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The role of marsupialization in the treatment of odontogenic keratocystic tumors: A clinical study of 21 cases

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Abstract

Odontogenic keratocyst is developmental lesion arise from the remnant dental lamina. They are differs from other odontogenic cysts in their aggressive behavior and high recurrence rate. Different surgical modalities were utilized in the treatment of odontogenic keratocystic tumors nevertheless this subject remains controversial. In this field conservative methods like enucleation, decompression and marsupialization can be done while other aggressive procedures include excision or resection of the tumor. Therefore this study was aimed to evaluate the results of marsupialization treatment of such lesions. This study included 21 patients from different age and sex groups identified to had odontogenic keratocystic tumors in the upper or lower jaw. The diagnosis was confirmed by clinical, radiographical and histopathological examinations All patients were treated by marsupialization of the tumor. The patients were followed clinically and radiographically regularly for 2-6 years after the treatment to evaluate their conditions according to the size of the tumor and recurrence of the lesion. Males were 16 (76.1%) and 5 (23.8%) females with average 33.8 years. Maxillary bone was involved in 3 (14.2%) cases while 18 cases (85.7%) affected the mandible mainly in the body and ramus which comprised 11 cases. Response to treatment was detected in 19 patients (90.4%) after minimum period of follow up for 2 years. Marsupialization plays an important role as conservative method in management of these tumors moreover further researches in molecular biology may provide knowledge about the aggressive behavior, recurrence of these lesions and appropriate treatment method.

Keywords: Marsupialization; Odontogenic keratocystic tumor; Decompression; Odontogenic cyst.

Introduction

Odontogenic keratocyst is developmental lesion arise from the remnant dental lamina firstly described by Philipsen 1956 which include odontogenic cysts that showed keratinization of epithelium. Pindborg and Hansen 1963 defined the histologic features essential to identify the odontogenic keratocyst [1, 2]. They are differed from other odontogenic cysts in their growth and clinical presentation with distinctive histological features. Owing to their aggressive behavior and high recurrence rate after treatment the World Health Organization (WHO) 2005 defined these lesions as benign tumors of odontogenic origin which are intraosseous multi or unicyst with parakeratinized stratified squamous epithelium lining. Based on molecular studies the WHO recommends using the term odontogenic keratocystic tumors to describe the neoplastic nature of these cysts [3, 4].

Histologically KCOT shows a uniform layer of parakeratotic stratified squamous epithelium

which is relatively thin, generally contain up to eight cell layers, with specific flat connective tissue border .The fibrous layer is normally without inflammatory infiltrate showed proliferations of odontogenic epithelium and formation of daughter or microcysts. This lesion is differed from orthokeratinized odontogenic cysts which considered as another lesions and not the part of the KCOT. It is thought that these cysts do not show destructive features like of KCOT [5].

OKCT form about 10 % of all jaw cyst affect patients of all age groups and it is common in the 20-30 years of life with male predilection. More than 70% of keratocysts are located in the mandible specially the angle and ramus. Not like other cyst of the jaw which induce bulge in the alveolar bone OKCT tend to grow in anterior posterior direction without clinical manifestation in some cases. Unless they are infected or cause complications they may reach to large size before diagnosis and sometimes they are discovered by radiographic examination done for other purposes [6,7].

Radiographically they are presented as unilocularor multilocular radiolucencies furthermore OKCT may be associated with unerupted teeth. Furthermore multiple KCOT are one of the characteristic features of Gorlin-Goltz syndrome which is rare autosomal dominant disorder associated with other features like hypertelorism, bifid ribs, frontal bossing and multiple basal cell carcinomas [7].

High recurrence rate and difficulty in treatment made OKCT of clinical importance whereas many researches focus on these topics. They have a tendency to recur in about 3-60% after the treatment which can be attributed to fragile lining and formation of daughter cysts.

Different surgical modalities were utilized in the treatment of OKCT nevertheless this subject remain controversial. In this field conservative methods like enucleation, decompression and marsupialization can be done while other aggressive procedures include excision or resection of the tumor can also be adopted for treatment purposes. Other adjunctive means like cryotherapy and Carnoy's solution have been used with enucleation to decrease the recurrence rate of these lesions [6-9]. Therefore this study was aimed to evaluate the results of marsupialization treatment of OKCT.

Materials and Methods

This article included 21 patients from different age and sex groups identified to had OKCT in the upper or lower jaw. The diagnosis was confirmed by clinical, radiographical and histopathological examinations. The study was done in the oral and maxillofacial surgery department of Sulaimani teaching hospital during the period from 2008 - 2016. The informed consent was obtained from each patient and the work approved the ethical committee of the institution.

Patients information includes age, sex, site of the tumor and general condition were also considered. The treatment was completed under local or general anesthesia depended on the tumor size, age of the patient and the affected area of the jaw bone. For all patients orthopantogram (OPG) X-ray was done to determine the tumor size.

All patients were treated by marsupialization of the tumor cavity with evacuation of its content followed by irrigation and iodoform packing for 2 weeks. Obturators were made for the patients during the period of follow up which reduced in size gradually according to the size of the lesion.

The patients were followed clinically and radiographically regularly for 2-6 years after the treatment to evaluate their conditions according to the size of the tumor and recurrence of the lesion.

Results

In the presented study 21 patients were included, 16 (76.1%) were male and 5 (23.8%) female with range of 10-56 years (average 33.8 years). Maxillary bone was involved in 3 (14.2%) cases while 18 cases (85.7%) affected the mandible mainly in the body and ramus which comprised 11 cases. Response to treatment was detected in 19 patients (90.4%) after minimum period of follow up for 2 years however recurrence was occurred in 2 cases (9.5%) which treated later on

by peripheral ostectomy. Complete ossifications of lesion cavity were detected in the patients who responded to the treatment according to clinical findings and radiographical examination. Table (1) summarized the results of the study also see Figures (1-5).

Table 1: **Distribution of OKCT according to the clinical presentation with follow up period and response to the treatment**

No.	Age	Sex	Site	Size of tumor on OPG (width x height) cm	Follow up in months	Response
1	52	M	Ant. Mandible	8 x 4	26	Yes
2	39	M	Ramus. Mandible	6 x 4	37	Yes
3	16	M	Ant. Mandible	9 x 4	57	Yes
4	38	M	Body. Mandible	2 x 3	24	Yes
5	29	F	Rt. Maxilla	7 x 3	25	Yes
6	31	M	Ant. Mandible	7 x 4	49	Yes
7*	40	M	Ant. Mandible	11 x 3	26	No
8	10	M	Ant. Mandible	8 x 3	28	Yes
9	56	M	Body. Mandible	3 x 4	38	Yes
10	33	M	Ant. Maxilla	2 x 2	25	Yes
11	44	F	Ramus and body of mandible	7 x 3	24	Yes
12	55	F	Ant. Mandible	7 x 4	36	Yes
13	35	M	Ramus and body of mandible	4 x 7	70	Yes
14	36	M	Ant. Mandible	6 x 3	69	Yes
15	26	M	Rams and body of mandible	3 x 5	24	Yes
16	29	F	Body. Mandible	3 x 3	48	Yes
17	23	M	Rt. Maxilla	3 x 2	25	Yes
18#	37	M	Multiple. Mandible	3 x 4 , 2 x 2	49	Yes
19*	21	M	Body. Mandible	2 x 2	50	No
20	38	F	Body. Mandible	2 x 3	67	Yes
21	39	M	Body. Mandible	3 x 3	66	Yes

** In both cases (7and 19) the cystic lesion was decreased in size but there was recurrence after 2 years. Further treatment by peripheral ostectomy was done for both of them.*
 # This case showed non syndromic multiple OKCT affected right side of the mandible in the ramus and body.

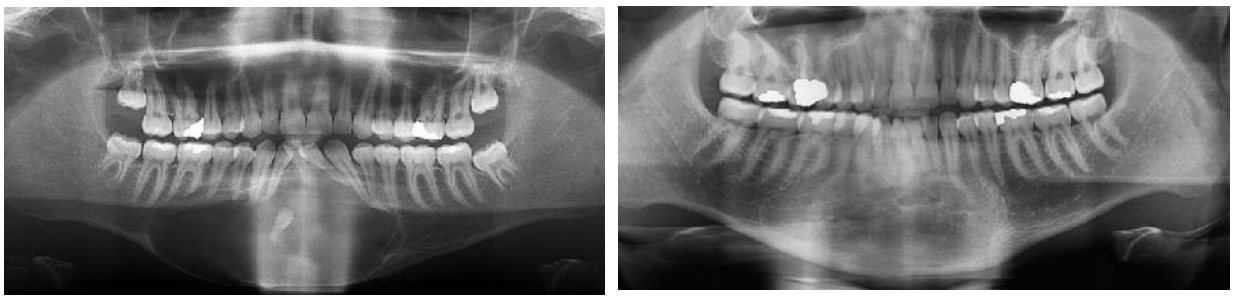

Fig. (1): OKCT of 16 years male patient affects both sides of the mandible showed complete healing of the bone after 4 years of treatment.

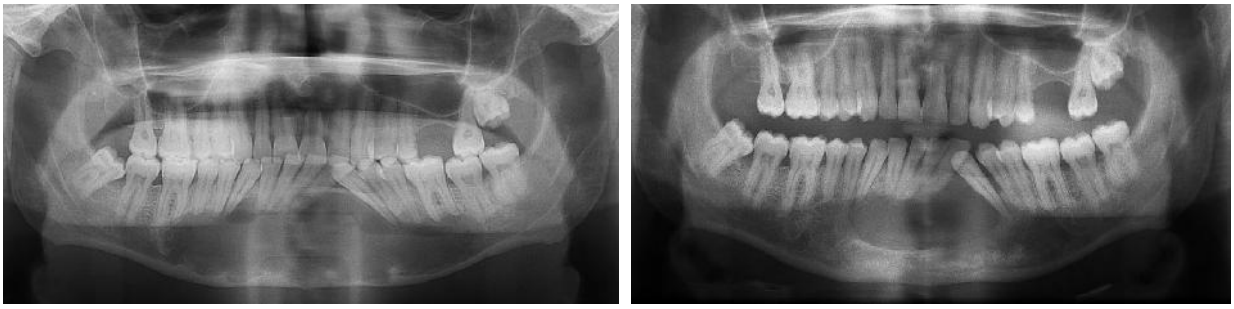

Fig. (2): Patient no.7 with OKCT which not responded to the treatment after 2 years and treated by peripheral ostectomy but the size of the tumor was reduced.

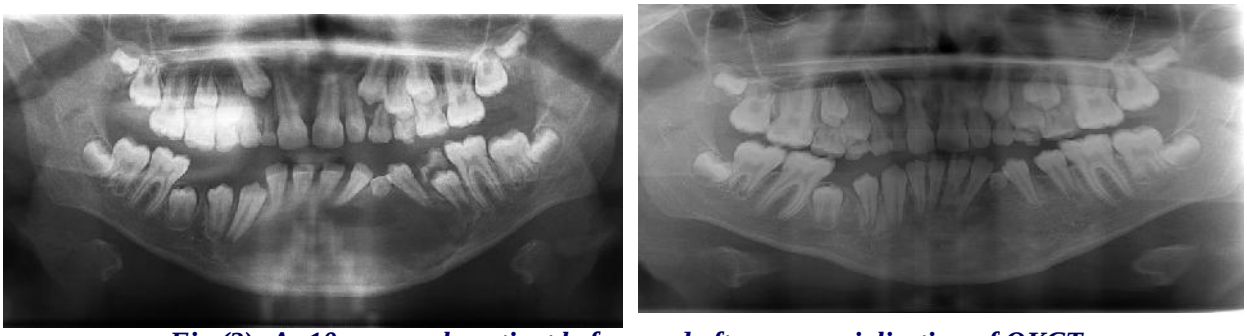

Fig.(3): A -10 -yrs. male patient before and after marsupialization of OKCT.

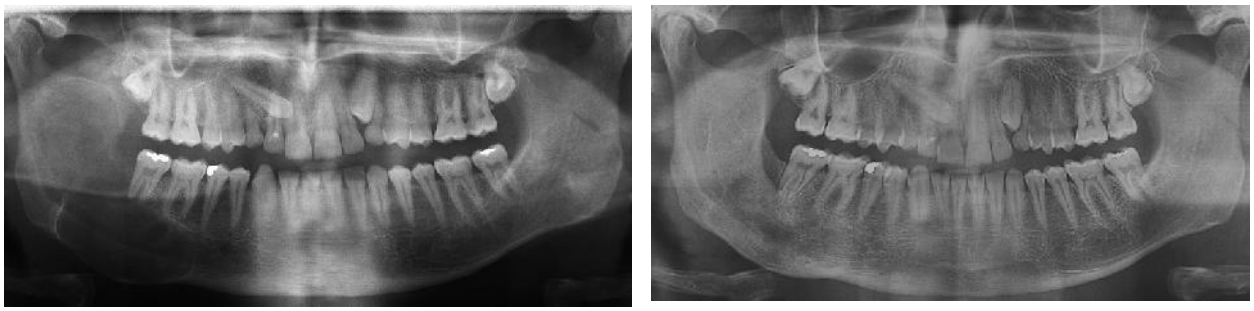

Fig.(4) OKCT of the ramus after 3 years of treatment showed ossification of the bone.

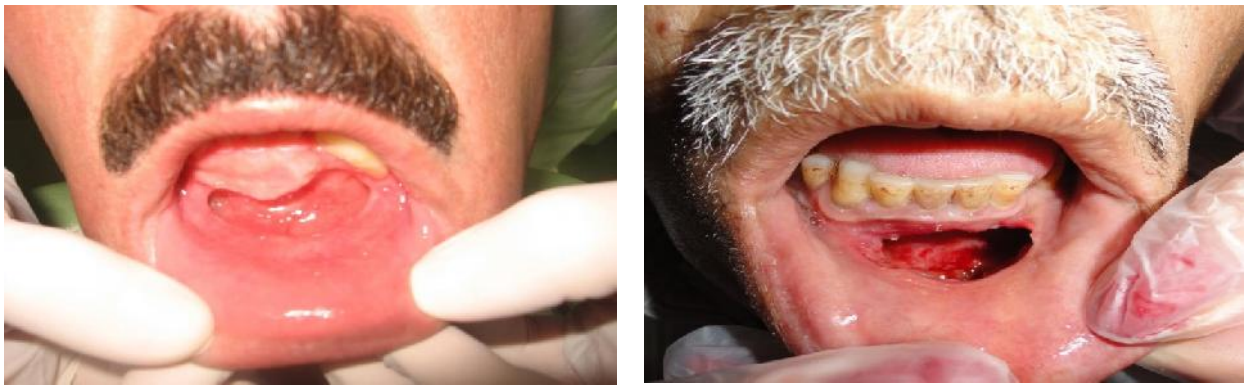

Fig.(5): Showed the lining of the tumor cavity of 2 patients after marsupialization Look like oral mucosa which may affects the behavior of the lesions.

Discussion

The aims of treatment of OKCT are to preserve the affected bone and prevent recurrence as these lesions have destructive infiltrative behavior. Accordingly some authors recommended radical treatment likes ostectomy or enblock resection of the bone to assure the healing and prevent recurrence of the tumor. On the other hand conservative managements of OKCT were reported in literatures as an option of treatment like decompression, marsupialization and enucleation with or without adjuvant therapy [9, 10].

In spite of many investigations were done to evaluate different surgical methods for treatment of OKCT still there is a lot of discussion about this subject. In the field of treatment of these lesions the literatures didn't provide adequate proof for determining which surgical technique achieves low morbidity and inhibit recurrence of tumor.

Many factors will affect the recurrence of OKCT includes the size of the tumor, age, location, histopathology and the method of the treatment Browne used three different procedures, marsupialization enucleation with primary closure and enucleation with packing open for treatment of these lesions. The author found an approximately the same rate of recurrence with all three techniques of treatment. He concluded that recurrence is due to the nature of the cystic lesion itself and that it is not correlated to the method used for treatment [11]. Whereas according to Kuroyanagi et al. the size or extent of the lesion will not affect the recurrence of OKCT it is mainly related to the type of the surgical methods [12].

Marsupialization considered the most conservative technique of management of KCOT which involves of changing the cystic cavity into a pouch that can be cleaned on regular intervals. This method has been designated for complete healing of benign odontogenic cysts. It was originally planned by Partsch at the end of 18 century as an ultimate treatment method for cystic lesion [13]. Genetic and immunohistochemical researches now a day play a significant role in understanding the pathogenicity of OKCT which give a clear depiction about management and prognosis of these lesions. To explain the biological potential and histological features of OKCT several studies was done of the expression of proliferative and anti-proliferative markers such as Ki-67, p53, PCNA and Bcl-2. Evidence of PTCH gene mutations in KCOT came from genetic studies which made the tumors to be targets of further genetic modifications and assisting tumor progression [12, 14]. A study made by Kuroyanagi et al. concluded that mutation of p53 play an important role in the development of OKCT but not the prognosis or recurrence of the lesions [15.] The interleukin (IL-1α) is one of the inflammatory cytokines highly expressed in the epithelial cells of OKCs and has significant relationship with tumor growth, invasion and recurrence.

It was found that the expression of IL-1α mRNA by epithelial cells of OKCT will be reduced significantly by marsupialization which leads to decrease in proliferation of epithelial cell as the IL-1α plays an important part in growth of the OKCT. Also the lining of OKCT will be changed by decompression or marsupialization from frank keratocyst to non keratocyst [16,17].

Generally the results of this study were in agreement of other reports regarding the treatment of OKCT by marsupialization. More than 90% of the patients in this study showed complete healing of the affected area with the period of follow up which is 2-6 years. Although recurrence of OKCT may occur within the five years, studies reported the recurrence after 9 years [18-20]. At this point one may ask is the period of follow up is enough or what is the time for follow up? The immunohistochemical and genetic study may answer of this question as mentioned above.

Decompression of cystic cavity in the form of marsupialization or cystectomy is a common procedure approved for management of many odontogenic cystic lesions as a solitary method or in combination with enucleation. Technically it is an easy method can be done in most cases under local anesthesia besides good results were achieved for treatment of OKCT as individual cases or in case series studies. It had been accepted that marsupialization or decompression is the first choice for the treatment of large cystic lesion whatever the diagnosis of such lesion. Nevertheless marsupialization have several disadvantages which includes the long treatment time and follow up in addition to patients cooperation which considered an important factor in this type of treatment[17].

The mandible was affected more the maxilla specifically the ramus and body so marsupialization of OKCT in these regions will preserve the integrity of the inferior alveolar nerve which may be affected by the use of other surgical modalities. Two cases didn't responded

to the treatment in this study but the size of the lesions in 2 cases was reduced which made further treatment by ostectomy easier and less morbidity for the patients. On the other hand marsupialization is a good option of treatment when deals with dentate region or in children, this considered less destructive treatment which can be tolerated by most of the patients. As a treatment modality of OKCT marsupialization can be done at any site of the affected jaw bone. Some cases in this study were presented with large lesions extended to both sides of the mandible so the advantages of marsupialization in these cases were preservation of the esthetic and function of the oral cavity. En block resection of such lesion will leave the patients with severe facial deformity and need other surgical intervention for reconstruction. It's better to try this treatment method as a first option of treatment of OKCT particularly large one or for lesions which involved the vital structures.

In conclusion the results of this report showed that marsupialization play an important role as conservative method in management of OKCT moreover further researches in molecular biology may provide knowledge about the aggressive behavior, recurrence of these lesions and appropriate treatment method.

Conflict of Interest: None to declare.

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