

# ASSESSMENT OF RECURRENCE OF ODONTOGENIC KERATOCYSTS TREATED WITH DIFFERENT SURGICAL MODALITIES — NINE YEARS FOLLOW UP OF DISEASE

<sup>1</sup>ARSLAN MANZOOR

<sup>2</sup>WASEEM AHMED

<sup>3</sup>MUHAMMAD UMAIR

<sup>4</sup>GULZAR ALI BUKHARI

<sup>5</sup>MUHAMMAD OMAR NIAZ

## ABSTRACT

*The purpose of this study was to determine the Recurrence rate of odontogenic keratocysts (OKC), and association of recurrence with various surgical modalities. It was cohort analytical study. Eighty-two odontogenic keratocysts cases treated at Oral and Maxillofacial department Armed Forces Institute of Dentistry, Rawalpindi formed the study subject, and were followed for a period of 9 years from 2004 to 2013.*

*Recurrence of OKCs were evaluated from 1-9 years after surgical treatment by clinical and radiographic means and correlated with various treatment modalities. The most frequent site affected by OKCs was the posterior mandible including body, angle and ramus. Patients were followed for recurrence. 16 patients were lost to follow up and this journal was 14 cases of recurrence out of 66 were treated with varying modalities (21.2%). Patients treated conservatively with marsupialization or enucleation alone demonstrated higher recurrence rates (100% and 50% respectively).*

*Odontogenic keratocyst is an aggressive cyst associated with high recurrence. Surgeons should decide on various modalities as per requirement of the case. Although resection carries least risk of recurrence, it should be limited to recurrent and aggressive OKCs. The outcomes of enucleation combined with peripheral ostectomy and chemical cauterization is reasonably acceptable.*

**Key Words:** Keratocysts, Treatment modalities, Recurrence.

## INTRODUCTION

Odontogenic Keratocyst (OKC) is an aggressive odontogenic cyst arising from dental lamina and is unique in its innate growth potential like tumors and association with high recurrence.<sup>1</sup> It accounts for ap-

proximately 12-14% of all odontogenic cysts of jaws, has slight male predilection and occurs mostly in 2nd and 3rd decade.<sup>2</sup> 60-80% lesions occur in the mandible with body and ramus being predominant sites of involvement.<sup>3</sup> OKC although tends to grow in medullary spaces without significant bony expansion, but it can cause extensive bone destruction.<sup>4</sup> Radiographically, it may present as unilocular or multilocular radiolucency with well-defined corticated margins, and can occur in dentigerous relationship causing tooth displacement or sometimes even resorption. In 25-40% of cases, impacted teeth are present in the lesion.<sup>5</sup> Microscopically, OKC is characterized by 8-10 cell layers thick stratified squamous epithelium with hyperchromatic and palisaded basal cell layer (characteristic of true OKCs). The luminal surface has flattened, parakeratotic epithelial cells exhibiting a corrugated appearance.<sup>6</sup> Orthokeratinized odontogenic cyst does not show palisading of basal cells and is histologically different from parakeratinized OKC showing less aggressive behavior and destruction.<sup>5,6</sup> OKCs may be suspected on clinical and radiographic

<sup>1</sup> Arslan Manzoor, BDS, FCPS, Assistant Professor of Oral Pathology, Foundation University College of Dentistry, Islamabad  
E-mail: drarslanmanzoor@hotmail.com Cell no. 092-3156897073

**Correspondence:** Foundation University College of Dentistry, Foundation University, Islamabad

<sup>2</sup> Waseem Ahmed, BDS, FCPS, Commandant & Head of Department of Oral & Maxillofacial Surgery, Armed Forces Institute of Dentistry, Rawalpindi

<sup>3</sup> Muhammad Umair, BDS, MCPS, Senior Registrar, Department of Oral Medicine, Foundation University College of Dentistry, Islamabad

<sup>4</sup> Gulzar Ali Bukhari, BDS, FCPS, Consultant, Department of Oral and Maxillofacial Surgery, Armed Forces Institute of Dentistry, Rawalpindi

<sup>5</sup> Muhammad Omar Niaz, BDS, MDPH, DDPH, Senior Registrar, Department of Community Dentistry, Foundation University College of Dentistry, Islamabad

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grounds, however histopathological confirmation is required for the diagnosis.<sup>5</sup>

OKCs have an aggressive behavior and a tendency to recur. This together with the fact that they are caused by the inactivation of a tumour suppressor gene has led to reclassify them as “neoplasms”. According to the WHO 2005 classification of odontogenic tumours, OKC has been classified as “Keratocystic odontogenic tumour”.<sup>7</sup>

Although various therapies for OKC have been documented in literature, the universally accepted approach remains undecided. These range from conservative methods such as enucleation, marsupialization, curettage, chemical cauterization, peripheral ostectomy to much aggressive treatments such as marginal or segmental resection.<sup>8</sup> Recurrence rate varies from 2.5 to 62.5% with much literature suggesting approximately 30%.<sup>1,3,5</sup> This much varied recurrence is due to varying periods of follow up, inclusion of orthokeratinized odontogenic cyst or not, and also on treatment modality adopted. The purpose of this study was to highlight higher rates of recurrence of OKCs and its association with different surgical modalities.

## METHODOLOGY

The study was conducted at Oral and Maxillofacial Surgery Department of Armed Forces Institute of Dentistry, Rawalpindi from 2004 to 2013 after obtaining permission from Ethical Committee of AFID. Eighty-two cases were studied retrospectively in a cohort analytical study and the sampling technique was consecutive non-probability. Parameters including age, gender, anatomical site of lesion, treatment modality and recurrence were evaluated. Recurrences were analyzed over a period of 1-9 years using clinical and radiographic evidence of bone destruction and tooth displacement or resorption and confirmed on biopsy. Three Basal cell naevus syndrome patients were excluded because of higher recurrence rates of syndromic cysts. Histological diagnosis of orthokeratinized odontogenic cyst was excluded from study because of different biological behavior. Patients with incomplete data were also excluded from the study. Ethical approval was obtained from ethical committee AFID.

The quantitative data of age were presented as mean and standard deviation. Percentage and frequencies were calculated for qualitative data including gender and recurrence of OKCs with various surgical treatment modalities.

## RESULTS

This series consisted of 82 lesions in 73 patients. 41 patients (56.2%) were male and 32 (43.8%) were female with male to female ratio of 1.3:1. Age ranged from 7

to 83 years (mean  $38.6 \pm 2.4$ ) with a predominance in 3rd and 4th decade. 67 lesions (81.7%) involved the mandible out of which 49 lesions (73.1%) were located in posterior mandible and 18 (26.9%) were located in symphysis, parasymphysis and anterior body region. 15 lesions (18.3%) involved maxilla, maxillary sinuses, pterygopalatine fossa or floor of the nose. (Table 1)

All these 82 lesions were treated with varying surgical modalities. These include conservative procedures such as marsupialization, enucleation, enucleation with chemical cauterization and/ or peripheral ostectomy to aggressive treatments such as resection. Two (2.4%) cases were treated with marsupialization alone; 7 (8.5%) with enucleation alone, 53 (64.6%) with enucleation and chemical cauterization/ peripheral ostectomy, 15 (18.3%) cases received marginal resection and 5 (6.1%) cases were treated with segmental resection.

The number of patients appeared for follow up was 66, out of which 14 cases of recurrence (21.2%) were found on follow up of 1st to 9th year. Most frequent

TABLE 1: DISTRIBUTION OF OKCS BY ANATOMIC LOCATION AND RECURRENCE

Sites	No. of cysts	Follow up	No. of recurrence	% age of recurrence
<b>Mandible</b>				
Anterior	18	13	2	15.4%
Posterior	49	42	9	21.4%
<b>Maxilla</b>				
Anterior	7	5	1	16.7%
Posterior	8	5	2	40%

Anterior mandible and maxilla includes regions in incisor and, canines and Posterior maxilla and mandible involves premolar and molar region, ramus, tuberosity and pterygopalatine fossa

TABLE 2: SURGICAL MODALITIES AND OUTCOME OF OKCS

Surgical Modality	No. of cysts	Follow up	No. of recurrence	% age of recurrence
Marsupialization	2	2	2	100%
Enucleation	7	6	3	50%
Enucleation + cautery/	53	44	8	18.2%
Peripheral ostectomy	15	10	1	10%
Marginal resection	5	4	0	0%

recurrences were observed in the posterior maxilla and mandible (40% and 21.4% respectively) as compared to anterior maxilla and mandible (16.7% and 15.4% respectively) and mostly occurred in cases treated conservatively i.e., 100% recurrence after marsupialization and 50% after enucleation alone; as compared to aggressive treatment modalities like 18.2% recurrence after enucleation+ cautery/peripheral ostectomy, 10% in marginal resection and 0% in segmental resection (Table 2).

## DISCUSSION

The mean age calculated in this study was  $38.6 \pm 2.4$  years which ranged from 7 to 83 years. The age of distribution of OKC in this study is consistent with international literature, with a peak incidence in 3rd and 4th decade. In a study conducted by Brannan<sup>1</sup>, OKCs occurred most frequently in 3rd and 4th decades and Blanas and colleagues demonstrated the peak incidence in 4th and 5th decade<sup>9</sup>. OKCs are rare in extremes of age i.e. under 10 and above 70. Male predilection was noticed in this study with male to female ratio of 1.3:1 which also conforms to the previous studies by Blanas and colleagues.<sup>9</sup> OKCs predominantly affect the mandible especially posterior mandible including body, angle and ramus region. This is quite similar to results of our study in which mandible was involved in 82% cases. (Table 1)

Treatment of OKCs of Jaw still remains a controversial topic and the choice of one best treatment modality is debatable. Higher recurrence rate has been documented with conservative procedures such as marsupialization and enucleation of the lesion and aggressive therapies, like resection, significantly lowers the recurrence and improves prognosis. In the study by Auluck and Pai, recurrence rate was found to be 62.4% after enucleation alone but it was significantly lower after more aggressive therapy like curettage, cauterization and resection (14.2%).<sup>2</sup> In this study, we found 11 cases of recurrences with enucleation and enucleation + curettage/peripheral ostectomy but only 1 case of recurrence with resection. Recurrence rate was 50% after enucleation (3 cases), 18.2% after enucleation + curettage/peripheral ostectomy (8 cases) and only 7.1% after marginal and segmental resection (1 case). (Table 2)

Segmental resection offers a high cure rate but produces significant morbidity such as loss of jaw continuity, facial disfigurement and need for reconstruction. In two different studies conducted by William and Conner; and Blanas and colleagues; resection was recommended to be the best treatment of choice only in cases of recurrence (3 or more times), in very large and aggressive lesion, in cases of insufficient residual cortical bone or extensive soft tissue involvement.<sup>9,10</sup>

Marsupialization alone as sole treatment modality is not recommended for OKC. However decompression reduces cystic volume and allows removal of 1L-1@ and cytokeratin 10 which is related to expansion of cyst. This causes metaplasia and thus thickening of cyst lining allowing easy removal.<sup>11</sup>

Brannon has, suggested 3 mechanisms for recurrence of OKC.<sup>1</sup> (i) Incomplete removal of cyst walls or epithelial islands of dental lamina associated with OKC (ii) cortical perforation and adherence with soft tissues and presence of daughter cysts within bone (iii) cystic change in dental lamina initially not associated with cyst. Auluck and Pai demonstrated 38% recurrence<sup>2</sup> and Brannon depicted 32.5% recurrence<sup>1</sup> after surgical cure of OKCs. In this study, recurrence occurred in 14 patients with recurrence rate of 21.2%. Recurrence rate is known to be higher after simple enucleation (50% in our study) but treatment with Carnoy's solution or peripheral ostectomy eliminates epithelial islands and microcysts within the peripheral bone.<sup>1,2</sup> These adjuncts when used with enucleation significantly reduce recurrence rate (18.2% in our study).

Carnoy's solution is a mixture of absolute alcohol, glacial acetic acid, chloroform and ferric chloride. It causes chemical cauterization of peripheral bone and soft tissues and its depth of penetration mainly depends on length of application (approx 1.54 mm after 5 minutes application).<sup>2</sup> Peripheral ostectomy should be used with caution as it may disperse microcysts embedded in peripheral bone to cause recurrence.<sup>3</sup> Recurrence is strongly related to presence of cortical perforation and adherence to soft tissues. Supraperiosteal dissection with/without excision of overlying mucosa is indicated in presence of perforation.<sup>1,2,3</sup>

## CONCLUSION

Odontogenic keratocyst is an aggressive cyst, recently categorized as tumour and is associated with high recurrence. Single appropriate treatment is yet to be decided. Surgeons should decide on various modalities as per requirement of the case. Although resection carries least risk of recurrence, it should be limited to recurrent and aggressive OKCs. Enucleation and adjunctive peripheral procedures can be considered optimal for most of the cases.

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#### CONTRIBUTION BY AUTHORS

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|------------------------------|---|
| <b>1 Arsalan Manzoor:</b>    | Main paper writing after data collection. |
| <b>2 Waseem Ahmed:</b>       | Supervisor.                               |
| <b>3 Muhammad Umair:</b>     | Helped in data collection.                |
| <b>4 Gulzar Ali Bukhari:</b> | Read the article and made improvements.   |
| <b>5 Muhammad Omar Niaz:</b> | Helped in data collection.                |