
SURGICAL MANAGEMENT OF A KERATOCYSTIC ODONTOGENIC TUMOR IN MANDIBLE: A CASE REPORT WITH REVIEW OF LITERATURE

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ABSTRACT:

Odontogenic keratocyst which is now called as Keratocystic Odontogenic Tumor, from 2005 by WHO is an odontogenic lesion, known for its recurrence. Commonly it affects the mandibular posterior region followed by premaxillary region. The treatment options of keratocystic odontogenic tumor varies from conservative approaches like marsupialisation, marsupialisation with enucleation, enucleation with peripheral ostectomy, enucleation with mucosal excision, enucleation with chemical cauterisation, enucleation with peripheral ostectomy followed by chemical cauterisation, enucleation with cryosurgery to a radical approach like resection. Each modality has its own advantages and disadvantages. Here we discuss a case of odontogenic keratocyst in the posterior mandible with relevant review of literature and its management.

Keywords: Marsupialisation, Enucleation, Peripheral ostectomy, Carnoy's solution



INTRODUCTION:

The term Odontogenic keratocyst was first described by Philipsen in 1956. Due to its aggressive nature, high recurrence and specific histopathologic feature it was reclassified under cystic tumors by WHO in 2005 and was renamed as Keratocystic Odontogenic Tumor.^[1] The keratocystic odontogenic tumor is a benign, uni or multicystic, intra osseous lesion predominantly in posterior mandible followed by premaxilla and posterior maxilla with male predominance of ratio 1.7:1 occurring in second and third decades of life.^[2]

There are two theories about the origin of Odontogenic keratocyst dental lamina theory and basal cell theory. The dental lamina theory states that odontogenic keratocyst originates from the cell rests of dental lamina.^[3] And according to the basal cell theory the odontogenic keratocyst originates from the basal cell of oral mucosa.^[4] Binkley and Johnson first reported a case of multiple keratocyst with basal epithelioma in 1951 and named it as Basal cell nevi syndrome.^[5] Later, the syndrome was found to have other characteristic features like basal cell nevi and bifid ribs as described by Gorlin and Goltz. They named the

syndrome as Nevoid Basal cell carcinoma syndrome or Gorlin Goltz syndrome.^[6]

Clinical features of keratocystic odontogenic tumor include pain, swelling, tooth displacement and root resorption rarely paresthesia and pathologic fracture.^[7] Some lesion may remain asymptomatic and revealed during radiographic examination.^[8] Radiologically the keratocystic odontogenic tumor appears as a well defined radiolucency with sclerotic margins which may be unilocular or multilocular but some part of borders may be diffuse as well.^[9]

Histopathologically the KCOT is parakeratinised stratified squamous epithelium with polarised basal cell in picket fence appearance.^[10] The aim of this report is to discuss a case of KCOT which was managed with enucleation and peripheral ostectomy along with carnoy's solution fixation with relevant review of literature.

CASE DETAIL:

A 25 year old male patient reported to department of Oral and Maxillofacial surgery with the complaints of painful swelling in left lower back tooth region of jaw for past 4 months. On examination patient showed intra oral swelling with obliteration of mucobuccal fold in relation to 37, 38. The margins were diffuse and surface of swelling was normal and tender on palpation. The orthopantomogram revealed an unilocular radiolucency

with sclerotic borders extending from the distal aspect of 36 to distal aspect of 38 (Figure 1). The cystic content aspirate was creamy white in colour with protein content of 4.7gm/dl.

Initially under local anaesthesia, extraction of involved teeth 37, 38 followed by decompression and the cavity was packed with iodoform gauze. A small portion of cystic lining was sent for histopathologic examination. The result of histopathologic examination confirmed as keratocystic odontogenic tumor. The proposed treatment plan was cyst enucleation followed by peripheral ostectomy and fixation with Carnoy's solution. Under general anesthesia the cystic lining was enucleated in toto and peripheral ostectomy was done and the cavity was cauterised with Carnoy's solution. Figure 2 shows enucleated specimen of size 2.2cm x 1.5cm x1cm. Figure 3 shows the extent of cystic cavity after enucleation of cystic lesion. Figure 4 shows application of Carnoy's solution. The surgical site was closed primarily and healing was uneventful.

DISCUSSION:

The KCOT comprises a total of 11% of cysts occurring in the maxillofacial region.^[11] The KCOT is regarded the most aggressive because of mural growth expansion. Biochemical markers like Ki67, PCNA, Cyclin D and genetic markers like PTCH mutation, bcl 2, gp 38, p53 over expression are associated with extensive growth rate of cystic epithelium and development

of satellite cysts.^[12] The satellite cysts are often embedded in the soft tissue when cortical perforation occurred. This is very important factor for recurrence. The thin and friable epithelium, innate infiltrative potential are other factors in recurrence rate which varies from 30-60%.^[13]

The treatment of this odontogenic keratocyst is based on the above aggressiveness of the lesion. Treatment approaches include

- Marsupialisation^[14]
- Marsupialisation with Enucleation^[15]
- Enucleation^[16]
- Enucleation with Carnoy's fixation^[17]
- Enucleation and Peripheral ostectomy^[18]
- Enucleation and Peripheral ostectomy with Carnoy's fixation.^[19]
- Enucleation and Cryotherapy^[20]
- Resection^[21]

Marsupialisation is preferred in patients of younger age and cysts which are large to avoid deformity and to preserve the vital structures which

are in close proximity. But the recurrence rate is 0-100%.^[17] Pogrel et al^[22] reported that the cystic lining changes to oral epithelium after marsupialisation. And according to Shear et al.^[23] the factors like interleukin 1 α and cytokeratins which are responsible for bone resorption are lost after marsupialisation and alter the behaviour of lesion to less aggressive form. However authors like Voorsmit et al,^[17] Kondell & Wiberg^[24]; Stoelinga & Bronkhorst;^[25] Nakamura et al;^[26] Stoelinga.^[27] found no difference in recurrence rate while comparing marsupialisation and simple enucleation. After the reduction of size of the cyst by marsupialisation, enucleation can be performed. It allows the complete removal of the cystic lining. If the cystic cavity is large after marsupialisation or enucleation, there is increased chance of post operative complications like wound dehiscence, infection due to lack of bony support for the suture to stay. Zobriel et al.^[28] showed that the process of bone grafting in to the cystic cavity decreased the complications. However the disadvantage of radio opaque appearance in follow-ups occurs with grafting. Autologous bone graft is commonly used. Other than this a various bone grafts like allogenic, xenogenic and alloplastic materials are also used.

Table I : Reviews comparing Marsupialisation alone with Marsupialisation and Enucleation

S.No	Study	Year	Procedure	No. Of cyst	Recurrence rate in percentage
1.	Pindborg and Hansen ²⁹	1963	Marsupialisation and Enucleation	16	62.5%
2.	Pogrel and Jordan ²²	2004	Marsupialisation followed by Cystectomy	10	0%
3.	Maurette, Jorge and De Moraes ⁷	2006	Marsupialisation followed by Enucleation	30	14%
4.	Browne ³⁰	1970	Marsupialisation	12	25%
			Enucleation and primary closure	42	26.2%
			Enucleation and packing open	30	20%

From the review of literatures in Table 1, it is clear that recurrence rate for marsupialisation is comparable with the recurrence rate of marsupialisation followed by enucleation except for the

Pinborg and Hansen.^[29] According to them the recurrence rate of marsupialisation followed by enucleation was as high as 62.5%.

Table II – Reviews comparing the type of Enucleation with Recurrence rate.

S.No	Study	Year	Procedure	No. Of cyst	Recurrence rate in percentage
1.	Kondell and Wiberg ²⁴	1988	Enucleation	29	24%
2.	Voorsmit, Stoeltinga and Van Haelst ¹⁷	1981	Enucleation	50	14%
			Enucleation, excision of overlying mucosa and Camoy's solution	42	2.5%
3.	Forssell, Forssell and Kahnberg ³²	1988	Enucleation in single piece	28	18%
			Enucleation in multiple piece	41	56%
			Marsupialisation	5	60%
4.	Partridge and Towers ³²	1987	Marsupialisation	2	0%
			Enucleation and primary closure	30	36.7%
			Radical curettage periosteal removal and packing	11	9%
			Resection with immediate bone graft		
5.	Schmidt and Pogrel ³³	2001	Enucleation and Cryotherapy	26	11.5%

Enucleation and curettage is the process by which the entire cystic lining is curetted out from the cystic cavity.

From the review of literature in table II it is said that enucleation does not eliminate recurrence, as the recurrence

rate remains in between 24% to 36.7%. Forssell and Kahnberg,^[32] stated recurrence also depends in the form in which the cystic lining is enucleated as when the lining is enucleated as multiple pieces the recurrence rate

increases to 56% when compared to the single piece enucleation with the recurrence rate of 18%. According to Partridge and Tower enucleation resulted in recurrence with the recurrence rate of 36.7%.

Table III – Reviews comparing Enucleation and the type of adjuvant procedures with Recurrence rate

S.No	Study	Year	Procedure	No. of cyst	Recurrence rate in percentage
1.	el-Hajj and Anneroth ³⁴	1996	Enucleation	63	29%
			Enucleation and Cryosurgery	16	38%
			Enucleation and Peripheral ostectomy	1	0%
			Enucleation and cryosurgery and Peripheral ostectomy	2	50%
			Resection	3	0%
2.	Morgan, Burton and Quian ¹⁸	2005	Enucleation	11	55%
			Enucleation with Peripheral ostectomy	11	18%
			Enucleation with Peripheral ostectomy and carnoy's solution	13	0%
			Enucleation and carnoy's solution	2	50%
			Resection	3	0%
3.	Zachariades ³⁵	1985	Enucleation	13	31%
			Resection	1	0%
			Marsupialisation	1	0%
			Decompression and enucleation	1	0%
4.	Madras J and Lapointe H ³⁶	2008	Curettage	16	37.5%
			Marsupialisation	3	0%
			Resection	2	0%
5.	Irvine and Boweman ³⁷	1985	Enucleation	7	16.7%
			Radical enucleation	6	0%
			Resection	2	0%

The review of literature from table III shows that additional procedures like chemical cauterisation and cryosurgery and peripheral ostectomy the recurrence rate decreases to 2.5% and 11.5% and 18% respectively. Due to the advent of chemical cauterisation, the treatment of KCOT has been improvised. Carnoy's solution was introduced by Culter and Zollinger in 1933.^[38] for tissue fixation. The properties of carnoy's solution are

powerful fixatives, hemostatic and cauterising agent which penetrate through the cancellous bone for devitalising the tumor cells.^[39] The precautions are followed in its application as the solution is caustic and cause mucosal injuries and damages adjacent vital structure like inferior alveolar nerve and maxillary sinus.^[27] The contents include 60% - absolute alcohol, 30% - chloroform, 10%- glacial acetic acid and 1gm of

ferric chloride. The alcohol shrinks and hardens the tissue while the glacial acetic acid prevents over hardening, the chloroform increases the speed of fixation and ferric chloride act as a dehydrating agent.^[40] Due to the carcinogenic potential of chloroform, the modified carnoy's solution and Farmer's solution was used for cauterization of tumor cells. The Farmer's solution excludes the ferric chloride and chloroform and the modified carnoy's solution excludes the chloroform from its constituents.^[41] The time dependent penetration of the solution is enhanced by glacial acetic acid, which allows the penetration into the connective tissue and bone.^[40]

The average depth of penetration by carnoy's solution is 0.3 mm in 3 minutes.^[42] and 1.54mm in 5 minutes.^[43] The Steolinga protocol in 2003 states that after peripheral ostectomy the carnoy's solution was applied in the cystic cavity, twice for 5 minutes each along with excision of surrounding mucosa.^[27] The latest modification include the use of carnoy's solution for 3 minutes each for 3 times reducing the damage to neural tissues. According to Morgan, Burton and Quian the recurrence rate reduces to 0% if the carnoy's solution is used after peripheral ostectomy and enucleation. However some authors advocate conservative modalities like marsupialisation, decompression and simple enucleation for smaller cysts of 1cm in size as per wait and see protocol.^[44] And radical treatment like

Carnoy's and peripheral ostectomy, resection are indicated in larger cystic lesion, recurrent lesion and association with Nevoid Basal Cell Carcinoma syndrome. The recurrence occur within period of first 5 years, so a post operative follow up of 5 years is required.^[45]

CONCLUSION:

The only modality with nil recurrence is surgical resection, the debate continues between conservative surgical management and aggressive surgical resection in the management of Keratocystic odontogenic tumor. Though the carnoy's solution and other adjuvant procedures have risk of sinus and neural injuries, to avoid deformities and in less aggressive cases the adjuvant procedures like enucleation followed by peripheral ostectomy and chemical cauterisation with carnoy's solution.^[18] and cryosurgery decreases the probability of recurrence drastically. Thus the benefits of using these adjuvant procedures clearly outweigh the risks.

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FIGURES:



Figure 1: preoperative orthopantomogram showing cyst in relation to 37,38 region



Figure 2: enucleated specimen



Figure 4: application of carnoys solution in the cyst cavity

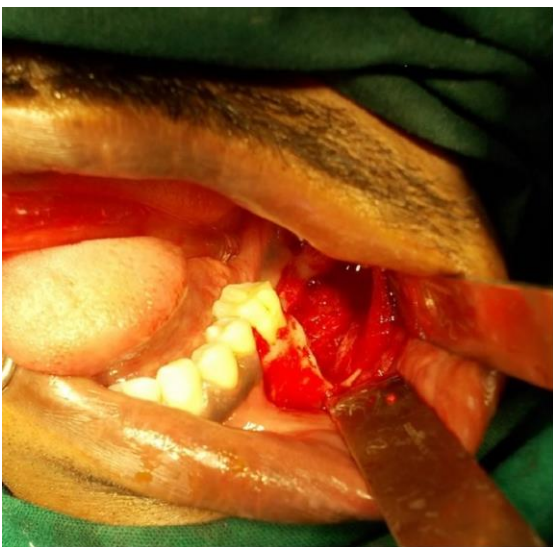


Figure 3: cystic cavity post enucleation of the cyst