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Case Report

Management of Odontogenic Keratocyst by Enucleation-Follow Up for 5 Year

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Abstract

OKC is considered to be enigma for oral surgeon in terms of management and debatable nomenclature.in 2017 WHO again renamed KCOT to OKC. Here we are presenting case of 36-year-old male patient initially diagnosed as dentigerous cyst clinically and radiographically. Management done was only enucleation but final diagnosis made was OKC based on histopathology. Follow up done for 5 year with no evidence of recurrence. In discussion various surgical modalities was described with their chances of recurrence & special emphasis was given on future trends and ray of hope on medicinal management of OKC, but medicinal management require drug trail.

Keywords: OKC; KCOT; PTCH

Abbreviations

OKC: Odontogenic Keratocyst; KCOT: Keratocystic Odontogenic Tumor; WHO: World Health Organization.

Introduction

OKC is developmental odontogenic cyst arises from dental lamina [2,3]. It is known for its recurrence & controversial nomenclature [10]. WHO defined it as ''a benign uni- or multi-cystic, intraosseous tumors of odontogenic origin, with a characteristic lining of Parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behavior [5]. The term 'keratocyst' was introduced in 1956 by Philipsen based on its histopathologic appearance of cystic lining [5]. Posterior mandible is most commonly involved followed by posterior maxilla, anterior maxilla, anterior mandible [8,9]. 82% of OKC occur in tooth bearing portion of jaw and 27% of OKC associated with impacted tooth [8].

Multiple OKC may be associated with Gorlin–Goltz syndrome Nevoid basal cell carcinoma syndrome (NBCCS). The prevalence of NBCCS about 1 in 60000 live births and associated with mutation of PTCH Gene present on long arm of chromosome 9 [1]. 5% cases of multiple OKC are non-syndromic [2]. Radiographically OKC grows antero-posteriorly within medullary cavity and showing less bony expansion as compared to another developmental cyst [5].

Broad spectrum of treatment modalities is available for management of OKC based on histopathologic variant. Starting from conservative like marsupialization or enucleation and chemical cauterization with carnoys solution to resection. feasibility of management is described in detail in discussion section of this article.

Case Presentation

Presenting case report of 36year old male patient presented to clinic with chief complain of pain and swelling with respect to

mandibular anterior region. Clinically patient showing expansion of buccal cortical plate from 36 to 45. All teeth are non-vital, missing right mandibular canine.no evidence of paresthesia. No contributary relevant past medical, dental, family history.

Investigation

- **Aspiration**: Aspiration done with 18g needle showing clear yellowish fluid
- **OPG/CBCT**: AAs shown in figure 1 and figure 2 unilocular radiolucency extending from 36 to 45 with impacted 43. displacement of 33. No evidence of perforation of buccal and lingual cortical plates but there is expansion of buccal cortical plate present in region of pathology.
- Blood investigation was within normal limit.

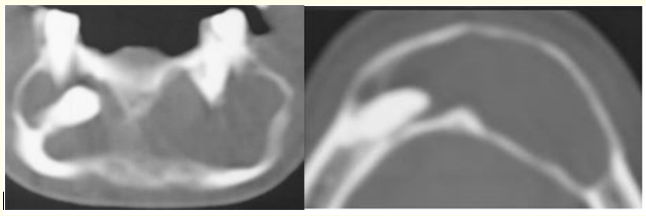

Figure 1: Pre operative CBCT showing expansion of buccal cortical plate and association of OKC along with impacted mandibular right canine.

Figure 2: Pre operative and post-operative OPG after follow up of 5year.

Histopathology

Final histopathology gives following impression

- Corrugated parakeratinised stratified squamous epithelium (Figure 3a)
- The epithelium- connective tissue interface is flat, desquamated keratin in the lumen (Figure 3b)
- Tombstone/picket fence appearance- 8 to 10-layer thick epithelium with palisading of basal cell layer (Figure 3b)

Figure 3: 3a-Corrugated parakeratinised stratified squamous epithelium, 3b-Tombstone/picket fence appearance.

Differential diagnosis

- Points favouring the provisional diagnosis as dentigerous cyst includes association with impacted teeth(canine), straw coloured aspiration fluid, expansion of buccal cortical plate with displacement of teeth.
- OKC-point favouring the differential diagnosis as OKC are antero-posterior extension of lesion is generally associated with minimal expansion of cortical plate but in this case this statement is seems to be contradictory as this case associated with expansion of buccal cortical plate as mentioned in investigation section of this article.

- Radicular cyst was ruled out as none of teeth was carious although all teeth are non-vital.
- Unicystic ameloblastoma -point favouring diagnosis of UA is association of lesion with impacted teeth as UA associated with impacted 3rd molar in 50-80% [8].

Management

In All non-vital teeth root canal treatment was done. Patient was taken into OT and patient was scrubbed and draped as per standard surgical protocol fullthikness mucoperiosteal flap was raised.

Impacted canine along with cystic lining was enucleated irrigation with betadine Normal saline was done haemostasis was achieved and surgical site was closed with 3-0 vicryl using sling suture Figure 4. Follow up of case was done at every 3month for 1st year. Every year for next five year.

Figure 4: Intra-operative view.

Discussion

OKC is considered to be dilemma for both surgeon and pathologist as in 2005 WHO classifications of head and neck tumour's renamed OKC to keratocystic odontogenic tumour KCOT [2]. OKC is locally aggressive histologically showing mitotic figures in basal layer of mucosa and genetically associated with PTCH gene [3,4,6]. In 2017 WHO classifications of head and neck tumours again renamed KCOT to OKC. So in this case report we will consider as OKC only.

Odontogenic keratocystic contribute 12-14% of odontogenic cyst. Most common occurs in mandible as compared to maxilla (5:1) of which 25-40% associated with impacted or semi impacted teeth most common impacted teeth associated with OKC is 3rd molar [5,8,9]. In this case its associated with impacted canine.

Patient must be ruled out for gorlin-goltz syndrome, condition is autosomal dominant its association with multiple OKC in 75% of cases [1]. In our case OKC was non-syndromic based of criteria by Evan., *et al* [1,6] and Kimonis., *et al* [1]. Other syndromes associated with multiple odontogenic keratocyst are noonan syndrome, Ehler-Danlos syndrome, orofacial digital syndrome, Simpson-Golabi-Behmel syndrome. Syndromic odontogenic keratocyst is more hostile than non-syndromic OKC and higher chance of recurrence [2].

Genetics

Mutation of human homologue of the Drosophila polarity Patched gene (PTCH) and P^{53} is associated with syndromic and sporadic OKC [6]. Presence of cell surface carbohydrate like epithelial membrane antigen and carcinoembryonic antigen in Para keratin layer, cell surface glycoprotein gp38 associated with more aggressive OKC, overexpression of Ki-67, monoclonal antibody of IPO, podoplanin are also associated with aggressive OKC [2,3]. Mutation of BRAF V600E is more commonly associated with Para keratinized odontogenic keratocyst (63.2%) then Ortho keratinized (9.1%) type [10].

Treatment options

Various treatment options are available for the management of OKC. As describe in table 1 and their recurrence rate [5,9,11]. Chemical cauterization with carnoys solution is used since 1933 and showing lowest rate of recurrence and low morbidity as compared to recurrence. Modified carnoys solution is used now a days because of carcinogenicity of chloroform in older preparation. according to animal study depth of penetration of carnoys solution in cancellous bone is 1.-0.54 mm. Carnoys solution must not be applied to the vital structure more than 3min [5,4]. As literature accepts the enucleation of OKC as least accepted modality for treatment due to higher recurrence but in present case the treatment modality used was enucleation only and follow up was done for 5 yr. With no recurrence in follow up period.

Treatment modality	Recurrence rate
marsupialization	32.3%
Enucleation	23.1%
enucleation with peripheral ostectomy	17.4%
decompression followed by residual cystectomy	14.6%
enucleation plus liquid nitrogen cryotherapy	14.6%
enucleation and Carnoy's solution	11.5%
Resection	8.4%

Table 1: Treatment modality used and recurrence of OKC.

Choice of treatment modalities is depending on type of histopathology obtained in biopsy. Two variants are associated with OKC. Parakeratinized, Orthokeratinized.Parakeartinized variants is more aggressive & syndromic cases are generally associated with Parakeratinized OKC [3,4]. Orthokeratinized odontogenic cyst (OOC) require less aggressive treatment & associated with low chance of recurrence ranging from 0-2%.

Future trend in management

Recently some researchers are working on the non-surgical management of odontogenic keratocyst.

- In 2000, Taipale., *et al.* shown that a plant derived teratogen cyclopamine shows the potent antitumor activity by blocking SHH pathway [7].
- In 2006, Zang., *et al.* was first to pointed out that intracystic injection of cyclopamine can be used as nonsurgical therapy for management of odontogenic keratocyst [7].
- vemurafenib, dabrafenib, and trametinib are BRAF inhibitors can be used in management of OKC but above all drugs requires clinical trial [10].

Conclusion

Various pathology are associated with impacted teeth. When radiologic evidence suggest association of pathology with impacted teeth case must be ruled out for various pathology like dentigerous cyst, Unicystic ameloblastome, odontogenic karatocyst, calcifiying odontogenic cyst, ameloblastic fibroma, adenomatoid odontogenic tumour in anterior maxilla [8] and appropriate management must be given as per histopathology report as in our case provision diagnosis made based on clinical and radiographic finding was dentigerous cyst but final diagnosis following enucleation was OKC. Only enucleation must be considered as management of OKC as compared to other more invasive options including use of carnoys solution as carnoys solution also damages vital structures like inferior alveolar nerve when used more than 3min. Furthers enucleation preserves normal anatomic structure of bone.

Bibliography

1. Satheesh Chandran., *et al*. "Odontogenic Keratocysts in Gorlin-Goltz Syndrome: A Case Report". *Journal of International Oral Health* 7 (2015): 76-79.
2. N Kargahi and M Kalantari. "Non-Syndromic Multiple Odontogenic Keratocyst: A Case Report". *Journal of Dentistry* (Shiraz). 14.3 (2013): 151-154.
3. María del Carmen González Galván., *et al*. "Orthokeratinized Odontogenic Cyst: A Report of Three Clinical Cases". *Case Repots in Dentistry* (2013): 672383.
4. BP RajeshKumar., *et al*. "Carnoy's in Aggressive Lesions: Our Experience". *Journal of Oral and Maxillofacial Surgery* 12.1 (2013): 42-47.
5. Walid Ahmed Abdullah. "Surgical treatment of keratocystic odontogenic tumour: A review article". *The Saudi Dental Journal* 23 (2011): 61-65.
6. Elizabeth A Jones., *et al*. "Basal Cell Carcinomas in Gorlin Syndrome: A Review of 202 Patients". *Journal of Skin Cancer* (2011): 217378.
7. Zhang L., *et al*. "Inhibition of SHH signaling pathway: molecular treatment strategy of odontogenic keratocyst". *Med Hypotheses* 67.5 (2006): 1242-1244.
8. Hamed Mortazavi and Maryam Baharvand. "Jaw lesions associated with impacted tooth: A radiographic diagnostic guide". *Imaging Science Dentistry* 46.3 (2016): 147-157.
9. Sánchez-Burgos R., *et al*. "Clinical, radiological and therapeutic features of keratocystic odontogenic tumours: a study over a decade". *Journal of Clinical and Experimental Dentistry* 6.3 (2014): e259-264.

10. Cha YH., *et al*. "Frequent oncogenic BRAF V600E mutation in odontogenic keratocyst". *Oral Oncology* 74 (2017): 62-67.

11. Al-Moraissi EA., *et al*. "What surgical treatment has the lowest recurrence rate following the management of keratocystic odontogenic tumor?: A large systematic review and meta-analysis". *Journal of Cranio-Maxillofacial Surgery* 45.1 (2017): 131-144.

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