Case Report

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Aggressive Surgical Management of Giant Size Maxillary Odontogenic Keratocyst with Conservative Approach: A Case Report

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ABSTRACT

Background: The odontogenic keratocyst is a developmental odontogenic cyst most commonly occurring in the jaws. They are unique odontogenic lesions that have the potential to behave aggressively, that can recur and can be associated with the nevoid basal cell carcinoma syndrome. The mandible is invariably affected more frequently than the maxilla. Here we are presenting a case report of a maxillary giant size odontogenic keratocyst exhibiting the aggressive behaviour of a tumour and its treatment with subsequent follow-up.

Keywords: Maxillary Odontogenic keratocyst, surgical management, conservative approach.

INTRODUCTION

The term odontogenic keratocyst was first used by Philipsen in 1956, who used the term to describe jaw cysts exhibiting keratinization of their epithelial linings.[1] Pindborg and Hansen in 1963 describe the essential feature of this type of cyst.[2] It is named keratocyst because the cyst epithelium produces so much keratin that it fills the cyst lumen. The cyst may occur at any age, but the peak incidence is in the 2nd and 3rd decade of life, with a gradual decline thereafter. In addition, a predilection for the male gender and the posterior mandible has been reported, with a male to female ratio of 2:1 and 65% to 83% of the cases involving the posterior mandible.[3] The mandible is invariably affected more frequently than the maxilla with (65% versus 35%) as reported by Brannon & (79% versus 21%) by Browne. In the mandible, the common site of occurrence is the ramus-3rd molar area. In the maxilla, the most common site is the 3rd molar area, followed by the cuspid.

The choice of treatment should take into account various factors, including patient age, size and location of the cyst, soft tissue involvement, history of previous treatment, and a histological variant of the lesion.[4-5] The goal of this paper is to choose the treatment modality that preserves maximum possible vital structures while ensuring the lesion is removed completely.

CASE REPORT

A 25 years old patient was referred to us with a history of swelling of the right side of the face and palate for one and half years (Fig.1). He had been previously operated on twice for the same with unsuccessful results. Considering the aggressive behaviour of the lesion, a CT of the face was

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planned. CT scan reveals a hypodense lesion involving the right maxilla (Fig.2). The internal structure is cystic in nature with no evidence of calcification. The lesion extends superiorly till the right frontal sinus, ethmoidal sinus and touching almost the floor of the orbit; inferiorly till the right maxillary alveolus and floor of the maxillary sinus; medially crossing the midline deviating the nasal septum and involving the superior and middle turbinates; laterally till the right lateral wall of the maxillary sinus and starting of the zygomatic arch; anteriorly till the right anterior alveolus and anterolateral wall of the maxillary sinus; posteriorly is defined by the posterior wall of the right maxillary sinus not involving the pterygoid plates(Fig.3).

The clinical and CT scan findings suggested a probable diagnosis of Odontogenic Keratocyst(OKC). To confirm the same, an incisional biopsy was taken, which turned out to be an OKC. Weber Ferguson incision was given to gain surgical access to the lesion site. The lesion was enucleated in toto along with peripheral ostectomy to prevent further recurrence in future. Antibiotic impregnated ribbon gauze packing was done for 48 hours to prevent dead space. The specimen was sent for histopathological examination and was confirmed as parakeratinized Odontogenic Keratocyst. Subsequent follow-ups were done after 1 week, one month, six months and one and half years. The lesion has not recurred, and the patient has restored normalcy with improvement in the quality of life, with the appreciable cosmetic result.

DISCUSSION

OKC requires special consideration amongst the cysts afflicting the jaws during the treatment because of its known aggressive behaviour and tendency to recur. Clinical evidence of its aggressive behaviour is supported by reported cases penetrating the cortical bone and involving adjacent soft tissues, as well as extending to the skull base from the mandible or to the orbit and infratemporal fossa from the maxilla.[6,7,8]

In the latest World Health Organization classification, this lesion is added to the benign odontogenic tumours category. The new term is "keratocystic odontogenic tumor" (KCOT). [9] World Health Organization Working Group has classified the KCOT as a benign tumors with odontogenic epithelium and mature, fibrous stoma without odontogenic ectomesenchyme. Several theories of the development of the KCOT have been presented. According to one theory, KCOTs develop from the remains of the dental lamina. According to other theories, KCOTs emanate from the basal cell layer of the oral mucosal epithelium or from the stellate reticulum of the enamel organ.[10]

Intraluminal hyperosmolality; active epithelial proliferation; collagenolytic activity in the cyst wall; synthesis of interleukin (IL)-1 and IL-6 by keratinocytes along with tumour necrosis factor, causing raised levels of prostaglandin; and increased expression of parathyroid hormone-related protein is thought to influence the expansion of OKC within the bone and thus facilitate its unremitting growth.[11]

The most important evidence that KCOT is a neoplasm comes from genetic studies demonstrating loss of heterozygosity of the tumour suppressor genes in KCOT cases, e.g., p53 and p16.[12]

The signs and symptoms reported are pain, swelling, expansion, drainage and bony perforation, but KCOTs were sometimes symptom-free and were found incidentally during routine radiographic examination. [13]

On the radiography, KCOTs appear as well-defined radiolucencies, which can be either unilocular or multilocular. [14] Unilocular lesions occurred with a greater frequency (73,7%) than the multilocular ones (26.6%) . Unilocular lesions can be located periapically, simulating periapical cysts; surrounding the crown of unerupted teeth, simulating dentigerous cysts (26,7% in the study of Haring and van Dis)[15]; between the roots of teeth, simulating lateral periodontal cysts or lateral radicular cysts; or in the maxillary midline, simulating nasopalatine duct cysts. Large unilocular KCOTs can be indistinguishable from cystic ameloblastoma.[14] The lesion's proximity to the tooth may cause displacement and/or root resorption, although displacement (28.3%) is more commonly seen than resorption (5%).[15] Conventional radiographic imaging, such as panoramic views and intraoral periapical films, in most cases are adequate to determine the location and estimate the size of the KCOT. Advanced

imaging techniques like computerized tomography and magnetic resonance imaging can be useful in large cases involving the maxillary sinus and the rare cases that extend to the skull base. [14]

KCOT is characterized by a uniform epithelial layer that lacks rete ridges. In addition, it has a corrugated parakeratinized luminal layer and a prominent basal cell layer. [14] Haring and Van Dis. [15] reported epithelial separation in 98% of cases they reviewed. Satellite cysts were evident in 33.3% of the KCOTs reviewed by them. No statistical significance existed between the radiographic appearance of the lesions and the presence of satellite cysts.[15] Some histologic findings of KCOT, including the presence of 1 or more daughter cysts and the budding of the basal cell layer of the lining epithelium, have been reported to be related to high recurrence rates.[13] Inflammation was present in 98% of the KCOTs reviewed by the same authors. The inflammation, predominantly a chronic inflammatory cell infiltrate, was most often in a generalized pattern of distribution.[15] A statistically significant relationship was found to exist between the severity of the inflammation (mild, moderate, severe) and the radiographic appearance of the lesion (unilocular versus multilocular). The multilocular lesions were shown to have more severe inflammation than the unilocular ones. A lesion may be stimulated to grow in response to inflammation.[15]

The treatment of the KCOT remains controversial. Treatments are generally classified as conservative or aggressive. Conservative treatment generally includes simple enucleation, with or without curettage, using spoon curettes or marsupialization. Aggressive treatment generally includes peripheral ostectomy, chemical curettage with Carnoy's solution, and resection.[15]

The goal of treatment is to eliminate the potential for recurrence while also minimizing surgical morbidity. Based on the aggressive nature of the lesion and the review of available kinds of literature, we would like to propose that if a lesion is suspecting KCOT, then irrespective of the size, a preliminary biopsy is mandatory. Many times conservative approach or incomplete removal of the lesion paved the way for recurrence, thereby hampering the patient to lead a quality life. Although conservative approach like marsupialization enables the preservation of vital structures like nerves and arteries, an aggressive approach like enucleation or resection with peripheral ostectomy followed by chemical cauterization can only prevent a recurrence. Whichever method of treatment is adopted by the operator, the success of the treatment lies in eradicating the lesion completely, even at the expense of a few vital structure sacrifices. A long term follows up of 5 years can only rule out disease-free healthy life.

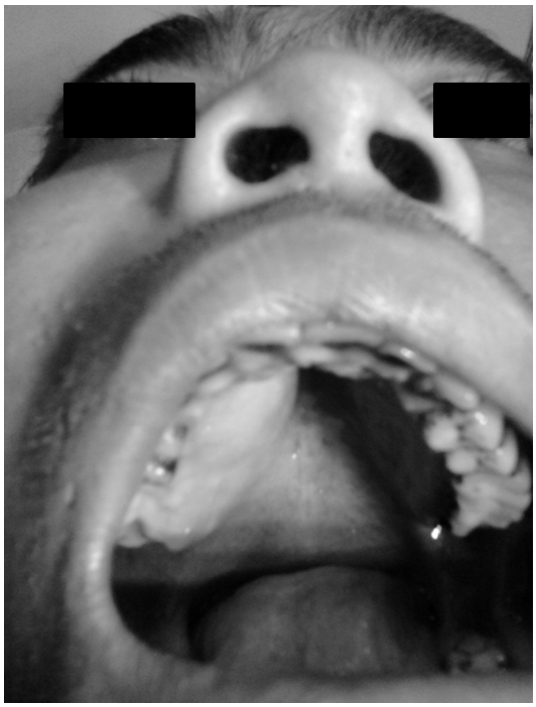

Fig 1 Preoperative photograph of the patient.

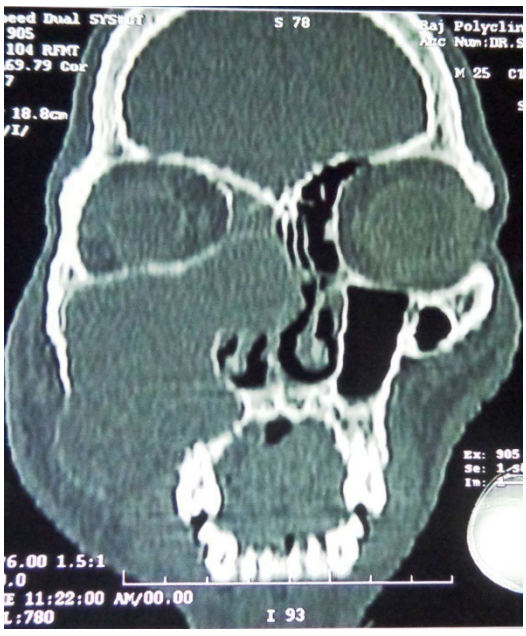

Fig 2: Preoperative coronal view of the CT scan

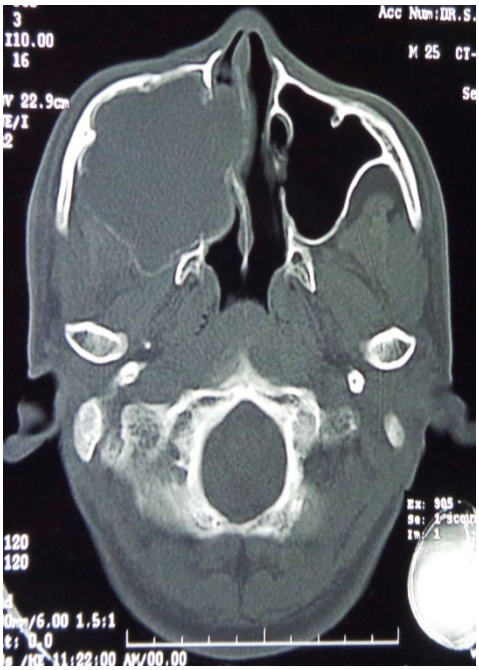

Fig 3: Preoperative axial view of the CT scan

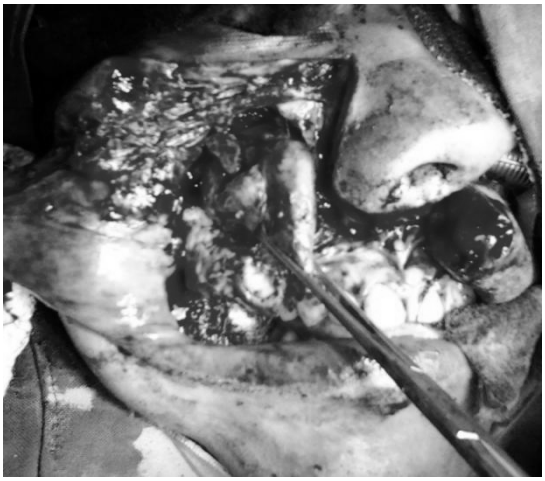

Fig 4: Intraoperative photograph of the patient.

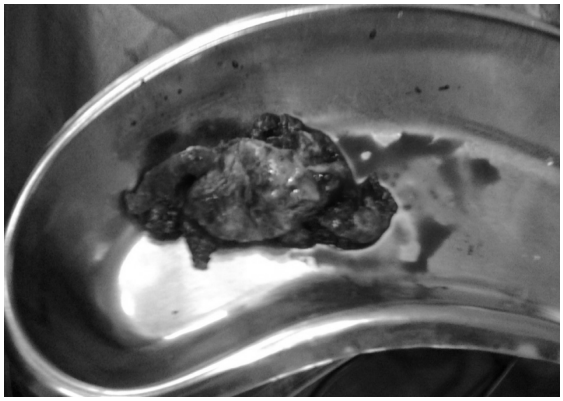

Fig 5: Photograph of the lesion enucleated.

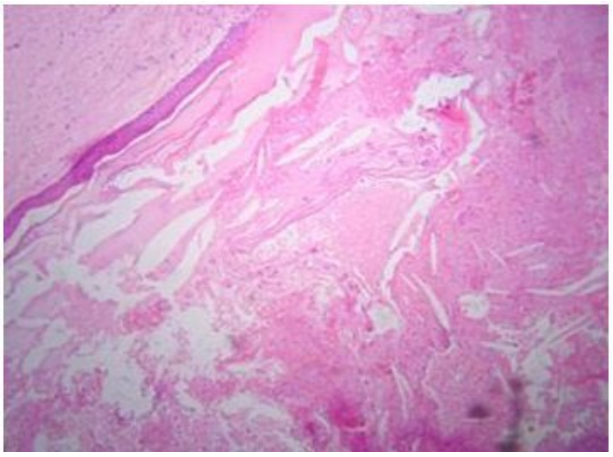

Fig 6: Histopathological examination.

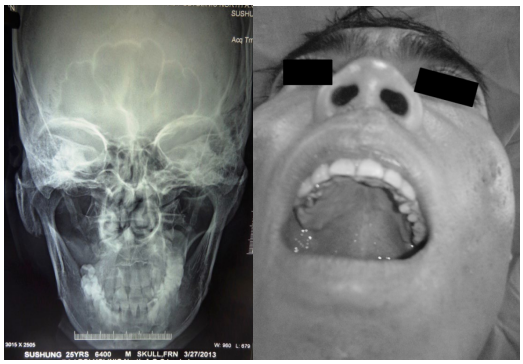

Fig 7: Post operative radiograph and pictures.

CONFLICTS OF INTEREST

The authors declare they have no potential conflict of interest regarding this article.

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