Conservative Treatment Protocol of Odontogenic Keratocyst: A Preliminary Study

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Purpose: The objective of this study was to report our experience with the treatment of 30 odontogenic keratocyst (OKC) patients with a conservative treatment protocol based on decompression with reference to the recurrence rate.

Patients and Methods: Twenty-eight patients (19 females, 9 males) with 30 OKCs attended the OMS Department of the Piracicaba Dental School of Campinas State University between 1995 and 2003. Age range was 13 to 69 years (mean, 30 years of age). Initial biopsy was carried out in all patients and the OKCs were diagnosed after histological examination by the Oral Pathology Department. The cases were treated according to the treatment employed in this department, consisting mainly of decompression and curettage of the remaining lesion. The average follow-up for the 28 cases was 24.89 months (±9.74).

Results: The majority of the lesions (16 patients, 53.3%) occurred in the angle of the mandible and mandibular ramus. The most common histological pattern of OKC was parakeratinized (66.6%) and 13 of 28 patients presented impacted teeth associated with the lesion. The mean time for decompression was 9.27 months. Recurrence occurred in 4 patients (14.3%) with 4 OKCs. These patients were treated initially with decompression and curettage (2 cases), or with decompression only (2 cases). All the cases were monitored continuously with panoramic radiographies and clinical evaluations.

Conclusions: The treatment protocol for OKC based on decompression offers a conservative and effective option with low morbidity and similar recurrence rates to those reported in the literature. The systematic and long-term post-surgical follow-up is considered to be a key element for successful results.

The odontogenic keratocyst (OKC) is a cystic lesion of odontogenic origin, which is classified as a developmental cyst derived from the dental lamina. This lesion was first described in 1956 by Phillipsen¹ and it is well known for its high recurrence rate.² For this reason, extensive research regarding this lesion has been carried out over the last 48 years.¹ ³

The OKC involves approximately 11% of all cysts in the jaws and is most often located in the mandibular ramus and angle. This lesion can be associated, although not in all cases, with an impacted third molar. Radiographically, it appears as a unilocular or multilocular lesion with a scalloped contour. These characteristics are suggestive but not considered an unequivocal proof for the definitive diagnosis of OKC because other lesions may exhibit similar features.⁴

The rates of recurrence vary enormously, from a maximum of 62% to a minimum of 0%.⁵ The majority of recurrent cases occur within the first 5 years after treatment.⁵ ⁹ For this reason, most surgeons advocate complete removal with extension margins or meticulous curettage of the surrounding tissues.⁵ The enucleation alone is associated with the highest recurrence rates (range, 17% to 56%), usually when the cyst is removed in a fragmented fashion. To decrease the recurrence potential, various adjunctive therapies have been tried, including peripheral ostectomy or the use of Carnoy’s solution, cryotherapy, or electrocautery.⁵ ⁹

Decompression or marsupialization seem to be more conservative options in the treatment of OKC.¹⁰ ¹¹ Marsupialization was first described by Partsch in 1882¹² ¹³ for the treatment of cystic lesions. This technique is based on the externalization
of the cyst, through the creation of a surgical window in the buccal mucosa and in the cystic wall. Their borders are then sutured to create an open cavity that communicates with the oral cavity. This procedure relieves pressure from the cystic fluid, allowing reduction of the cystic space and facilitating bone apposition to the cystic walls.\(^{10\text{-}15}\)

Decompression and marsupialization are very similar techniques. The main difference between them lies in the creation of a surgical window in the oral mucosa and cystic membrane,\(^{14}\) and in using a cylindrical device\(^{11}\) (like the rubber of a dropper) or a surgical rigid drain to prevent mucosal closure. This is done with the objective of maintaining a continuous communication between the oral cavity and the interior of the cyst. The decompression technique allows the permeability of the cystic cavity because the union of the cyst epithelial wall with the mucous membrane results in the externalization of the lesion. In addition, the intraoral device facilitates the irrigation of the cavity. This helps avoid food impaction and microorganism accumulation in the area, which could lead to an undesired secondary infection. In addition, after the surgical intervention, the cystic covering tends to become thicker, which facilitates its complete removal in a second surgery.

The use of this technique is not new among possible OKC treatments. In 1971, Browne\(^{16}\) described marsupialization as a technique for the treatment of OKC. In 1976, Tucker\(^{14}\) first described the use of decompression and secondary enucleation as a first-line treatment option for OKC.

In 1991, Brøndum and Jensen\(^{11}\) reported a recurrence rate of 18% in 51 OKC patients during a 13-year period. Thirty-two of these patients were treated with decompression of the lesion. Of these cases, 8 presented recurrence of the lesion. Brøndum and Jensen\(^{11}\) described the use of cylinders of polyethylene fabricated by a drainage stem that are introduced into the cyst cavity and maintained for a period of 1 to 14 months. The patient was instructed to irrigate the cavity with a 0.12% chlorhexidine solution using a plastic syringe. Additionally, when achieving a significant reduction of the lumen—which can be confirmed through radiographic imaging—a secondary cystectomy was justified to prevent recurrence of the lesion.

Those who criticize the use of marsupialization\(^{5}\) or decompression for the treatment of OKC argue that this technique does not allow a complete removal of the whole cystic covering, which would lead to a continuation of epithelial proliferation and facilitate an increment of the recurrence.\(^{5}\) Brøndum and Jensen\(^{11}\) were not in agreement with this argument because they did not observe recurrences in patients treated by decompression with subsequent enucleation.

Several reports describe the use of decompression to decrease the size of the cyst, after which it is definitively enucleated.\(^{10\text{-}16}\) Use of these techniques alone is not reported commonly when a complete resolution of the OKC has been achieved.\(^{15\text{-}16}\)

Regarding the remaining epithelium after decompression of the lesion, August et al\(^{19}\) reported the differentiation of the OKC epithelium once treatment is carried out. Through histochemical analyses based on Cytokeratin-10 tests, August et al\(^{19}\) accomplished the preoperative identification of the lesion in 14 OKCs. After surgery, the same analysis was carried out again in the cystic epithelium to determine whether the marsupialization/decompression technique results in epithelial modulation, which is associated with lower recurrence rates. It was observed that 64% of the patients did not present Cytokeratin-10 in the epithelium analyzed, which shows the differentiation of this tissue, and therefore the lower rates of recurrence.

Pogrel and Jordan\(^{17}\) reported the use of marsupialization as a definitive treatment of OKC. In this study, 10 patients were treated exclusively with marsupialization and decompression, achieving resolution of the lesions with a recurrence rate of 0%.

The purpose of this study was to report our experience with the surgical treatment of 30 OKCs by the use of a defined protocol, based on decompression and posterior enucleation or curettage with reference to the recurrence rate.

### Patients and Methods

Twenty-eight patients (19 females, 9 males) with a total of 30 OKCs attended the OMS Department of Piracicaba Dental School at Campinas State University between 1995 and 2003. Basal cell nevus syndrome patients with multiple OKCs were not included in our trial. Two patients with 2 OKCs in different anatomic locations, without clinical features of Gorlin Syndrome, were included in the sample. In this study, the age range was 13 to 69 years (average, 30 years). All the OKCs were diagnosed by histologic examination by the Oral Pathology Department.

All lesions were discovered by radiographic images and the preoperative diagnoses of OKC were obtained with biopsied specimens, confirmed postoperatively by histopathologic report. Our treatment protocol for cystic lesions consists of carrying out the initial biopsy and decompression of the lesion on the same day if possible.

Under local anesthesia (2% lidocaine with epinephrine 1:200,000), an aspirative biopsy was made with a 10-ml syringe and #18 needle. A yellow, serous liquid or semi-solid content was obtained, matching the typical description of a cystic lesion content. Posteriorly, a #15 scalpel was used to carry out an elliptical inci-
sion in the attached gingival tissue of the alveolar ridge. Fragments of the cystic capsule, mucosa, and bone were taken and introduced into an iodoform solution for histologic analyses. A sterile rubber foam dropper was performed, fitted in the surgical windows, and fixed with non-absorbable suture (nylon 4.0). Rubber cylinders were used to allow permanent communication between the cyst and the oral cavity. The postoperative care included the use of paracetamol via oral for pain control. In addition, daily irrigation of the cystic cavity with saline solution and 0.12% chlorhexidine was carried out to prevent a secondary infection of the cystic cavity. Antibiotic therapy was indicated only in those cases in which the cyst presented an infection and was based on amoxicillin or clindamycin (in penicillin-allergic patients) orally. The irrigation procedure was made with a 20-ml syringe with no needle active point to prevent tissue injury. Irrigations were carried out 3 times/day with 60 ml of the irrigation solution. This was started the same day of the surgery. Rubber cylinders were removed after 2 weeks and daily irrigation was maintained for another 6 to 9 months. Careful monitoring was based on monthly panoramic radiographies and clinic visits to determine lesion size regression as an effect of decompression and bone formation (Fig 1).

Secondary curettage of the surrounding tissues was carried out after the decompression phase, once radiographic evaluation confirmed a size decrease of the lesion. Under local anesthetic, a horizontal incision was made in the alveolar ridge with a #15 scalpel. A mucoperiosteal flap was obtained with exposure of the cystic cavity. The secondary curettage of the cavity was carried out with a Lucas’s bone curette and the cavity was irrigated with saline solution. Finally, the flap was closed with 4.0 silk suture (Johnson & Johnson, Ethicon, Brazil). Postoperative care included oral paracetamol, celecoxib, amoxicillin or clindamycin (in penicillin-allergic patients). The use of topical 0.12% chlorhexidine solution twice a day was indicated for 2 weeks.

In those cases where treatment of the lesion consisted of enucleation only, a procedure similar to the one described above was used.

Patients receive follow-up with clinical and serial panoramic radiographs at 7, 15, and 30 days. Ninety days after the procedure is done, patients are monitored periodically every 6 months.

Results

Of 28 patients, diagnoses of OKC were more prevalent in white (57.1%) young (20–29 years) females (19 patients, 67.9%). Most of the lesions (16 lesions, 53.3%) occurred in the angle of the mandible and mandibular ramus. The most common histologic pattern of OKC was parakeratinized (70%), and 13 of 28 patients presented impacted teeth associated with the lesion. Five OKCs presented satellite cysts and had the pattern parakeratinized. Twenty OKCs (68%) were treated by decompression and curettage of the remaining lesion and 10 OKCs (32%) by enucleation and curettage only. The mean time for decompression was 9.27 months with 3 months being the smallest time of decompression and 14 months the longest time. Recurrence occurred in 4 patients (14.3%) with 4 OKCs. These patients were treated initially with decompression and curettage (2 cases), or with decompression only (2 cases). All recurrence cases were submitted to enucleation/curettage and peripheral osteotomies of the remaining bone cav-
ity. The average follow-up for the 28 cases was 2 years (24.89 ± 9.74 months).

Discussion

Many authors have shown the successful treatment of large OKCs using the technique of decompression and irradiation.\(^{10,11,15,16}\) This treatment does require a cooperative patient who will irrigate the cyst on a regular basis and will follow up regularly. For this reason, only a select group of patients may be suitable for this treatment.

The benefit of this treatment over more conventional approaches (enucleation, en bloc resection) lies in the minimal surgical morbidity. In addition, associated structures such as the inferior alveolar nerve and developing teeth are less vulnerable to damage.\(^{10,12,17}\)

The decompression and marsupialization techniques were based in the exteriorization of the cystic cavity and result in communication with the oral cavity.\(^{11,14}\) These procedures relieve pressure of the cystic fluid, allowing shrinkage of the cystic space and the apposition of bone to the cystic walls. Several reports describe the use of the decompression technique to decrease the size of the cyst, after which it was definitively enucleated. The use of marsupialization for the treatment of cystic lesions is not new and was originally described by Partsch in the late 1800s.\(^{12,15}\) However, it is not common and there is relatively little in the literature on the use of this technique alone for carrying out the complete resolution of OKC.\(^{11,15,17}\)

In our sample, recurring lesions were small in dimension. The secondary treatment, based in curettage associated to the peripheral ostectomy, has been accomplished in all of the cases, reducing the chance of new recurrences.

The main advantage of the conservative treatment is the preservation of bone structure, woven soft and teeth associated OKC, fact that it is covered of great importance if we consider that most of the patients is young. These procedures are less traumatic for the patient, eliminating medication and hospitalization expenses, and in most cases, avoid the need to accomplish reconstruction through grafts or extensive reconstructions.

In most of the cases in which the recession is the elected treatment, the need of accomplishing the reconstruction of the jaw through grafts of autogenous bone is imperative. Usually these reconstructions are accomplished in a second surgery, which translates into larger discomfort for the patient, and increase of the morbidity, increments in the costs of the treatment, and time of recovery, among others. Additionally, in these cases, there can exist a need to put on some type of reconstruction plate. This adds possible complications with the use of rigid fixation material such as the exhibition of the plate, dehiscence and infection, among other complications.

<table>
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<th>Author(s)</th>
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Time of duration of the decompression treatment (1 to 14 months) is one of the disadvantages of this technique. In fact, this is one of the main causes of abandonment of the treatment by the patient because of loss of interest in proper irrigation treatment and attendance of periodic controls. In spite of being a technique that requires prolonged postoperative treatment and special considerations (like the ones mentioned above), and even a second surgical procedure in order to curette the remaining cystic cavity, it is a technique that allows the professional to offer the correct treatment and save hospital expenses that would increase with other, complicated procedures that require general anesthesia and hospitalization.

The technique is then an ideal alternative procedure for the treatment of odontogenic keratocysts that, in addition to being conservative (if compared with the enucleation technique), is well-adapted to the Latin American reality, making the “cost-benefit” of the technique one of its more important advantages.

The recurrence rate observed among our sample was 14.3%. In comparison with other important published studies\(^4,9,11,16,25-40\) (Table 1), we obtained re-
sults within the average with a more conservative approach. If we sum up the advantages of a conservative approach, like the one we suggest, versus a more radical treatment, the decomposition treatment protocol stands up as an equally effective, cheaper, and simpler procedure for the treatment of OKC if the patient can be closely monitored and periodically evaluated (clinically and radiographically).

The mean time of the follow-up in our sample was 24.89 months (±9.74), a relatively short time, as described by Stoeilinga and August. These authors explain the necessity to observe a long follow-up to consider a complete resolution of the OKC. In 2003, August et al examined the nature of the cyst lining and after decompression with cytokeratin stains and reported positive cytokeratin-10 staining in the predecompression biopsy and negative cytokeratin-10 stains in the postdecompression specimen. In 3, 6, 9, and 12-month samples, a return to more normal oral epithelium with 9 months of decompression treatment was indicated.

Several studies suggest that the largest number of recurrences of OKC occur during the first 5 years after the initial treatment period (about 70%). For this reason the annual radiographic control of these patients is recommended for an undetermined time.5,23,24 We conclude that OKC treated with decompression presents similar recurrence rates to those reported previously.4,9,11,18 This shows that this method of treatment for OKC offers a conservative and effective option with low morbidity and similar recurrence rates. Close follow-up and frequent control is important, however, for successful treatment.

References
33. Eversole LR, Sabes WR, Rovin S: Aggressive growth and neo-