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Case Study

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ORTHOKERATINIZED ODONTOGENIC KERATOCYST: A CASE OF UNCOMMON HISTOLOGY IN A COMMON LOCATION

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ABSTRACT

Background: Orthokeratinized odontogenic cyst (OOC) is a rare developmental cyst of odontogenic origin, considered a distinct entity from the more common parakeratinized odontogenic keratocyst. It presents predominantly in young adult males and typically affects the posterior mandible. **Case Presentation:** A 19-year-old male presented with a swelling in the lower left posterior region of the jaw present for one year. Clinical examination revealed a solitary, non-tender swelling in the mandibular left buccal vestibule. Radiographs showed a unilocular radiolucency with haziness, root resorption of 36, and distal displacement of 37. Surgical excision with extraction of 36 and 37 was performed. Histopathological analysis revealed orthokeratinized epithelium with a granular cell layer and daughter cysts, confirming the diagnosis of orthokeratinized odontogenic keratocyst. **Conclusion:** This case highlights the importance of correlating clinical, radiographic, and histopathological findings in diagnosing OOC. Due to its lower recurrence rate and less aggressive nature compared to parakeratinized variants, accurate identification is essential for appropriate management and long-term follow-up.

KEYWORDS: Posterior mandible, Unilocular radiolucency, haziness, Orthokeratinized epithelium.

INTRODUCTION

Odontogenic keratocyst (OKC), first described by Philipsen in 1956, is a developmental cyst arising from remnants of the dental lamina.[1] It accounts for approximately 10–15% of all odontogenic cysts and commonly affects the posterior mandible, particularly the angle and ramus region.[2,3] OKCs are notable for their potential for local aggressiveness, tendency for recurrence, and capacity to grow extensively along the medullary cavity with minimal buccolingual expansion.[4]

Two histological variants are recognized: parakeratinized and orthokeratinized. The orthokeratinized odontogenic cyst (OOC) is less common and considered less aggressive, with a lower recurrence rate and minimal association with nevoid basal cell carcinoma syndrome.[5] Histologically, OOCs exhibit an orthokeratinized stratified squamous epithelial lining with a prominent granular cell layer and lack the palisading basal cells characteristic of the parakeratinized variant.[6]

This case report describes an orthokeratinized odontogenic keratocyst in a 19-year-old male, emphasizing its clinical presentation, radiographic characteristics, and histopathological features.

CASE REPORT

A 19-year-old male reported with a complaint of swelling in the lower left back region of the jaw for the past one year. The patient was apparently normal until he noticed a swelling in the left mandibular posterior region. The swelling was small (peanut sized) and did not increase in size. There was no history of pain, trauma, pus discharge (intraoral or extraoral), fever, cough, or mobility of teeth. On extraoral examination, a mild, solitary, oval, diffused swelling measuring approximately 2×1 cm was seen on the left lower border of the mandible. The overlying skin appeared normal, and there was no draining sinus. On palpation, the swelling was firm to hard in consistency, non-tender, with normal surface temperature, and there was no paraesthesia of the lip. Intraoral examination **(Figure 1)** revealed a solitary, diffuse, dome-shaped swelling in the left posterior buccal vestibule, extending from the mesial

aspect of tooth 36 to the mesial aspect of tooth 37, measuring approximately 1.5×1 cm. The buccal vestibule was obliterated, the overlying mucosa appeared normal, and on palpation the swelling was soft to firm and non-tender, with no mobility in teeth 36 and 37. The provisional diagnosis of fibrous dysplasia wr t 36,37 was made considering the age and location with differential diagnosis of Odontogenic keratocyst. IOPA **(Figure 2)** and Orthopantamogram **(Figure 3)** showed a well-defined unilocular radiolucency (2×2.5 cm) with haziness, smooth borders. Epicentre is above the mandibular canal. Drifted 37 is seen distally. Distal root resorption and diversion of root with 36. A mandibular occlusal radiograph **(Figure 4)** showed a radiolucency buccally and lingually from the mesial aspect of 37 up to the distal aspect of 37, suggestive of minimal bony expansion with sclerotic borders, with buccal expansion greater than lingual. Drifting of 37 was also noted. Distal root resorption and diversion of the root of 36 were noted. Radiodiagnosis of Odontogenic keratocyst wr t 36-37 with differential diagnosis of Lateral Periodontal Cyst w r t 36,37. Based on these findings, an excisional biopsy **(Figure 5)** of the lesion along with extraction of teeth 36 and 37**(Figure 6)** was performed. Histopathological examination **(Figure 7)** showed orthokeratinized stratified squamous epithelial lining, 8–10 layers thick, exhibiting a classical tombstone appearance. The cystic capsule revealed inflammatory cell infiltration and vascular supply, along with the presence of daughter cysts, suggesting aggressiveness. These findings confirmed the diagnosis of an orthokeratinized odontogenic keratocyst.Patient is recalled after 15 days for follow up.

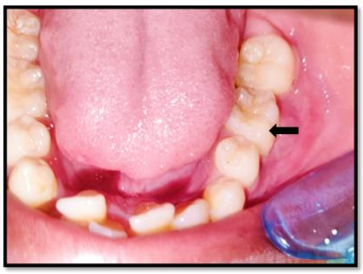

Fig. 1: Intraoral swelling seen on buccal aspect w r t 36, 37.

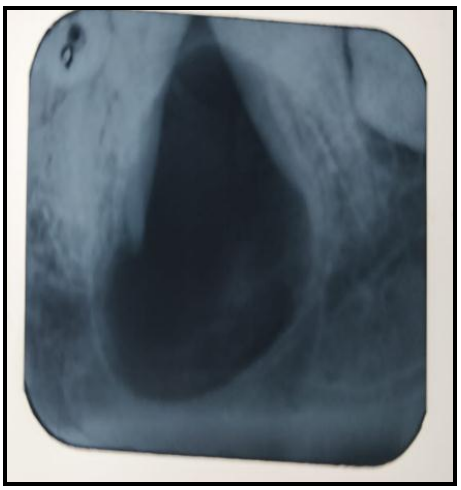

Fig. 2. IOPA w r t 36, 37 showing odontogenic keratocyst.

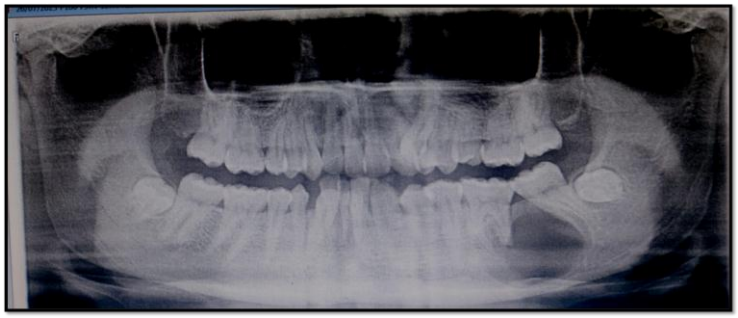

Fig. 3: OPG showing odontogenic keratocyst w r t 36, 37.

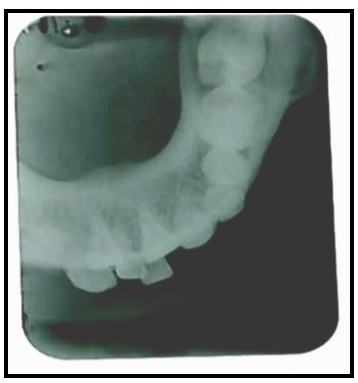

Fig. 4: Mandibular Occlusal radiograph showing minimal bony expansion w r t 37.

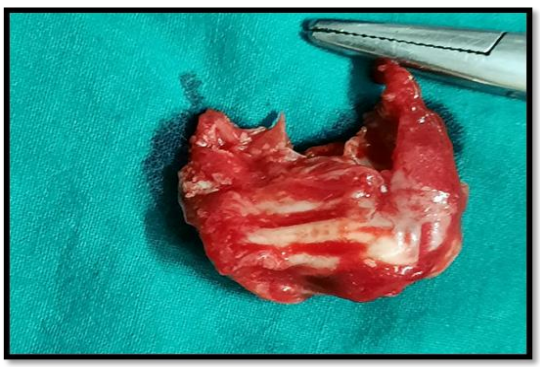

Fig. 5: Excisional biopsy of the lesion.

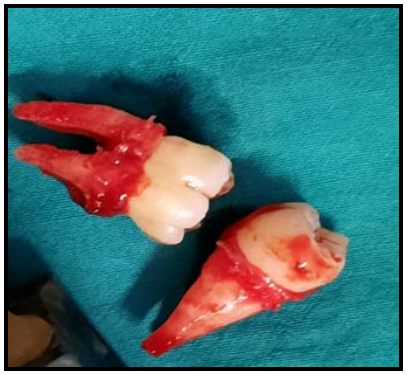

Fig. 6: Extraction with 36, 37.

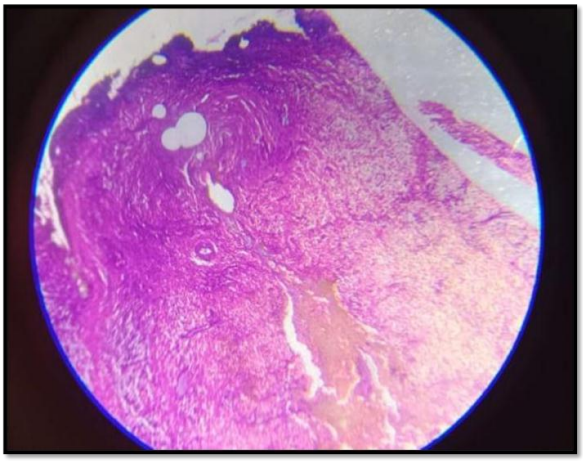

Fig. 7: Histopathological slide showing the features of Orthokeratinized odontogenic keratocyst.

DISCUSSION

Orthokeratinized odontogenic cyst (OOC) is a rare developmental odontogenic cyst considered a distinct entity from the parakeratinized variant of OKC. Although OKC was briefly reclassified as a keratocystic odontogenic tumor (KCOT) by the WHO in 2005 due to its neoplastic potential, the term OKC was reinstated in 2017 owing to lack of conclusive evidence supporting neoplastic behavior in all cases.[7]

Clinically, OOC occurs more commonly in males and typically presents in the second to third decades of life, often as an incidental radiographic finding or a slowly enlarging, painless swelling of the posterior mandible.[8,9] The present case aligns with this typical demographic and location. Radiographically, OOCs are often unilocular with well-defined margins, and may exhibit tooth displacement or root resorption in larger lesions. In the present case, the panoramic radiograph revealed a unilocular radiolucency with haziness, distal drift of 37, and root resorption of 36, which are in agreement with literature reports on larger orthokeratinized cysts.[10]

Histologically, OOCs demonstrate a thin epithelial lining composed of orthokeratinized stratified squamous epithelium, a well-defined granular layer, and non-palisaded basal cells. The presence of daughter cysts, as noted in this case, although more typical of parakeratinized variants, may reflect more extensive epithelial proliferation and warrants careful surgical planning.[11,12]

The standard treatment for OOC involves conservative surgical enucleation with or without extraction of involved teeth. Unlike the parakeratinized variant, which shows recurrence rates as high as 30%, OOCs have recurrence rates as low as 2–4%.[13] Complete excision, as done in the present case, is typically curative, and long-term prognosis is favorable. Follow-up remains essential due to occasional reports of recurrence, particularly in large or multilocular cases.[14]

CONCLUSION

Orthokeratinized odontogenic cyst is a less aggressive but clinically significant variant of the odontogenic keratocyst. Its presentation may mimic other cystic jaw lesions; therefore, thorough clinical and radiographic evaluation, followed by histopathological confirmation, is critical for diagnosis. Surgical enucleation remains the treatment of choice, with recurrence being uncommon. This case underscores the need for awareness of OOC's distinctive features to avoid misdiagnosis and overtreatment.

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