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BILATERAL ODONTOGENIC KERATOCYSTS – A CASE REPORT AND REVIEW

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ABSTRACT Odontogenic keratocysts are benign odontogenic cysts. They account for about 10% of all cysts of odontogenic origin. It is an enigmatic developmental cyst that requires special attention. They arise from the dental lamina and have a cystic space containing desquamated keratin with a uniform lining of para-keratinized squamous epithelium. They have a characteristic aggressive behaviour and relatively high recurrence rate. The aggressive nature of the cysts are often associated with syndromes. Radiological imaging such as computed tomography (CT) and magnetic resonance imaging (MRI), plays an important role in the diagnosis and management of OKCs. The present case emphasizes on a case of odontogenic keratocyst involving the ramus of mandible bilaterally and also about the nature and characteristics of them.

KEYWORDS : Bilateral, Odontogenic keratocyst, Keratocystic odontogenic tumor, Odontogenic cysts, Non – syndromic

INTRODUCTION

The 4th edition of classification of head and neck tumors of World Health Organization (WHO) which was published in January 2017 has reclassified keratocystic odontogenic tumour (KCOT) as odontogenic keratocyst (OKC)[1]. It was first described by Phillipsen in 1956[2]. The etiology of OKC is that it arises from the dental lamina and originates in tooth bearing regions with predilection to mandible than maxilla (2:1)[3]. They can occur at any age but are commonly seen from 20 to 40 years of age with slightly male predilection. They have locally infiltrative behaviour and can be incidentally discovered during routine dental radiographic procedures[4]. This report presents the findings of a patient with bilateral OKC and also discusses on the literature review.

CASE REPORT

A 20 year old female patient came to our hospital with the chief complaint of pain in upper and lower right and left region for past 4 months. History of presenting illness revealed that pain was acute, intermittent in nature and was aggravated during eating and relieved on medication. Past dental history revealed history of consultation for the same before 2 days. All routine investigations were done. The medical, social and family histories were remarkable. On general examination, the patient was afebrile, calm, cooperative, conscious, well oriented, well built and nourished. Facial asymmetry was seen on extra oral examination. A diffuse swelling of size app. 2x2 cms was seen on right lower region of face extending superiorly till the ala tragal line, inferiorly till the inferior border of the mandible, medially till the angle of the mouth and laterally along the angle of mandible. On palpation, it was warm and tender. Intraorally, all third molars were missing. The routine hemogram and dermatological examination did not reveal any abnormalities. On radiographic examination, Orthopantomograph revealed a mixed radiolucent, radiopaque tooth like structure on the right side which was app. 0.5cms from the anterior border of ramus of mandible which is surrounding the entire tooth by well defined radiolucency and surrounding sclerotic border in 38. A mixed radiolucent radiopaque tooth like structure was seen on the right side along the angle the angle of mandible which is surrounding the crown portion of 48 by well defined radiolucency and surrounding sclerotic border in 48. A mixed radiolucent radiopaque tooth like structure was seen within the left maxillary sinus suggestive of impacted 28. The long axis of 18 was distally inclined suggestive of distally tilted 18 (Figure 1).

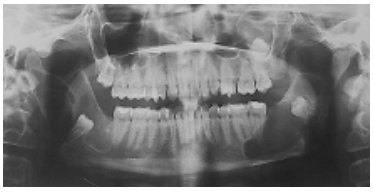

Figure 1 : Preoperative radiographic image

Based on clinical and radiological findings, a provisional diagnosis of bilateral dentigerous cyst in relation to 38, 48 was made. The lesions in relation to 38 and 48 were surgically curetted followed by chemical cauterization with Carnoy's solution along with surgical removal of impacted 28 under general anesthesia (Figure 2).

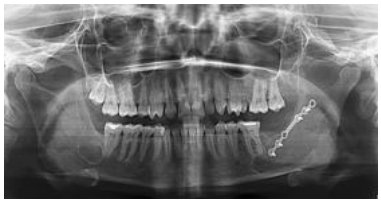

Figure 2 : Post operative radiographic image

The specimen was sent for histopathological examination. Hematoxylin and Eosin stained section of specimen showed cyst line by multi layered stratified squamous epithelium with a corrugated surface and nuclear palisading of the basal layer enclosing keratinous debris. The underlying stroma was collagenous in nature and showed dense infiltrates of foamy histiocytes, lymphocytes and areas of calcifications. At places the epithelium is retracted from the underlying stroma creating subepithelial clefts with focal budding odontogenic

epithelial rest (Figure 3). Corelating the clinical, radiological and histopathological findings a final diagnosis of bilateral non-syndromic odontogenic keratocyst was made.

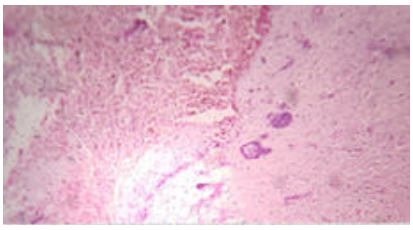

Figure 3 : Histopathological image (10X)

DISCUSSION

The WHO defined KCOT as benign uni- or multicystic, intraosseous tumor of odontogenic origin with a characterstic lining of parakeratinized stratified squamous epithelium and potential for aggressive, infiltrative behaviour[5]. The term odontogenic keratocyst was reinserted as keratocystic odontogenic tumor as WHO recommends the term KCOT reflects its neoplastic nature better. It was first described by Mickuliz in 1826 as a part of familial condition that affects the jaw[6]. Then it was referred as cholesteatoma in 1926. Robinson in 1945 mentioned this as a primodial cyst and was named as odontogenic keratocyst by Phillipsen in 1956. Toller in 1967 suggested it to be benign neoplasm. And due to its aggressive behaviour it was redesignated as KCOT in 2005.

The common complaints of the patients with OKC are pain, swelling and occasional pus discharge. Paresthesia of lower lips can be seen in some rare conditions[7]. It is usually asymptomatic till it attains a large size or has pathological fractures. Like other jaw cysts, it causes displacement of teeth and root resorption of adjacent teeth is often rare. It is commonly seen in tooth bearing areas (82%) and are associated with atleast 1 impacted teeth. In the mandible, the most common location is the posterior teeth, the angle or the ramus. In contrast, the anterior teeth and the third molar region are the most common sites in the maxilla. The factors that were considered for KCOT which was pointed out by Madras et al. [5] are high recurrence rate, localized destruction, budding of basal layer into connective tissues, association with mutation of tumor suppressor PTCH gene on chromosome 9q22.3q31 and presence of mitotic figure in suprabasal layer[8].

Radiographically, it presents as a unilocular / multilocular lesion. In a conventional radiograph, a hazy radiolucent lumen can be seen suggestive of proteinaceous substance such as keratin. Histologically, it has 3 variants- orthokeratinized, parakeratinized or both. But according to WHO classification of KCOT (2005) only parakeratinized variant has been included[8]. Histopathologically, it includes a uniform cell lining, palisading and hyperchromatic basal cells and wavy parakeratin production. Most specific features of OKC are its appearance of satellite cysts/islands of odontogenic epithelium. KCOT is most commonly associated with Gorlin-Goltz syndrome which is autosomal dominant inherited syndrome. It is characterized by its association of basal cell epitheliomas, jaw cysts and bifid ribs.

Treatment modalities such as curettage, enucleation, radical enucleation, marsupialization, resection are used for the management of OKCs[9]. Recurrence occurs for several reasons. It varies from 2.5% to 62%[10]. It is commonly due to incomplete removal of cystic lesions leading to formation of new cysts. It can also occur due to nevoid basal cell carcinoma syndrome.

ABBREVIATIONS

WHO – World Health Organization
KCOT – Keratocystic odontogenic tumor
OKC – Odontogenic keratocyst

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