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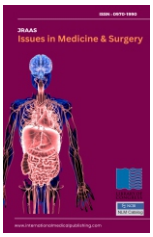

Case Report

Section: Radiodiagnosis

Odontogenic Keratocyst of Mandible in 20 Years Old Male Patient

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HIGHLIGHTS

- Twenty-year-old male with jaw swelling.
- Radiolucent lesion found in mandible.
- Histopathology confirms odontogenic keratocyst.
- Lesion exhibits aggressive local behavior.
- Surgical enucleation planned with follow-up.

Key Words:
Odontogenic keratocyst
Mandible
CT imaging
Enucleation
Recurrence

ABSTRACT

Odontogenic keratocyst (OKC) is a benign yet potentially aggressive cystic lesion of odontogenic origin, commonly affecting the posterior mandible in young adults. It is characterized by a high recurrence rate and distinctive histopathological features. This case report presents a 20-year-old male who reported right mandibular pain and swelling following a road traffic accident. Non-contrast computed tomography (NCCT) revealed a large, well-defined multiloculated radiolucent lesion extending to the condylar base with cortical expansion but no soft tissue invasion. Ultrasonography confirmed a hypoechoic cystic lesion, and fine-needle aspiration cytology was inconclusive. Histopathological examination revealed parakeratinized stratified squamous epithelium with palisading basal cells, consistent with OKC. Surgical management involved enucleation, peripheral ostectomy, and application of Carnoy's solution. The procedure was completed under general anesthesia via an intraoral approach, with care taken to preserve vital structures, including the inferior alveolar nerve. Postoperative recovery was uneventful, with resolution of symptoms within a week, no infection or nerve injury, and normal masticatory function. Follow-up radiographs at one and three months showed progressive bone healing, and no recurrence was noted at six months. This case emphasizes the importance of detailed imaging, histopathological correlation, and a multidisciplinary surgical approach in successfully managing OKC. The use of adjunctive chemical cauterization and vigilant follow-up contributed to the favorable short-term outcome. The findings support evidence in the literature regarding individualized surgical planning and long-term monitoring to prevent recurrence and ensure optimal recovery in young patients.

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INTRODUCTION

Odontogenic keratocyst (OKC) is a locally aggressive developmental cyst of odontogenic origin, arising from the remnants of the dental lamina, particularly the epithelial rests of Serres. First described in 1956 by Hans Philipson, OKC has long intrigued clinicians and pathologists due to its high recurrence rate, potential for significant local destruction, and characteristic histological features. While initially classified as a cyst, the lesion was reclassified in 2005 by the World Health Organization (WHO) as a keratocystic odontogenic tumor (KCOT) in recognition of its neoplasm-like behavior. However, subsequent genetic and clinical studies revealed that not all cases exhibit consistent tumorigenic properties, leading to its reclassification as a cyst once again in the 2017 WHO classification of odontogenic lesions [1, 2].

OKCs represent approximately 10% to 14% of all odontogenic cysts, with a strong predilection for the mandible, especially the posterior region involving the angle and ramus. They are most commonly diagnosed in patients between the second and fourth decades of life and show a slight male predominance. Although many OKCs are solitary and sporadic, multiple lesions are frequently observed in patients with nevoid basal cell carcinoma syndrome (Gorlin-Goltz syndrome), an autosomal dominant condition caused by mutations in the PTCH1 gene on chromosome 9. In such syndromic cases, OKCs tend to appear at a younger age and may be accompanied by cutaneous basal cell carcinomas, skeletal anomalies such as bifid ribs, and intracranial calcifications [3, 4].

Clinically, OKCs often remain asymptomatic for long periods and are frequently discovered incidentally during routine radiographic evaluations. When symptoms do occur, they may include painless jaw swelling, facial asymmetry, mild discomfort, or signs of secondary infection such as discharge. In advanced cases, lesions may cause tooth displacement, cortical plate thinning, or even pathological fractures. Radiologically, OKCs present as well-defined radiolucent lesions that may appear unilocular in small lesions and multilocular in larger ones, often with scalloped or smooth borders. One distinguishing radiographic feature is their tendency to expand extensively in the anteroposterior direction without marked buccolingual expansion [1, 5].

Histologically, the cyst wall of OKC is lined by a uniform layer of parakeratinized stratified squamous epithelium that is typically 6–10 cell layers thick. The surface is corrugated, and the basal layer displays palisaded, hyperchromatic nuclei. The fibrous connective tissue capsule is generally thin and non-inflamed unless infected. One of the key reasons for the high recurrence rate of OKCs is the frequent presence of satellite cysts or daughter cysts and epithelial remnants within the fibrous wall. These microscopic remnants can remain after . surgical excision and later give rise to recurrent lesions '[1, 6].

Treatment options for OKC vary depending on the lesion's size, location, and recurrence status. For small, well-circumscribed lesions, enucleation with or without peripheral ostectomy is the preferred approach. Application of chemical fixatives like Carnoy's solution may be used to reduce recurrence. In larger lesions, marsupialization or decompression is often performed initially to reduce cyst size and preserve surrounding structures before final enucleation. In recurrent or aggressive cases, especially those associated with syndromic presentations, resection may be warranted. Regardless of the treatment modality, long-term follow-up is essential, as recurrence can occur even after several years. Regular clinical and radiographic monitoring over a 5 to 10-year period is recommended to ensure early detection of recurrence. Early diagnosis and appropriate individualized treatment planning are critical to minimize morbidity and optimize outcomes in patients affected by odontogenic keratocyst of the mandible -[7, 8].

REVIEW OF LITERATURE

Reported a rare case of an odontogenic keratocyst (OKC) demonstrating transformation into a unicystic ameloblastoma in a young patient. The lesion was large and aggressive, extending from molar to molar within the mandible. OKCs are known for their aggressive and recurrent nature, often mimicking ameloblastomas in terms of location, age of presentation, and multilocular radiographic features, which complicates diagnosis. While combined lesions are documented, such transformations are exceedingly uncommon. This case highlighted the importance of long-term follow-up, meticulous surgical planning, and comprehensive rehabilitation to monitor recurrence and restore optimal quality of life [9].

Rported a case of a 13-year-old with a large odontogenic keratocyst (OKC) involving the left mandibular second and third molars. Decompression was performed to preserve the second molar and the inferior alveolar nerve, while the third molar was extracted. Over ten months, the cyst reduced significantly in size, allowing the second molar to erupt spontaneously. The residual cyst was later treated with enucleation. At 18 months follow-up, the second molar had fully erupted to the occlusal plane and was functioning normally, with no evidence of recurrence observed throughout the follow-up period [10].

Described a case involving a 37-year-old Indian male who presented with throbbing pain in a lower posterior tooth requiring endodontic therapy and a prior history of surgically treated odontogenic keratocyst (OKC) in the anterior jaw. Although OKC is typically benign, its aggressive behavior and resemblance to periapical lesions, especially in the anterior region, posed diagnostic challenges. Radiographic evaluation confirmed the diagnosis, and successful endodontic treatment was completed without affecting masticatory function. The case underscored the need for a multidisciplinary approach in OKC

management and demonstrated a favorable outcome with timely clinical and radiographic intervention [11].

Reported a rare case of a 31-year-old man with an isolated odontogenic keratocyst (OKC) measuring 13×12×6 mm at the base of the mandibular condyle. Although OKCs typically arise in the mandibular molar-ramus region and often remain asymptomatic until expansion, condylar involvement is uncommon and usually necessitates ramus resection. In this case, the lesion was excised under general anesthesia through anterior mandibular shaving, preserving the condylar head. The cavity was managed using a packed open technique and an obturator. At 20-month follow-up, there was no recurrence, demonstrating the effectiveness of conservative surgical management [12].

Described a case involving a 19-year-old male who presented with swelling in the posterior left mandible, initially presumed to be a dentigerous cyst based on clinical and radiographic findings. Surgical enucleation revealed a thick, firm cystic membrane, and histopathological examination confirmed an odontogenic keratocyst (OKC) with parakeratinized squamous epithelium and inflammation. OKCs are locally aggressive with high recurrence rates and often mimic other jaw cysts radiographically, making diagnosis challenging. The patient's postoperative recovery was uneventful, with no recurrence observed at six months. The case highlighted the importance of imaging, histology, and individualized treatment planning '[13].

Presented a case involving a 20-year-old male diagnosed with an odontogenic keratocyst (OKC) in the right mandible, notable for the rare presence of dystrophic calcification within the cystic wall. Since its original description in the twentieth century, OKC has remained a topic of debate due to its aggressive nature and uncertain optimal treatment. Dystrophic calcification is an uncommon histopathological feature in OKCs, with only a few reported cases. This report examined the possible etiopathogenic mechanisms behind such calcifications and contributed valuable insight into their clinical and diagnostic implications [14].

conducted a study in Central India to assess the prevalence, sex distribution, and treatment outcomes of odontogenic keratocysts (OKCs) by screening 2,900 patients across multiple centers. They identified 49 cases, with the highest incidence in the third and fourth decades-mean age 28 in males and 31 in females. The mandibular angle and ramus were the most commonly affected sites. Lesions were radiographically categorized as initial, moderate, or advanced, which aided in tailoring effective treatments with minimal recurrence. The study highlighted the importance of routine clinical, radiological, and histopathological evaluations for early diagnosis and reduced morbidity [15].

CASE DESCRIPTION

The patient, a 20-year-old male, reported no significant prior dental or medical issues before the current presentation. He had no history of pain, swelling, or dental extraction in the region of interest. There was no known history of trauma or systemic illness related to bone or soft tissue pathology. The patient was in generally good health, with no previous hospital admissions or surgeries. His dental history was unremarkable, with no history of impacted teeth or known jaw cysts. He was a non-smoker and non-alcoholic. There was no family history of jaw lesions or neoplasms. The chief complaint emerged following a road traffic accident (RTA), after which he developed headache, right lower jaw pain, and body aches. On clinical examination, localized tenderness and mild swelling in the right mandibular region were noted. This incident led to further imaging and diagnostic workup, which ultimately revealed an underlying jaw lesion unrelated to the trauma.

Initial imaging via non-contrast computed tomography (NCCT) of the head was performed due to the patient's RTA-related complaints. Thin-section axial, coronal, and sagittal bone window reconstructions revealed a large, well-demarcated radiolucent lesion in the posterior mandible on the right side. The lesion extended up to the base of the condylar process and displayed expansion of the cortical bone with focal erosion, though no breach of soft tissue was observed.

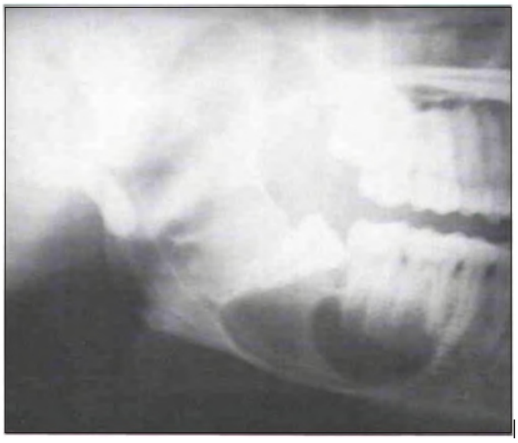

Figure 1: Lateral Oblique Radiograph Showing Radiolucent Lesion of Odontogenic Keratocyst in Posterior Mandible

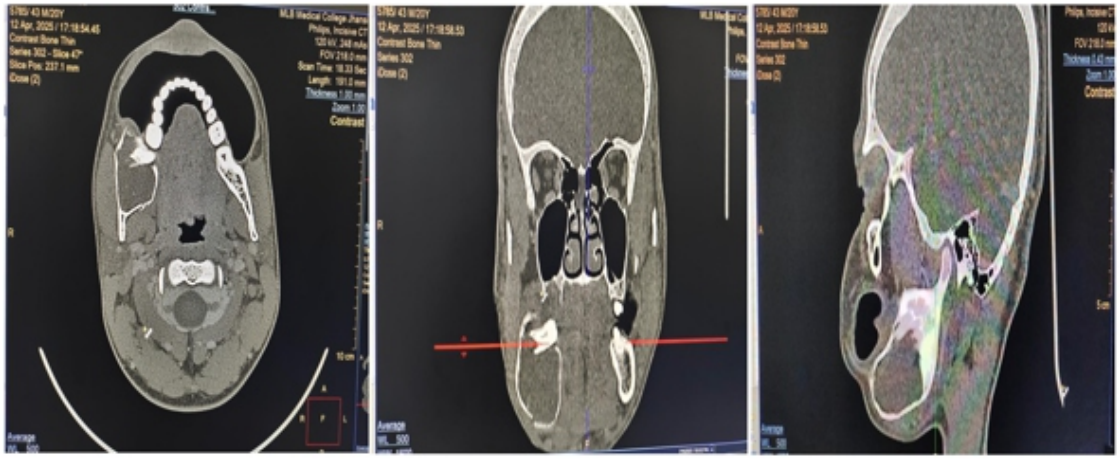

Figure 2: CTImaging Showing Odontogenic Keratocyst of Right Mandible (Axial, Coronal, and Sagittal Views)

The internal structure showed multiple thin septa, giving a multiloculated, soap-bubble-like appearance. Importantly, the lesion was not associated with dental caries or an unerupted tooth. No associated calcification or periosteal reaction was seen. Ultrasonography, though less informative for bony lesions, confirmed a hypoechoic cystic structure with no significant vascularity, supporting the impression of a non-inflammatory lesion. Fine-needle aspiration cytology (FNAC) was inconclusive, prompting a biopsy. Histopathology revealed features characteristic of odontogenic keratocyst-parakeratinized stratified squamous epithelium with a palisading basal cell layer and inflammation in the subepithelial connective tissue. No malignancy was detected.

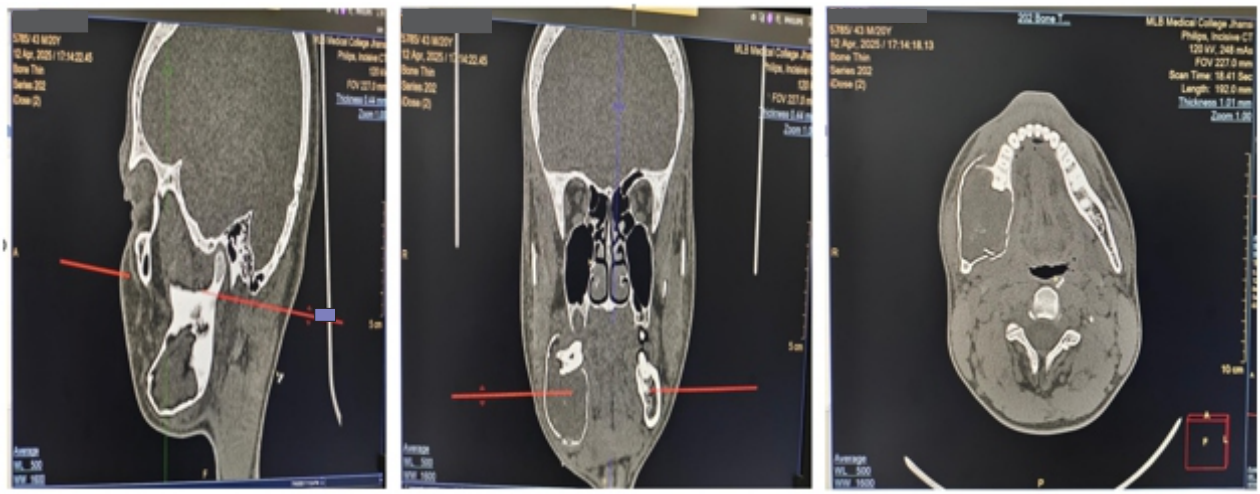

Figure 3: Additional NCCT Views Demonstrating Multiloculated Odontogenic Keratocyst in the Right Mandible

The patient underwent surgical enucleation of the odontogenic keratocyst under general anesthesia. A mucoperiosteal flap was raised via an intraoral approach to access the lesion. The cystic lining was meticulously dissected and completely removed from the surrounding bone to minimize recurrence. Peripheral ostectomy was performed to remove any residual epithelial remnants and satellite cysts commonly associated with OKCs. Care was taken to preserve vital structures, including the inferior alveolar nerve. Since the lesion had extended close to the base of the condyle, close intraoperative monitoring ensured complete excision without compromising mandibular function. Chemical cauterization using Carnoy's solution was applied to the bony cavity to reduce recurrence risk. The surgical site was closed with resorbable sutures, and the patient was prescribed postoperative antibiotics and analgesics. The excised tissue was submitted for histopathological confirmation. Follow-up included periodic clinical examinations and radiographs to monitor healing and detect early signs of recurrence.

The patient tolerated the surgical procedure well and had an uneventful postoperative recovery. Pain and swelling subsided within a week, and there were no signs of infection or nerve involvement. Follow-up radiographs at one- and three-months post-surgery showed progressive bone healing and no signs of residual or recurrent lesion. Mouth opening and masticatory function remained normal, with no temporomandibular joint dysfunction. The patient was advised regular long-term follow-up due to the high recurrence rate associated with odontogenic keratocysts. At six months, no recurrence had been observed, indicating a favorable short-term outcome following complete

surgical excision with adjunctive therapy.

DISCUSSION

Our finding shows that a 20-year-old healthy male, with no prior dental or systemic illness, presented with right mandibular pain and localized swelling following a road traffic accident. Imaging and clinical workup incidentally revealed a jaw lesion unrelated to trauma. This presentation closely parallels the observations of Razmara F et al. (2019), who analyzed three cases of mandibular traumatic bone cysts (TBC) in young individuals with a mean age of 20 years. One of their cases had a trauma history, while others were discovered incidentally, with imaging typically showing unilocular radiolucencies with scalloped margins. They emphasized the diagnostic challenge and need for clinical-radiographic correlation. Similarly, Thelekkat Y and Basheer SA (2022) reported a 19-year-old male with mild jaw pain and altered sensation, where orthopantomogram and CBCT revealed a well-defined unilocular radiolucency in the mandibular ramus, confirmed as TBC. Their study highlights the difficulty in distinguishing TBC from other aggressive jaw lesions, aligning with our case –[16,17].

Our finding shows that NCCT revealed a well-defined, multiloculated radiolucent lesion in the right posterior mandible extending to the condylar base, with cortical expansion and focal erosion but no soft tissue breach. The soap-bubble-like internal structure and absence of dental involvement suggested a non-inflammatory lesion. Ultrasonography confirmed a hypoechoic cystic area without vascularity, and FNAC was inconclusive. Histopathology confirmed odontogenic keratocyst (OKC). This aligns with Petchiammal SM et al. (2024), who reported detailed imaging features on CT/CBCT in OKC and highlighted the role of FNAC, enucleation, peripheral ostectomy, and Carnoy's solution in reducing recurrence, with FNAC showing high diagnostic yield. Though our FNAC was inconclusive, the imaging features and histopathology matched their findings. Strokov S et al. (2024) emphasized the importance of clinicoradiologic-pathologic correlation and noted features like multiloculated appearance and cortical expansion-findings consistent with our case-underlining their role in improving diagnostic accuracy and guiding surgical planning [18,19].

Our finding shows that the patient had an excellent postoperative recovery, with pain and swelling resolving within a week, no signs of infection or nerve involvement, and normal mouth opening and function maintained. Follow-up radiographs at one and three months confirmed progressive bone healing, and no recurrence was observed at six months, indicating a favorable short-term outcome. This aligns with Dioguardi M et al. (2024), who analyzed various OKC treatment strategies and found that factors such as multilocularity, young age, and large lesion size increase recurrence risk. However, treatments like enucleation with peripheral ostectomy and adjunctive therapies such as 5-fluorouracil demonstrated low recurrence rates, consistent with our case. Similarly, Stoelinga PJ et al. (2023) emphasized the need for standardized, long-term follow-up protocols due to the variability in reported recurrence rates and durations, supporting our approach of advising ongoing monitoring to ensure effective management and early detection of recurrence.

CONCLUSION

This case highlights the importance of early detection, accurate diagnosis, and comprehensive management in odontogenic keratocysts. The incidental finding of a multiloculated radiolucent lesion during post-trauma imaging led to a timely intervention that included enucleation, peripheral ostectomy, and Carnoy's solution application-resulting in a favorable outcome with no recurrence at six months. Correlation with imaging and histopathological features confirmed the diagnosis, guiding appropriate surgical planning. This case reinforces the value of vigilant long-term follow-up in preventing recurrence and underscores the necessity of individualized treatment strategies to optimize patient outcomes in mandibular OKCs.

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