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Odontogenickeratocyst Clinically Mimicking Osteomyelitis – A Rare Case Report

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Abstract:- Odontogenickeratocyst is infrequent and benign ,but locally aggressive neoplasm. Highly varied clinical presentation makes its clinical diagnosis difficult. This is a case report of rare presentation of odontogenickeratocyst with large extra oral swelling and multiple sinus openings which resemble osteomyelitis clinically. Timely diagnosis and accurate treatment plan is mandatory for better prognosis of patient and reduces the risk of recurrence.

Keywords:- *odontogenickeratocyst, osteomyelitis, CBCT, Recurrence.*

I. INTRODUCTION

In 2017,world health organisation[WHO] reassessed classification of tumors of head and neck and notions on the clinical, radiological and biological features of keratocysticodontogenictumor and retitled it again as odontogenickeratocyst[OKC].[1]Since 1950,the term odontogenickeratocyst was first used to define all the odontogenic cyst which contain keratin. In 2005 it was classified as neoplasm and termed it as keratocysticodontogenictumordue to its destructive behaviour, high recurrence rate, association with nevoid basal cell carcinoma and mutations in the patched (PTCH) tumor-suppressor gene. In 2017,WHO reclassified it as odontogenickeratocyst since evidence from many paper showed that PTCH mutation could also be found in other non -neoplastic lesions[2].

OKC usually grow within medullary cavities of bone without obvious bone expansion [3].Initial lesions are asymptomatic. Local swelling occurs when cyst perforates thinner region of cortical bone[4].Multiple sinus openings are rare in OKC. Here is a rare case report of OKC with large extra oral swelling and multiple sinus opening which resemble osteomyelit is clinically. Accurate diagnosis is essential because it leads to successful therapy, which improves patients' prognosis and quality of life.

II. CASEREPORT

A 39-year-old male patient reported with the complaint of pus discharge from the chin region since 9 days. He gave a history of trauma 15 years back to the chin, after which he suffered from mild, intermittent pain. Occasional pus discharge was seen in the lower anterior teeth and in the chin region since one year which was continuously seen since 9 days. No contributory medical history was reported.

On extra-oral examination, there was a mild facial asymmetry with diffuse swelling seen on the right lower third of the face, measuring approximately 5x5 cm in size, extending antero-posteriorly from 1 cm left to the midline, crossing the midline upto the right posterior border of ramus and supero-inferiorly from the commissure of the right lip to 2 cm below the lower border of the mandible. Surface of the lesion had a sinus opening with crusted appearance below the chin region, with pus discharge.[figure 1] It was tender on palpation with little pus exudation on compression, bonyhard in consistency with local rise in temperature. Bilateralsinglesub-mandibular lymph nodes werepalpable, measuring 1x1 cm in size, non-tender, firm in consistency, and not fixed to underlying structures.

On intra-oral examination, gingiva was soft, erythematous, and edematous in the lower anterior region with multiple sinus openings[figure 2].It was tenderon palpation with little pus exudation on compression in the region of 43. Vestibular obliteration present from the region of 33, crossing the midline till 46, suggestive of buccal cortical plate expansion.

Electric pulp testing was done which elicited delayed response to no response from teeth 33-48.Based on the history of trauma 15 years back to the chin and the clinical findings we arrived at the provisional diagnosis of Chronic suppurative osteomyelitis with extra-oral draining sinus irt lower anterior teeth.Differential diagnosis given was Chronicperi-apical abscess with extra-oral draining sinus i.r.t.43.

On aspiration, a straw coloured fluid along with bloody discharge was obtained Mandibular occlusal radiograph was done to evaluate the cortical plate expansion which revealedbucco-lingual cortical plate expansion[figure 3]

OPG revealed well defined, multi-locular radiolucency with sclerotic borders from the distal aspect of 33,crossing the midline to the ramus of the mandible on the right side, 1cm below the coronoid notch. Superiorly running between the roots of the mandibular teeth and inferiorly to the lower border of mandible. Well defined radiolucency present at the apical region of 45 and 46 suggestive of bone windowing. Inferior alveolar canal is moved inferiorly by the lesion and distal migration of the roots present i.r.t.-31 and 33[figure 4]

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CBCT images confirmed the bucco lingual expansion of the lesion. The size and extent of the lesion was measured. The dimension of the cyst was approximately superoinferiorly16.5 mm, anteroposteriorly 23.1mm, mediolaterally 65.6 mm.[figure 5,6,7]

Biopsy was taken which showed that the epithelium is stratified squamous parakeratinised ,having tall columnar basal cells in a tome stone pattern. The suprabasal cells showed inconsistent thickness in different areas and range from cuboidal to squamous cell. Few calcified areas were seen in the connective tissue .some proliferating and some resting odontogenic islands were seen focally. All the features were suggestive of Parakeratoticodontogenickeratocyst. Final diagnosis given was infected odontogenickeratocyst of the mandible.

Since the lesion was huge extending to ramus of mandible posteriorly and inferior border of mandible inferiorly there was poor prognosis and marsupialisation of the cyst was performed. Patient was recalled periodically for review.

III. DISCUSSION

OKC is a infrequent and benign but locally aggressive developmental cystic neoplasm.OKC constitutes of 7.8% of all cyst of the jaw. The incidencevary from 4-16.5%. It occurs at all ages with the peak incidence in 2nd and 4th decade oflife[4]. The most common location is in the mandible in the angle- ramus region. In the present case, the swelling crossed the midline and extended to the ramus region.

The common symptoms presented are pain ,swelling ,discharge, infection. Multiple sinus openings are rare in OKC which was seen in our case. Highly varied clinical presentation of OKC makes its clinical diagnosis difficult.

Commonly OKC appear as oval shaped uniformly radiolucent lesion extending along the body of the mandible with little mediolateral expansion with well defined and corticated margin. Important characteristic of OKC is its propensity to grow through the bone without bone expansion . This tunnelling type of growth pattern with minimal expansion occur throughout the body of mandible except ramus and coronoid process[5].In many occurrences, patients are surprisingly asymptomatic until the cyst have reached large size involving ascending ramus including condyle and coronoid process .Similar pattern was observed even in our case too.

Knowledge about the extent of lesion and the amount of destruction of bone is mandatory before surgery. CBCT provides a 3 dimensional information about the extent of lesion which makes the work of surgeons easier and presurgical assessment efficient.[6]

Histopathological assessment by means of an incisional biopsy is the best way to diagnose the OKC before the surgery, when suspicion has arisen from the clinical and radiographic presentation. Enucleation, marsupialization, resection are the main treatment modalities in OKC. In the present case, marsupialization was the treatment plan.

12% to 58.3% is the recurrence rate of OKC. Incomplete removal of primary lesion with thin epitheliallining ,epithelial remnants and satellite cyst can lead to recurrence. So regular periodic follow up and radiographic examination is essential to identify lesion before symptoms occur. Periodic follow up are mandatory torule out recurrence[7]

IV. CONCLUSION

OKC appears with varied clinical presentation which results in misdiagnosis. Timely diagnosis and accurate treatment planning is essential. Advanced diagnostic modalities like CBCT aids in better treatment planning which helps in the better prognosis of the patient and reduces the risk of recurrence.

Fig. 1: Diffuse swelling seen on the right lower third of the face

Fig. 2: Multiple sinus openings in lower anterior region

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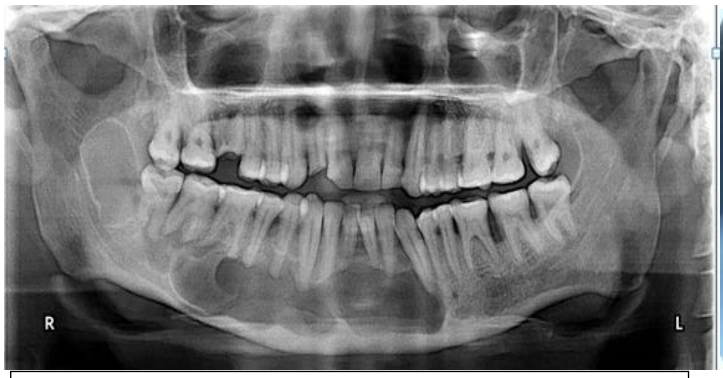

Fig. 4: OPG revealed well defined, multi-locular radiolucencv with sclerotic borders

Fig. 3: Occlusal radiographrevealed bucco-lingual cortical plate expansion

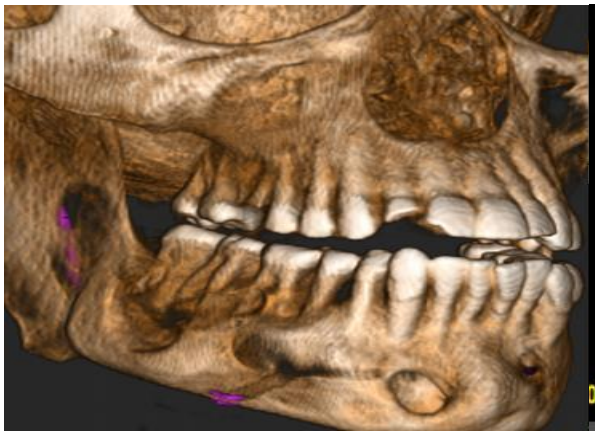

Fig. 5: 3D view of CBCT

Fig. 6: Coronal view of CBCT

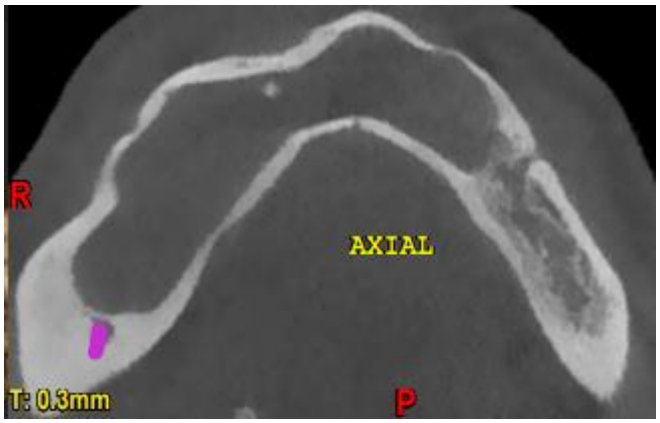

Fig. 7: Axial view of CBCT revealed the extent of lesion

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