



RESEARCH ARTICLE

ODONTOGENIC KERATOCYST: A DEMON IN DISGUISE

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ABSTRACT

The maxillofacial region is one of most commonly affected region by the cysts, compared to any other part of the body. Odontogenic keratocysts (OKC) are aggressive odontogenic cysts due to their high recurrence rate and locally aggressive nature. These are commonly seen in the 2nd to 3rd decade of life with mandibular posterior region being frequently involved. Here we report a case of 14yr old boy with follicular OKC that was clinically misdiagnosed as a dentigerous cyst. This case report emphasises on the role of histopathological evaluation and differential considerations of various cystic lesions.

INTRODUCTION

An Odontogenic Keratocyst (OKC) is a histologically distinct lesion that is thought to arise from the dental lamina (Neville, 2009). It was first described by Hans Phillipson in 1956 in a Danish publication (Phillipsen, 1956). Later Pindborg described its essential histological features in 1963 (Guru Prasad, 2010) Previously termed as primordial cyst, it may occur in place of a tooth where cystic degeneration of enamel organ epithelium may occur before the development of dental hard tissue (Heamvathy, 2011). It accounts for 3-11% of all the odontogenic cysts and is one of the most aggressive ones owing to its high recurrence rate and its tendency to invade the adjacent tissues. Because of its aggressive growth pattern and neoplastic behaviour WHO in 2005 redefined OKC and named it as Keratocystic odontogenic Tumor (KCOT) (Ali, 2003). It is defined as "a benign uni or multicystic, intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behaviour (Madras, 2008)." 60% of OKC's are seen in the 2nd to 3rd decade of life with a slight male predilection (Neville, 2009 and Mohammad, 2003). 60-80%cases are seen in the mandible with a high predilection for posterior body and ramal regions. In the maxilla these are commonly seen in the molar and the canine regions (Neville, 2009 and Mohammad, 2003). These can present as solitary or multiple lesions of the jaw.

Usually multiple lesions are associated with nevoid basal cell carcinoma syndrome in children (Meng-Ling Chiang, 2004). Usually asymptomatic these hidden demons are routinely seen on a routine radiographic examination. It can be painful in case these get secondarily infected. Associated with soft tissue and bony swellings, large cysts may often cause pathological bone fractures, loss of permanent tooth bud, airway obstructions and bony deformities (Neville, 2009). OKCs are well-defined radiolucencies, which are either unilocular or multilocular. The OKC typically enclosing the crown & attached to the neck of the tooth was termed as Follicular OKC by Altini and Cohen (Mohammad Ali, 2003). Odontogenic Keratocysts consists of fluid varying from a clear yellow liquid to a semi-solid cheese-like mass. Difficulties in the preoperative diagnosis of KCOT have enthused attempts to find a biochemical or immunological marker in aspirates of cyst fluid (Madhvika Patidar, 2015). Hereby we present a case which initially mimicked a Dentigerous cyst and was later diagnosed to be an OKC.

Case Report

A 14 yr old boy reported to our department with the chief complaint of a painless swelling in the left maxillary canine region that has increased in size since the past 10 days (FIG 1). Patient was clinically healthy with no significant past medical or dental history. Intra oral examination revealed a bony swelling obliterating the left mucobuccal fold from distal of 21 to mesial of 26. A palatal swelling was also seen in the same region. Overlying mucosa was normal.

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On palpation the swelling was found to be well defined and firm with egg shell crackling. 63 had Grade II mobility with no tenderness on percussion.



Figure 1. Pre – operative (clinical)



Figure 2. Occlusal Radiograph

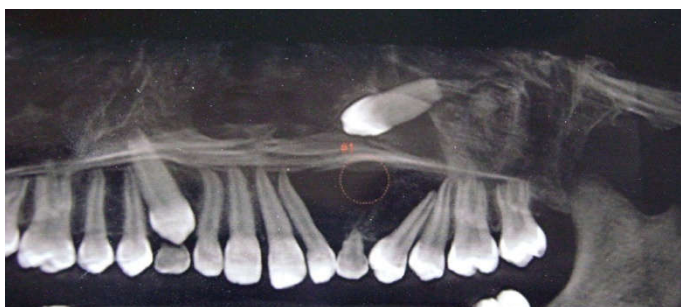


Figure 3. Opg Image

The patient was advised an occlusal radiograph and an OPG. Occlusal radiograph (FIG 2) revealed a thin sclerotic border surrounding well defined radiolucent area in relation to 22, 63, 24, 25, 26.

- Displacement of 21, 22, 24 and 25 was also seen
- Root resorption was also seen in relation to 22, 24 and 25, 23 was seen close to the maxillary sinus in the OPG and thus to get a clearer view a CBCT was advised.
- On CBCT (FIG 3) Lesion was noted to approximate the sinus floor with the dimensions of 31.48× 29.09× 29.72mm.
- Considering all the points a clinic-radiographic provisional diagnosis of Dentigerous cyst was made.
- An Enucleation was planned under GA after all the routine investigations.



Figure 4. Creamy exudate from the site



Figure 5. Enucleation of the lesion along with the permanent canine

As soon as 63 was extracted a thick, creamy white fluid oozed (FIG 4) out after which the buccal cortical plate was removed and the cyst was enucleated enmasse along with 23 attached to it (FIG 5). It was sent for histopathological examination later in 10% formalin (Fig 6). On Histological examination specimen revealed a cystic lining of uniform thickness with para and orthokeratinization at various places with basal layer

giving the characteristic “Picket Fence Appearance”(FIG 7). Connective tissue showed fibroblasts and collagen with vessels.



Figure 6. Macroscopic appearance of the lesion

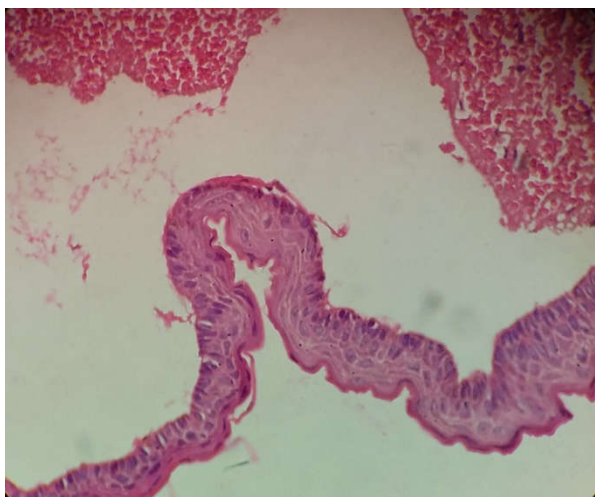


Figure 7. Picket fence appearance



Figure 8. Nance palatal arch

Based on these characteristic histological findings a diagnosis of odontogenic keratocyst was made. After this the patient was placed on a strict recall. 7 days postoperatively the patient

showed good signs of healing with reduction in swelling and discomfort. Instructions were also given regarding the long term effects of the lesion. After 3 weeks the patient was again recalled and the retained deciduous teeth i.e 53 and 72 were extracted prophylactically. After 5 weeks a significant space loss was seen in relation to the left canine region and a Nance palatal appliance (FIG 8) was planned to maintain the space till further prosthetic rehabilitation could be carried out.

DISCUSSION

OKC is one of the most aggressive jaw lesions. Toller in 1967 suggested that OKC may be regarded as benign neoplasm rather than a cyst based on its clinical behaviour. In 1984 Ahlfors suggested that “if OKC were to be recognized as true benign cystic neoplasm the question of modified treatment schedule would be raised” Odontogenic keratocysts are routinely discovered on radiographic examination and may simulate various non aggressive lesions like Dentigerous cyst. In a study by Brannon et al he reported that 8.5% of 1850 dentigerous cyst to be OKC’s (Shear, 1992). Mohammad et al in a series of 398 OKC’s reported only one follicular OKC in the canine region which makes this case a rare entity (Mohammad Ali, 2003). Typical histologic features of OKCs have been well characterized by Philipsen and Browne as: a regular layer of columnar basal cells with nuclear palisading giving a picket fence or tomb stone appearance (Li, 2011). According to Patidar et al there is difference in the content of total proteins, phosphates and keratinocytes in between OKC and other cysts that can be used as a diagnostic marker pre surgically (Li, 2011). According to Morgan there was a higher recurrence rate in case of enucleation when compared to ostectomy and no difference was seen when Carnoy’s solution was used (Teresa, 2005) while Voorsmith et al reported a decreased recurrence with Carnoy’s solution. Forsell et al reported a good healing with decompression in cases of a large cyst. Guler et al reported no statistically significant difference following enucleation, marsupialization or use of carnoy’s in the rate of recurrence (Nurhan, 2012). Other treatment modalities include Cryotherapy with liquid Nitrogen and en bloc resections (Schimdt, 2003 and Stoelinga, 2005). The treatment modalities thus are varying with no explicit age defined treatment protocol, that can ensure long term positive results of this aggressive and recurrent cyst.

Conclusion

This case report stresses on the need of FNAC evaluation of cystic lesions before any surgical management and also on the utilization of histopathological examination for the final diagnosis of the lesion. It also establishes the need for consideration of follicular odontogenic keratocyst as a differential in cases of a dentigerous cyst like lesion in young adults due to the vast difference in their clinical behaviours, treatment and prognosis

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