Conservative Approach to the Treatment of Keratocystic Odontogenic Tumor

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ABSTRACT

The odontogenic keratocyst, also known as the keratocystic odontogenic tumor, is an aggressive, intraosseous lesion of odontogenic origin that presents a high rate of recurrence. Treatment modalities include aggressive surgical procedures and more conservative approaches that significantly influence the lesion's recurrence potential. The purpose of this case report was to demonstrate a conservative approach in the treatment of an extensive keratocystic odontogenic tumor, located in the mandible's posterior region, using decompression and enucleation.

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The odontogenic keratocyst (OKC) is now designated by the World Health Organization (WHO) as a keratocystic odontogenic tumor (KCOT) and is defined as "a benign uni- or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized, stratified, squamous epithelium and potential for aggressive, infiltrative behavior."¹ It occurs more frequently in the second, third, and fourth decades of life (54%), with some reports in the first decade of life.²⁻⁴ In a total of 183 KCOTs found, both genders were affected almost evenly³ (male-to-female ratio=1.05:1), more frequently affecting the posterior region of the mandible and associated with an impacted third molar.^{3.5-7} Radiographically, it is shown

to be similar to other lesions; it can cover extensive unior multilocular areas, and has well-defined edges.⁸

The literature refers to extremely variable recurrent rates of OKC, occurring from 62% to 0% according to the type of treatment selected.^{5,9-11} In most cases described, the recurrence potential is higher in the first 5 years, which does not discard the possibility of occurrence in longer periods.^{8,10} Stoelinga12 justifies the high recurrence for 3 reasons: (1) incomplete removal of the epithelium lining the cyst; (2) the presence of satellite cysts; and (3) development of a new cyst. In studies by González-Alva et al in 2008³ that analyzed 183 KCOTs, a recurrence rate of approximately 13% was found. All of the recurrent tumors were parakeratinized. In 3 of the 5 patients whose tumors recurred more than once, the tumors were associated with nevoid basal cell carcinoma syndrome.3 The same association was found by Habib et al.⁴

The treatment modalities include more conservative or aggressive procedures and even a combination of both types. The surgical treatment options consist of:

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- 1. curettage with high recurrence rates¹³;
- simple enucleation, which is difficult to perform due to the OKC lining's thin and friable layer, with high recurrence rates ranging from 17% to 56%²;
- 3. enucleation followed by peripheral ostectomy, which consists of removal of a layer of bone tissue via drills or the application of chemical substances (Carnoy's solution and liquid nitrogen cryotherapy), which promotes a safety margin through necrosis of the epithelial remains or satellite cysts, thus reducing the possibility of recurrence^{5,7,8,12-14}; and
- 4. marginal or segmentary bone resection, in which the former preserves the continuity of the bone structure and the latter removes the block of bone entirely, requiring reconstruction procedures. In spite of presenting low recurrence rates, this type of treatment causes esthetic and functional alteration to patients; therefore, it is more commonly used in extensive recurrent lesions.^{9,13}

Decompression or marsupialization are presented as more conservative options in the treatment of OKC, as they diminish the size of the cyst and facilitate its complete removal in a second time of enucleation surgery.^{5,7,8,14} Pogrel and Jordan⁷ reported 10 cases of OKCs treated exclusively with marsupialization, without signs of recurrence in up to 2.8 years.

Adequate treatment should reduce the recurrence potential and concurrently diminish surgical morbitidy.^{2,6} Therefore, several factors must be analyzed, such as the: patient's age; cyst's size and location; soft tissue involvement; previous treatment history; and histological variant of the lesion.¹³

The purpose of this case report was to demonstrate a conservative approach in the treatment of an extensive keratocystic odontogenic tumor, located in the mandible's posterior region, using decompression and enucleation.

CASE DESCRIPTION

Without complaints of pain, an 11-year-old boy sought the ambulatory surgery clinic of the Araçatuba School of Dentistry, São Paulo State University, Araçatuba, São Paulo, Brazil, with a panoramic radiograph that had been requested by an orthodontist. The presence of an extensive radiolucent area, with its entire outline delimited by a radiopaque margin, was verified. It covered the mandibular left first and second molars, angle, ramus, coronoid process, and mandibular left condyle, with displacement of the left mandibular third molar to the condyle (Figure 1). The lesion presented as completely asymptomatic, without altering shape. The teeth involved showed pulp vitality, and the mandibular left third molar was retained.

Under local anesthesia (2% lidocaine with adrenalin 1:100.000), puncture aspiration was performed with a 20-ml syringe, which showed evidence of the presence of a whitish liquid. Immediately afterward, cystic de-



Figure 1. Initial panoramic radiograph. A mandibular left cyst can be seen covering the region of the first and second molars angle, ramus, coronoid process and mandibular condyle, with displacement of the third molar to the condyle.

compression was performed by means of an elliptic incision and removal of mucosa, bone, and cystic capsule, favoring the placement of a rigid rubber drain fixed to the mucosa with a suture made with nonabsorbable thread, and kept in place for 14 days.

The removed material was sent for histopathological examination, and the diagnostic hypothesis of a KCOT was confirmed. Microscopically, the thin cyst wall was shown to be without inflammatory infiltrate. The epithelial limiting was composed of a uniform lining of 6 to 8 cells, a palisaded basal layer of cuboidal/columnar cells, and the presence of a corrugated layer of parakeratin on the epithelial surface. Frequently, the epithelialconnective tissue junction was flat, though the surface roughness was imperceptible. The detachment of the epithelial limiting from the fibrous capsule was commonly observed (Figure 2a-b).

Antibiotic (amoxicillin) and analgesic (paracetamol) were prescribed in the immediate postoperative period. Instructions were given to irrigate the cavity 3 times a day with physiological solution and 0.12% chlorhexidine gluconate for the purpose of preventing secondary infection in the cystic cavity.

The patient remained under fortnightly ambulatory control and, later, monthly control, with panoramic radio-graphs taken in the first, third, sixth, and ninth months (Figures 3 and 4), in which significant reductions were observed in the lesion's size and the bone tissue's formation. At this time, 9 months after decompression, the patient received cystic enucleation followed by curettage. Under local anesthesia, the lesion was removed in a single piece together with the retained third molar and the mucosa was sutured with 6-0 nylon thread. Postoperative instructions included the prescription of antibiotic (amoxicillin), anti-inflammatory (nimesulide), and analgesic (paracetamol) medication. The part was sent for histopathological examination, which, in spite of showing evidence of significant alterations in the epithelium and cystic capsule, suggested the same result based on the previous biopsy.



Figure 2. First histologycal examination: A) The circle indicate the detachment of the epithelial limiting from the fibrous capsule (hematoxylin-eosin staining). B) the arrow (a) indicate the presence of a corrugated layer of parakeratin on the epithelial surface; the arrow (b) indicate presence of cuboidal cells in the basal layer; a flat epithelial-connective tissue junction and the detachment of the epithelial limiting from the fibrous capsule interface (hematoxylin-eosin staining). Second histologycal examination showed evidence of significant alterations in the epithelium and cystic capsule: C) The circle indicates a hyperplasic process with evident epithelial ridges with increasing number of cell layers (hematoxylin-eosin staining). D) The arrow indicate a superficial epithelial layer edema; the square indicate the presence of a mild inflammatory infiltrate and scattered fair-epithelial (hematoxylin-eosin staining).



Figure 3. Follow-up panoramic radiograph 3 months after the second surgery.



Figure 4. Follow-up panoramic radiograph 9 months after the second surgery.

The second histopathological exam showed a hyperplasic process forming evident epithelial crests; increase in the number of cell layers; presence of mild inflammatory infiltrate; and tight epithelial dispersion. The superficial layers of the epithelium were shown to be edemaciated (hydropic degeneration; Figure 2c-d).

Clinical and radiographic follow-up was conducted on the seventh, 15th, 30th, and 90th postoperative days and, llater, every 6 months. The patient has remained under control for 7 years and has presented no signs of recurrence (Figure 5).



Figure 5. Follow-up panoramic radiograph 7 years after the second surgery.

DISCUSSION

The WHO'S reclassification of this lesion from cyst to tumor underscores its aggressive nature and should motivate clinicians to manage the disease in a correspondingly aggressive manner. The most effective treatments are enucleation supplemented with Carnoy's solution or marsupialization with later cystectomy. Future treatment may involve molecular-based modalities, which may reduce or eliminate the need for aggressive surgical management.¹

The conservative treatment of extensive OKCs via decompression or marsupialization techniques has been widely used by various authors.^{2,5,7,14-16} This technique consists of exteriorization of the cystic cavity, resulting in communication with the oral cavity.¹⁷ This causes reduced cystic pressure and bone formation on the cyst walls, facilitating its removal via a second surgical time, which is the principle used by the authors for treatment of the mentioned case.

According to Maurette, Jorge, and Moraes,⁵ the main advantage of conservative treatment would be preservation of bone tissue, soft tissue, and teeth and avoiding damage to the adjacent anatomic structures (inferior alveolar nerve). These are significantly important facts concerning young patients. Moreover, there is a reduction in costs, as hospitalization and reconstruction procedures with the use of either bone grafts or fixation materials become unnecessary.

Nevertheless, only a select group of patients can be submitted to this type of treatment by means of compression or marsupialization. This is because this technique requires exhaustive cooperation from the patient or family members due to the need for constant irrigation of the cavity and frequent returns to the ambulatory clinic.^{5,17} Although the patient in this report was a child, he was extremely cooperative and his parents were very dedicated and attentive.

One reason why patients abandon this type of treatment is the long period necessary for decompression, which may last up to 19 months.^{7,15,17} This is the time required for histological and immunohistochemical alterations to occur in the epithelial linings of the OKC, such as epithelial hyperplasia, inflammatory infiltrate, and absence of cytokeratine 10 expression.¹⁵ In the reported case, the compression period was 9 months, the minimum period for significant cellular alterations to become established.

The patient follow-up period submitted to OKC treatment has not been clearly defined, as there are reports of recurrence in long periods.⁸ It is known that a large number of the recurrences are concentrated in the first 5 years.^{8,10} Moreover, the recurrence of KCOT may not necessarily be the result of surgical management; instead, it is probably a reflection of the nature of the lesion itself. Thus, long follow-up periods are suggested for the tumor.³

Therefore, decompression followed by enucleation is a good treatment option for large OKCs, minimizing the possible negative factors of aggressive surgical procedures with morbidity and costs.

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