Case Report

Bilateral Infected Maxillary Odontogenic Keratocyst: A Case Report

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ABSTRACT

Odontogenic keratocyst (OKC) is a benign intraosseous cystic lesion with a high recurrence rate. It is commonly found in the mandibular posterior region, but it can also be present in the maxilla. Multiple OKCs are usually found associated with Gorlin–Goltz syndrome. This is a rare case report of bilateral OKC at the maxillary posterior region. A 40-year-old male patient presented with a chief complaint of pus discharge from the right upper back tooth region. Radiographically, a well-defined radiolucent area in relation to the impacted 18 and 28 was appreciated. Based on histopathological findings, a final diagnosis of bilateral infected maxillary OKC was made. However, multiple or bilateral OKCs are rare findings and have been reported in only 5% of patients. Hence, careful diagnosis and follow-up are required for such cases.

Key words: Odontogenic keratocyst, Gorlin–Goltz syndrome, Nevoid basal cell carcinoma, Protein patched homolog gene

INTRODUCTION

The remnants of the dental lamina give rise to a benign but aggressive intraosseous cyst known as an odontogenic keratocyst (OKC).[1] OKC makes up to 3–11% of all jaw cysts[2] and primarily affects individuals between 30 and 50 years of age group.[1]

Patients with early-stage OKC usually do not exhibit any recognizable symptoms or signs as despite the size of the lesion, it does not cause any significant enlargement of the alveolar bone.[2] On secondary infection, the patients may have symptoms such as swelling, pain, abnormal sensation, pus outflow, and loose teeth.[3] The histopathological features of the cyst include a cystic cavity lined by a parakeratinized stratified squamous epithelium consisting of six to ten cell layers, a palisaded basal cell layer comprising of columnar cells, and a flat epithelial-connective tissue interface.[4,5] Basal cell budding/off-shoots, satellite/daughter cysts, and odontogenic epithelial proliferations are sometimes seen in the connective tissue wall, indicating that the epithelium is genetically predisposed to enhanced proliferation.[6] The aggressiveness of cystic epithelium is shown by its high recurrence rate and enhanced mitotic activity.[7]

Occurrence is more common in the posterior region of the dental arch involving the angle or the ramus of the mandible (66–77%) than in the maxilla.[3,8]

CASE REPORT

A 40-year-old male patient reported to the outpatient department of the Institute of Dental Sciences, Bareilly, Uttar Pradesh, with the chief complaint of pus and cheesy material discharge from the right upper back tooth region for since 6 months. The patient had no significant medical history and presented a positive dental history for the diagnosis and treatment of some cystic lesions 10 years back. On clinical examination, round and oval bilateral swellings were present at the maxillary posterior region in relation to 18 and 28 alveolar regions, respectively. However, both the maxillary 3rd molars were missing clinically. The tenderness and discharge were also present in relation to 18 region. No abnormality was detected extra orally.

Orthopantomograph showed two bilateral expansile cystic lesions in maxillary bone with imperceptible cortex at multiple places and impacted teeth in relation to 18 and 28 within the lesion [Figure 1]. Considering this, the working diagnosis of "Bilateral infected dentigerous cysts of maxilla" was made.

An excisional biopsy from the bilateral maxillary posterior region was performed for histopathologic confirmation. On grossing of the specimen, it was found that five pieces of tissue

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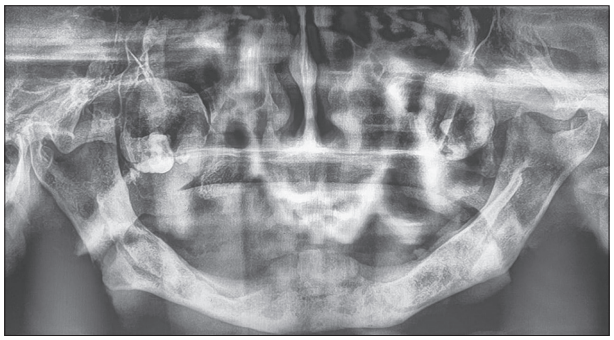

Figure 1: Orthopantomograph shows two bilateral expansile cystic lesions in maxillary bone with imperceptible cortex at multiple places and impacted teeth in relation to 18 and 28 within the lesion

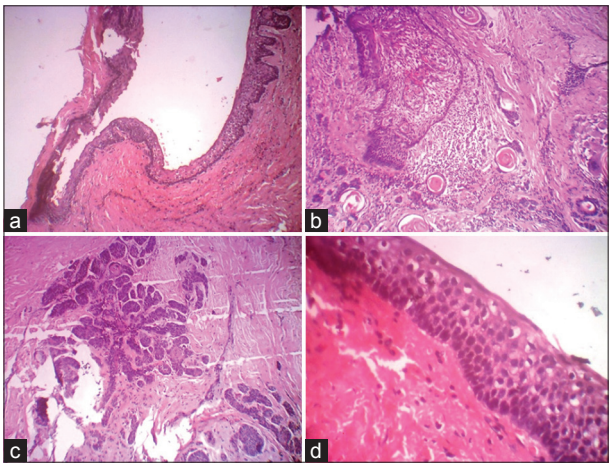

Figure 2: (a-d) the photomicrograph under hematoxylin and eosin-stained sections shows: (a) areas depicting irregular cystic epithelium with supporting inflamed hemorrhagic connective tissue capsule encircling cystic lumen (×40), (b) multiple daughter cysts with aberrant keratin deposits in the center (×100), (c) odontogenic islands (few with keratin formation) embedded in the underlying connective tissue (×100) and (d) stratified squamous parakeratinized cystic epithelium depicting 5–8 cell layers and palisaded basal cell layer of tall columnar cells with hyperchromatic elongated nuclei (×400)

received from the left maxilla which were blackish-brown, soft to firm in consistency, and rough in texture, with the largest tissue measuring 1.3 cm × 2.5 cm. From the right maxilla, a single piece of tissue was received which was brown-gray, soft in consistency, rough in texture, and measuring 2.0 cm × 2.0 cm in maximum dimensions. Two extracted third molars from each side were also received.

On histopathologic evaluation, under the scanner and low power view, areas depicting irregular cystic epithelium with supporting inflamed hemorrhagic connective tissue capsule encircling cystic lumen were revealed [Figure 2a]. Multiple daughter cysts with aberrant keratin deposits in the center and odontogenic islands (few with keratin formation) embedded in the underlying connective tissue are also appreciated [Figure 2b and c].

Under higher magnification, the cystic epithelium is stratified squamous parakeratinized type and 5–8 cell layers thick demonstrating basilar hyperplasia in few places. The epithelium shows a palisaded basal cell layer of tall columnar cells with hyperchromatic elongated nuclei [Figure 2d]. The connective tissue stroma comprises a loose to dense arrangement of collagen fibers with plump to spindle-shaped fibroblasts. The stroma depicts chronic inflammatory infiltrate predominantly comprising lymphocytes and zones of extravasated red blood cells.

The final histopathological diagnosis of bilateral infected OKC (parakeratinized type) was made.

DISCUSSION

OKC is a developmental odontogenic cyst with 31.3% of them occurring in the maxilla compared to the mandible, that is, OKCs of the maxilla are less prevalent than those of the mandible, making them more challenging to diagnose.[8] The present case illustrates the relatively rare incidence of OKC in the maxilla.

According to histopathology, OKC is distinguished by a cystic lumen lined with six to eight uniformly thick layers of corrugated, parakeratinized stratified squamous epithelium. Tall columnar basal cells with palisading nuclei and polarity reversal are characteristics of lining epithelium.[2-5] The epithelial connective tissue interphase is flat and satellite cysts or odontogenic epithelial islands may be present in the connective tissue wall.[7] In addition to these common characteristics, the present case demonstrates basilar hyperplasia, daughter cysts, and odontogenic islands embedded in the underlying stroma.

It has been reported that OKC has a tendency to recur so frequently. Furthermore, during enucleation, the delicate, thin epithelial cyst lining of OKC can rupture easily, resulting in insufficient clearance and recurrences. The development of small satellite cysts could be another factor.[9] In the present case, the thick cystic lining was seen in relation to 18 and 28 regions and on histopathology, it was revealed to be an aggressive lesion due to the presence of multiple daughter cysts and odontogenic islands in the supporting connective tissue. The possibility was suspected of any syndromic association, that is, Gorlin–Goltz syndrome which was to be ruled out clinically as parakeratinization; intramural epithelial remnants, and satellite cysts were a more frequent finding among Nevoid basal cell carcinoma (NBCCs) associated OKCs than among solitary OKC.[8,9] OKCs are one of the most frequent features of NBCCs. It is linked with a mutation in the protein-patched homolog gene. Partial expression of the gene may result in the occurrence of multiple recurring OKC.[9] However, bilateral OKC is a rare finding and has been reported in only 5% of cases.[10] It is important to carefully consider the clinical manifestation and diagnostic aspects for clinical management of the lesion. Available treatment options that are more conservative including the Carnoy solution, cryotherapy, or decompression accompanied by a subsequent enucleation may be considered.

CONCLUSION

The present case report highlights a rare bilateral maxillary cystic lesion. Even though parakeratinized OKCs are uncommon in the maxilla, particularly in the posterior region, the clinician needs to accurately assess whether the syndrome's characteristics are present. To monitor and prevent recurrence, the patient must be under strict post-operative clinical follow-up.

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