

A RATIONAL MANAGEMENT APPROACH TO AN ODONTOGENIC KERATOCYST (A LONG TERM FOLLOW-UP)

¹AQIB SOHAIL, BSc, BDS, FCPS (Oral Surgery)

²TAYYABA RAFIQ, BDS, FCPS II Resident (Oral Surgery)

ABSTRACT

Odontogenic keratocyst (keratocystic odontogenic tumor) is a destructive jaw lesion with the propensity to recurrence. The atypical symphyseal odontogenic keratocyst was managed with a defined protocol which entailed diagnosis, treatment with enucleation along with peripheral ostectomy and rehabilitation. A long term follow-up schedule was provided to the patient to observe the recurrence behavior of this cyst. In post operative phase, no complication was noticed regarding wound healing and recurrence.

Therefore, defined treatment protocol and long term follow-up may provide better means to manage cysts with such indistinct recurrence behavior. Future treatment may involve molecular-based modalities which may reduce or eliminate the need for aggressive surgical management.

CASE REPORT

An 18 year old male presented to Oral Surgery department, Lahore Medical & Dental College in April 2004 with the chief complaint of gradual drifting of lower anterior teeth, which he noticed three month before his visit to the department. He has had no history of trauma. Three year back, he had got his left lower 2nd premolar and 1st molar extracted. Medical history was insignificant.

Extra oral examination revealed less pronounced swelling that off centered the chin. (Figure 1A) Swelling was non tender to palpation. Neither the lymph nodes nor impairment of labial sensation could be demonstrated. Oral examination revealed oral mucosa with slight erythematous appearance. (Figure 1A, B) The labial cortex of mandible was expanded and crepitus was also appreciated. All anterior teeth were displaced but were firm and non tender to percussion. (Figure 1C)

On radiographic examination, a fairly large multilocular radiolucent area with distinct margins was seen extending from right 1st molar to the mesial aspect of the left 2nd molar. The lower border cortex was thin mainly in between the mental foramina. The

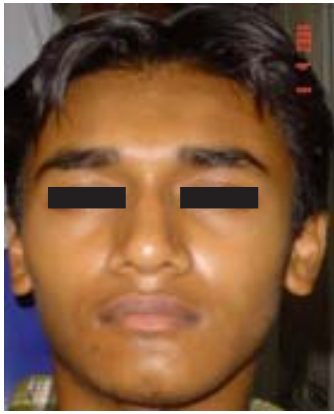
inferior alveolar canal was pushed down to the lower border of mandible. (Figure 2)

All mandibular teeth were vital with electric pulp tester. Thick, creamy, odorless fluid was observed on aspiration with a disposable syringe that may indicate odontogenic keratocyst. Incision biopsy was also performed. Aspiration cytology and histopathology reflected keratinized desquamated squamous epithelial cells and parakeratinized squamous epithelium with no signs of malignancy respectively. (Figure 3)

It was decided to perform enucleation with peripheral ostectomy under general anesthesia. Preservation of all involved mandibular teeth except broken down roots of right 1st molar was also warranted. Root canal therapy was offered to involved teeth i.e. from right 2nd premolar to left 1st premolar. In July 2004, enucleation with peripheral ostectomy was performed with the salvage of lower border. Apicoectomies of all involved teeth were also completed. The patient was strictly reviewed to observe recurrence. Missing teeth were rehabilitated with porcelain bridges. To date, post operative follow up has been uneventful regarding wound healing and recurrence. (Figure 4A-D) Lower anterior teeth have gradually aligned without orthodontic mechanics. (Figure 5, 6).

¹ Associate Professor & Head Department of Oral and Maxillofacial Surgery, Lahore Medical and Dental College, Lahore

² Lahore Medical and Dental College & Ghurki Trust Teaching Hospital, Lahore



A



B



C

Fig 1: Pre operative photographs

- A: Frontal view showing slight swelling on the chin
- B: Upper dental arch, occlusal view
- C: Lower dental arch, showing labially proclined incisors, lingually inclined and rotated left canine and 1st premolar. Right premolars also show malalignment

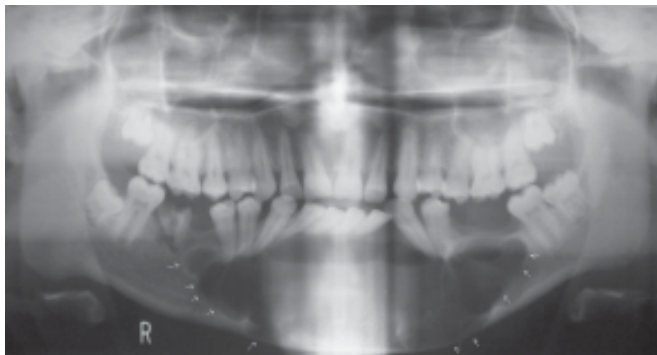


Fig 2: Pre-operative OPG.
Multilocular radiolucency extending from left 2nd molar to right 1st molar with a thin lower cortical border

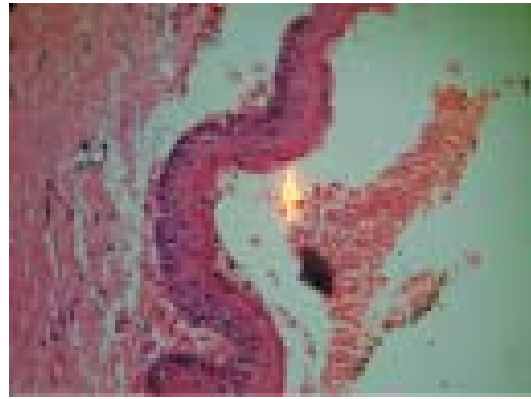


Fig 3: H & E Histologic slide (40x)
Parakeratinized epithelium.
Palisaded basal cell layer.
Fibrous capsule.

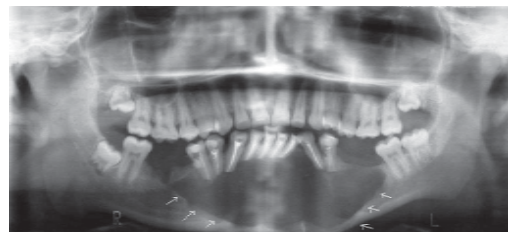


Fig 4A

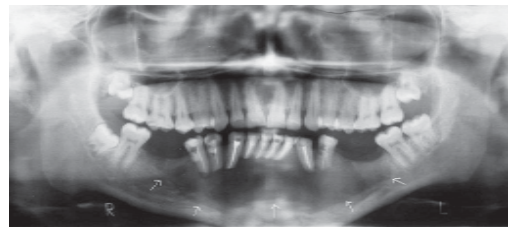


Fig 4B

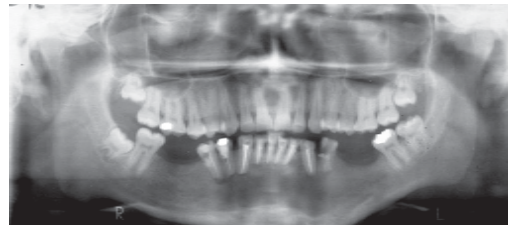


Fig 4C



Fig 4D

Fig 4: Post operative OPG
A: At 3 months follow up
B: At 6 months follow up
C: At 12 months follow up
D: At 28 months follow up



Fig 5: Mandibular occlusal view at 28 months follow up



Fig 6: Clinical situation, 24 months follow up

DISCUSSION

The odontogenic keratocyst is a “benign uni- or multicystic, intraosseous tumor of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behavior”.¹ Though it has a tendency to occur in any part of mandible and maxilla, majority almost 70%, arise in the posterior body of the mandible and 6.9% at the symphyseal region.² Peak incidence in both genders occur in the third decade.³ World Health Organization (WHO) has reclassified odontogenic keratocyst as keratocystic odontogenic tumor (KCOT).⁴ Cases have been reported in nevroid basal cell carcinoma syndrome (NBCCS)^{5,6} and Simpson- Golabi- Behmel Syndrome associated patients.⁷ It is suggested that KCOT associated with NBCCS is usually observed in younger patients with presentation at multiple sites. Heparanase expression is correlated with the invasive properties of NBCCS associated KCOT.³ Studies have also shown association of cytokeratin 17 expression with this syndrome.⁸

Although treatment modalities range from marsupialization followed by enucleation to resection,^{4,9,10} W.H.O’s reclassification of this lesion from cyst to tumor underscores its aggressive nature, therefore motivating clinicians to manage the disease in a correspondingly aggressive manner.^{1,8}

This case was managed with enucleation along with peripheral ostectomy. Although KCOT with a parakeratinized epithelial lining has a higher recur-

rence, resection at this young age would have been similar to an amputation leading to disability. Marsupialization however was against patient’s compliance. As the patient presented to us with no neural impairment, enucleation with chemical cauterization was not opted. Carnoy’s solution is reported to be responsible for some postoperative sensory disturbances.¹¹ In our patient, peripheral ostectomy could therefore be justified.

Lingual cortex was intact and no damage to the periosteum was observed. All lower involved teeth were preserved although that is a constant threat for recurrence. The authors believe that in a lesion of symphyseal area; anterior teeth can be easily apicectomised now and may be extracted in future, if required. Reconstruction with bone graft was not opted since dental implants were not in the rehabilitation plan. A close post operative review is as important as the primary treatment, especially for conditions with such different biological behavior.

According to Taipale and colleagues¹² cyclopamine, a plant-based steroidal alkaloid, blocks the activation of sonic hedgehog (SHH) pathway therefore makes it a potential “mechanism-based” therapeutic agent for those human tumors whose pathogenesis involves excess SHH pathway activity. Zhang¹³ postulated that antagonists of SHH signaling factors could also be effectively used to treat KCOT. The suggested strategies include reintroduction of a wild-type form of PTCH (a tumour suppressor gene), inhibiting SMO molecule

(an oncogene) by synthetic antagonists and suppressing the downstream transcription factors of the SHH pathway. They suggest that intracystic injection of an SMO protein-antagonist has the greatest potential as a future treatment option.¹³ Future treatment may involve molecular-based modalities, which may reduce or eliminate the need for aggressive surgical management.⁴

REFERENCES

- 1 Philipsen HP. Keratocystic odontogenic tumor. In: Barnes L, Eveson JW, Reichart PA, Sidransky D, World Health Organization classification of tumours. Pathology and genetics of the head and neck tumours. Lyon, France: IARC Press: 2005; p. 306–7
- 2 Oda D, Valiente R. Odontogenic Keratocyst The Northwestern USA Experience. The Journal of Contemporary Dental Practice. 2000;1(2): 1-10
- 3 Steolinga P.J.W. Long term follow up on keratocysts treated according to a defined protocol. Int J Oral Maxillofac Surg. 2001; 30: 14-25
- 4 Madras J, Lapointe H. Keratocystic Odontogenic Tumour: Reclassification of the Odontogenic Keratocyst from Cyst to Tumour. JCDA. 2008; 74(2): 165a-165h
- 5 Katase N, Nagatsuka H. Analysis of the neoplastic nature and biological potential of sporadic and nevoid basal cell carcinoma syndrome associated keratocystic odontogenic tumor. J Oral Pathol Med: 2007; 36: 550–4.
- 6 Cohen, Jr M. M. Nevoid basal cell carcinoma syndrome, molecular biology and new hypotheses. Int J Oral Maxillofac Surg 1999; 28: 216-223
- 7 Krimmel M, Reinert S. Multiple odontogenic keratocyst in mentally retardation over growth (Simpson- Golabi- Behmel Syndrome) Br J Oral Maxillofac Surg 2000; 38(3): 221-2
- 8 John G. Meara, Ben J. Cytokeratin expression in the odontogenic cyst. J Oral Maxillofacial Surgery. 2000; 58: 862-865
- 9 Pogrel M A, Jordan R C K. Marsupialization as a definitive treatment for the odontogenic keratocyst. J Oral Maxillofac Surg 2004; 62: 651–655.
- 10 Maurette PE, Jorge J, de Moraes M. Conservative treatment protocol of odontogenic keratocyst: a preliminary study. J Oral Maxillofac Surg. 2006; 64(3): 379-83
- 11 Loescher A. R., Robinson P P. The effect of surgical medications on peripheral nerve function. Br J Oral and Maxillofac Surg 1998; 36: 327-332
- 12 Taipale J, Chen JK, Cooper MK, Wang B, Mann RK, Milenkovic L, Scott MP, and others. Effects of oncogenic mutation in Smoothed and Patched can be reversed by cyclopamine. Nature 2000; 406(6799): 1005–9.
- 13 Zhang L, Sun ZJ, Zhao YF, Bian Z, Fan MW, Chen Z. Inhibition of SHH signaling pathway: molecular treatment strategy of odontogenic keratocyst. Med Hypotheses 2006; 67(5): 1242–4. Epub 2006 June 27.