Case Report

Odontogenic keratocyst involving the maxillary antrum: A case report

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ABSTRACT

Odontogenic keratocyst (OKC) is a benign neoplasm of the jaw with a common occurrence in the mandibular ramus area characterized by aggressive behavior and a high recurrence rate. This lesion rarely occurs in the maxilla. The aim of this report is to discuss the case of a 27-year-old male who suffered from OKC in the left maxillary antrum, which includes surgical management of the case with cyst enucleation and chemical cauterization.

Key words: Odontogenic cyst, Maxillary antrum, Keratocyst, Enucleation, Cauterization

Odontogenic keratocyst (OKC) was first described by Philipsen in 1956 and later renamed as keratocystic odontogenic tumor by the World Health Organization in 2005. These cysts are described as developmental, benign, but locally aggressive lesions characterized by invasive growth into the neighboring structures with a high rate of recurrence [1-3]. Clinically, the cyst presents with localized asymptomatic swelling associated with mobility of the teeth [1,2]. Frequent occurrence of cysts is seen in the mandible, of which, about 90% occur posterior to the canines and 50% in the ascending ramus of the mandible [4-6]. Less than 1% of cases have been reported in the maxillary antrum [1]. Due to unspecific clinical and radiographic features, it may be confused as an ordinary cystic lesion, leading to underdiagnosis and under-treatment, resulting in unnecessary recurrences. Successful treatment depends on a precise diagnosis, adequate surgical procedure, and follow-up [7,8].

The objective of the report is to present a rare case of occurrence of OKC in the left maxillary antrum in a male patient and surgical management of the case with enucleation and follow-up.

CASE REPORT

A 27-year-old male patient reported to the department of oral surgery with the chief complaint of uncharacterized painless swelling on the left side of the face. On extraoral examination, there was diffuse swelling on the left side of the face involving the left maxillary sinus region, which was tender on palpation. Extraorally, the swelling was 4 × 2 cm in size, extending from the midline to the left canine region anteroposteriorly and from the philtrum to the upper lip superior-inferiorly. Intraorally, the swelling was 2 × 2 cm in size, extending from the depth of the labial vestibule to the attached gingiva with respect to upper incisors superoinferiorly and from the midline to the left canine region anteroposteriorly. The lymph nodes were not palpable on both sides. On palpation, mild tenderness was present.

Radiological evaluation was done with non-contrast computed tomography (NCCT) and 3D computed tomography (CT) showing a 2 × 2 cm radiolucent lesion in the left maxillary antrum involving the incisor-canine region and roots of the teeth (Fig. 1).

The cytological examination was done with fine-needle aspiration. Cytological examination suggested a cystic lesion with a lining comprising parakeratotic squamous epithelium, along with fragments of bone, fibro-collagenous tissue with focal areas of chronic inflammation, and mucous glands. On histopathological examination, the submitted tissue was suggestive of a cystic lesion lined by para-keratinized stratified squamous epithelium of uniform thickness of 6–8 cell layers. The basal cells were hyperchromatic showing palisaded arrangement and the underlying connective tissue component was collagenous in nature. The histopathological examination was suggestive of OKC (Fig. 2).

Under local anesthesia, a sulcular incision was made from the left side of the maxilla between the central incisor and the first molar (Fig. 3a). Multiple extractions of the involved teeth were done. The approach to maxillary antrum was made over the canine fossa. The anterior wall of the maxillary antrum was

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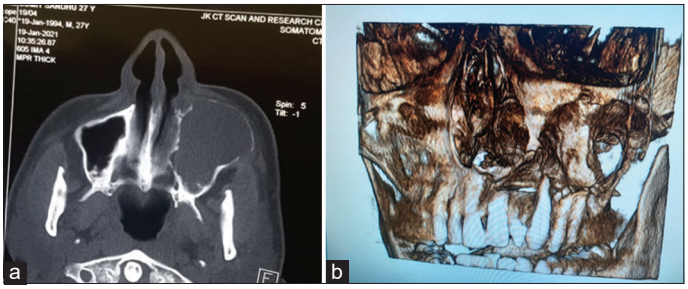

Figure 1: (a) Cross-sectional computed tomography (CT) showing involvement of left maxillary antrum and (b) 3D CT showing a lesion involving incisor-canine region

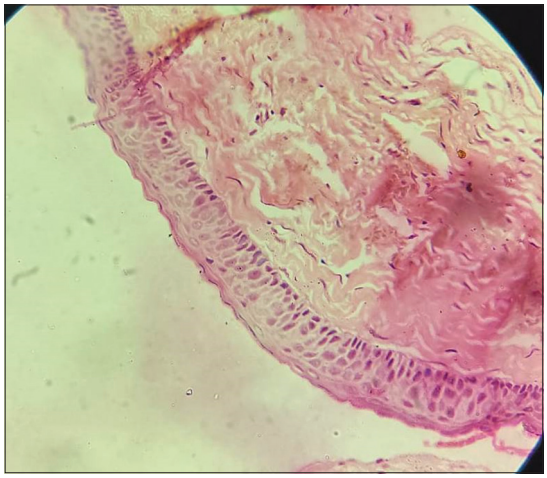

Figure 2: Photomicrograph with histopathological features suggestive of odontogenic keratocyst

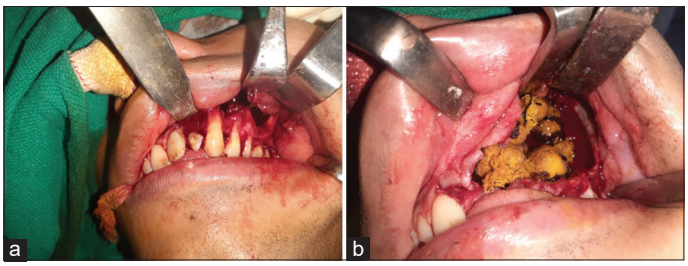

Figure 3: Intraoperative images (a) showing incision to approach maxillary sinusand (b) showing enucleation of cyst followed by cauterization

eroded by the cyst, which was enucleated and submitted for histopathological evaluation. Enucleation of the cyst followed by chemical cauterization was done (Fig. 3b). The flap was placed in its original position and sutured. The site was examined thoroughly in the follow-up visits by radiographs (Fig. 4).

DISCUSSION

The main characteristics of OKC are the high recurrence rate and potentially aggressive nature [1,9]. This cyst occurs significantly more in the posterior region of the mandible affecting people in the second and third decades of life with a predilection for males [10]. Recurrence incidence is found to be higher in cases of

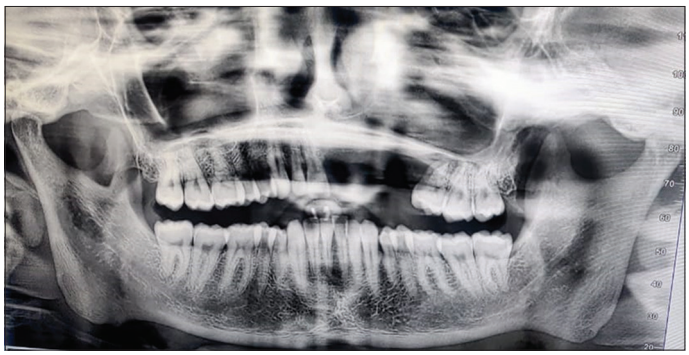

Figure 4: Follow-up visit radiograph of the patient

multilocular lesions and the lesions located in the ramus region, compared with the other regions [11]. Less than 1% of occurrence is found in the maxilla with sinus involvement [8]. An unusual case of invasive maxillary sinus OKC into the orbital floor, pterygoid plates, and hard palate has also been reported [12]. Our presented case presents with this rare occurrence in the maxillary antrum with sinus involvement. OKC located in the maxillary antrum is found in correlation with the impacted wisdom tooth or canine, which was not found in this case. In the maxilla, it is seen most commonly in the canine area, followed by third molar tuberosity and anterior maxilla. In most cases, it presents as a periapical lesion, as seen in this case [10].

The origin of OKC in the maxillary sinus is presumably from the entrapment of odontogenic epithelium from the developing dental lamina of canine, due to its close anatomic relationship with the floor of the sinus [8]. Multiple lesions are also associated with the nevoid basal cell syndrome (Gorlin-Goltz syndrome); however, the present was a single lesion in a non-syndromic patient [11]. The peak incidence is in the second and third decades of life and is found more frequently in males. Our case, however, was a female in the third decade of life. Radiographically, OKC appears as a well-defined radiolucency which may be unilocular or multilocular [5]. In the present case report, the radiographic examination and NCCT showed obliteration of the left maxillary sinus with involvement of periapices of the incisor and canine teeth.

The treatment options for OKC vary from simple curettage, enucleation (in combination with cryotherapy or Carnoy's solution), marsupialization, decompression and secondary enucleation, and resection (marginal or segmental) [1,2,13]. The treatment management and the follow-up period are the two prominent factors affecting the recurrence rate of the cyst. Simple enucleation was associated with a higher recurrence rate [11-14], while resection and enucleation with bone curettage presented lower rates. Due to a higher rate of recurrence found in the dentate area, special follow-up is recommended in the dentate area if enucleation is chosen as a treatment option [11]. Enucleation with chemical cauterization was the treatment option used in our exhibited case with no recurrence in the follow-up period. However, a long-term follow-up is needed to avoid any unnecessary complications, such as recurrences of the cyst, which might be noted even ten years after the treatment [15].

CONCLUSION

The appearance of OKC is rare in the maxillary region, so the misinterpretation of the radiographic image is common. Due to unspecific clinical and radiographic features, it may be confused with ordinary cysts, leading to underdiagnosis and under-treatment, resulting in unnecessary recurrences. Computed tomography and biopsy specimen examination was done in our case to rule out the misdiagnosis and provide the most effective treatment to avoid recurrence.

ETHICAL APPROVAL

The ethical clearance certificate was obtained from the Institutional Ethical Committee as per the standard guidelines.

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