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Non-syndromic bilateral odontogenic keratocyst

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Abstract

The odontogenic keratocyst (OKC), formerly known as keratocystic odontogenic tumour, is one of the most controversial orofacial disorders, emerging from the dental lamina rest or the oral epithelium's basal cells, or reduced enamel epithelium of the dental follicle. It has a high rate of recurrence due to inadequate clearance of the original cyst lining and the formation of a new cyst from a small satellite cyst. The WHO has defined Odontogenic Keratocyst as an Odontogenic cyst in its most recent classification of head and neck malignancies in 2017. Patients with OKC may be asymptomatic, and the condition is discovered by chance on routine radiographs. Alternatively, it could be accompanied by discomfort, swelling, or discharge. The Gorlin-Goltz syndrome is associated with the presence of multiple Odontogenic keratocysts.

Mostly surgical treatment is considered the standard treatment for odontogenic cyst, but in the last years, a combined therapy has shown more success rates with the use of multi modal approach such as decompression, use of Carnoy's solution, mar supialization, cryotherapy after conservative surgical management.

Here we present a case report of a 26-year-old female patient with bilateral OKC in posterior mandible which was not associated with Gorlin-Goltz syndrome.

Keywords: Gorlin - Goltz syndrome, keratocyst, odontogenic tumor.

Introduction

The odontogenic keratocyst (OKC) is a developing cyst of the jaws that is epithelial in nature. Because of its propensity for substantial expansion, extension into neighbouring tissues, and fast development, this lesion is often encountered in the maxilla and mandible, and can get quite large.(1) The main features of odontogenic keratocysts were described by 'Pindborg' and 'Hansen' in 1963 although the term had earlier been used by 'Philipsen' in 1956.(2)

In 1992, the World Health Organization introduced the term 'odontogenic keratocyst', synonymous with

'primordial cyst', to denote benign cysts of odontogenic origin and specific histological appearance. In 2005, considering a high risk of recurrence, aggressive clinical course, mutations in the tumor suppressor gene (PTCH1), the occurrence of satellite cysts, and the association with the Gorlin–Goltz syndrome, WHO reclassified this pathology as a benign keratocystic odontogenic tumor (KCOT). In 2017, though, WHO released a new classification of head and neck Tumors. As there was insufficient evidence to categorize the abovementioned pathology as a neoplastic lesion, KCOT was moved back into the cyst category under the name of odontogenic keratocyst (OKC). [3]

The origin of OKC is not clear though it is presumed to be either primordial, including dental lamina remnants, basal cells of the overlying epithelium, or dentigerous, which implies reduced enamel epithelium of the dental follicle. It is more frequent in the mandible, and the maximum frequency in people aged ten to thirty years old with a mild male preponderance.

Keratocysts are most commonly discovered by chance during routine radiographic examinations. [4] They may present as either a multilocular or unilocular radiolucent lesion. In 25% to 40% of cases, there is an unerupted tooth involved in the lesion. [5] The Para keratinizing lesions are characterised clinically by rapid development and a tendency to recur following surgical therapy. In addition, multiple OKC have been linked to nevoid basal cell carcinoma syndrome.

Although various therapies have been reported in the literature, ranging from conservative methods such as enucleation (with or without curettage), decompression, and mar supialization to aggressive treatments such as peripheral ostectomy with rotary instruments, cryotherapy with liquid nitrogen, and Carnoy's and jaw resection, the universally accepted approach remains undecided. [6]

Case report

A 26-year-old female reported to the department of oral medicine and radiology with the chief complaint of pain in right lower back tooth region since 15 days which was spontaneous in onset, severe in intensity for which she took analgesic medication prescribed by a local physician but she did not get any relief and this was accompanied by swelling in the same region along with reduced mouth opening. Extra oral examination revealed no visible asymmetry on right and left half of face (Figure:1). Intraoral examination revealed initial occlusal caries i.r.t 37 with vestibular tenderness on buccal vestibule and localised swelling on gingiva on lingual aspect i.r.t 37. (Figure:2,3). A provisional diagnosis of periapical abscess was given i.r.t 37 and an orthopantomography was advised which showed unilocular bilateral well-defined radiolucency extending from 46 up to the sigmoid notch and from symphysis region up to 3rd molar tooth on right and left side respectively with displacement of 35 on distal aspect (Figure:3)

FNAC was done which procured a cheesy aspirate and was send for histopathological examination which showed numerous in flammatory cells chiefly poly morphonuclear leukocytes and lymphocytes in a haemorrhagic back ground with few desquamated epithelial cells suggestive of inflammatory lesion. All the blood reports were within normal range except haemoglobin which was below normal for her age range.

CBCT was carried out and the axial and sagittal view demonstrated an area of well-defined hypodensity with corticated borders extending from root apex of 32 to apical region of 38 on left side with displacement of roots of 34 and 35 also involving inferior alveolar nerve

canal with minimal cortical expansion (as seen in axial view). Right side axial and sagittal view also shows an area of well-defined hypo density with corticated border extending from root apex of 46 up to the right sigmoid notch, involving inferior alveolar nerve with minimal cortical expansion (as seen in axial view) giving a radio graphic impression of multiple odontogenic keratocyst with differential diagnosis as dentigerous cyst, ameloblastoma and simple bone cyst. (Figure: 4,5-a, b, c, d). An incisional biopsy was done and was sent for histopathological examination which revealed cystic space lined by 8–10-layer thick Para keratinised stratified squamous epithelium and underlying fibrous connective tissue wall with dense infiltrate composed of lymphocytes with plasma cells. Thus based upon the history, clinical examination and investigations, a final diagnosis of odontogenic keratocyst involving right and left side of mandible was given.

Discussion

Philipsen originally described the OKC in 1876 and named it in 1956. [2] It is a unique epithelial developing odontogenic cyst that accounts for roughly 11% of all jaw cysts. [7]

The advent of fresh data about their morphology and biological activity led to the reclassification of OKC from benign odontogenic tumours to odontogenic developing cysts. A mutation of the PTCH1 gene, for example, has been proven to not be OKC-specific, as it also occurs in follicular cysts. PTCH1 changes are seen in 30–85 percent of OKCs, depending on whether the lesion is spontaneous or linked with the Gorlin–Goltz syndrome. It's also been discovered that OKCs retreat after decompression or mar supialization, and that their lining transforms spontaneously into normal oral epithelium. All these characteristics prohibit this clinical entity from being classified as a neoplastic lesion, justifying the change of the lesion's nomenclature to OKC. [3] The aetiology of developmental odontogenic cysts is unrelated to inflammatory stimuli and arises from epithelial remains of various stages of odontogenesis. There are seven types of cysts, with the dentigerous cyst and the keratinizing odontogenic cyst, also known as Keratocyst, being the most frequent. Unlike other odontogenic cysts, odontogenic keratocysts arising from remains of the dental lamina have an aggressive clinical behaviour and a high recurrence rate. [8]

Several processes have been proposed to explain recurrence. Three explanations have been proposed: inadequate removal of the original cyst lining, formation of a new OKC from satellite cysts or odontogenic epithelial remnants left over following surgical therapy, and the development of an unrelated OKC in an adjacent jaw area that is interpreted as a recurrence. Because of the cyst's placement in inaccessible locations, adhesions, or the thin, friable cystic epithelium, incomplete evacuation is thought to be a result of technical problems. Due to the possibility of recurrence many years after first treatment, regardless of the treatment administered, a life-long follow-up regimen, every year for the first 5 years and every 2 years thereafter, is recommended. [9]

OKCs are a common benign jaw tumour, although bilateral OKCs are uncommon. There have been several reports of single OKCs in the literature. Bilateral or multiple cysts are uncommon and may be linked to conditions such as Gorlin syndrome. In 1960, Gorlin and Goltz described Gorlin–Goltz syndrome as a condition, comprising the principal triad of multiple BCC, odontogenic keratocysts (OKC) and skeletal anomalies. The present case has bilateral OKC but was not associzted with Gorlin Goltz syndrome. In the literature,

OKC has been documented to transform into squamous cell carcinoma and Amelo blastomas. Amelo blastomas transformation of the OKC is very rare.

The recommended treatment for OKC is curettage with peripheral Ostectomy, cryosurgery (curettage with liquid nitrogen therapy), curettage plus application of Carnoy's solution, localized en bloc resection and occasionally, mandibular segmental resection, enucleation with post-operative intra-oral suction and rinsing the bone defect with 3% hydrogen peroxide to detect and remove eventual remains of the capsule. [10]

Conclusion

Multiple OKC are mostly associated with Gorlin-Goltz syndrome, but in some cases like the one presented above it is not associated with the syndrome which was confirmed by chest x ray to evaluate the ribs in which no anomaly was detected, and skull CT which did not show any calcifications. OKC are recognised to be locally aggressive and prone to recurrence thus aggressive follow up should be done every year for the first 5 years and every 2 years thereafter.

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Legend Figure

Figure 1: Extraoral examination revealed no visible asymmetry.

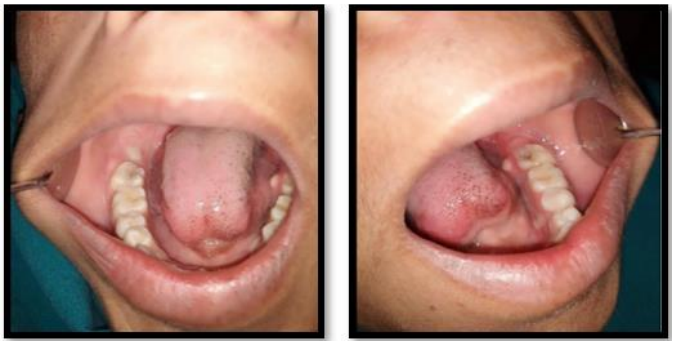


Figure 2: Intraoral examination showing localised swelling on lingual aspect of 37 and the right side appears to be normal.

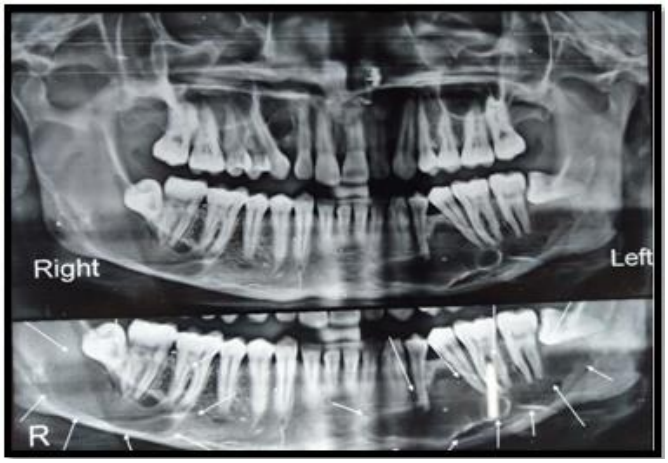
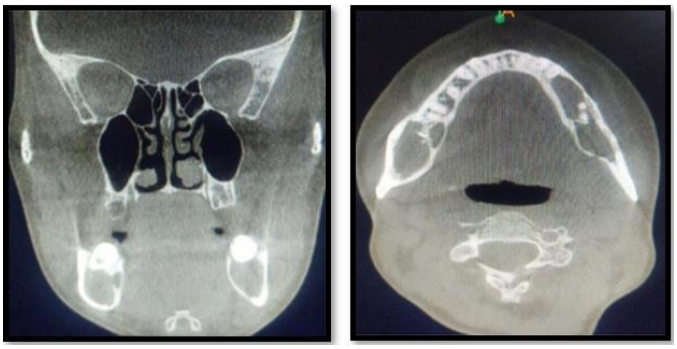
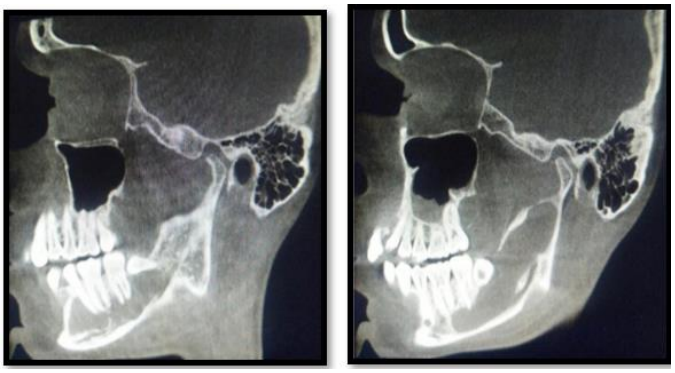


Figure 3: OPG showing well defined radiolucency on right and left side of mandible.



(a) (b)

Figure 4: (a) CBCT coronal view and (b) Axial view showing well defined hypodensity on right and left side with minimal cortical expansion.



(a) (b)

Figure 5: (a) CBCT left sagittal view (b) right sagittal view showing well defined hypo density with dis placement of 34 and 35.