NON SYNDROMIC MULTIPLE KERATOCYSTIC ODONTOGENIC TUMORS IN A PATIENT IN HIS LATE 50'S: A CASE REPORT

Sanaa Ahmed[1✉], Talha Bin Saeed[2], Zahid Ali[3]

ABSTRACT

Odontogenic keratocystic tumors (KCOT) are found over a wide range of age and are associated with syndromes when found in younger age and in multiple number. Only 5 % are non-syndromic cases. We are reporting a case of non-syndromic multiple KCOT in 58 year old patient.

Key words: Odontogenic Keratocystic Tumor, Odontogenic Tumors (MeSH), Neoplasms (MeSH).

THIS ARTICLE MAY BE CITED AS: Ahmed S, Saeed TB, Ali Z. Non syndromic multiple keratocystic odontogenic tumors in a patient in his late 50's: A case report. Khyber Med Univ J 2015;7(2): 81-83.

[1✉] MS Oral Surgery Trainee, Abbasi Shaheed Hospital, Karachi, Sindh, Pakistan
Address: C-147 Block C North Nazimabad, Karachi Sindh, Pakistan.
Cell: 0213-6686057, 0333-2924028
Email: drsanaaumair@gmail.com
[2] MS Oral Surgery Trainee, Abbasi Shaheed Hospital, Karachi, Sindh, Pakistan
[3] Associate Professor, Oral and Maxillofaciaol Surgery Department, Abbasi Shaheed Hospital, Karachi, Sindh, Pakistan

Date Submitted: December 25, 2014
Date Revised: June 02, 2015
Date Accepted: June 05, 2015

INTRODUCTION

Odontogenic keratocyst was first named by Philipson in 1956 due to the presence of keratin in the cyst lumen. Defined as "the cyst derived from the remnants of the dental lamina, with a biologic behavior similar to a benign neoplasm with distinctive lining of six to ten cells in thickness which exhibits a basal cell layer of palisaded cells and a surface of corrugated parakeratin".[1] Due to its aggressive behavior and high recurrence rate its being renamed to keratocystic odontogenic tumor.[2]

It constitutes around 3.1% of all cysts.[3] Exhibits male predilection. Frequently affects mandible ramus, third molar region followed by first, second molar then anterior mandible while in maxilla most commonly found in third molar and cuspid region. Multiple KCOT are associated with Gorlin Goltz, Oro-Facial Digital, Ehler Danlos, Noonan and Simpson Golabi Behmel Syndromes. Only 5.8% are non-syndromic cases with multiple odontogenic keratocystic tumor.[3,4]

There are few cases reported up to 40 years of age of non-syndromic Odontogenic tumors,[3-7] we are reporting here a unique case of two odonotgenic keratocystic tumors in a 58 yr old patient.

CASE REPORT

A 58 year old male presented to Oral and maxillofacial surgery clinics of Karachi medical and dental college complaining of swelling and pain on lower left side of the face since 3 months (Figure 1a, 1 b). Swelling gradually increased in size with no pus or blood discharge. Patient had no history of any co-morbid conditions or surgical procedures. On examination 1*1 cm swelling on lower left side of face was observed causing bucco-lingual expansion intra-orally in region of lower left central incisor to third molar region of same side. Oral hygiene was adequate with upper right second pre-molar missing. Generalized gingival recession was also noticed.

An Orthopantomogram (OPG) {Figure 2} was advised which showed 2 multilocular radiolucent areas with scalloped margins. Anterior radiolucency had margins from lower left central incisors up to the first molar while the posterior radiolucency was limited between the second lower left molar to the angle of mandible on the same side with the third molar root in the posterior cavity. Both cavities were separated by a thin bony wall. All base line investigations were done which were within normal limits.

Differential diagnosis consists of Odontogenic Keratocystic Tumor and Ameloblastoma. Patient was treated under general anesthesia. Enucleation of cyst was done through intra-oral incisions, treating both cysts separately

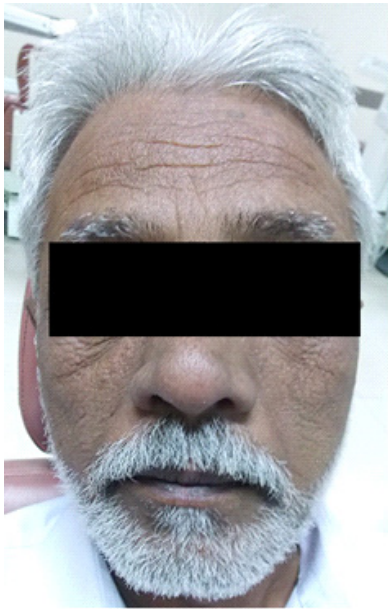

Figure1: Extra-Oral View showing asymmetry, swelling on left side of face.

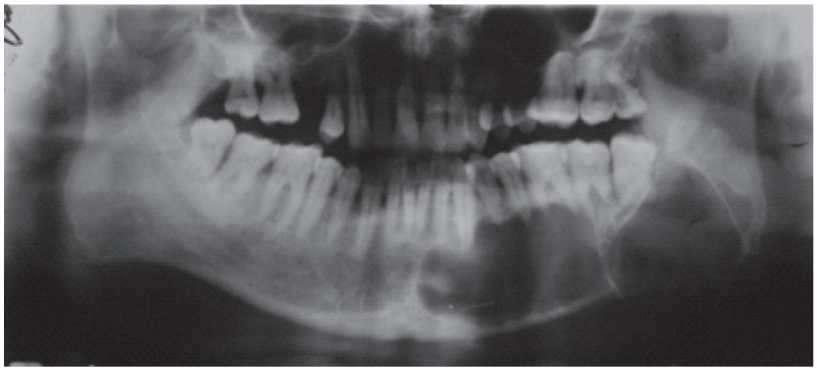

Figure 2: Pre-operative OPG showing two radiolucencies in lower left side of mandible, separated by thin bony wall

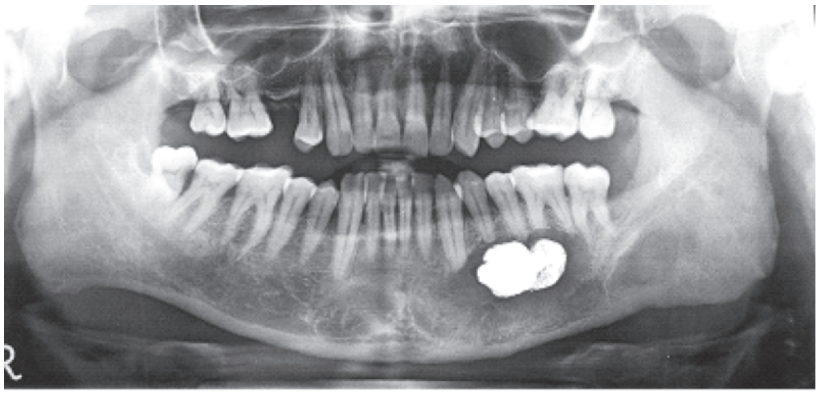

Figure 3: Post-operative OPG 8 months showing posterior cavity completely healed and radio-opaque BIPP in anterior cavity surrounding by healing bone

and placing a BIPP in bony defects. The posterior Cyst was small in comparison to the anterior one and had lower left partially impacted third molar associated with it. Third molar was also removed with the cyst lining. Patient was put on antibiotic 5 day regime post-operatively and after 2nd post-operative day was discharged having no complain of pain, blood or pus discharge. Recalled every 15 day to change the Bismuth iodine paraffin paste pack.

Patient has been coming for follow to get Bismuth iodine paraffin paste pack removed. Sample was sent for biopsy which confirmed the diagnosis of odontogenic keratocystic tumor showing characteristic parakeratin squamous epithelium with prominent basal columnar epithelium along with inflammatory infiltrates. The Bony defect in the angle of the mandible is healed fully while we are planning for soft tissue closure in the anterior Bony defect which has almost filled. Post-operative orthopantomogram (Figure 3) after 1 year showed healed posterior defect and good healing in anterior defect.

DISCUSSION

Odontogenic keratocystic tumor are known for their aggressive behavior and high recurrence rate. It can occur from 10-80 years of age showing 2:1 male to female ratio. 1 60-80% occur in mandible occurring in 2nd and 3rd decade of life.[5] It has been shown that 25-40% are associated with a un-erupted tooth.[3]

Mechanism of formation is still unknown but PTCH mutation is the link. Chronic inflammation causes remnants of dental lamina to undergo Cystic changes. Features include Pain, intra-oral bucco-lingual bone expansion, Soft tissue swelling, drainage, root resorption of adjacent teeth, paresthesia of the lip or teeth.[1] On radiograph it appears as radiolucent expansile lesion with sclerotis margins. If unilocular it can mimic Dentigerous cyst as a well defined radiolucency surrounding the unerupted tooth while if multiloculated it resembles ameloblastoma. In current case it appeared multiloculated on OPG. It has the tendency to change into malignancy.[8,9]

On histopathology it appears as ortho or parakeratinized squamous epithelium with prominent basal layer of columnar or cuboidal cells. Cystic cavity is filled with keratin. In our case keratin flakes with acute and chronic inflammatory infiltrates was reported on histopathology.[1]

Multiple KCOT are found in Gorlin Goltz syndrome commonly but on rare occasions sporadic cases are there. Young patients with multiple cyst are key candidates for ruling out any associated syndromes.

Treatment is still debatable as resection is the curative treatment while enucleation with currettage being least requirement.[3-9] Patients are usually kept on long term follow up.

Our case is the first to report multiple KCOT in non syndromic patient in late 50 years. Both cyst were found in lower left mandible separated by a thin bony wall which were treated separately. It was associated with partially erupted lower left third molar. Cyst in the anterior mandible is from the lower left central incisor to first molar while second cyst is found posterior to first molar. Patient is kept on follow up.

REFERENCES

1. Shafer, Hine, Levy. Shafer`s Text book of Oral Pathology. (ed 5). Churchill livingstone; 2005. p. 357-432.
2. Barnes L, Eveson JW, Reichart P, Sidransky D. Pathology and Genetics of Head and Neck Tumours", WHO Classification of Tumours, Volume 9. WHO. Geneva. 2005.
3. Parikh NR. Nonsyndromic Multiple Odontogenic Keratocysts: A Case Report". J Adv Dental Research 2010; 2(1): 71-4.

4. Bartake AR, Shreekanth NG, Prabhu S, Gopalkrishnan K. Non-Syndromic Recurrent Multiple Odontogenic Keratocysts: A Case Report. J Dent (Tehran) 2011 Spring; 8(2): 96-100. Epub 2011 Jun 30.

5. Guruprasad Y, Chauhan DS. Multiple odontogenic keratocysts in a nonsyndromic patient. J Cranio-Maxillary Dis 2012; 1: 36-40.

6. Nirwan A, Wanjari SP, Saikhedkar R, Karun V. Multiple non-syndromic odontogenic keratocysts in three siblings. BMJ Case Reports 2013; doi:10.1136/bcr-2012-007503.

7. Kurdekar RS, Prakash J, Rana AS, Kalra P. Non-syndromic odontogenic keratocysts: A rare case report. Natl J Maxillofac Surg 2013; 4(1): 90-3.

8. Tamgadge S, Tamgadge A, Modak N. Bhalerao S. Primary intraosseous squamous cell carcinoma arising from an odontogenic keratocyst: A case report and literature review. Ecancermedicalscience. 2013 May 9; 7: 316.

9. Siar CH, Ng KH. Squamous cell carcinoma in an orthokeratinised odontogenic keratocyst. Int J Oral Maxillofac Surg 1987; 16(1): 95-8.

AUTHOR'S CONTRIBUTION

All authors have made substantial contributions to the manuscript.

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

CONFLICT OF INTEREST

Authors declare no conflict of interest

GRANT SUPPORT AND FINANCIAL DISCLOSURE

NIL

KMUJ web address: www.kmuj.kmu.edu.pk

Email address: kmuj@kmu.edu.pk