Odontogenic Tumors Treatment Paradigm with Long Term Follow-Up Case Series

Jacob John1, DhanyaPrakash G2, Mubarak Aziz3, Lipin Peter4, Milind Wasnik5, Milind Atulkar6

1-MDS, Professor, Department of Oral and Maxillofacial Surgery, Azeemia College of Dental Sciences and Research, Meyannoor, Kollam, Kerala, India. 2,4-Junior Resident, Department of Oral and Maxillofacial Surgery, Azeemia College of Dental Sciences and Research, Meyannoor, Kollam, Kerala, India. 3-MDS, Senior Lecturer, Department of Oral and Maxillofacial Surgery, Azeemia College of Dental Sciences and Research, Meyannoor, Kollam, Kerala, India. 5,6-MDS Post Graduate Student, Department of Pediatric & Preventive Dentistry, Swarigya Dadasaheb Kalmegh Smruti Dental College Nagpur Maharashtra India.

Correspondence to:
Dr. Jacob John, MDS, Professor, Department of Oral and Maxillofacial Surgery, Azeemia College of Dental Sciences and Research, Meyannoor, Kollam, Kerala, India.
Contact Us: www.ijohmr.com

ABSTRACT

Purpose: A variety of treatment modalities has been suggested for the management of keratocystic odontogenic tumours and unicystic ameloblastoma of the mandible. The purpose of this follow-up study was to evaluate the decrease in recurrence rate of odontogenic tumours of the posterior mandible with less morbid surgical management. Materials and Methods: A retrospective study of 21 patients with the pathologic diagnosis of Keratocystic odontogenic tumour (KCOT) and unicystic ameloblastoma (intraluminal) involving the posterior mandible, treated with enucleation, curettage, peripheral ostectomy and chemical catarization under general anaesthesia, at Azeemia Medical College, Kollam from March 2006 to April 2015. Clinical, radiographic and histological data were collected for each patient, and five-years postoperative follow-up was done for each case. Results: Out of twenty-one cases of odontogenic tumours 17 cases of KCOT and 4 cases of unicystic ameloblastoma were evaluated in detail, none of the cases showed recurrence during the five-years follow-up period. Conclusion: Based on the review reports and our study details, we can conclude that enucleation, curetage, peripheral ostectomy and chemical catarization using carnoy’s solution provides the best, less aggressive surgical treatment protocol to the patient.

KEYWORDS: Keratocystic Odontogenic Tumour, Enucleation, Carnoy’s Solution, Peripheral Ostectomy

INTRODUCTION

An odontogenic lesion may require surgical management because of its physical presence, biologic behaviour, biologic activity or growth potential. The goal of the treatment is eradication of a lesion with the least morbidity possible and the preservation or restoration of form and function.1,2

The keratocystic odontogenic tumour is a benign neoplasm of odontogenic origin, is most often unilocular or multilocular well-circumscribed radiolucent lesion, surrounded by smooth or scalloped margins with sclerotic borders. KCOT frequently occur in 310 and 410 decades of life in the posterior region of the mandible with high incidence in male patients. Unicystic ameloblastoma is a single cystic lesion with ameloblastomatous lining, first introduced in 1977 as a separate entity than a solid ameloblastoma. Most common presentation is associated with severely displaced and impacted third molar. Next commonly involving areas are premolar region or at the ramus region without any relationship with the tooth, mostly occurring at 410 and 510 decades of life.

Keratocystic odontogenic tumour and ameloblastoma are noted for its locally aggressive nature and its recurrence rate1. According to current literature, a broad range of recurrence rates for various surgical treatment options of about 0% - 62%, with most presenting within five years of treatment.4,5 As this a nonmalignant disease, resection to achieve a cure will be more morbid than the disease. Enucleation along with any or more of the adjunctive treatments, like curettage, peripheral ostectomy, chemical catarization (carnoy’s solution), curettage and decompression and longitudinal irrigation along with subsequent residual cystectomy is a less morbid treatment procedure, but the recurrence reported to be very high.6,7,8

Here we report our management protocol and five-years follow-up details of twenty-one cases of odontogenic tumours of mandible [keratocystic odontogenic tumour &unicystic ameloblastoma (intraluminal)] reported at our centre (Azeemia College of Dental Sciences And Research, Meeyannoor, Kollam, Kerala, India ) from March 2006 to April 2015.

CASE REPORT

In this follow-up study, we have included the cases of odontogenic tumours of the posterior mandible, which have been reported to our centre. Out of twenty-one cases, 17 cases were keratocystic odontogenic tumours, and four cases were unicystic ameloblastoma, and all...
those were large lesions involving the body, angle, extending towards the ramus, condyle and coronoid process. Most of the patients were of average age 25-40 yrs, and about 14 males and seven females. As the part of the preoperative evaluation, OPG and CT scan of the mandible (axial and coronal cuts) were advised in all cases. An incisional biopsy and aspiration of the cystic contents were done in all the cases to confirm the preoperative diagnosis. All the cases were operated by the same surgeon in the same paradigm.

Under general anaesthesia, in all the cases a submandibular approach (modified Risdon and Hind’s) was performed, (fig.1) and in cases of larger lesion modified retromandibular approach (Hinds) was performed. The initial incision was carried through skin and subcutaneous tissues to the level of the platysma muscle, incision of platysma, superficial layer of deep cervical fascia, ligation of the facial artery, retraction of the marginal mandibular branch of facial nerve and dissection of the pterygomasseteric sling was done to reach the inferior border of mandible and to expose the site. Peripheral ostectomy was performed to deroof the lesion. After enucleation and thorough curettage, peripheral ostectomy of about 1-3mm (mechanical debridement) was performed. (Fig 2,3) Irrigation and debridement was followed by the application of carnoy’s solution[Ferric chloride-1 gram (1 parts), Chloroform - 3ml (3 parts), Glacial acetic acid - 1ml (1 parts), Absolute alcohol - 6ml (6 parts)] for 5 minutes was performed. Application of carnoy’s solution over the involved areas of the nerve and soft tissue were done (Fig 4). After thorough irrigation and debridement, small vacuum drain was placed and wound closed in layers. In one case where the lesion was extending from one angle crossing the midline to opposite angle, we performed a transmucosal approach, and the treatment protocol remained the same. Postoperative period was uneventful, and all the cases were discharged on the second postoperative day.

Transient paresthesia of the inferior alveolar nerve noted for few cases (5 cases of KCOT and in 2 ameloblastoma cases) and became normal within 1-3 months of time. Neuropraxia of marginal mandibular nerve noted in 3 cases (recovered within 1-2 months of time). In all the cases healing was satisfactory.
Fig 5 shows preoperative CT axial cuts of another patient with a lesion (keratocystic odontogenic tumour) involving the posterior mandible. Fig 6 shows post operative five years follow up OPG of the same patient.

Fig 7 shows OPG with large multilocular lesion extending from (R) angle crossing the midline extending into the (L) angle of a patient.

Fig 8(A) shows Transmucosal approach to expose the lesion (B) immediate postoperative view of the lesion. Fig 9 shows (a) Six months postoperative OPG. 9(b) 4 year follow up OPG of the same patient.

The data collected from 21 cases were tabulated and analysed (Table 1).

Table 1: Descriptive statistical analysis of the data collected.

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DISCUSSION

The ameloblastoma and keratocystic odontogenic tumour are among the most aggressive odontogenic tumours, due to its relatively high recurrence rate, its relatively fast growth, and its tendency to invade adjacent tissues. The unicystic ameloblastoma usually appears as a cystic lesion with either an intraluminal proliferation of the cystic lining. In cases of intraluminal or plexiform pattern, in which no penetration of fibrous capsule by ameloblastic cells, enucleation, and curettage suffices. But if there is a mural component that extends up to the wall or the level of interface with the bone, the bony recession is necessary to ensure adequate removal. Similarly, it becomes difficult to rule out mural invasion of the fibrous tissue wall with the single incisional biopsy because of the potential for achieving nonrepresentative sample.

Scharfetter et al. demonstrated both slowly and rapidly proliferating areas in different parts of the KCOT epithelium, and its invasive growth probably resulted from the active growth of the connective tissue wall. Other possible explanations for its high recurrence rate are increased fibrinolytic activity in the cyst wall, increased mitotic activity and epithelial proliferation in connective tissue, and residual dental lamina with subsequent new cyst formation. The bone resorption is mediated by activation of osteoclast-like cells and biologically active collagenases.

A review of the biological behaviour of this aggressive pathological entity, the molecular (growth factors, p53,
PCNA and bcl-2) and genetic alterations associated with this lesion, strengthen the current concept that the KCOT should be regarded as a neoplasm. Markers known to be rapidly induced in response to growth factors, tumour promoters, oncogenes, hormones and shear stress, such as COX-2, may explain the development of these benign but sometimes aggressive neoplasms of the jaws. Fig 10 shows H&E – 40x showing parakeratinized the stratified squamous epithelium of 6-8 cells thickness (KCOT). Fig 11 shows delicate connective tissue capsule surrounded by odontogenic epithelium (ameloblastoma). Fig 12 shows ameloblastic lining showing luminal and intramural growth.

Treatment of KCOT and ameloblastoma (unicystic) remains controversial, the choice of treatment approach should be based on the age of the patient, the size of the cyst, its location, recurrence status, and radiographic evidence of cortical perforation. Radical surgery includes resection (with or without continuity defects), has been advocated for larger KCOTs, aggressive and recurrent lesions. In cases of resection with continuity defects, reconstruction with bone grafts (preferably autogenous) or transport distraction osteogenesis using indigenous distractors is yet another affordable option for reconstruction of mandibular defects.

Resection offers the high cure rate but produces significant morbidity and facial disfigurement. There fore should be reserved only for aggressive or recurrent lesions, for the patients who cannot be closely followed-up after not aggressive treatments. On the other hand, radical treatment would adversely affect the psychosocial condition of the young patients and also would inhibit growth potential especially if done in the first decade of life. Less aggressive surgical procedures would be preferred in young patients because of low morbidity rate and conserving the teeth adjacent to the lesion.

Another treatment as described in the reports of Brondum, Jensen and Marker et al., is that decompression is accomplished by opening the cystic cavity and establishment of drainage. The disadvantages are the requirement of subsequent enucleation, two surgical procedures and the treatment time needed is comparatively long. August et al. Epithelial differentiation and loss of cytokeratin-10 production observed in 64% of patients treated with the cyst decompression or irrigation after a nine-month average treatment time. Advocates of conservative treatment suggest that marsupialization is the exteriorization of cyst or an enclosed cavity by resecting a portion of its wall followed by approximating the cut edges to adjacent soft tissue, thereby creating a pouch, and they suggest that marsupialization yields the comparable results to those obtained with more extensive surgery. Dredging Method is a conservative surgical procedure in which, after enucleation, repeated peripheral ostectomy is carried out to accelerate new bone formation by removing out the scar tissue from the bony cavity. Dredging is done in every 2-3 month's interval, and the follow-up begins when the tumour cells are not identified in two consecutive microscopic examinations of the scar tissues. Continuous and regular follow-up is an essential part of the treatment.

Enucleation of the KCOTs followed by open packing has been suggested as another conservative method of surgical treatment. The resulting cavity was irrigated with the mixture of normal saline and chlorhexidine gluconate followed by packing with iodoform gauze impregnated with bacitracin ointment to minimize the risk of recurrence. Currently, a total enucleation, with or without a “peripheral ostectomy” presents the most common surgical procedures used to treat most of the KCOTs.

About 940 cases of keratocystic odontogenic tumour analyzed in two separate systemic reviews by Johnson and Blanas et al. during the period between 1970 and
2010 have helped to make recommendations as to which management techniques can best minimize the chance of recurrence. Johnson in his systemic review demonstrated that, other than resection, enucleation with Carnoy solution in most of the cases, when compared with other measures shows high recurrence control. Blanas et al. reported that simple enucleation, curettage with adjunctive measures without Carnoy solution resulted in a recurrence rate of 27.8% and 30.8%, followed by marsupialization alone 18.2%. The use of Carnoy’s solution along with enucleation and curettage reduced the recurrence rate to 4.8%, and the rate for resection is about 1.85%. Chirapathomsakul et al. reported one recurrence in 5 patients (20%) but did not disclose whether chloroform was included in the carnoy’s solution. In another study conducted by Stoelinga et al., demonstrated that original formulation of carnoy’s solution was used and the recurrence rate was remarkably low (0 recurrences in 20 cases and 0 recurrences in 5 cases in another study). Voorsmit et al. reported one recurrence in 40 cases (2.5%) in their study. Carnoy’s solution was first used as a medicament in surgery by Cutler and Zollinger in 1933. It is a powerful haemostatic, fixative and cauterizing agent which can penetrate cancellous spaces in the bone and also devitalizes and fixes the remaining tumour cells. Its average depth of penetration is 1.54mm after five mins of application. However, Blanas et al., states that Carnoy’s solution will not cause damage to the inferior alveolar nerve, even after 3 minutes application. The most common procedures done when definitively treating a KCOT was Enucleation + Mechanical Curettage (Curettage and Peripheral ostectomy) followed by Enucleation + Mechanical Curettage + Chemical curettage (Carnoy’s solution). Carnoy solution described by Voorsmit (1981) contains ethanol (100%), chloroform and glacial acetic acid in a 6:3:1 ratio along with ferric chloride. This composition provides a tissue fixation property and tissue cauterization property and is utilized in chemical curettage.

Four treatment modalities for unicystic ameloblastoma were identified. According to a structured, systemic review by S L Lau and N. Samman, the recurrence rates for the treatment procedures for ameloblastoma were, 30.5% for enucleation, 3.6% for resection, 16% for enucleation with application of carnoy’s solution and 18% for marsupialization with/without other treatment in a second phase. Enucleation resulted in highest recurrence rate and treatment by marsupialization cannot be fully evaluated because most cases were followed by a second stage surgery.

**CONCLUSION**

Among the different options for the treatment of odontogenic tumours, resection offers highest cure rate but produce significant morbidity. In this study, we have reviewed all the conservative options for the management of keratocystic odontogenic tumours and intraluminal unicystic ameloblastoma. Based on the review reports and our study details, we can conclude that enucleation, curettage, peripheral ostectomy and chemical cauterization using carnoy’s solution provides the best less morbid surgical treatment protocol to the patient.
33. Fixatives for plant cytogenetics, wheat genetic and genomic resources centre, Kansas University, April (2007)

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