

FROM INCIDENTAL FINDING TO DEFINITIVE DIAGNOSIS: MULTIMODAL RADIOGRAPHIC ASSESSMENT OF A PARAKERATINIZED ODONTOGENIC KERATOCYST OF THE MANDIBULAR RAMUS: A CASE REPORT

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ABSTRACT

Background: Odontogenic Keratocyst (OKC) is a developmental odontogenic cyst characterized by a distinctive parakeratinized epithelial lining and a tendency to grow extensively within the medullary spaces of the jaws. It demonstrates a predilection for the posterior mandible and ramus and may remain clinically silent despite considerable radiographic extension.

Case presentation: A 54-year-old male presented for routine dental evaluation, during which an asymptomatic multilocular radiolucency was incidentally detected in the right mandibular ramus. Panoramic, periapical and Donovan's radiographs demonstrated a well-defined corticated lesion with buccal cortical expansion associated with impacted 48. CBVI revealed cortical thinning and lingual breach. Histopathology confirmed Parakeratinized Odontogenic keratocyst.

Conclusion: This case emphasizes that OKC may remain asymptomatic and be discovered incidentally on routine radiographic examination. Careful interpretation of conventional radiographs supplemented by specialized intraoral projections such as Donovan's technique can provide useful information regarding lesion extent and cortical expansion. Histopathological examination remains essential for definitive diagnosis.

KEYWORDS: Odontogenic keratocyst; parakeratinized odontogenic keratocyst; mandibular ramus; Donovan's technique; multilocular radiolucency; cone-beam computed tomography

INTRODUCTION

Odontogenic keratocyst (OKC) is a developmental cystic lesion of odontogenic origin that is recognized by its characteristic epithelial lining and its capacity to extend within the cancellous bone of the jaws. Following its original description by Philipsen in 1956, OKC has remained of considerable clinical interest because of its characteristic growth behavior, potential for recurrence, and radiographic overlap with other intraosseous radiolucencies. In the fourth edition of the World Health Organization classification, the lesion was designated as a keratocystic Odontogenic tumor, largely on the basis of its proliferative characteristics and the reported involvement of alterations in the PTCH signaling pathway. Later evidence concerning its biological characteristics and clinical response to conservative management led to its reclassification as an Odontogenic cyst in the 2017 WHO classification. The 2022 WHO classification continues to recognize OKC as a distinct Odontogenic cystic lesion. [1–4]

Published epidemiological series generally place OKCs at around one-tenth of Odontogenic cysts, although the proportion varies among individual reports and reviews. Although OKC can occur across a broad age range, many epidemiological reports describe a greater frequency during the second to fourth decades. A clear mandibular predilection has been reported, with the posterior body, angle and ascending ramus representing commonly involved regions. [1, 5–7]

The clinical presentation is variable. Small lesions may produce no recognizable clinical signs and are therefore sometimes detected unexpectedly on radiographs taken for other dental indications with increasing size, patients can develop pain or swelling, and larger lesions may be accompanied by tooth displacement, cortical expansion or sensory changes. One notable feature of OKC is that it can spread along the antero-posterior axis of the jaw before producing conspicuous bucco-

lingual expansion. Further growth may progressively thin and bow the cortical plates outward, with cortical perforation becoming possible in more advanced lesions. [5, 8]

On conventional radiographs, OKC is usually recognized as a well-demarcated radiolucency, often bordered by a distinct corticated rim. Its internal appearance may be unilocular or multilocular, depending largely on lesion size and the presence of internal bony septations. When an unerupted tooth is involved, the imaging appearance may overlap with that of other Odontogenic lesions, particularly a Dentigerous cyst, Ameloblastoma or Odontogenic Myxoma. Involvement of the posterior mandible, including the ramus, represents a characteristic distribution of the lesion. [5,7,9]

Although panoramic radiography and cross-sectional imaging are routinely used in the assessment of posterior mandibular lesions, supplementary intraoral projections can provide additional information regarding the cortical boundaries and spatial extent of a lesion. Donovan's projection is a modified form of mandibular occlusal radiography developed to improve visualization of the posterior mandible, particularly the region surrounding the mandibular third molar. In this projection, the receptor is positioned to include the posterior mandibular region, while an obliquely directed X-ray beam is used to obtain additional visualization of this anatomically challenging area. This modified positioning helps overcome the limited representation of the posterior mandibular region that may occur with a standard mandibular occlusal projection. [10,11]

The present report describes an incidentally detected Parakeratinized OKC involving the right mandibular ramus in an asymptomatic 54-year-old male. Particular emphasis is placed on its radiographic presentation and the additional information obtained using Donovan's technique.

Case Report

A 54-year-old male patient reported to the Department of Oral Medicine and Radiology for a routine dental examination. His chief complaint was not related to the mandibular lesion, which was discovered incidentally during radiographic evaluation.

The patient stated that he had remained apparently healthy until approximately five months prior to presentation, when endodontic treatment of 44 and restoration of 46 had been performed at a private dental clinic. An orthopantomogram was advised as part of the postoperative evaluation, during which a radiolucent/cystic lesion was incidentally identified in the right posterior mandible. He did not report pain, swelling, discharge, or any other local symptoms attributable to the affected region.

The patient's medical history was significant for type II diabetes mellitus and hypertension of approximately four years' duration, for which he was receiving allopathic medication. His dental history included root canal treatment with respect to 44 and occlusal restoration with respect to 46. The patient reported a history of occasional smoking for approximately 30 years but had discontinued the habit one year before presentation.

Clinical examination

Extra-oral examination did not demonstrate gross facial asymmetry or an extra-oral draining sinus (Figure 1). Palpation of the facial region was non-tender, with no local rise in temperature. Regional lymph nodes were non-palpable, non-mobile and non-tender. Temporomandibular joint movements were smooth, with straight opening and closing movements. Intraoral examination revealed a generally preserved dentition with a prosthesis associated with 44 and an occlusal restoration associated with 46 (Figure 2). Teeth 27, 28, 38 and 48 were clinically absent.



Figure 1: Extra-oral examination



Figure 2: Intra-oral examination

Radiographic examination

Orthopantomogram

A panoramic radiograph was obtained using a Carestream CS 9600 unit at 73 kVp and 8 mA. The principal area of interest was the right ramus of the mandible.

The radiograph demonstrated a well-defined multilocular radiolucent lesion involving the right mandibular ramus, extending superiorly from the angle of the mandible toward the sigmoid notch. The lesion was associated with a horizontally impacted mandibular right third molar (48). The lesion exhibited a sharply demarcated outline with a thin, smooth and corticated peripheral margin. The internal aspect was predominantly radiolucent, with several curved bony septa creating a multilocular, soap-bubble-like configuration. No calcified components or discrete radiopaque structures were evident within the lesion (Figure 3).

The inferior alveolar canal appeared displaced inferiorly in relation to the lesion. Tooth 48 was horizontally impacted and displaced from its normal eruptive pathway. No significant root resorption of the adjacent mandibular second molar (47) was evident. Based on the radiographic appearance, an Odontogenic keratocyst was considered among the principal diagnostic possibilities, with Dentigerous cyst, Ameloblastoma, lateral periodontal cyst and Odontogenic Myxoma included in the differential diagnosis.



Figure 3: Panoramic radiograph showing a well-defined radiolucent image.

Intraoral Periapical radiograph

An intraoral periapical radiograph of the mandibular right posterior region demonstrated a well-defined round radiolucency extending distal to the impacted 48. The mesial aspect of 48 was seen impinging upon the cervical region of the distal aspect of 47. The occlusal restoration associated with 46 was also evident (Figure 4).



Figure 4: Intraoral Periapical radiograph showing radiolucency distal to impacted tooth 48

Donovan's technique

A mandibular oblique occlusal radiograph was subsequently obtained using Donovan's technique to further evaluate the posterior mandibular region.

On Donovan's projection, the lesion was visualized as a multilocular radiolucency occupying the right posterior mandibular region and extending superiorly toward the ramus. The lesion consisted of multiple radiolucent compartments separated by internal bony septa. Its margin was well-defined and well corticated. A notable finding on this projection was the outward bowing of the buccal cortical plate, accompanied by evident cortical thinning. The cortical boundary remained appreciable in this projection without definite evidence of perforation (Figure 5).



Figure 5: Donovan's technique showing buccal cortical expansion

Cone-beam computed tomography

Cone-beam volumetric imaging was performed using a Carestream CS 9600 system with a field of view of 12×10 cm at 120 kV and 5 mA. CBVI was performed to delineate the three-dimensional boundaries of the lesion and evaluate its proximity to the surrounding anatomical structures.

The panoramic reconstruction demonstrated a well-defined multilocular radiolucency involving the right mandibular ramus and extending superiorly from the angle toward the sigmoid notch and subcondylar region. The lesion measured approximately 28.0×17.7 mm and was associated with the horizontally impacted 48. A thin, smooth corticated margin surrounded the lesion. Several curved osseous septa were present within the radiolucency, resulting in a characteristic soap-bubble-like internal architecture. No internal calcifications or radiopaque foci were identified (Figure 6).

The inferior alveolar canal was displaced inferiorly. No significant root resorption involving 47 or 48 was observed. Axial sections demonstrated an approximate buccolingual dimension of 8.3×19.2 mm at the level of the ramus (Figure 7). Evaluation in the coronal plane showed attenuation of both cortical plates, with focal discontinuity of the lingual cortex indicating cortical perforation (figure 8).

Based on the imaging findings, an Odontogenic keratocyst involving 48 and the right mandibular ramus was considered the leading radiographic diagnosis. Ameloblastoma, Dentigerous cyst associated with 48 and Odontogenic Myxoma were considered as differential diagnoses.

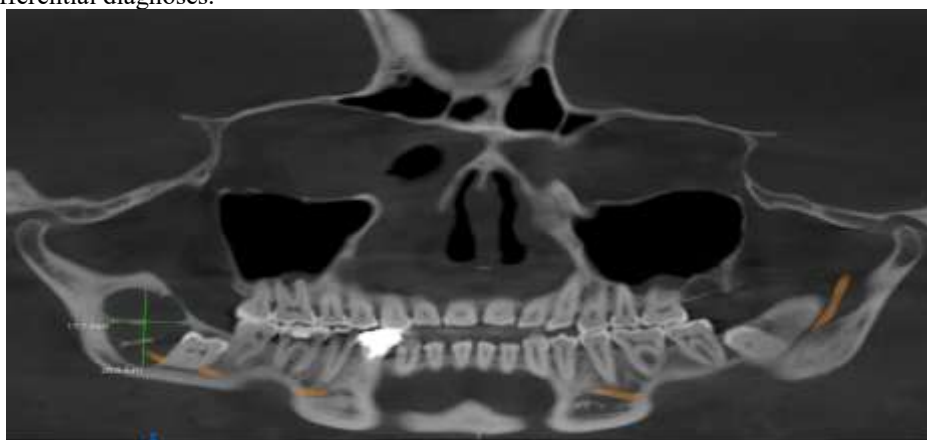


Figure 6: Panoramic view showing well defined lesion in the right ramus of the mandible



Figure 7: Axial view showing bucco-lingual dimension

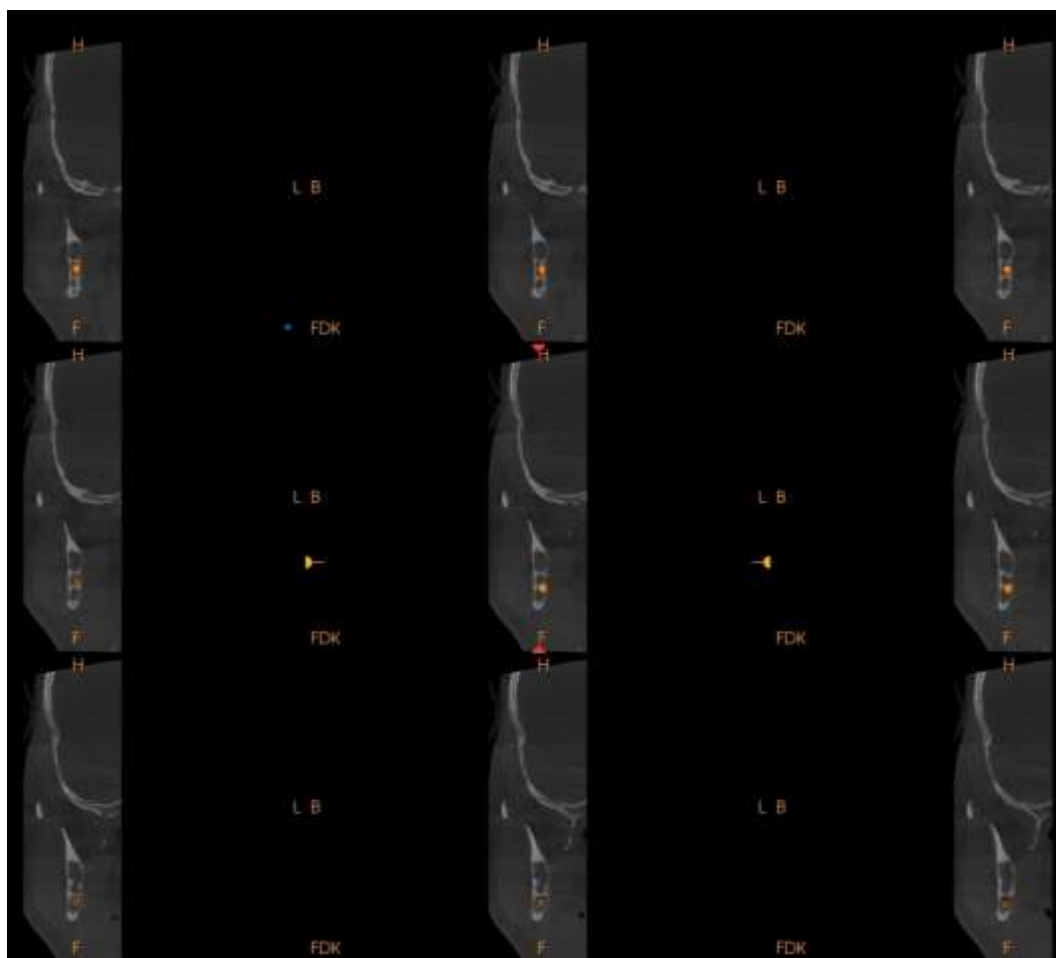


Figure 8: Coronal view showing involvement of inferior alveolar canal

An incisional biopsy was obtained from the cystic lesion involving the posterior mandible. Gross examination revealed a single greyish, irregular, soft cystic tissue fragment measuring approximately $0.5 \times 0.3 \times 0.3$ cm.

Histological evaluation showed a cystic lumen lined by approximately 6–8 cell layers of corrugated Parakeratinized stratified squamous epithelium. The basal epithelial layer displayed the characteristic nuclear palisading pattern. The cystic wall consisted predominantly of fibrous connective tissue with areas of moderate inflammatory cell infiltration. (Figure 9)

Taken together, the microscopic findings supported the diagnosis of a Parakeratinized Odontogenic keratocyst.

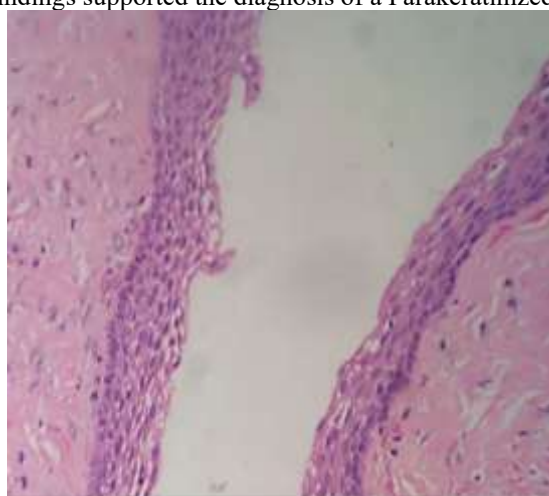


Figure 9: Histological examination showing a cystic lumen with 6–8 cell layers of corrugated Parakeratinized stratified squamous epithelium

Based on the clinical, radiographic and histopathological findings, a final diagnosis of Parakeratinized Odontogenic keratocyst involving the right mandibular ramus and associated with impacted tooth 48 was established.

Considering the location and extent of the lesion, a surgical treatment plan consisting of segmental mandibulectomy with en bloc resection of the lesion with appropriate margins and reconstruction using a free flap was proposed. Intermaxillary

fixation was planned to provide mandibular stability and facilitate healing. Oral prophylaxis and periodic clinical and radiographic follow-up were also planned.

DISCUSSION

Odontogenic keratocyst (OKC) warrants particular clinical attention because its behavior differs considerably from that of several other developmental Odontogenic cysts. Although histologically benign, an OKC may become extensive within the bone, eventually compromise the cortical boundaries, and show recurrence following treatment. Its clinical importance consequently stems not merely from its frequency, but also from the possibility of substantial progression in the absence of obvious clinical symptoms. [1,5,8]

Epidemiological studies have reported that OKC constitutes approximately 10% of Odontogenic cysts, whereas some reviews estimate its contribution to be approximately 10–20%. Although OKC may occur throughout life, epidemiological studies have frequently demonstrated greater occurrence during the second and third decades, with older patients also being affected. A slight male predominance has been observed in several large series. [1,5,6] At 54 years of age, the patient represents an older age group than that typically emphasized in epidemiological descriptions, further demonstrating that OKC is not restricted to younger individuals.

The mandible is affected considerably more often than the maxilla. Within the mandible, the posterior region, angle and ramus are recognized as characteristic sites. A systematic review and meta-analysis demonstrated a substantial representation of OKCs in the posterior region of the mandible, while more recent systematic evidence has similarly identified the mandibular ramus and posterior mandible among the most frequent locations. [5,7] The location in the present case is therefore highly compatible with the established distribution of OKC, with the lesion extending from the mandibular angle into the ramus.

The clinical course of OKC has several features that distinguish it from many other Odontogenic cystic lesions. Because early lesions may produce little or no clinical disturbance, some OKCs are first recognized on radiographs obtained for an unrelated dental problem. As the lesion enlarges, it may cause swelling, pain, tooth displacement, cortical expansion or sensory symptoms. The silent enlargement observed in some extensive lesions may reflect their ability to progress through cancellous bone while producing relatively limited cortical alteration during the earlier stages. [5,8] In the present case, the lesion was detected incidentally during postoperative radiographic assessment following endodontic treatment. The patient denied pain, swelling or discharge, emphasizing the importance of radiographic examination in detecting clinically silent lesions.

Radiographically, OKC can present as either a single-compartment or multiloculated radiolucency and commonly has a sharply defined, frequently corticated margin. Internal osseous septa may divide larger lesions into multiple compartments, giving the radiolucency a multilocular or soap-bubble appearance. The lesion may be associated with an impacted tooth and can resemble a Dentigerous cyst when it surrounds the crown. From a radiographic differential perspective, Ameloblastoma and Odontogenic myxoma are among the principal lesions that may demonstrate overlapping features. [5,9] In the present patient, these characteristic radiographic features were collectively evident, including a sharply corticated margin, multilocular architecture with curved septations, and intimate association with the horizontally positioned 48.

An interesting radiographic feature in this case was the inferior displacement of the inferior alveolar canal. Despite the relatively limited expansion that may accompany early OKC growth, progressive enlargement can result in displacement of neighbouring structures and compromise of the cortical boundaries. Cross-sectional imaging permits more precise evaluation of the lesion in relation to neighbouring anatomical structures and provides better visualization of the integrity of the buccal and lingual cortices. In this patient, CBVI revealed bilateral cortical thinning together with focal lingual cortical perforation, findings that were not fully appreciable on panoramic imaging.

Role of Donovan's technique

The use of Donovan's technique represents an important radiographic aspect of the present case. The modified occlusal projection attributed to Donovan was originally introduced to improve radiographic assessment of the mandibular third-molar region. In this projection, modification of receptor positioning and patient orientation facilitates visualization of the posterior mandibular region that may otherwise be incompletely represented. Donovan's original report, published in 1952, described its application in occlusal imaging of the mandibular third-molar area. [10]

The posterior extent of the mandible may be inadequately captured on a conventional mandibular occlusal projection because of the anatomical configuration of the ramus and surrounding soft tissues. Donovan's modification addresses this limitation by positioning the receptor toward the ascending ramus and directing the central beam obliquely. Although principally associated with third-molar imaging, the projection can also serve as an adjunctive conventional view when evaluating pathological changes involving the posterior mandible. [10,11]

In the present case, Donovan's projection demonstrated the lesion as a multilocular radiolucency with a well-defined, corticated margin and internal bony septations. More importantly, the projection provided appreciable evidence of buccal cortical expansion and thinning, with outward bowing of the cortical outline. Although CBVI provided the most comprehensive three-dimensional assessment, Donovan's projection complemented the cross-sectional findings by demonstrating the buccal cortical response on a conventional radiograph.

The radiographic findings in this case also demonstrate the difficulty of establishing a specific diagnosis based on imaging alone, as several Odontogenic lesions may produce overlapping features. The combination of multilocularity and cortical expansion can mimic Ameloblastoma, while the relationship of the lesion to the impacted 48 may raise consideration of a Dentigerous cyst. Odontogenic myxoma may also demonstrate multilocularity and internal trabeculation. Accordingly,

imaging findings should be correlated with the clinical presentation and confirmed, when required, by histopathological examination. [5,9]

Microscopically, the Parakeratinized variant typically shows a thin epithelial lining with a corrugated keratin surface and a palisaded basal cell layer. The biopsy specimen in the present case demonstrated these characteristic microscopic findings. The histopathological diagnosis therefore established the definitive diagnosis of Parakeratinized OKC.

Differentiation between the Parakeratinized and Orthokeratinized variants has clinical relevance, as the Parakeratinized type has traditionally been regarded as having a higher recurrence propensity. Recurrence has been attributed to several factors, including the delicate nature of the epithelial lining, residual lesion following treatment, satellite cyst formation and the characteristic growth pattern of OKC. Reported recurrence rates vary widely depending on treatment modality, follow-up duration and lesion characteristics. [8,12,13] Lesions that are larger, multilocular, associated with cortical perforation or located in the posterior mandible may present additional management challenges.

Management of an OKC should be tailored to the individual lesion, taking its size, site, extent and biological behavior into consideration. Depending on its extent and biological behavior, management may involve conservative procedures such as enucleation or marsupialization, while more extensive lesions may require adjunctive measures or surgical resection. Factors such as lesion dimensions, anatomical location, cortical involvement, extension into adjacent soft tissues, previous recurrence and the patient's general clinical status contribute to treatment selection. Current evidence indicates that no single treatment approach can be uniformly applied to all cases of OKC. [12–15]

In the present case, the lesion's ramus location, multilocular morphology, cortical expansion and lingual cortical breach were important considerations in planning definitive management. The proposed treatment was segmental mandibulectomy with en bloc removal and reconstruction, reflecting the concern regarding the extent and behavior of the lesion.

From an oral radiology standpoint, the case is particularly relevant because a relatively extensive lesion was identified in the absence of local symptoms. Sequential evaluation with panoramic and intraoral radiography, supplemented by Donovan's projection and CBVI, progressively clarified the lesion's extent and effects on adjacent structures, while histopathological examination provided diagnostic confirmation. It therefore reinforces the value of a systematic radiographic approach when an incidental posterior mandibular radiolucency is identified.

CONCLUSION

This case emphasizes the need to include OKC in the differential diagnosis of a well-demarcated posterior mandibular radiolucency, particularly when an impacted third molar is present. Despite its considerable radiographic extent, the lesion remained clinically silent and was identified incidentally. Donovan's projection provided supplementary information regarding buccal cortical expansion, whereas CBVI enabled assessment of the three-dimensional extent and cortical integrity. Histopathological examination ultimately confirmed the Parakeratinized nature of the lesion. A systematic combination of conventional and cross-sectional imaging, followed by histopathological correlation, is therefore valuable in the evaluation and definitive diagnosis of posterior mandibular radiolucencies.

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