Evaluation of Decompression in the Treatment of Odontogenic Keratocysts : A Clinical and Radiographic Study

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ABSTRACT

Background and Objectives: Keratocyst is the most aggressive entity of odontogenic cysts with a high tendency to recure after enucleation. This objective of this study was to assess the effect of decompression in inducing cyst shrinkage before enucleation.

Materials and Methods: A prospective non-randomized clinical study was conducted on 11 patients with odontogenic keratocyst presenting to the department of maxillofacial surgery, university-affiliated teaching hospital, between February 2016 to March 2021. There were 8 males and 3 females, with a mean age of 57 years. Tube decompression was conducted as a preliminary treatment, before enucleation. The size of the cyst was measured orthopantomographically pre- and post-decompression. The percentage reduction was estimated, as was the correlation of cyst shrinkage with the initial size of the lesion and age of the patient.

Results: The mean duration of decompression was 208 days (152-316). The cyst size before and after decompression was 14.51 cm^2 (5.63 cm^2 -32.37 cm^2) and 6.69 cm^2 (1.76 cm^2-14.81 cm^2), respectively. The total and monthly percent shrinkage of the cysts were 54.39% (37.29% -76.37%) and 7.84% (4.45% -11.27%), respectively. The initial size of the lesion did not correlate significantly with the percentage of reduction, whereas the age of the patients showed a significantly negative correlation with the percentage of reduction.

Conclusions: Decompression is a well-tolerated minimally invasive surgical treatment that leads to a significant reduction in the size of the cyst. It induces the cyst to shrink away from anatomical structures to reduce the morbidity associated with subsequent enucleation.

Keywords: Decompression, Enucleation, Image J, Keratocyst, Orthopantomograph

INTRODUCTION

Odontogenic keratocyst (OKC) is highly aggressive and infiltrative developmental odontogenic cyst, first described by Philipsen in 1956[1], and comprises about 10% of jaw cysts[2]. The cyst originates from the dental lamina residues or basal layer of oral epithelium[3] and occurs twice as common in the mandible as in the maxilla[4] with a predilection for the angle- ramus region[5]. The recorded age distribution is usually broad, however, the highest incidence is recorded in the third decade of life[6] with a slight predilection for males[7].

Odontogenic keratocysts have a very thin fibrous wall coated by a homogeneous parakeratinised stratified squamous epithelium of 6 to 8 cell layers, with a columnar basal layer that is notably palisaded and corrugated luminal surface. The epithelial connective tissue interface is markedly flat, allowing basal epithelial cells to bulge into the underlying connective tissue and create daughter cysts[6].

Despite their aggressive and infiltrative nature, OKCs produce modest osseous enlargement due to the propensity for extending antero-posteriorly through the medullary cavities, and may be detected by chance during regular radiologic investigations[8]. Extremely large OKCs, on the other hand, may cause substantial cortical plate erosion, painful swelling, and involvement of adjacent tissues[9]. The OKC might appear on radiographs as a unilocular or multilocular radiolucency with smooth or scalloped corticated borders[10].

The third edition of WHO classification of head neck tumours, which was held in 2005, has renamed the OKCs as keratocystic odontogenic tumour (KCOT) due to the immunohistochemical findings that supported its neoplastic behaviour, primarily over expression of proliferative markers and inactivation of the tumour suppressor genes[11,12]. However, the fourth edition of the WHO Classification of Head and Neck Tumours, which was released in 2017, has recategorized this pathological condition as a cyst[13].

The main goals in the treatment of OKCs are to decrease the likelihood of recurrence, while minimizing the morbidity to the patients. Different surgical operations have been performed in the treatment of OKCs including, marsupialization, decompression, marginal or segmental resection. Furthermore, additional adjunctive measures have been used to reduce recurrence, such as osteotomy of the cavity and treating the cyst bed with Carnoy's solution or nitrogen cryotherapy[14]. The recurrence rate of OKCs after enucleation was reported to be about 30%, as compared to 18% by marsupialization alone. Carnoy's solution reduces the recurrence rate to 8%. Marginal or segmental resection, is associated with a notably low recurrence rate, however, it is reserved for the treatment of recurrent cysts, because of its high surgical morbidity[15].

The cyst is fenestrated in marsupialization by excising portion of the cyst wall and suturing the lining to the neighboring oral mucosa, allowing direct connection between the oral cavity and the cyst lumen. Any treatment that reduces the intraluminal pressure of a cystic cavity by maintaining an opening into the oral cavity is termed to as

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decompression[16]. Partsch reported in the late 1880s that exposing a cyst into the oral cavity stopped it from enlarging, a technique known as cystostomy[17]. Von Neuschmidt[18], a German surgeon, used a rubber tube to decompress odontogenic cysts in 1942, and Earle Thomas[19], an American surgeon, popularized it in 1947.

Decompression of OKC is accomplished by catheterizing the cyst cavity with a tube. The use of red rubber catheters, nasopharyngeal airways, intravenous tubing, nasal cannula tubing, and tuberculin syringes have been used for this purpose[20]. The purpose of decompression is to decrease intracystic pressure by continuous drainage of the cystic fluid, which encourage new bone formation.

Decompression has two key benefits: it preserves tissues by reducing the risk of damage to surrounding anatomical structures, and it avoids inpatient expenditures and procedures[21]. Decompression, on the other hand, has a number of drawbacks, including a longer treatment time, tube dislodgment, and the necessity for a compliant patient. Furthermore, epithelial lining remains might cause recurrence, necessitating further surgical intervention[22].

This study was conducted to evaluate the effect of decompression as a preliminary treatment of keratocysts before attempting enucleation.

MATERIALS AND METHODS

This is a prospective nonrandomized observation clinical study which included patients in whom tube decompression was implemented as the primary treatment method for the management of odontogenic keratocytes of the mandible. The study was conducted at the department of maxillofacial surgery, university-affiliated teaching hospital, between February 2016 to March 2021. The Declaration of Helsinki on medical protocol and our institutional ethical committee guidelines were strictly followed. A signed written informed consent was obtained from each patient to undergo this treatment.

Patients with large OKCs,of ≥ 3 cm^2, of the posteriormandible orramus were selected. Exclusion criteria were previously treated cysts, the presence of acute infection or draining fistula, any systemic illness that can impede healing, pregnancy and Gorlin-Golts syndrome. The patients had a panoramic x-ray examination of the cyst and size of the lesion was measured with the Image J software. By examining the light-dark contrast of the image, the border of the lesion was defined and followed. The size was measured before, monthly during compression, and after completion of decompression process by multiplying the maximum length height by the maximum height.

After administering local anesthesia, a small incision and a small bony access (approx. 1 cm diameter) were made on the buccal side of the deepest and most mesial part of the cyst. A sample of the cyst lining was removed for biopsy, then a 2.0 cm long and 0.5 cm wide polyethylene decompression tube was inserted into the cyst cavity and secured to the nearest tooth or the edge of the bony window with a stainless-steel wire. To avoid clogging of the tube with its thick contents, which could obstruct drainage, the thick contents were evacuated by gentle curettage and copious irrigation with saline. The soft tissue wound was closed with 4/0 silk sutures and the patient was instructed to flush twice daily with saline for the duration of decompression. One week after surgery, the patient was recalled for suture removal and confirmation that the biopsy results were consistent with the diagnosis of keratocyst. Thereafter, the patients were examined monthly to reassess the size of the cyst on the panoramic view and to check for any dislodgment of the tube. When the cyst had shrunk away from the anatomical structures, decompression was ceased and the cyst was enucleated (Figure 1).

SPSS 20.0 (Statistical Package for the Social Sciences, IBM Inc., Chicago) software program was used to perform the statistical analyses. The linear function between the percentage of reduction and both the initial size of the lesion and the age of the patients was assessed by simple linear regression. The P value 0.05 was considered to be statistically significant.

Figure 1: Odontogenic keratocyst, before and after decompression

RESULTS

The sample of this study consisted of 11 patients, 8 males (72.72%) and 3 (27.28%) females with a mean age of 43.67 years ±10.92 years (range 18-57 years). Six patients (54.55%) presented with painless swelling and in one patient the cyst was accidentally discovered during radiographic examination. (Table 1) shows the demographics of patients and the presenting signs/symptoms. (Table 2) shows the duration of decompression, pre- and post-decompression size of OKCs, and total and monthly percentage of reduction in size. The mean duration of decompression period was 208 days (152-316). The pre- and post-decompression size of the cysts was 14.51 cm^2 (5.63 cm^2 -32.37 cm^2) and 6.69 cm^2 (1.76 cm^2-14.81 cm^2), respectively. The total and monthly percentage of reduction in the size of the cysts was 54.39% (37.29%-76.37%) and 7.84% (4.45%-11.27%), respectively.

The initial size of the lesion has no significant correlation with the percentage of reduction (P=0.8105), (Figure 2). The patient's age showed a significant negative correlation with the percentage of reduction (P=0.0202), (Figure 3).

Table 1: Demographic data and presenting signs/symptoms

Variables	Results
Patients (n=11)	
Male	8(72.72%)
Female	3(27.28%)
Age (years)	
Mean ±SD	43.67 ±10.92
Range	18-56
Signs/symptoms	
Swelling	6 (54.55%)
Pain	2 (18.18)
Unerupted tooth	1(9.09%)
Tooth mobility	1(9.09%)
Incidental finding	1(9.09%)

DISCUSSION

The purpose of this clinical and radiological study was to evaluate the effects of decompressing OKCs with a polyethylene tube on

Table 2: Duration of decompression, size of the cyst and percentage of reduction

Patient	Duration of decompression (days)	Initial size (cm^2)	Post-decompression size (cm^2)	Total percentage of reduction	Percentage of reduction per month
1	182	25.46	9.37	63.19	10.45
2	210	14.40	4.56	68.33	9.67
3	156	8.74	3.62	58.58	11.27
4	227	18.85	11.43	39.36	5.20
5	187	32.37	14.81	54.24	8.70
6	212	5.63	1.94	65.54	9.27
7	224	7.45	1.76	76.37	10.29
8	316	13.60	7.23	46.83	4.45
9	193	8.34	5.23	37.29	5.80
10	152	15.34	8.32	45.76	9.03
11	233	9.44	5.39	42.90	5.52
combined	208	14.51	6.69	54.39	7.84

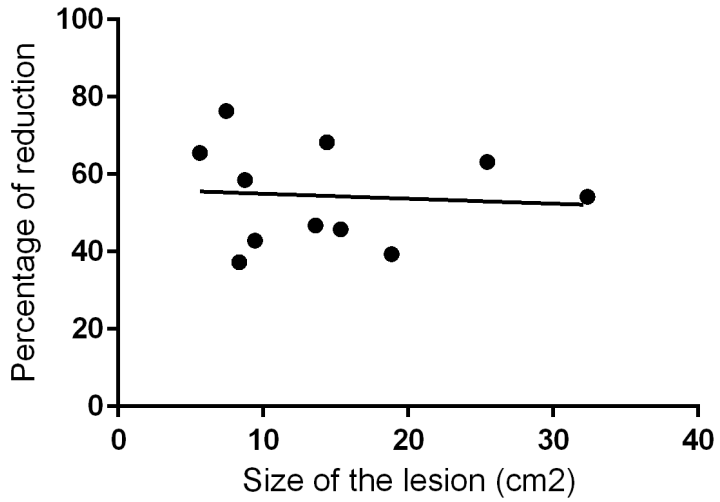

Figure 2: Regression graph between the independent variable size of the lesion and the dependent variable percentage of reduction

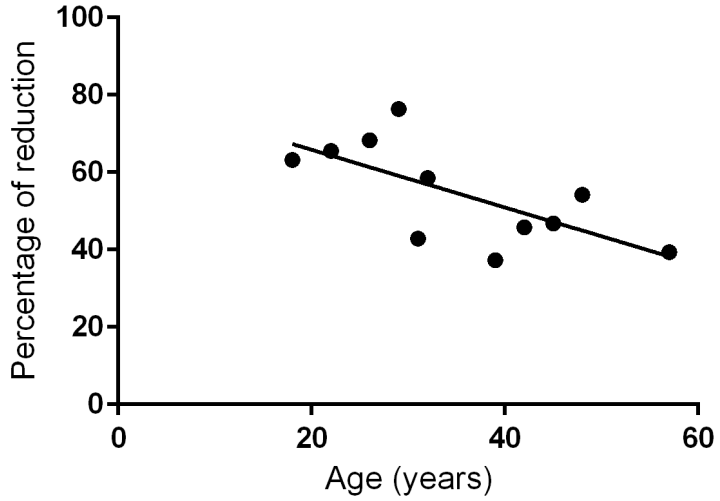

Figure 3: Regression graph between the independent variable age and the dependent variable percentage of reduction

their shrinkage before attempting surgical enucleation. In addition, the possible influence of patient age and initial size of the cyst on the percentage of shrinkage were also evaluated. In this study, an orthopantomograph rather than a CT scan was used to measure the size of the cyst before and during subsequent decompression period. Yoshiura et al[23] found that the size of the OKCs in the posterior mandible in panoramic images has a linear relationship to the volume of the lesion in the CT, because cysts in the posterior mandible rarely expand buccolingually. Although a CT scan would be an accurate three-dimensional imaging, nevertheless it is expensive and expose the patient to an excessive radiation.

The aggressive behavior of OKCs and their tendency to recur should be taken in consideration when selecting the treatment modality. Other factors that possibly influence the treatment plan are the patient's age and general health, the size and location of the lesion and proximity to vital structures such as the inferior alveolar nerve. Furthermore, involvement of overlying soft, whether the lesion is previously treated and recurrent, and the pattern of keratinization could also affect the selection of treatment method[24].

Odontogenic keratocysts are characterized by their tendency to recur after less aggressive treatment. Many factors may be responsible for recurrence including the extremely thin and crumbly epithelial lining, which it is easily torn during attempted removal, leaving fragments of epithelial residue. The presence of satellite cysts in the connective tissue wall and microcysts in the overlying mucosa could probably lead to relapse[25]. The recurrence rate of OKCs is highly variable and is greatly affected by the treatment modality. The overall recurrence rate of OKC was reported to be 23.15%. The recurrence rates were followed by treatment modalities were 0% for radical surgery with resection, 0% for enucleation with peripheral ostectomy and application of Carnoy's solution, 18.18% for enucleation with peripheral ostectomy, 26.09% for enucleation alone, 40% for marsupialization alone[26].

Because it is a non-invasive and well-tolerated surgery, decompression with subsequent enucleation of OKC may be the initial therapeutic choice in terms of functional and cosmetic repair. It is based on the rationale that relieving the intraluminal cystic pressure causes the lesion to shrink in size by gradual bone apposition from the periphery. In addition to relieving pressure inside the cyst cavity, decompression could also lead to histologic changes in the epithelial lining and connective tissue capsule. Chronic inflammatory process associated with decompression, may induce the epithelial lining to undergo metaplasia into a hyperplastic, stratified, non-keratinizing squamous epithelium[27]. Furthermore, the connective tissue capsule usually becomes dense, thick and fibrotic. These histological changes simplify the enucleation process and facilitates complete removal of the cyst wall[28].

The results of the present study obviously shows that large odontogenic keratocysts can be initially treated by micromarsupialization through a tube, followed by surgical enucleation. After an average decompression period of 6.9 months, the cysts showed marked reduction in size by more than 54% (range of 37-68%).

The reported response of OKCs to decompression is variable. After an average decompression of 9.8 months, Oh et al[27] detected that the size of OKCs on panoramic radiographs are reduced by 59±21%. Park et al[29] also found that the size of OKCs is reduced by 58.83% after decompression of 17 lesions for an average period of 9.9 months. Over 103, 270, and 727 days, the volume of OKC undergoing decompression was reduced by 25, 50, and 75 percent, respectively. Atusay et al[30] attempted a six month decompression on 16 patients with OKCs and obtained a 46.72% reduction in the cyst volume.

In the present study, the age of the patient was negatively correlated with the rate of reduction in cyst volume after decompression. This finding comes in accordance with that of Park et al[29] This finding is rational as younger patients have better healing abilities. In contrast to this study, Kubota et al[31] didn't found any correlation between the velocity of cyst shrinkage and age of the patients. These differences among studies can be attributed to different ethnic background of the patients and variation in the inclusion criteria and methods of estimating the cyst collapse.

The limitations of this study are fairly small size of the sample and the absence of follow-up of the lesions after enucleation to detect any recurrence. In addition, no comparison was made in the histological appearance of the lesions before and after decompression.

CONCLUSION

The decompression of keratocysts is a non-invasive surgical procedure that leads to marked reduction in the size of the lesion with shrinkage of the cyst far off the anatomical structures, decreasing the morbidity associated with subsequent enucleation. The time needed to collapse the cyst volume by 50% is about 6.9 months.

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Competing Interest: None

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REFERENCES

1. Philipsen HP. About keratocysts (cholesterol atoms). Tandialge bladet 1956;60:963-71.
2. Antonoglou GN, Sandor GK, Koidou VP, et al. Non-syndromic and syndromic keratocystic odontogenic tumors: systematic review and meta-analysis of recurrences. J Craniomaxillofac Surg 2014;42(7):364-71.
3. Ahlfors E, Larsson A, Sjogren S. The odontogenic keratocyst: A benign cystic tumor? J Oral Maxillofac Surg 1984;42(1):10-9.
4. Harmon M, Arrigan M, Toner M, et al. A radiological approach to benign and malignant lesions of the mandible. Clin Radiol 2015;70(4):335-50.
5. MacDonald D. Lesions of the jaws presenting as radiolucencies on cone-beam CT. Clin Radiol 2016;71(10):972-85.
6. Bilodeau EA, Collins BM. Odontogenic cysts and neoplasms. Surg Pathol Clin 2017;10(1):177-222.
7. Avril L, Lombardi T, Ailianou A, et al. Radiolucent lesions of the mandible: a pattern-based approach to diagnosis. Insigh Imag 2014;5(1):85-101.
8. Scarfe WC, Toghyani S, Azevedo B. Imaging of benign odontogenic lesions. Radiol Clin North Am 2018;56(1):45-62.
9. Eryilmaz T, Ozmen S, Findikcioglu K, et al. Odontogenic keratocyst: an unusual location and review of the literature. Ann Plast Surg 2009;62(2):210-2.
10. Sanchez-Burgos R, Gonzalez-Martin-Moro J, Perez-Fernandez E, et al. Clinical, radiological and therapeutic features of keratocystic odontogenic tumours: a study over a decade. J Clin Exp Dent 2014;6(3):259-64.

11. Gomes CC, Diniz MG, Gomez RS. Review of the molecular pathogenesis of the odontogenic keratocyst. Oral Oncol 2009;45(12):1011-4.
12. Shear M. The Aggressive nature of the odontogenic keratocyst: Is it a benign cystic neoplasm? Part 1. Clinical and early experimental evidence of aggressive behaviour. Oral Oncol 2002;38(3):219-26.
13. Soluk-Tekkesin M, Wright JM. The World Health Organization classification of odontogenic Lesions: A summary of the changes of the 2017 (4th) edition. Turk Patoloji Derg 2018;34(1):1-18.
14. Kaczmarzyk T, Mojsa I, Stypulkowska J. A systematic review of the recurrence rate for keratocystic odontogenic tumour in relation to treatment modalities. Int J Oral Maxillofac Surg 2012;41(6):756-67.
15. Johnson NR, Batstone MD, Savage NW. Management and recurrence of keratocystic odontogenic tumor: a systematic review. Oral Surg Oral Med Oral Pathol Oral Radiol 2013;116(4):271-6.
16. Pogrel MA, Jordan RC. Marsupialization as a definitive treatment for the odontogenic keratocyst. J Oral Maxillofac Surg 2004;62(6):651-5.
17. Partsch C. For the treatment of jaw cysts. Dtsch Mschr Zahnheilkd 1892;10:271-304.
18. Neuschmidt V. Drainage method of jaw cysts. Dtsch Zahnärztl Wschr 1942;21:287.
19. Thomas EH. Cysts of the jaws; saving involved vital teeth by tube drainage. J Oral Surg 1947;5(1):1-9.
20. Pogrel MA. Treatment of keratocysts: the case for decompression and marsupialization. J Oral Maxillofac Surg 2005;63(11):1667-73.
21. Maurette PE, Jorge J, de Moraes M. Conservative treatment protocol of odontogenic keratocyst: a preliminary study. J Oral Maxillofac Surg 2006;64(3):379-83.
22. Swantek JJ, Reyes MI, Grannum RI, et al. A technique for long term decompression of large mandibular cysts. J Oral Maxillofac Surg 2012;70(4):856-9.
23. Yoshiura K, Higuchi Y, Araki K, et al. Morphologic analysis of odontogenic cysts with computed tomography. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1997;83(6):712-8.
24. Terranova F, Trevisiol L, Nocini PF, et al. Keratocyst, conservative treatment: Case report. Minerva Stomatologica 2013;62(8):71-8.
25. Stoelinga PJW. The treatment of odontogenic keratocysts by excision of the overlying, attached mucosa, enucleation, and treatment of the bony defect with Carnoy solution. J Oral Maxillofac Surg 2005;63(11):1662-6.
26. Kaczmarzyk T, Mojsa I, Stypulkowska J. A systematic review of the recurrence rate for keratocystic odontogenic tumour in relation to treatment modalities. Int J Oral Maxillofac Surg 2012;41(6):756-67.
27. Oh JS, You JS, Kim SG. Clinical and histomorphometric evaluation of decompression followed by enucleation in the treatment of odontogenic keratocyst. J Dent Sci 2018;13(4):329-33.
28. Awni S, Conn B. Decompression of keratocystic odontogenic tumors leading to increased fibrosis, but without any change in epithelial proliferation. Oral Surg Oral Med Oral Pathol Oral Radiol 2017;123(6):634-44.
29. Park JH, Kwak EJ, You KS, et al. Volume change pattern of decompression of mandibular odontogenic keratocyst. Maxillofac Plast Reconstr Surg 2019;41(1):2-6.
30. Asutay F, Atalay Y, Turamanlar O, et al. Three-Dimensional Volumetric Assessment of the Effect of Decompression on Large Mandibular Odontogenic Cystic Lesions. J Oral Maxillofac Surg 2016;74(6):1159-66.
31. Kubota Y, Imajo I, Itonaga R, et al. Effects of the patient's age and the size of the primary lesion on the speed of shrinkage after marsupialization of keratocystic odontogenic tumours, dentigerous cysts, and radicular cysts. Br J Oral Maxillofac Surg 2013;51(4):358-62.