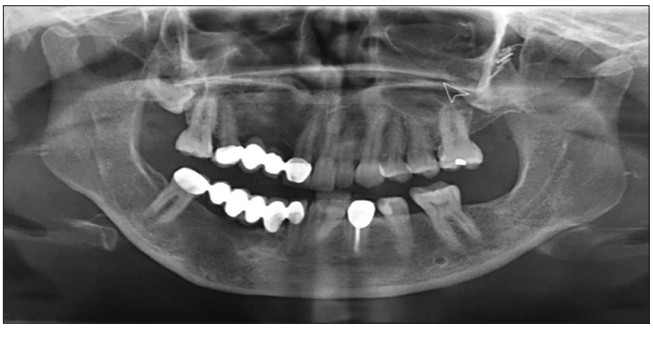

Figure 2: Panoramic radiographic image demonstrating bone formation after a 14-year observation period

DISCUSSION

The term keratocyst odontogenic was first introduced in 1956 by Philipsen.[9-11] The odontogenic keratocystic tumor, formerly known as the KCOT, received its new designation to better characterize its neoplastic nature.[12] It is a benign developmental odontogenic tumor with many distinguishing clinical and histologic features including (i) a potential for locally destructive behavior, (ii) a relatively high recurrence rate, and (iii) designation as a consistent finding in the nevoid basal cell carcinoma syndrome or Gorlin syndrome. This cystic lesion most frequently presented in the second, third, and fourth decades of life at the posterior mandible of male patients, which corroborate our findings. This could be elucidated by the hypothesis that KCOTs originate from the basal layer of oral epithelium, or the remnants of the dental lamina and these epithelial residues may be associated to the formation of a KCOT.[13] It accounts for approximately 12–14% of all odontogenic cysts of the jaws. It has a high recurrence rate with reports ranging from 20% to 60%.[14,15]

Radiographically, KCOT can appear the unilocular or multilocular lesion. Small unilocular cysts can be confounded with periapical, dentigerous, lateral periodontal cysts, or gingival cysts, and larger unilocular KCOT can mimic ameloblastoma. A unilocular KCOT appears as a well-defined radiolucent lesion. Root resorption, extrusion of erupted tooth, or displacement of impacted erupted teeth may be evident. Histologically, the KCOT is characterized by a uniform, usually corrugated parakeratinized epithelium, thick cells presenting a flat basal surface lining, called the fibrous wall. The histology of the KCOT is pathognomonic: The cystic cavity is lined with a thin layer of connective tissue covered by orthokeratinized or parakeratinized stratified squamous epithelium.[12-16] Pindborg *et al*.[13] established the following histopathologic criteria for this lesion: (i) The epithelium lining is usually very thin and uniform in thickness with little or no evidence of ridges, (ii) there is a well-defined basal cell layer, and the component cells are cuboidal or columnar in shape and often in a palisaded arrangement, (iii) there is a thin spinous cell layer which often shows a direct transition from the basal cell layer, (iv) the cells of the spinous cell layer frequently exhibit intracellular edema, (v) keratinization is predominantly parakeratotic, but it may be orthokeratotic; (vi) the keratin layer is often corrugated; and (vii) the fibrous cyst wall is generally thin and usually uninflamed. A recent study[16] evaluated 112 mandibular KCOTs in 109 patients who had undergone surgical enucleation. The authors showed a recurrence rate of 28 tumors treated by this technique, where seven

had multiple recurrences. However, another study[12] sustains that 60% of KCOTs recurrences are from treatments using only simple enucleation, especially due to the difficulty in removing all cystic epithelium, and due to the involvement of some anatomical structures as the maxillary sinus, dental roots, and inferior alveolar canal, which could be facilitated by decreasing the pre-enucleation cystic volume. For these reasons, it is important to emphasize the need for post-treatment follow-up for at least 15 years.

CONCLUSION

The use of the marsupialization technique followed by enucleation for the treatment of KCOTs was an effective and conservative approach for the management of the KCOT, enabling the reduction of the initial lesion, the preservation of anatomical structures and teeth, and allowing quicker return to function. No signs of recurrence after 14 years were observed.

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