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Case Report

Conservative surgical management of large bilateral mandibular odontogenic keratocyst: a case report and literature review

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ABSTRACT

Odontogenic keratocyst (OKC) have a been a controversial topic within the medical, research and taxonomy community. It is because of these controversies this requires our special attention. From a clinician's view, its varied presentation and high recurrence rate makes it special. One possible explanation for such high recurrence is the presence of anti-apoptotic gene; BCL-2 in high concentration in the epithelial lining of OKCs. Even rarer is its presentation bilaterally, we present one such case of a young male. After facing multiple misdiagnosis and any significant improvement, he was finally diagnosed on the basis of biopsy and underwent serial marsupialization followed by enucleation. We present this case to stress on the fact that conservative surgical management by marsupialization under local anaesthesia itself is enough without the need of costly reconstruction in cases with unilocular cyst involving the mandible bilaterally with review of relevant literature.

Keywords: Odontogenic keratocyst, Bilateral, Marsupialization, Mandible, Cyst, Local anaesthesia

INTRODUCTION

Odontogenic cyst is a relatively common occurring lesion with a varied presentation, this can be a small benign lesion or an aggressive lesion with a malignant potential. Amongst the aggressive ones, the most important is odontogenic keratocyst. OKC is a rare developmental cyst which has a very aggressive growth as well as high recurrence rate. The lesion was first described by Mikulicz in 1876 as a dermoid cyst which was later classified by Philipsen in 1956 as 'odontogenic keratocyst'.[1]

It was then considered as a developmental cyst. However, it was first known as 'cholesteotoma' in 1926.[2] It was later in 1962, Pindborg and Hansen gave the histological diagnostic criteria. After many attempts to classify, the WHO in 1992 settled on odontogenic keratocyst (OKC); which was again challenged in 2002 and after the Editorial and Consensus Conference, held at Lyon, France in 2003 the consensus was formed on 'keratocystic odontogenic tumour' (KOT).[3] Although again, in 2017 the WHO has renamed KOT as OKC.[4] They arise from the proliferation of the epithelial dental lamina in both maxilla and mandible. OKC have high recurrence rate, mitotic counts and epithelial turnover rates making it a tumour requiring quick diagnoses and prompt and aggressive treatment. They can occur at all ages, but also show a peak in the second and the third decade of life, with a predilection for white males.[5,6] As rare as they are (2.5% to 62%), even rarer is their presence bilaterally.[7] Such a presence is more commonly associated with syndrome like basal cell nevus syndrome rather than isolated occurrence. This report is of such an isolated occurrence.

CASE REPORT

This is a 17-year-old male patient, came to the department with the chief complaint of swelling in the left posterior

region of the lower jaw for past 15 days for which he visited another tertiary care centre. Further, this swelling was initially small but gradually increased to present size. He gave no history of trauma or discharge, along with no previous difficulty in chewing food. His past medical history revealed presence of no systemic illness. His dietary choices included non-vegetarian food with no history of any addictions like smoking, tobacco, chewing betel nut or alcohol. On visual inspection, there was a diffuse swelling extending from the left angle of mandible to the right angle of mandible posteriorly. extending superiorly to the angle of the mouth; while inferiorly to the lower border of mandible approximately 2×8 cm (Figure 1). Further, the colour of the swelling was the same as that of the surrounding skin.

On palpation it was soft-to-firm in consistency, non-tender, lesion extensions on one side of face. non-compressible, non-fluctuant and afebrile. Intraoral examination revealed an obliteration of buccal vestibule near the third quadrant molars by the swelling, which had the same colour as the surrounding mucosa. No teeth were missing and there was presence of generalised calculus and stains.

The lesion was aspirated, which came out to be straw coloured fluid, following which a differential diagnosis of generalised chronic periodontitis, odontogenic keratocyst and unicystic ameloblastoma was considered. Intraoral periapical (IOPA) radiograph was taken which showed a radiolucency in the premolar and molar region of the third quadrant, extending from second left lower premolar anteriorly to second left lower molar region posteriorly, while superiorly it extended from the alveolar ridge of second premolar and first molar of the third quadrant.

An orthopantomogram (OPG) was taken which showed well-defined radiolucency extending from the third quadrant molar to the fourth quadrant molar (Figure 2).

This was ill-defined in shape with ill-defined corticated borders involving the roots ranging from lower left molar to right lower molar, thus giving a floating root appearance along the superior border; inferiorly though, thin alveolar bone is appreciated. The radiographic evidence narrowed us towards a provisional diagnosis of odontogenic keratocyst with a differential diagnosis of unicystic ameloblastoma, odontogenic myxoma or simple bone cyst.

For the purpose of full disclosure, the patient was provided with all the options treatment, as he was not very financially strong, he couldn't afford reconstruction. Hence, after taking an informed consent, he underwent marsupialization after extraction of the following teeth: central incisor (third quadrant), central and lateral incisor (fourth quadrant), canine (fourth quadrant); under local anaesthesia.

During extraction the wall of the cyst associated was removed and sent for histopathological analysis. The cyst was packed with betadine gauze and started on prophylactic antibiotics with painkillers for a week upon discharge (Figure 3). The histopathological report stated presence of stratified squamous epithelium with regimentation of basal layer without rete pegs, transformed into hyper plastic and secondarily inflamed epithelium. It also showed ulceration and vascular proliferation with thin ameloblastic epithelium. This confirmed the diagnosis of inflamed odontogenic keratocyst.

Patient was then recalled every week for assessment of patency of the window, iodoform dressings and serial marsupialization's. After 6 months of regular follow up, continuous dressing and window assessment; for better compliance, after an informed consent another extraction was done; under local anaesthesia of the second and third molars in the third quadrant (Figure 4).

Marsupialization was done again to create a window and the cyst lining was taken out. Immediately after, betadine gauze was used to prevent bleeding and later packed with iodoform dressing. Further, over the coming months serial marsupialization's were done to decompress the cystic cavity. This led to the shrinkage of cystic cavity also some bone formation as was confirmed by the OPG.

Around 17 months later, after confirmation of a significant reduction in the size of the cyst, finally enucleation was done to remove the remaining cystic lining. The patient was followed up on a weekly basis for constant evaluation and irrigation. OPGs were taken during every visit, after a year of enucleation being done, the presence of bone can be abundantly appreciated (Figure 6). The windows created for decompression were closed using mucosal flaps.

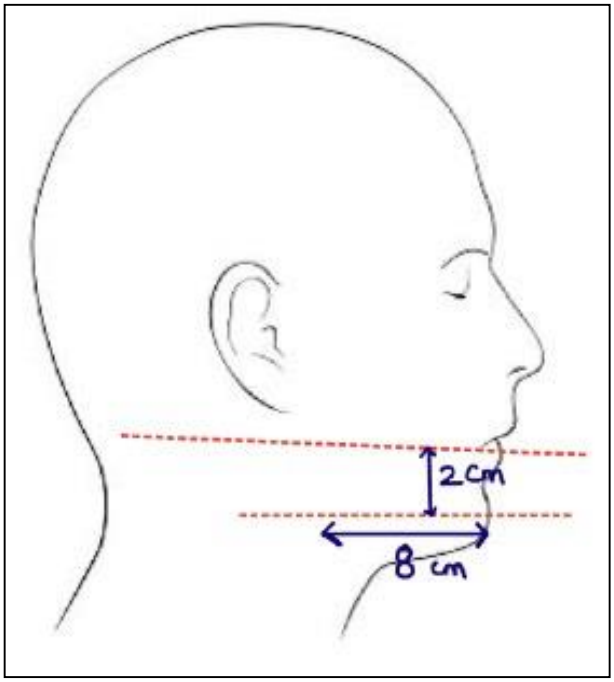

Figure 1: Lesion extensions on one side of face.

Figure 2: OPG showing radiolucency across the body of mandible.

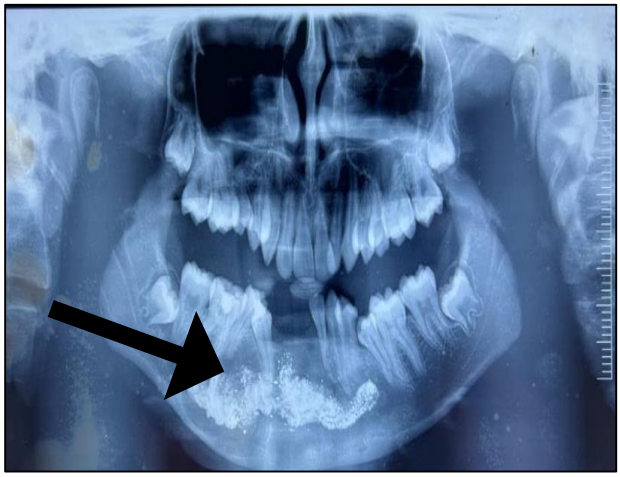

Figure 3: OPG showing extraction of mandibular incisor and canines with iodoform gauze (arrow).

Figure 4: Extracted molars with associated cystic lining.

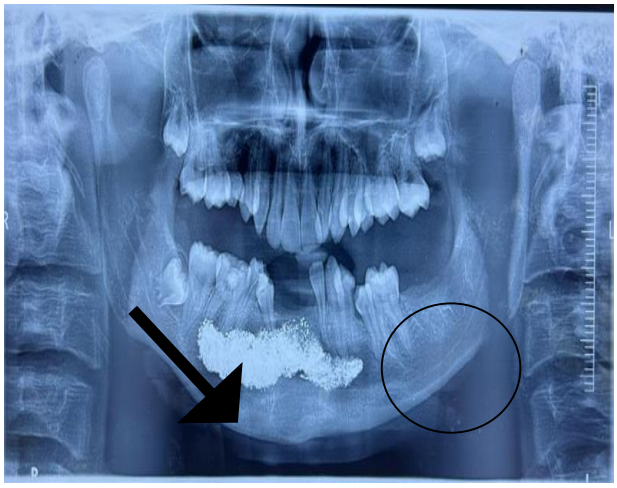

Figure 5: OPG showing extraction of left mandibular molars(circle) with some bone formation(arrow).

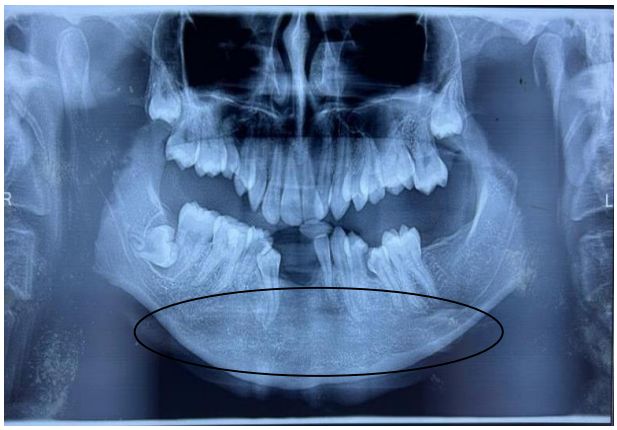

Figure 6: OPG showing presence of new bone.

DISCUSSION

The first mention of 'odontogenic keratocyst' can be traced back to 1956 in a paper by Philipsen.[1] He described it as any jaw cyst in which keratin was formed excessively. It was since that all such jaw cysts were classified as OKC, whether it be parakeratinized or ortho keratinized. It was much later after extensive clinical research, realised that there are two above mentioned variants that exhibit differences in behaviour, recurrence, association with nevoid basal cell carcinoma syndrome (NBCCS), histological characteristics and chemical markers. So, in 2005 the WHO classified the parakeratinized variant as 'keratocystic odontogenic tumour'.

Occurrence of OKC is a rare phenomenon in itself, over and above its bilateral occurrence is even rarer as can be seen in the article by Pimpalkar et al.[9] They are believed to arise from the odontogenic epithelium as the dental lamina and its remnants after the enamel organ has served its purpose.[10] But the presence of epithelial lining in the mucosa over the OKC in the third molar region points that there may be a role of basal cell layer of epithelium as well.[11-13] While choosing the appropriate management and

treatment protocol it is important to remember that OKCs have high recurrence rates. According to various studies this can go as high as 62.5% with the majority occurring within 5 years of start of initial treatment.[14,17,18] There has various studies to establish a link between factors and risk of recurrence, some studies consider multilocular lesions and daughter cysts to have high risk of recurrence.[19]

Due to such high recurrence rates, it is important to choose effective management protocols. This can range from conservative surgical approaches like- enucleation, marsupialization, and curettage; to aggressive treatments like- segmental and marginal resection, and hemi-mandibulectomy. Conservative surgical approaches have their advantages such as being more cosmetically appealing by preserving the bony and dentate structures as was favoured by Meiselman et al.[20] Also this allows to evade the morbidity associated with a mandibulectomy such as adjunct neurological defects as mentioned by Eyre et al.[21]

The first treatment that was recommended for such cysts was decompression followed by marsupialization and termed as Partsch I procedure given in the 19th century; while Partsch II procedure was enucleation with primary closure.[22] The conservative surgical approach follows the concept of decompression as explained by Pogrel 'any technique that relieves the pressure within the odontogenic cyst, thus restricting its growth'.[23] This is achieved during marsupialization by creating a window in the cyst thereby releasing the osmotic pressure within and allowing a reduced release of bone absorbing factors such as prostaglandins, interleukins and other growth factors.

In case of maxillary cysts this window can open into the nose, maxillary sinus or the oral cavity while for mandibular cysts, intraoral opening is the sole option. As a big window is created in the bony cyst with the cystic lining inside being sutured to outer normal mucosa, the need for placing a drain to keep the patency is redundant. The defect created in the form of window can be later covered by mobilisation of mucosal flaps. There have been opponents of marsupialization technique for the treatment of OKCs. Their reasoning has been that due to the leaving behind of cystic lining there are high chances of recurrence owing to epithelial proliferation.[24,25] On the other hand, there have been a vast number of studies showing that marsupialization can achieve complete resolution with no recurrences, it may be even the optimal approach.[23,26]

In a study done by Pogrel et al and Jordan et al it was reported that following marsupialization the epithelial lining appeared to be thicker than the initial 5-6 cells layer thick and resembled normal oral mucosa thus making it easier to enucleate.[23] Also on immunohistochemical examination initial biopsy reported high levels of anti-apoptotic bcl-2 markers which was not observed in the histology of tissue after marsupialization. In another study, they used ELISA to measure expression of interleukin-1(IL-1) alpha in the epithelial lining of the keratocyst; they reported that after marsupialization, the expression of IL-1 alpha was significantly reduced thereby inhibiting epithelial proliferation.[27]

This case underwent treatment for a period of two years with a follow up period of another two years where no recurrence was observed.

CONCLUSION

After doing an extensive search, there have been no cases reported with such large unilocular cyst involving the mandible ramus-to-ramus that were treated by enucleation following marsupialization under local anaesthesia. The patient's healing was uneventful and the follow up OPG showed good healing of the cyst with proper formation of bone. Therefore, we recommend that Thus, there is a need for more extensive study on the use of marsupialization technique for larger odontogenic keratocyst under local anaesthesia thereby reducing the need for radical surgeries.

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