

**CONSERVATIVE APPROACH FOR MANAGEMENT OF ODONTOGENIC
KERATOCYST - A CASE SERIES AND REVIEW OF LITERATURE****Dr. Anchal Varshney**

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Abstract: Odontogenic Keratocyst is characterized by a unicystic or polycystic intraosseous lesion of odontogenic origin. The choice of treatment approach is multifactorial and different conservative and surgical treatment options have been discussed in literature with variations in recurrence. The correct diagnosis and treatment plan deserves consideration in patients to avoid greater mutilation in these individuals. Odontogenic keratocyst should be considered as a differential diagnosis in the cases of proliferative and osteolytic lesions in gnathic bones. Herein we report a case series of seven patients diagnosed with odontogenic keratocyst and submitted to marsupialization followed by enucleation showing no recurrence so far. Out of these one uncommon case of OKC in a six year old and another in maxillary sinus was also reported.

Key-words: Odontogenic Keratocyst, Marsupialization, Conservative management, Recurrence.

Key Message:

- We report a case series of seven patients diagnosed with Odontogenic keratocyst.
- It includes two rare cases of OKC, one involving maxillary sinus (with incidence <1%) and another case in a paediatric patient.
- All the patients were treated conservatively with marsupialization followed by enucleation with no recurrence till date with average period of follow up from 3 to 5 years

Introduction:

Odontogenic Keratocysts (OKC) have long been known for their variegated origin and development, aggressive behavior, high recurrence, treatment modalities, association with syndrome (NBCC), transformation to ameloblastoma & squamous cell carcinoma and debatable nomenclature lately. They were titled as Keratocystic Odontogenic Tumor (KCOT) by the World Health Organization (2005) owing to its recurrent and aggressive¹. In 2017, WHO put it back into the cystic category based on well documented data that it regresses well following decompression². We report a case series of OKCs including two unusual cases with one in a pediatric patient and another in maxillary sinus.

Case Reports

Case 1

A six year old male patient reported with complaint of a hard, gradually increasing, painful swelling in the left mandibular premolar region with no history of trauma. Anamnesis was non-contributory. A solitary, diffuse, bony hard, tender, swelling with intact mucosa was present in the left mandibular premolar region, which approximately measured 2 X 1 cms in diameter. The associated teeth (74 and 75) were mobile. Orthopantomogram (OPG) showed a smooth, unilocular radiolucency with corticated borders measuring 3 X 2.5 cms in region of 73, 74, and 75 with root resorption in relation to 74 and 75 along with unerupted 33, 34, 35 (Figure-1). FNAC revealed dirty yellowish, cheesy, viscid material with total soluble protein content less than 4gm/dl. Surgical management included marsupialization with extraction of 74 and 75. The specimen was sent for Histopathologic Examination (HPE) which reported it to be OKC (Figure-2a). After a year and four months, it was enucleated along with peripheral ostectomy. The histopathology of the enucleated specimen revealed it to be normal oral mucosa (Figure-2b). The subsequent OPGs show satisfactory bone formation with well developing permanent teeth below (Figure-3). We advocate the procedure has maintained good quality life of the patient, who is under a close follow up with no recurrence since three years.

Case 2

A sixty-four-year-old male patient reported with the complaint of white discharge from the left maxillary premolar area which caused a sour taste. On inspection, a solitary, soft, fluctuant, compressible swelling was palpated in the region of chief complaint. Patient was completely edentulous and was a complete denture wearer since four year. OPG revealed unilocular radiolucency of 3 X 4 cms in diameter in the anteroposterior direction not crossing the midline. FNAC yielded white granular viscid material with protein content less than 4gm/dl which pointed the lesion to be OKC. Marsupialization of the lesion was done and was sent for the HPE which confirmed it to be OKC. Enucleation was done after the reduction of size of the residual lesion. The case showed no recurrence till the present day.

Case 3

A forty-two-year-old male reported with a history of mild pain associated with the left mandibular premolar molar region along with mild tingling sensation in the lower left half of lower lip since two months. On examination, a solitary swelling in relation to 34,35,36,37 was present. The swelling was diffuse, firm to hard on palpation with no visible discharge. OPG revealed a multilocular radiolucency present overlying the inferior alveolar nerve with two loci, the superior one measuring approximately 4 X 3 cms and the inferior one measuring 1 X 0.7 cms. The lesion was marsupialized and the HPE reported it to be OKC. Six months post treatment, the neurologic deficit returned to normal and after an year enucleation was done. The case is under close follow up and has not reported any recurrence yet.

Case 4

A twenty-one-year-old male, complained of pain in the right mandibular anterior region with pus discharge. The pain was dull, intermittent in nature which aggravated on chewing. On examining intraorally, a solitary swelling was evident in 43, 44 region. A draining sinus in between 43 and 44 on the buccal side from which a thick, yellow and foul smelling discharge was present. OPG revealed a unilocular radiolucent lesion measuring 3 X 4 cms extending from 42 to 44 in proximity with the lower border of the mandible. The lesion was marsupialized and the specimen was sent for HPE and was diagnosed to be OKC. Antibiotic coverage was ensured for the patient. Enucleation was done after ten months. The follow up OPGs have not yet revealed any recurrence so far.

Case 5

A nineteen-year male, reported with chief complaint of pain and swelling in the right maxillary anterior region for past eight months with numbness over the right cheek. Extraorally, a diffuse swelling measuring 4 X 3 cms with ill defined borders was present over the right cheek region. Intraorally, the swelling corresponded to right maxillary canine eminence and the teeth 13,14 obliterating the buccal vestibule. OPG revealed unilocular radiolucency measuring 3 X 2 cms approaching the right maxillary sinus. PNS view showed haziness with thick lining in the right maxillary sinus. Using Caldwell Luc approach the lesion was marsupialized and sent for HPE which confirmed it to be OKC. Complete resolution of numbness occurred after four months of the treatment. Enucleation was done after eight months when the size of the swelling reduced to less than a centimetre. The case is under regular follow up with no recurrence till date.

Case 6

A thirty-eight-year-old male patient came for routine dental check up. On examination, a pea sized swelling was noticed in relation to 36. OPG revealed a unilocular radiolucency measuring 2 X 3 cms from 35 to 37. Marsupialization was done saving all teeth. The cavity was horizontally packed using iodoform gauze which was changed every other day till the cavity reduced in size to less than half. The histopathologic report gave a diagnosis of OKC. Enucleation was done after seven months. The case has healed well with no recurrence till date.

Case 7

A thirty-seven-year-old male, reported with chief complaint of swelling and pain in lower right back tooth region since two years. On clinical examination, no significant finding was found. However OPG revealed a unilocular radiolucency measuring 2 X 3 cm extending from mesial surface of right mandibular second premolar till distal surface of the right mandibular first molar was noted. The lesion was marsupialized and was sent for HPE which specified it to be OKC. Enucleation was done after eight months. The case has not shown any recurrence till date.

Discussion

The Odontogenic Keratocyst has been known in the literature for a long time. Philipsen was the first to describe it in the year 1956. It is defined as “a benign uni- or multicystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized

stratified squamous epithelium and potential for aggressive, infiltrative behaviour"¹. The genetic basis for this was a tumour suppressor gene, PTCH ("patched") which is involved in both sporadic KCOTs and NBCCS occurring on chromosome 9q22.3-q31 and mutation in PTCH results in the upregulated proliferation-stimulating effects of SMO (smoothened)³. OKC comprises approximately 3-11% of odontogenic cysts⁴. Mostly they occur in the posterior mandible and radiographically present as unilocular or multilocular radiolucency. Multiple OKCs are considered to be a part of Nevoid Basal Cell Carcinoma Syndrome⁵. They have been reported to transform into ameloblastoma and primary intraosseous squamous cell carcinoma (PIOSCC)⁶. They tend to grow in the anteroposterior direction without causing obvious bone expansion which is why they are observed late by the patients⁷. The recurrence rates of OKC variegate a lot ranging from 0% to 62.5% with the majority of cases showing recurrence in the first five years⁸. As per Morgan et al, recurrence rate of 78% occurred in five years or less while 22% occurring after five years of initial treatment⁹. It presents mostly in the second to fourth decades of life (54.2%), but the cases have also been reported in first and the ninth decade of life¹⁰⁻¹².

The treatment choice should depend upon various factors like the age of the patient, clinical extent of the pathology, location and proximity to vital structures, mobility of overlying teeth, radiographic appearance of the lesion, involvement of the soft tissues, variant of the lesion, presence of daughter cysts and recurrence rate. The various treatment modalities are debatable for their pros and cons. Conservative management includes simple enucleation or marsupialization with or without curettage. Aggressive treatment usually includes cryotherapy, electrocautery, peripheral ostectomy or enbloc/segmental resection. However, the treatment of the OKC still remains controversial.

Nakamura in his case series of 28 patients reported marsupialization as an effective treatment modality in treating large OKC with no recurrence¹³. de Molon showed no recurrence in an OKC case treated by the same modality with a follow up period of five years¹⁴. In a case series of 23 patients, Marker et al concluded that decompression with and without enucleation is a successful modality with low recurrence rate¹⁵. Less than 1% of OKC cases occur in the maxilla with sinus involvement¹⁶. Torres-Lagares, Ohki and Olson advocated marsupialization as a successful treatment modality for OKC in maxillary sinus¹⁷⁻¹⁹. We also report a case series of seven patients diagnosed with OKC with one case in a pediatric patient and the other in maxillary sinus both of which are not common to find²⁰. All the cases were treated successfully by marsupialization followed by enucleation with no recurrence till date with an average period of follow up ranging from three to five years. The technique of marsupialization as described by Pogrel, is to make a window of at least a cm into the cyst, and the outline of the cyst mucosa is sutured with the oral mucosa^{21,22}. Decompression relieves the pressure within the cystic cavity and thus augments new bone fill. It has been well documented that after decompression and marsupialization the cyst lining undergoes histological changes and is eventually replaced by oral nonkeratinizing epithelium^{23,24}. The same was observed in our cases also. As advocated by Stoeltinga, the bony defect can be additionally treated with carnoy's solution too to further limit the recurrence²⁵. Its average depth of penetration is 1.54 mm after five minutes of application²⁶. Iodoform as a dressing material is shown to exert antimicrobial activity after topical application to wounds, reduces oedema through fibrinolytic activity and decreases pain

by covering denuded bone surface²⁷. Weighing both pros and cons of marsupialization, one cannot deny its usefulness especially when followed by enucleation.

Conclusion

We advocate the conservative approach of decompression and marsupialization followed by enucleation for odontogenic keratocysts as it maintains the function & esthetics in adults and added to this the growth potential of the jaw in pediatrics. We also conclude that the lining of OKC changes to normal epithelium once it is exposed to the oral cavity after marsupialization. Out of the multiple treatment options available, the aim is to choose the treatment option that carries the minimum risk of recurrence along with the lowest morbidity. Thus, conservative management for OKC has proven its worth through the test of the time.

Declaration of interests: None

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Figures:



Figure 1

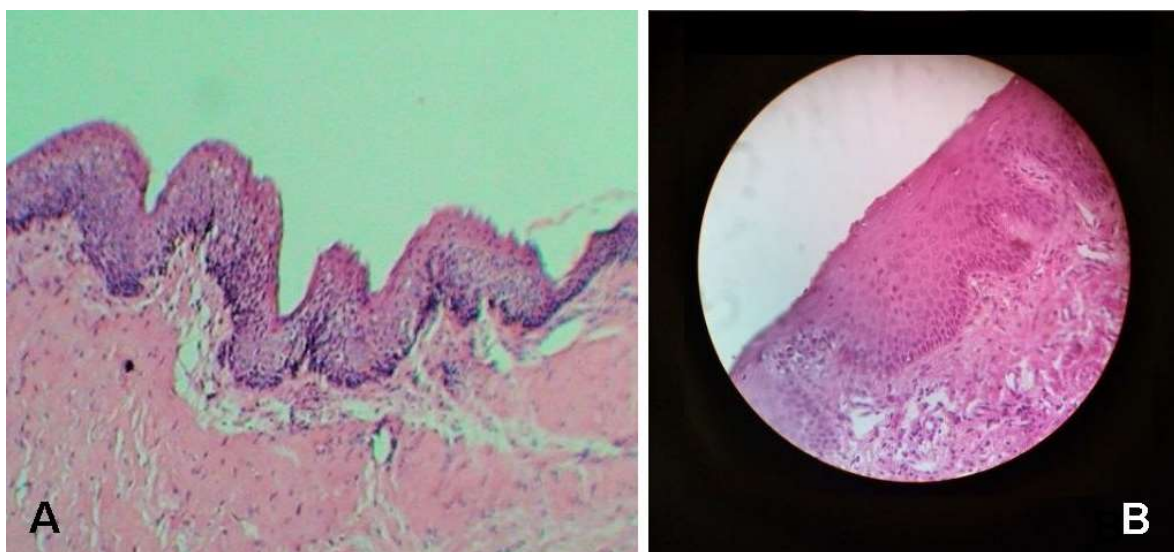


Figure 2



Figure 3

Figure legends:

Figure 1) Preoperative panoramic radiograph.

Figure 2a) Hematoxylin and Eosin staining 10X- Cystic Lining;

2b) Hematoxylin and Eosin staining 40X- Normal oral epithelium.

Figure 3) Two-year postoperative panoramic radiograph