The Treatment of Odontogenic Keratocysts by Excision of the Overlying, Attached Mucosa, Enucleation, and Treatment of the Bony Defect With Carnoy Solution

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Odontogenic keratocysts (OKCs) are known for their propensity to recur. The rate of recurrence is probably largely depending on the mode of treatment used. Unfortunately, there are only a few reliable studies available from which meaningful conclusions can be drawn with regard to the best possible treatment strategy.† The main drawback of most studies is the retrospective nature of the study, the variable follow-up time, the lack of information about the percentage of patients available for follow-up, and the lack of details about the actual surgery. Many papers are written by pathologists who obviously are depending on the written and often incomplete notes of surgeons, hence the link with the clinical parameters is often based on less than optimal information. It is the aim of this article to present a rational treatment protocol based on clinical and histologic studies.

Etiology and Pathogenesis

To understand the clinical presentation of OKCs, and their propensity to recur, it is necessary to have some knowledge of the etiology and pathogenesis of OKCs. The etiology is probably closely related to the development of the dental lamina and in particular remnants of it after this organ has served its purpose. OKCs that occur in the dentate areas of both the maxilla and mandible probably derive from those remnants. However, one must realize that epithelial islands that are derived from the dental lamina are mainly found in the gingiva and probably in the periodontal ligament. This explains the often seen lateral periodontal or lateral follicular presentation of these cysts. It is not known why keratocysts develop from such epithelial residues. It is also not clear why a keratocyst develops from one such epithelial island, while others remain dormant. The clinical implication may be, however, that if one removes such an OKC some of these epithelial residues may be left behind which may later give rise to a new keratocyst.

The frequently seen presentation of keratocysts in the ascending ramus of the mandible is less easily explained by this theory. Stoelinga and Peters‡ first pointed toward the course of the dental lamina forming the third molar and the unlikeliness of possible remnants or offshoots of this part of the dental lamina to be located in the mucosa posterior to the last molar. Yet, there is plenty of evidence that the majority of epithelial islands, as found in the wall of OKCs, are in fact located in the mucosa that is overlying the OKC and attached to it. This is the reason why it is thought that offshoots of the basal layer of the epithelium of the oral mucosa may also be involved in the etiology of OKCs.§,∥

Whatever the causative epithelial source may be, epithelial islands and/or microcysts are found in approximately 50% of the cases in the overlying mucosa. It is, therefore, of paramount importance to locate the area where the cyst is attached to the mucosa and to excise that part of the mucosa, preferably in conjunction with enucleation of the cyst. In failing to do so, in approximately 50% of cases one will leave behind possible sources of recurrent OKCs, or better to say newly formed cysts.

Clinical Presentation

One of the problems when discussing keratocysts is that there are in fact 2 clinical types. The first type is the commonly, relatively small, unilocular cyst in
the dentate area that often presents as a lateral periodontal or lateral follicular cyst. The clinician has no way of knowing in advance whether the lesions are to be considered an OKC. Excision of the attached mucosa is, therefore, usually not performed and most often the cysts are just enucleated. A prospective study including 82 OKCs showed that in approximately 40% of the cases a preoperative diagnosis was not made because of the specific presentation of the cyst. Recurrences will, therefore, be unavoidable but will present themselves in an area that is easily accessible. The second type is the frequently large, often multilocular or multilocular cyst in the posterior maxilla or angle and ascending ramus of the mandible. In these locations a high degree of alertness should be present and aspiration biopsies may be performed to ascertain the diagnosis. The differential diagnosis in all these cases should include unicystic ameloblastoma. Treatment of these 2 lesions should not be all that different. An algorithm on how to approach cystic lesions of the jaws has been presented elsewhere.

**Analysis of Nature of Recurrences**

Studies on recurrent OKCs have shown that in almost 100% of the cases epithelial islands and or microcysts are present in the overlying, attached mucosa. No microcysts were found in the surrounding bone when block resections were carried out on 5 recurrent OKCs. Therefore, this author is very critical of proponents of radical resections including continuity resections of the mandible or maxillectomies. To the best of my knowledge no studies or even case reports are available to prove that microcysts do occur in the bone surrounding the OKC. For this reason there is very little reason to perform these resections or to freeze the bone over the full thickness of the mandible.

If one wants to eliminate possible recurrences after treatment of an OKC it is of crucial importance that one realizes where these recurrences originate from. Considering the histopathology of the OKC with the active basal layer of its epithelial lining, there is no doubt that recurrences may arise if parts of the lining are left behind. All efforts should, therefore, be aimed at proper enucleation and elimination of possible remnants of the cyst wall in case the cyst ruptures and has to be removed piecemeal.

There is also the possibility that microcysts are present in the connective tissue of the cyst wall and that these are left behind after enucleation. The likelihood of this phenomenon is rather remote because a prospective study has shown that these microcysts are only sporadically found elsewhere in the cyst wall. The majority of islands and microcysts are to be found in the mucosa to which the cyst is attached. Some keratocysts show active budding of the basal layer of the epithelial lining that may reach to the periphery of the connective tissue wall and, therefore, may also be the source of a true recurrent cyst.

The third reason for a recurrent OKC is the development of a new keratocyst from an epithelial island or microcyst left behind in the mucosa. It is the author’s opinion that this probably is by far the most likely reason because most clinicians are keen to remove OKCs with great accuracy because they are aware of the tendency toward high recurrence. Even in a case where one has excised the overlying attached mucosa, this may still be the reason for a recurrence because it is not macroscopically visible whether one has included the whole area where these clusters of islands occur. In fact, the author has described a few recurrences showing epithelial islands in the overlying, attached mucosa, despite the fact that the mucosa was excised at the first operation.

**Rational Surgical Approach to Keratocysts**

Because it is not the aim of this article to explain the possible diagnostic procedures or to discuss the differential diagnosis, the reader is referred to the appropriate literature. When considering removal of a keratocyst, however, it is important to keep in mind the 3 possible reasons why an OKC could recur (explained above). Therefore, treatment should aim at elimination of possible vital cells left behind in the defect. Whether they are from the original lining or derived from microcysts in the wall, they are bound to be located rather superficially in the defect. For this reason a mild, not deeply penetrating, cauterizing agent such as Carnoy’s solution should be enough to do the job. In case the cyst was multilocular, care should be taken to eliminate the bony septae as to assure proper treatment of the whole bony cavity with Carnoy’s solution. In case the cyst has penetrated through the lingual or buccal cortex, one may use electrocauterization to avoid a recurrence in the soft tissues. Identification and retraction of the lingual nerve is recommended if the area is close to this structure.

Elimination of the epithelial islands and microcysts located in the overlying, attached mucosa should be assured by excising this part of the mucosa. For that reason the area needs to be identified. In the ascending ramus of the mandible it is almost always located at the anterior aspect of the retrotrigonal area. It can be palpated or defined using a needle. In the posterior maxilla one must assume that the OKC is attached to...
the thick mucosa of the tuberosity and one should include that part in the excisional biopsy. Preferably, the cyst with the attached overlying mucosa should be removed in 1 piece (Fig 1). That presents the pathologist an opportunity to examine that area for the presence of epithelial islands and microcysts. Detailed descriptions on how to perform these operations for both the maxilla and mandible are presented elsewhere.5,15

The overriding consideration of this approach is to do as little damage as possible and yet to reduce the chances of recurrence as much as possible. OKCs in the dentate area that were not preoperatively diagnosed as such should be followed up without drastic second interventions until a recurrence has become apparent. These recurrences are always amenable for relatively simple surgery without significant side effects.

Follow-Up

The recommended follow-up for OKCs is once a year the first 5 years postoperatively. The literature suggests that most recurrences will present the first 5 years after primary treatment. Because recurrences or newly developed OKCs may also present late, a follow-up once every 2 years thereafter seems a reasonable policy.5 Of course, much depends on the suspected cooperation of the patient and on geographic and socioeconomic factors. Proper information on why it is important to keep the patient under control is important as well. It is the opinion of this author that drastic operations, such as continuity resection, are not warranted because of patient-bound factors. The described method gives rise to a very low recurrence rate with little or no permanent morbidity.5

Recurrences

Recurrent OKCs may present as unilocular lesions but also often present as a multilocular lesion or even a separate unilocular lesion (Fig 2). The latter strongly
suggests that parts of the lining have been left behind. There is, however, very little systematic research performed on the presentation and histology of recurrences. This author in the late 1960s performed marginal block resections on 5 recurrent OKCs to ascertain their biological behavior.8 In none of these cases was there any sign of microcysts hidden in the surrounding bone. Instead, in all cases clusters of epithelial islands and microcysts were seen in the overlying attached mucosa (Fig 3). Since that time we have never treated recurrences by block resections, but just removed them with the overlying attached mucosa. Of the 9 patients with a recurrence included in the prospective study, 3 were located in the angle of the mandible or ascending ramus and primarily treated with excision of the overlying mucosa, while the defect was treated with Carnoy's solution. Microcysts and epithelial islands were always seen in the overlying attached mucosa.5 The recurrences occurred, despite this treatment, which probably points to incomplete excision of the overlying mucosa, because again these epithelial islands and microcysts were seen in the recurrent cysts.

In short, recurring OKCs deserve the same treatment as proposed for the primary OKC. There is no reason for any more drastic treatment and certainly no continuity resections.

**Alternative Treatment Options**

Considering the pathophysiology of the recurrent OKCs there are 3 alternative modes of treatment that deserve discussion. The first is decompression and marsupialization as proposed by Brondum and Jensen.16 This treatment can also be followed by removal of the cyst wall after this has changed by metaplasia.17,18 This treatment certainly has something to offer in case of large cysts particularly in old or medically compromised patients. When the overlying, attached mucosa is removed it also eliminates the source for newly formed cysts. There definitely is a need for long-term, preferably prospective studies to validate this treatment option. These studies should include histology of the tissue removed and also biopsies of the cyst wall that is left behind.

This author recently saw a dramatic recurrence as the result of such treatment (Fig 4). This certainly is not a completely safe method because one has no way of knowing how the active basal layer of the original cyst will behave.

The second option is the same as the described protocol but instead of the use of Carnoy’s solution, liquid nitrogen is used to freeze the defect.19,20 It remains to be seen whether this treatment provides the same good results as reported with Carnoy’s solution. The method is also much more complicated. It is also questionable whether it is needed to freeze deeply in the bone for reasons explained above. As the proponents of this method admit there might be indications in selected patients.20

The third option is block resection, with or without preservation of the continuity of the jaw.21,22 This is a drastic method, which creates considerable morbidity, particularly because reconstructive measures are necessary to restore jaw function and esthetics.

Resection can only provide adequate results when it includes the overlying attached oral mucosa. That entails, in most cases, contamination from intraoral to
extraoral. If the mucosa is not excised one can expect recurrences even in the bone grafts used to bridge the defect, as has been reported in several cases.

One has to wonder whether such aggressive therapy is warranted for a benign lesion that can be managed reasonably well with relatively simple means. This author has never found it necessary to use such treatment and finds it hard to justify carrying out continuity resections and complicated reconstructions in many patients to save a very few recurrences. The latter remains to be seen, however, because for this mode of treatment long-term follow-up studies are also lacking.

References


FIGURE 4. A, Computed tomography scan showing OKC in the left maxillary sinus, almost completely obliterating the sinus. B, A somewhat lower portion of the section showing destruction of the lateral sinus wall. C & D, Recurred OKC posteriorly and anteriorly (arrows). This OKC was primarily marsupialized 4 years previously. A year later the mucosa in the defect was removed, after which it healed by secondary intention. Unfortunately, there was no histology of the second intervention available.