REVIEW ARTICLE

Updates in the surgical management of odontogenic keratocyst

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Abstract

Odontogenic keratocyst (OKC) is the second most common odontogenic cyst in the oral cavity accounting for 10% of all cysts. Controversies regarding its tumor-like behavior, recurrences, and management have been the reason for this cyst to be exclusive unlike any other odontogenic cyst. In recent literature, the once named keratocystic odontogenic tumor has again been renamed as OKC, thus establishing it as a cyst with an aggressive behavior. In this article, we describe the conservative and radical treatment options for this tumor-like cyst.

Introduction

The odontogenic keratocyst (OKC) has been a controversial entity with regard to its terminology, recurrence tendency, and varied treatment options. OKC comprises 10% of all the odontogenic cysts.¹ Philipsen first described it in 1956 while Pindborg investigated its recurrence in 1963.²,³ In the 1960s, Gorlin and Goltz described the existence of a syndrome associated with multiple OKCs, basal cell carcinoma, and bifid ribs.⁴ OKC originates from the dental lamina or basal lamina. Controversies surrounding this cyst have been going on for a decade long when the WHO renamed it as keratocystic odontogenic tumor in 2005.⁵ This was due to its tumor-like aggressive behavior and presence of PTCH gene, which mainly existed in odontogenic tumors. However, again in 2017 edition of the WHO classification of odontogenic cysts and tumors reclassified it as a cyst.⁶ Studies showed that the lesion regressed following decompression, a quality not true to a neoplasm. Furthermore, it was concluded that the PTCH gene, which was found in 80% of OKCs due to which it was classified as a tumor, was found in the developmental cysts like dentigerous cyst.⁷,⁸ Clinical features of OKC include location mainly in the posterior body and ramus area of the mandible followed by posterior maxilla. Radiographically, it may present as a unilocular or multilocular radiolucency with sclerotic borders and may be associated with an unerupted tooth, more often an impacted third molar. Computed tomography and, in selected cases, magnetic resonance imaging are the imaging modalities which play a critical role in the diagnosis and management of OKCs. OKCs expansile nature is in an anteroposterior direction along the marrow spaces. This feature distinguishes it from an ameloblastoma. The cyst border becomes more irregular as the cyst size increases, thus making imaging and surgical approaches difficult. Recurrence of OKC has been attributed to the presence of a thin and friable epithelial lining, its complex location that is the posterior mandible and maxilla and presence of overlying epithelium, which is the probable precursor of OKC formation. Browne et al found that 25% of 139 odontogenic keratocysts that they studied, had satellite cysts.⁹ To overcome the recurrence of OKC, multiple treatment options have been put forth. Indications, advantages, and disadvantages of these treatment options have been described in this review.

Discussion

A simple enucleation alone may be an understated treatment plan. Recurrence rates for enucleation alone have been 9–62.5%.³,¹⁰
Varied treatment plans have been described in literature, which include decompression and marsupialization, enucleation, curettage/peripheral ostectomy, enucleation with chemical cautery by various agents such as Carnoy’s solution and liquid nitrogen, resection, either partial or segmental, and the recently introduced endoscopic enucleation. Treatment depends on the site, size, extent, and age of the patient. Controversies surrounding the treatment options have been the topic of discussion for many years. At present, no agreement exists concerning the best management of OKC. Brondum and Jensen proposed the terms decompression and marsupialization; while these terms are used interchangeably, they are performed differently. Decompression relieves the pressure within a cystic cavity, whereas marsupialization refers to relieving the intracystic pressure and converting it into a pouch by placing a drain into the cavity. These techniques have been indicated in large OKCs, proximal to inferior alveolar canal, erupting tooth buds, and cranial base. Marsupialization is often indicated in younger children and medically compromised patients. The previous studies were conducted which used decompression and marsupialization as a part of a two-stage operation called the Partsch II operation. These cysts were enucleated following marsupialization. Disadvantages of this technique include secondary operation and cooperation from the patient for regular follow-up. Hence, the selection of patients for these techniques is limited. Many authors have recommended against marsupialization as it leaves pathological tissue in situ, thereby leading to recurrence or rather persistence of the lesion. No published literature has proven that marsupialization leads to increased recurrence rate. On the contrary, Pogrel et al. did extensive studies where marsupialization was the sole treatment for resolution of 14 OKCs. Histological studies showed that the thin parakeratinized lining was converted to a thicker oral mucosa resembling lining. Recurrence rate for decompression has been recorded as 10%. A histological study showed the absence of bcl-2 protein in the healing bed of the cyst. Furthermore, in the immunohistochemical studies, the level of interleukin-1 reduced significantly following the marsupialization. Brondum reported a low recurrence rate in OKC following the use of a polyethylene drainage tube placed at the operation site and biopsied months before primary cystectomy. In the secondary stage, the cystic epithelium did not adhere to surrounding structures. No recurrences were observed although a limitation was the sample size of 12. In a follow-up study, Brondum et al. found two recurrences in 23 patients. Enucleation alone has shown to have the maximum recurrence rate due to retained fragments of the epithelial lining left behind and the presence of daughter cysts beyond the epithelial margins. Peripheral ostectomy involves scraping off bone using rotatory instruments to enable eliminating the residual cystic lining. The problem with this is that there is no quantitative measure of the amount of bone removed. Peripheral ostectomy is an outdated treatment option as an adjunct to enucleation. Voorsmit et al. compared enucleation alone and enucleation with excision of the overlying mucosa and treatment of the cyst cavity with Carnoy’s solution. Recurrence rates were found to be less, i.e., 2.5% in the second group, although, there was no confirmation if the lower recurrence rate was due to Carnoy’s solution or the excision of the overlying attached mucosa. In 1983, Bradley reported on the management of seven OKCs of the 25 jaw lesions with cryosurgery and observed no recurrences in an 8-year follow-up period. In 2001, 26 patients with OKCs treated with a combination of enucleation and liquid nitrogen cryotherapy were retrospectively evaluated by Schmidt and Pogrel. Findings suggested that liquid nitrogen decreased the recurrence rate for OKCs, particularly for recurrent lesions. Unlike Carnoy’s solution, liquid nitrogen maintains the osseous architecture to facilitate neoangiogenesis and hence new bone formation. Sensory recovery following liquid nitrogen application is significantly quick, due to its unique ability to devitalize bone in situ, while the inorganic framework remains intact. Cryosurgery lastly is indicated only for large, complex mandibular lesions, in which other treatment methods have been futile or management might involve vital structures such as the inferior alveolar nerve. Liquid nitrogen cryotherapy has more to offer an incompliant patient for definitive management. Resection has been one of the most aggressive treatment options of OKC; it may either partial or segmental resection. Morbidity associated with this procedure is dysfunctioning. Segmental resection creates a defect which, in particular, requires definitive reconstruction to restore jaw function and esthetics. Adequate results can be obtained only when resection includes the overlying attached oral mucosa. Although the recurrence rate has been cited as zero in case series, some authors believe this treatment to be too radical for a cyst. A marginal resection preserves continuity and a segmental resection violates continuity and requires additional reconstruction. A minimum of 5-mm bony margin is adequate to ensure satellite cyst removal and some authors feel that it is unnecessary to carry out segmental resection. Bataineh and Al Qudah reported marginal resections with 1-cm margin of surrounding bone carried out on 31 consecutive OKCs, which were followed up from 2 to 8 years with 0% recurrence rate. Transient inferior alveolar nerve paresthesia seems to be the only complication in resections. However, there are some absolute indications for resections, such as recurrent or extensive lesions, condylar involvement, pathologic fracture caused by an untreated cyst, or a malignant degeneration of OKC. Maxillary keratocysts are rare but have a greater potential to extend and cause destruction of the surrounding structures. Difficulty arises in planning treatment due to the presence of increased cancellous bone and the poorly demarcated anatomical boundaries that allow for rapid development of recurrence.
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progression of the OKC. Orbit, base of skull, and intracranial region are the most common areas for the maxillary OKC to extend into, as documented by Jackson et al. Marsupialization and resection are two main treatment options for OKC in maxilla. Enucleation using endoscopy has been previously described in literature for the management of OKC. Endoscopic technique holds a valuable advantage of avoiding facial incisions/transoral incisions. Reconstruction following endonasal techniques is also minimal. Endoscopic technique may be used for diagnostic purposes. Enucleation of large jaw cysts using an endoscope seems to be a possible option to replace all other modalities for managing non-malignant jaw cysts whatever its size. Advantages include better visualization, decreased intraoperative and post-operative complications, and ensuring complete cyst removal. Cysts of both the jaws are managed efficiently and safely with this procedure. Comparison of the endonasal techniques with transoral technique has not been undertaken in any of the studies. Furthermore, the shorter follow-up period has been a limitation.

Conclusion

A benign odontogenic cyst may be managed by relatively conservative treatment options, but it is worth questioning if radical treatment is warranted for an aggressive lesion like the OKC. Multiple treatment strategies have been described; the management of OKC ultimately depends on its characteristics such as site, size, extent of the lesion, perforation of overlying mucosa, and age of the patient. With any kind of treatment, a long-term follow-up is inevitable.

References


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