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ORIGINAL

Application of "Dredging Method" for the patients with odontogenic keratocyst.

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ABSTRACT :

When conservative treatment such as enucleation is applied, odontogenic keratocyst (OKC) is a well-known jaw lesion with a high recurrence rate. Other treatment modalities aimed at preventing recurrence have been used for managing OKCs. In our institute, an alternative conservative approach, known as the dredging method, is applied in OKC treatment. This paper describes and reports the characteristics and outcomes of conservative treatments involving the dredging method and enucleation in the management of patients with OKC.

Seventy-four patients with OKC or keratocystic odontogenic tumor (KCOT) presented to Oral and Maxillofacial Surgery at Hokkaido University Hospital between 1983 and 2017. They were treated and followed for more than 12 months. The patients' median age was 40 years (range 7–72 years) and the median longitudinal diameter of the lesion was 30 mm (range 7–128 mm). A total of 14 cysts (18.9%) were multilocular, as determined via radiographic evaluation. The surgical intervention consisted of enucleation alone in 17 cases (22.3%), deflation followed by enucleation in seven cases (9.5 %), and the dredging method in 50 cases.

Statistical significance associated with the size (P = 0.01) and locularity (P = 0.02) of the lesions was found between the cases treated with enucleation versus the dredging method. The median duration of follow-up was 40 months (range 12 to 405 months). Recurrence occurred in 9 of 74 cases (12.2%), including 2 of 17 cases (11.8%) treated with enucleation and 7 of 57 cases (12.3 %) treated with the dredging method. The recurrence period ranged from 12 to 131 months. All recurrence cases were managed by enucleation. There was no correlation between recurrence and patient sex or age, lesion size or locularity, the presence of a daughter cyst, or surgical approach. These results suggest that the dredging method is a successful conservative treatment option for large, multilocular OKCs. Long-term regular follow-up is essential to identify and manage recurrent cases.

Key Words : Odontogenic keratocyst, Keratocystic odontogenic tumor, Dredging method, Conservative treatment

Introduction

An odontogenic keratocyst (OKC) is a slow-growing, painless lesion that first presents radiographically as a large radiolucent mass. It is the third most common cyst of the jaws, representing 10 to 20% of all odontogenic cysts, and is most frequently localized in the mandible [1,2]. About 10% of cases involve multiple OKCs, half of which are diagnosed as arising from nevoid basal cell carcinoma syndrome [3,4].

The term *odontogenic keratocyst* (OKC) was first used by Philipsen in 1956 to describe an odontogenic developmental cyst arising from dental lamina remnants. Its classification has a complicated history [5]. In 2005, the World Health Organization (WHO)'s classification of head and neck pathology changed the entity's name to

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keratocystic odontogenic tumor (KCOT) to better reflect its neoplastic nature [6]. The WHO then reversed its decision in 2017 and reclassified OKC as an odontogenic cyst "characterized by a thin, regular lining of parakeratinized stratified squamous epithelium with palisading hyperchromatic basal cells" [7].

There is no consensus on the best treatment for managing OKC. High recurrence rates have been reported for most conservative treatment modalities, including enucleation alone (20.8 to 30.0%), enucleation with marsupialization (30.0 to 32.3%), marsupialization / decompression followed by enucleation (11.9 to 17.8%), and enucleation plus peripheral osteotomy (17.4 to 18.0%) [8-10]. The presence of daughter cysts and a multilocular pattern are two factors that have been associated with recurrence [11–13]. In the treatment of large lesions, resection provides the lowest recurrence rate but results in much higher morbidity than other treatment approaches, including deformity and dysfunction from loss of jaw bone support.

A conservative treatment protocol known as the *dredging method* offers an alternative to traditional treatment approaches. This method involves repeated dredging of the bony cavity after removing the cyst by enucleation alone or enucleation subsequent to deflation. The aim of the dredging method is complete removal of the lesion to accelerate the formation of new bone.

First developed by our institute to manage other benign mandible lesions, the dredging method has been effective in treating 50 cases of ameloblastoma, resulting in no deformity or loss of jaw bone support and a low recurrence rate 16% [14,15]. Based on these results, our institute's dredging method is now being used to treat other benign and/or cystic lesions in the mandible, including OKC [16].

The aim of this retrospective study is to evaluate the outcomes of patients with OKCs treated at a single institute with the dredging method.

Methods

1. Patients

This study was approved by the Hokkaido University Hospital Clinical Research Administration Center (IRB No. 015-0246). It is based on cases treated between 1983 and 2017 in the Oral and Maxillofacial Surgery at Hokkaido University Hospital. A total of 74 cases were identified as meeting the following inclusion criteria: (1) a histopathologic diagnosis of OKC or KCOT; (2) a preserved medical record; (3) regular follow-up of >12 months; and (4) the presence of only a single lesion (to exclude the potential influence of symptomatic disease, such as nevoid basal cell carcinoma syndrome).

Fig. 1. Treatment concept of Dredging method for cystic lesion.

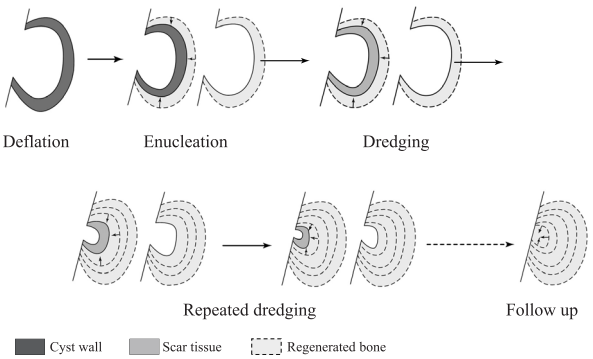

Fig. 1 Treatment concept of Dredging Method for cystic lesion.
Treatment procedures of "Dredging Method" for OKC. Deflation and enucleation are followed by repeated dredging. For a multilocular OKC, removal of the intracystic septum is applied in the deflation procedure (quoted from ref. 14 with modifications).

2. Surgical procedure

In our institute, the decision-making criteria for selecting a surgical procedure were as follows: (1) enucleation was selected for lesions with a clear bony outline; (2) marsupialization followed by enucleation or the dredging method was selected for lesions lacking a bony outline. Excision and reconstruction surgery were also presented as an option for large lesions, but a single patient did not accept it during the survey period. For the sake of convenience, seven cases treated with deflation followed by enucleation were included with the dredging cases in our analysis. Removal of newly formed bone adjacent to the cyst is common to both enucleation and the dredging procedure, as described below. Based on the information in the medical records, at least 15 surgeons performed the operations under the guidance of certified oral surgeons to ensure surgical procedure quality.

The dredging method is a conservative surgical procedure that involves repeated dredging of the cyst cavity after deflation and enucleation or enucleation alone (Fig. 1). The conceptual and clinical rationale of our treatment approach are as follows. (1) In cases where a large cystic lesion is lacking a bony outline around the cyst, deflation is used to release intracystic pressure and to facilitate the formation of the bony outline. (2) Enucleation includes removal of some of the surrounding

Fig. 2. Residual cyst in dredging procedure

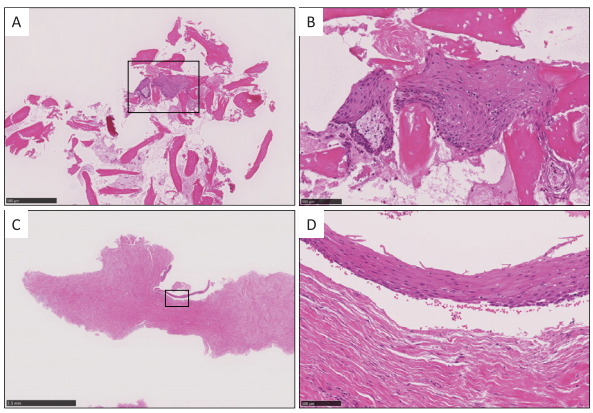

Fig. 2 Residual cyst in dredging procedure.

Residual cyst in removed newly formed bone (A, B) or scar tissue (C, D). A rectangle in A, C is magnified in B, D respectively. Scale bar in A: 500 μm, B, D: 100 μm and C: 2.5 mm. All specimens are removed from deep site of the lesions that are not continuous with the oral mucosa or deflated area.

bone for histopathologic investigation, as residual cyst wall–like squamous epithelium is observed in newly formed bone (Figs. 2A and 2B). The bony cavity remains deflated throughout treatment, and patients receive instructions on how to irrigate the cavity themselves. (3) Dredging follows enucleation to remove scar tissue that would hamper the formation of new bone. Based on how thick it becomes, newly formed bone is evaluated histopathologically to identify cyst remnants in the scar tissue (Figs. 2C and 2D). Dredging is repeated at 2- to 3-month intervals to accelerate new bone formation until the bony cavity is filled with new bone or devoid of cysts as confirmed histopathologically in the scar tissue obtained from two consecutive dredgings. (4) Long-term follow-up is essential to the dredging method's success because of the historically high recurrence rates associated with conservative treatment modalities.

3. Assessment of the lesion, clinical outcome, and prognosis of patients with recurrent OKC

The longitudinal diameter of the lesions was measured on orthopantomogram (OPG). A lesion was determined to be multilocular based on the presence of a septum inside the lesion on OPG or computed tomography images or in the surgical record. The presence of a daughter cyst in the specimens was determined histopathologically at enucleation. A recurrence was defined as a new lesion arising during the follow-up period, as identified radiographically, in an area where treatment was completed.

4. Statistical analysis

The data were analyzed with Mann–Whitney U test, chi-squared test, or Fisher's exact test as appropriate, with the aid of Statistical Package for the Social Sciences (SPSS ver. 26.0, SPSS Inc., Chicago, IL, USA). Probabilities of <0.05 were designated as significant.

Results

1. Patient characteristics

Of the 125 patients with OKC treated at our institute, 74 met the enrollment criteria of a histopathologic diagnosis, a preserved medical record, regular follow-up of >12 months, and the presence of only one lesion. Demographically, the patients ranged in age from 7 to 72 years (median 41 years) and the male to female ratio was 1.96:1. The lesion's longitudinal diameter on OPG ranged from 7 to 128 mm (median 30 mm). Multilocular lesions were observed in 14 cases (18.9%), and a daughter cyst was histopathologically detected in 17 cases (23.0%). Of the 74 patients enrolled in the study, 57 (77.0%) were treated with the dredging method at least one time and not more than five times (median two times). Duration of

Table 1. Patient's characteristics.

Sex		
Male	49	(66.2 %)
Female	25	(33.7 %)
Age, years		
Range	7-72	
Median	41	
Longitudinal diameter, mm		
Range	7-128	
Median	30	
Locularity		
Unilocular	60	(81.1 %)
Multilocular	14	(18.9 %)
Daughter cyst		
Positive	17	(23.0 %)
Negative	57	(77.0 %)
Treatment		
Enucleation	17	(23.0 %)
Dredging method	57	(77.0 %)
Number of dredging		
Range	1-5	
Median	2	
Follow-up, months		
Range	12-405	
Median	40	

Table 2. Clinical features in different treatment modalities.

Characteristics	Enucleation (n=17)	Dredging Method (n=57)	P-value
Sex			0.14[a]
Male	9	41	
Female	8	16	
Age, years			0.39[b]
Range	7-71	10-72	
Median	42	40	
Longitudinal diameter, mm			0.01*
Range	7-48	12-128	
Median	23	33	
Locularity			0.02**
Unilocular	17	44	
Multilocular	0	13	
Daughter cyst			0.22[c]
Positive	2	15	
Negative	15	42	

Statistical significance determined by Mann–Whitney U test*and Fisher's exact test**.

P-value was determined with Chi-squared test[a], Mann–Whitney U test[b] and Fisher's exact test[c].

follow-up ranged from 12 to 405 months with a median of 40 months (Table 1).

2. Clinical features of cases treated with different modalities

Because treatment selection was based on the presence of a bony outline around the lesion, clinical parameters were examined to identify any differences between the cases treated with enucleation alone or the dredging method (Table 2). Enucleation were performed in 17 cases (23.0%) and the dredging method in 57 cases (77.0%). The longitudinal diameter (P = 0.01) and locularity (P = 0.02) of the lesions showed statistical significance. There was no statistical significance between the cases treated by enucleation and the dredging method in the parameters of sex (P = 0.14), age (P = 0.40), or the presence of a daughter cyst (P = 0.22).

3. Characteristics of recurrences

Recurrence was observed in 9 (12.2%) of the 74 patients (Table 3). To elucidate the factors associated with recurrences, their clinical features were evaluated. The recurrence rate was higher in females (20.0%) than in males (8.2%), although the difference was not statistically significant (P = 0.14). The lesions with a daughter cyst had a higher recurrence rate (23.5%) than those without (8.8%), but statistical significance (P = 0.12) was undetected. Other factors such as age, longitudinal diameter, locularity, and treatment modality showed no correlation with recurrence.

4. Clinical features and prognosis of recurrent cases

Details of the recurrent lesions are shown in Table 4. For cases treated with the dredging method, radiographic evidence of a recurrence was found as soon as 12 months and as late as 131 months (median 26 months) after the final dredging procedure, which we officially designated as the start of the follow-up period. All recurrent cases were treated with enucleation.

Discussion

The purpose of this retrospective study was to investigate the clinical characteristics and outcomes of patients with OKC treated with conservative surgery, namely the dredging method, at a single institute. Our primary hypothesis was that differences in treatment approaches would influence the recurrence rate of OKCs. Our results showed no statistical difference in the recurrence rate of OKC treated with enucleation (11.8%) versus those treated with the dredging method (12.3%) (see Table 3).

A number of different surgical approaches have been used to reduce the recurrence rate of OKCs, including

Table 3. Characteristics of the recurrence.

Factors	Recurrence		Recurrence rate (%)	P-value
	+ (n = 9)	- (n = 65)		
Sex				0.14[a]
Male	4	45	8.2	
Female	5	20	20.0	
Age, years				0.50[b]
Range	12-71	7-72		
Median	36	41		
Longitudinal diameter, mm				0.22[b]
Range	12-128	7-95		
Median	48	30		
Locularity				0.54[a]
Unilocular	7	53	11.7	
Multilocular	2	12	14.3	
Daughter cyst				0.12[a]
Positive	4	13	23.5	
Negative	5	52	8.8	
Treatment				0.66[a]
Enucleation	2	15	11.8	
Dredging Method	7	50	12.3	

P-value was determined with Fisher's exact test[a] and Mann–Whitney U test[b].

Table 4. Clinical features and prognosis of recurrent cases

Patient	Sex	Age (Years)	DC*	Locularity**	Treatment	Time of recurrence (Months***)	Treatment for recurrence
1	Female	36	(-)	Uni-	enucleation	96	enucleation
2	Male	71	(+)	Uni-	enucleation	26	enucleation
3	Female	45	(-)	Uni	Dredging method	26	enucleation
4	Male	12	(+)	Uni-	Dredging method	12	enucleation
5	Female	17	(-)	Uni-	Dredging method	36	enucleation
6	Male	35	(-)	Multi-	Dredging method	12	enucleation
7	Female	43	(+)	Multi-	Dredging method	48	enucleation
8	Female	50	(+)	Uni-	Dredging method	24	enucleation
9	Male	20	(-)	Uni-	Dredging method	131	enucleation

*: Daughter cyst, (-): negative, (+): positive. **: Uni-; unilocular, Multi-; multilocular. ***: months after final surgery.

radical resection [17], simple enucleation, marsupialization/decompression with subsequent enucleation, and enucleation with or without Carnoy's solution or cryotherapy [8,18,19]. According to the literature, the recurrence rate of OKC managed with conservative treatment ranges from 11.5 to 32.3% [8–10]. Based on the results presented in this paper, the recurrence rate at our institute is equivalent to the lowest reported rate.

At our institute, decisions about which lesions to treat with enucleation and which to treat with the dredging method were primarily based on the presence of a bony outline around the lesion, which facilitates the lesion's complete removal. Based on this criterion, the dredging method was the treatment assigned to large and multilocular lesions (see Table 2).

Clinicopathologic features associated with OKC recurrence include lesion size and locularity (unilocular or multilocular), and the presence or absence of daughter cysts. The relationship between the lesion size and recurrence rate has not been determined [11–13, 20, 21]. No statistically significant association between lesion size and the recurrence rate was found in this study despite

the fact that large lesions were treated with the dredging method. This result strongly suggests that the dredging method is an effective modality for treating large OKCs. Regarding locularity, multilocular lesions are known to recur at a higher rate than unilocular lesions [11,12,20,21]. In this study, there was no association between recurrence rate and locularity even though all multilocular lesions were treated with the dredging method (see Tables 2 and 3). This result verifies the effectiveness of the dredging method in preventing recurrence in lesions associated with a high recurrence rate. The third feature possibly associated with recurrence, daughter cysts, remains under debate [6,13]. In our institute, OKC lesions with daughter cysts had a higher rate of recurrence (23.5%) than those without daughter cysts (8.8%), although the difference was not statistically significant (see Table 3). This result is consistent with the findings of other investigators [11,22]. Further studies with larger sample sizes are needed to elucidate the influence of each of these probable prognostic features.

The objective of the dredging method is twofold: to completely remove the lesion from the jaw and to accelerate the formation of new bone. To achieve this objective, dredging is performed every 3 to 4 months after the initial deflation of the cyst. At our institute, the median number of dredgings was two, which can be extrapolated to mean that in a single year, patients received a total of four surgical procedures comprising deflation, enucleation, and two dredgings (see Table 1). The number of surgeries and duration of this treatment approach, which might be seen as a disadvantage, is justified by the prioritization of clinical outcomes at our institute. Our study revealed that in regular follow-up, recurrences were detected between 12 and 131 months (median 26 months) after surgery, which corresponds to a previous study's results [23], and all recurrent lesions were managed by enucleation. This result demonstrates that regular long-term follow-up is an essential component of an alternative conservative treatment approach such as the dredging method for successful management of patients with OKC.

Due to this study's retrospective design, the apparent advantages of the dredging method as compared with enucleation were limited to the size and locularity of the treated lesions. Therefore, further prospective and multicenter studies are needed to obtain a fuller picture of the clinical outcomes of the dredging method in the treatment of OKC.

Conclusion

In this retrospective study, the recurrence rate of OKCs treated with conservative surgery in Oral and Maxillofacial Surgery at Hokkaido University was 12.2%. All large and/or multicystic lesions were treated with the dredging method instead of enucleation alone. The lack of statistical significance associated with recurrence rate between cases treated with enucleation versus the dredging method indicates that successful clinical outcomes are obtained in the treatment of large and multilocular OKCs when the dredging method is applied with a long-term follow-up.

Disclosure of conflict of interest

The authors declare no conflicts of interest associated with this study.

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