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CASE STUDY

ODONTOGENIC KERATOCYST OF MAXILLA RELATIVELY UNCOMMON SITE OF INVOLVEMENT: A CASE REPORT

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ABSTRACT

The Odontogenic keratocyst (OKC) is a benign lesion of the Oro-facial region having distinct clinicopathological features, high propensity for recurrence and characteristic aggressive behaviour. It commonly involves the mandible while maxilla is relatively unusual location. The clinico-histo-pathological characteristics and subsequent of OKC in a 30 years old male patient with maxillary involvement has been discussed herewith.

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INTRODUCTION

The proliferation of odontogenic epithelial remnants within the jaws to form either cysts or tumours---is a relatively common event. The jaw cysts usually arise from odontogenic remnants---most commonly cell rests of Malassez or cell rests of Serre or remnants of dental lamina (Nagraja *et al*., 2012). Odontogenic keratocyst (OKC) is an epithelial developmental cyst, which was first described by Philipsen in 1953. It is treated as a distinctive clinical entity due to high propensity for recurrence and aggressive behaviour. This has necessitated the reclassification of the lesion by WHO (2005) as Keratocystic Odontogenic Tumour (KCOT) (Barnes *et al*., 2005). It comprises 3-10.5% of all jaw cysts occurring mainly in the 2nd and 3rd decades of life. Majority of keratocysts occur in the posterior aspect of body and ascending ramus of the mandible but may also occur anywhere in the jaws. In the initial stages OKC does not produce any signs or symptoms. OKCs tend to grow in an anteroposterior direction within the medullary cavity of the bone without causing obvious bone expansion. Large lesions lead to facial asymmetry, thinning of the cortical plate, mobility of regional tooth and discharge of pus evidence of secondary infection (Chirapathomsakul *et al*., 2006). So the clinical features and radiographic appearance of OKCs are not very characteristic. Radiographic appearance of OKCs may range from small unilocular to a large multilocular radiolucencies. Hence it may resemble to ameloblastoma, dentigerous cyst, lateral periodontal cyst or even a radicular cyst (Mozaffari *et al*., 2007; Garlock *et al*., 1998). Histopathologically, OKC presents with a thin and friable epithelial lining enclosing a cystic lumen that may contain a clear fluid similar to transudate of serum or may be filled with cheesy substance of keratinaceous debris. The epithelial lining comprises of stratified squamous epithelium which is 6-8 cell layers in thickness with a wavy or corrugated luminal surface. Most significantly the basal layer composed of a palisaded arrangement of cuboidal to tall columnar cells having hyperchromatic nuclei giving a characteristic tomb stone appearance. The interface between epithelium and connective tissue is often flat and rete-ridges are inconspicuous. Presence of inflammatory cells infiltration and satellite cysts are also occasionally noted in the connective tissue stroma (www.quintpub.com; Sivapathasundharam, 2016; Neville BW *et al*., 2015). OKCs differ from other odontogenic cysts in that they have a biologically active lining epithelium, a tendency to extend along cancellous bone in an antero-posterior direction without causing obvious bony expansion and a considerable high rate of recurrence (Mervyn Shear *et al*., 2007). Successful treatment of OKC depends upon the precise diagnosis, adequate surgical procedure (marsupialisation / Enucleation) and close periodic follow-ups. Here, we present a case of OKC involving left maxilla of a 30 years old male patient with through clinical, radiological and histopathological details.

CASE REPORT

A 30 years old male patient from semi-urban area reported to the Department of Oral & Maxillofacial Pathology, GNIDSR,

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Kolkata, with a chief complain of swelling involving the left side of the face for the last four to five months. Initially the swelling was small which gradually enlarged and attained the present dimensions, causing difficulty in phonation and mastication. Extraoral examination revealed a diffuse firm to hard non-tender swelling involving left middle 3rd of face with obliteration of nasolabial fold leading to facial asymmetry without any history of spontaneous discharge or other secondary changes. Intra-oral examination revealed a well-defined round to ovoid, dome shaped, non-tender, non-pulsatile, non-compressible, non-reducible, swelling measuring about 4cm x 5cm extending from 23 to 27 region associated with expansion of buccal cortical bone with egg-shell crackling at places. History and examination did not reveal any association with Basal Cell Nevus Syndrome.

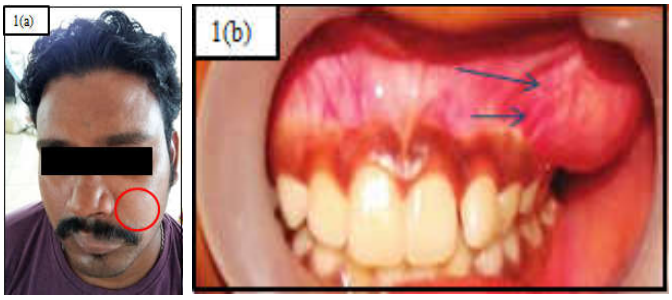

Fig.1.(a). Extra oral photograph of the patient showing diffuse swelling over the left cheek.
Fig.1.(b). Intraoral photograph showing a well-defined round to ovoid dome shaped swelling extending from 23 to 27 regions.

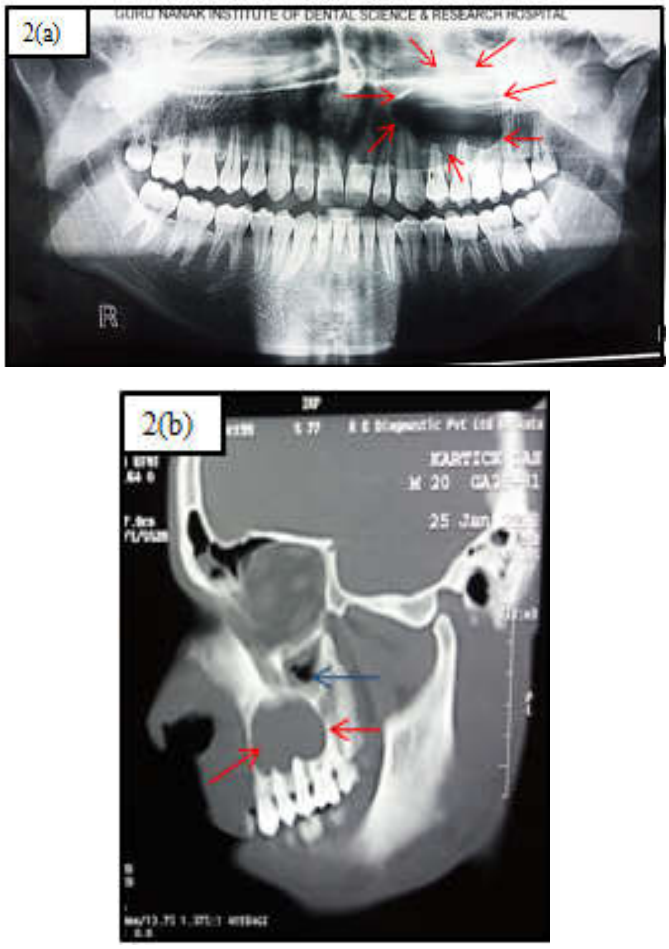

Fig.2.(a. b). The conventional OPG and CT revealed a well defined unilocular radilucent lesion in relation to 22 to mesial root of 27, encroaching the maxillary air sinus (blue arrow) without any displacement of regional teeth.

Fig. 3. Photograph showing aspirated straw colored fluid

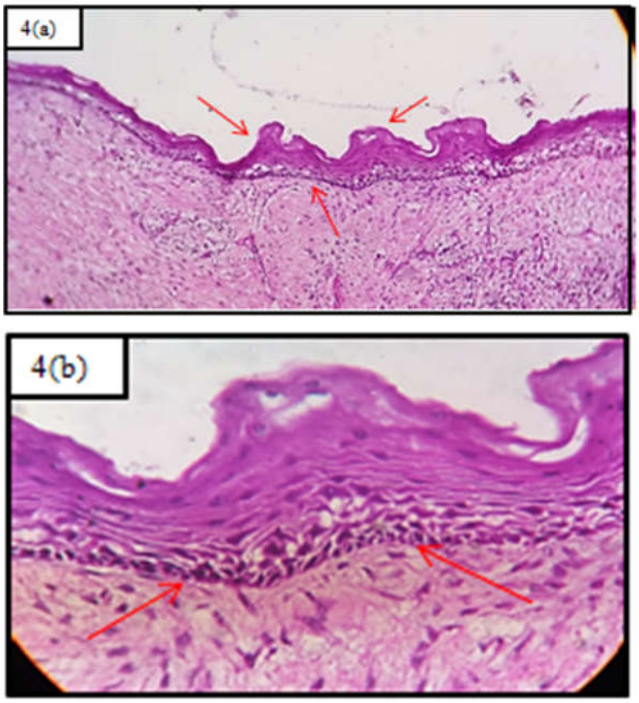

Fig.4.(a). Scanner view photomicrograph (4x) shows the cystic lumen, lined by epithelium being backed by fibro-vascular connective tissue wall with luminal surface corrugation at places and lack of rete-ridges. Fig 4. (b). High power photomicograph (40x) depicting the epithelium of six to eight cell layers thickness along with low columnar to cuboidal basal layer containing hyperchromatic palisaded nuclei giving a characteristic tomb stone pattern.

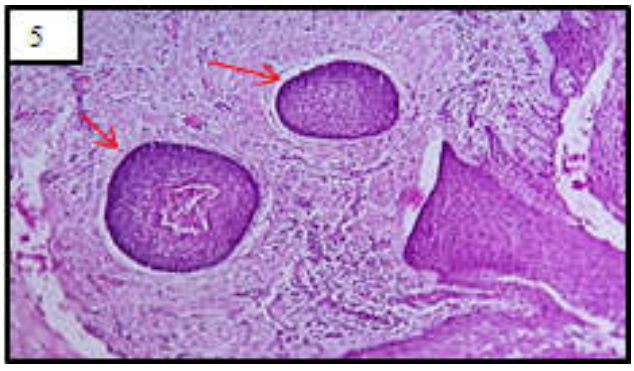

Fig. 5. Low power photomicrograph (10x) showing satellite cyst in the loosely arranged connective tissue stroma.

Based upon the clinical, radiological and aspiration findings a provisional diagnosis of Odontogenic cyst was made and the patient was referred to the Dept. of Oral & Maxillofacial Surgery for treatment and management. Enucleation of the lesion with placement of PRF (Platelet Rich Factor) was performed under general anaesthesia and gross specimen was sent to our Dept. for histoopathological evaluation. Gross examination revealed a greyish white soft tissue specimen measuring about 6.5cm x 4cm x 0.4 cm. A portion of the cystic lining was obtained for microscopic evaluation. Histopathological evaluation revealed the presence of a cystic lumen lined by parakeratinized stratified squamous epithelium

being backed by fibro-vascular connective tissue stroma devoid of significant inflammatory cell infiltrate. The epithelium revealed six to eight cell layers in thickness along with surface corrugation at places associated with lack of rete-ridges leading to a relatively flat epithelium-connective tissue interface. Artifactual separation of the lining epithelium from fibrous wall and epithelial foldings were noted at places. The basal layer revealed palisaded arrangement of low columnar to cuboidal cells having the hyperchromatic nuclei giving a "tomb stone" appearance. Strikingly, satellite cysts were also noted in the connective tissue stroma. The conventional OPG and CT revealed a relatively well defined unilocular radiolucent lesion in relation to 22 extending to mesial root of 27, encroaching the maxillary sinus without displacement of the regional teeth. Aspiration yielded a straw coloured fluid.

DISCUSSION

Odontogenic keratocyst (OKC) is a distinctive form of developmental odontogenic cyst that deserves some special consideration because of its unique histopathological features and clinical behaviour (Barnes *et al*., 2005). OKC have been reported to account for 3 to 10.5% of all the jaw cysts. They mainly appear during 2nd and 3rd decades of life showing slight male predilection than female. Generally OKC appear in the form of solitary lesion unless they are accompanied by Nevoid Basal Cell Carcinoma Syndrome (NBCCS). The case under discussion was a 30 years old male patient with the large diffuse firm to hard dome shaped swelling involving the left maxillary premolar region showing buccal cortical plate expansion and egg shell crackling at places. These clinical ssfindings were supported by the authors of previous studies (Garlock *et al*., 1998; Barnes *et al*., 2005; Chirapathomsakul *et al*., 2006; Mozaffari *et al*., 2007; Nagraja, 2012; www.qu intpub.com; Suma1 *et al*., 2015). The general physical examination was carried out to exclude the NBCC syndrome. Medical history of the patient was also non-contributory (Veena *et al*., 2011; Suma NK *et al*., 2015). The conventional OPG and CT revealed the well defined unilocular radiolucent lesion extending from 22 to 26 encroaching the maxillary sinus without displacement of roots of the regional tooth. These radiological findings of the present case are very much corroborative to the previous case series (Barnes *et al*., 2005; Chirapathomsakul *et al*., 2006; Mozaffari *et al*., 2007; Mervyn Shear *et al*. 2007; Veena *et al*., 2011; www.quintpub.com; Sivapathasundharam 2016). Based on clinical and radiological findings the case was provisionally diagnosed as odontogenic cyst. Enucleation was performed and the specimen was submitted for histopathological evaluation. The light microscopic features revealed the presence of a cystic lumen lined by parakeratinized stratified squamous epithelium being supported by fibrous connective tissue wall devoid of significant inflammatory infiltrate. The epithelium revealed six to eight cell layers in thickness characterized by surface corrugation at places and relatively flat epithelium-connective tissue interface. Artifactual separation from fibrous wall and epithelial foldings were noted at places. The basal layer revealed palisaded arrangement of low columnar to cuboidal cells having the hyperchromatic nuclei giving a "tomb stone" pattern. Satellite cysts were also evident in the connective tissue stroma. All these histopathological findings of our case were in accordance with the previous case studies (Veena *et al*., 2011; Nagraja *et al*., 2012; Pinky *et al*., 2015). The treatment of the OKC is generally classified into two groups-conservative and aggressive. Conservative treatment includes simple enucleation, with or without curettage, whereas aggressive treatment ranging from peripheral ostectomy, chemical curettage with Carnoy's solution, and resection (Banik S *et al*., 2011). The goals of treatment should involve eliminating the potential propensity for recurrence.

Conclusion

OKCs should be considered as one of the differential diagnoses for the periapical radiolucencies where vitality of the regional tooth is maintained. The thorough clinical, radiographic, and histopathological correlations are essential for proper management. This will avoid the further complications, since OKCs are highly aggressive, have high recurrence rate, and are associated with NBCCS.

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REFERENCES

Barnes, L., Eveson, JW., Reichart, P. and Sidransky, D. 2005. (Eds). Pathology and genetics of head and neck tumours. Lyon: IARC Press; WHO classification of tumours series.

Banik S, Banik S, Shaikh MH, Sadat SA, Mallick PC. Keratocystic odontogenic tumor and its radiological diagnosis by 3 dimensional Cone Beam Computed Tomography (CBCT). Update Dental College Journal. 2011;1(1):10-3.

Chirapathomsakul, D., Sastravaha, P. and Jansisyanont, P. 2006. A review of odontogenic keratocysts and the behavior of recurrences. *OralSurg Oral Med Oral Pathol Oral Radiol Endod.*, 101:5-9.

Cysts of the Oral and Maxillofacial Regions, Fourth edition, Mervyn Shear & Paul Speight, Black well Munksgaard, 2007.s

Garlock, J. A., Pringle, G. A. and Hicks, M. L. "The odontogenic keratocyst: a potential endodontic misdiagnosis," *Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontics,* vol. 85, no. 4, pp. 452–456, 1998.

Mozaffari E, Marmor DS, Alawi F. Odontogenic keratocyst with a misleading clinical and radiologic appearance. Quintessence International. 2007 Nov 1;38(10).

Nagraja A, Anigol PS, Kamath VV, Setlur KP. Keratocystic odontogenic tumor of the maxilla: Report of a rare case and review of literature. World Journal of Dentistry. 2012 Jan;3:100-8.

Neville BW, Damm DD, Allen CM, Chi AC. Oral and maxillofacial pathology. Elsevier Health Sciences; 2015 May 13.

Oral And Maxillofacial Pathology A Rationale for Diagnosis and Treatment Second Edition, Robert E. Marx DDS & Diane Stern DDS, MS, Quintessence Publishing Co, Inc 4350 Chandler Drive Hanover Park, IL 60133 www.quin tpub.com, Editor: Leah Huffman.

Suma NK, Pinky C, Babu NV, Jha S. Odontogenic keratocyst of maxillary premolar region: A case report. IJSS Case Reports & Reviews. 2015 Feb;1(9):38-41.

Textbook of Oral pathology; 8th Edition; Shafer, Hine & Levy; Edited by Sivapathasundharam, 2016, B. Elsevier.

Veena, K.M., Rekha Rao *et al*., 2011. Odontogenic Keratocyst Looks Can Be Deceptive, Causing Endodontic Misdiagnosis; Volume, Article ID 159501, 3 pages doi:10.1155/2011/159501.

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