Odontogenic keratocyst (OKC) is an uncommonly occurring jaw cyst that generally has a more aggressive nature and higher recurrence rate than other odontogenic and non-odontogenic cysts. Consequently, early diagnosis of the lesion, before it involves a larger portion of the bone, is important to reduce its likelihood of recurrence after therapy.

The radiologic appearance of OKC may vary from that of a small unilocular radiolucency to that of a large, expansile multilocular lucency. Most OKCs occur in the posterior regions of the jawbones. However, OKCs may also develop around the crowns of unerupted teeth, at the apices of teeth, between teeth, and in the midline of the maxilla. Thus, while OKCs are commonly mistaken for ameloblastoma, giant cell granuloma, and odontogenic myxoma, other lesions may be confused for OKC, such as dentigerous cyst, lateral periodontal cyst, radicular cyst, and nasopalatine duct cyst.

This report describes the case of an 82-year-old woman who presented with the chief complaint of a mild pain and gingival swelling of 2-months’ duration in the buccal aspect of her mandibular left premolar region. The condition was initially diagnosed as a periodontal abscess and was treated as such by the referring clinician. The initial periapical radiograph provided by the clinician demonstrated horizontal alveolar bone resorption associated with active periodontal disease, which appeared to be more severe between the 2 premolars adjacent to the swelling. After more detailed clinical evaluation of the area and examination of the adjacent teeth, including vitality testing, it was concluded that the condition was not related to dental or periodontal disease. Consequently, a biopsy specimen was taken, and the histopathologic examination of the lesion proved it to be an odontogenic keratocyst. The case is reported because of the unusual clinical and radiographic findings associated with this lesion. Early diagnosis and appropriate treatment of odontogenic keratocyst can reduce the possibility of recurrence. However, misinterpretation of such a condition as endodontic or periodontal disease may lead to incorrect treatment planning and possibly unnecessary complications. (Quintessence Int 2007;38:837–841)

Key words: differential diagnosis, odontogenic keratocyst, periodontal disease, radiology

Odontogenic keratocyst (OKC) is an uncommonly occurring jaw cyst that generally has a more aggressive nature and higher recurrence rate than other odontogenic and non-odontogenic cysts. Consequently, early diagnosis of the lesion, before it involves a larger portion of the bone, is important to reduce its likelihood of recurrence after therapy. The most important aspects of OKC that can contribute to misdiagnosis or confusion during diagnostic procedures are the variations of its radiologic and clinical presentations. This variability can lead to confusion of the condition with a great number of pathologic processes, including those associated with endodontic and periodontal disease.

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This report describes a case of a small OKC that appeared between and in the buccal aspect of the mandibular left premolars. The lesion presented with unusual clinical and radiologic features that initially caused confusion about the true nature of the pathology. The methods of differentiating between such a lesion and more commonly occurring dental and periodontal diseases are discussed.

CASE REPORT

An 82-year-old African-American woman reported to the emergency clinic of the dental school complaining of a mild but persistent pain associated with a gingival swelling in the mandibular left premolar area. Per the patient’s report, the swelling developed approximately 2 months before her presentation. She had initially seen her general dentist who “drained” the swelling and prescribed systemic antibiotics. However, the swelling persisted. At the time of presentation, a 7-H11003 5-mm firm mass was noted on the left mandibular buccal gingiva in the area of the second premolar tooth (Fig 1). The overlying mucosa had normal color, but the gingival margin adjacent to the lesion was slightly erythematous and thickened. Other significant clinical findings included generalized deposition of calculus around multiple teeth, as well as diffuse gingival inflammation and swelling.

A panoramic radiograph demonstrated generalized moderate periodontitis-related horizontal bone loss that appeared to be slightly more severe between the mandibular left premolar teeth where the swelling was noted. The periapical radiograph demonstrated a faint radiolucent shadow with an ill-defined border measuring approximately 8-H11003 4 mm between the premolar teeth (Fig 2). The crestal lamina dura between the 2 premolars was fuzzy and less clear than that of the adjacent teeth. The lamina dura and periodontal ligament space around the apical portion of the regional teeth appeared normal and intact, and all teeth tested vital. Periodontal or odontogenic abscesses are expected to have soft to fluctuant consistency and are often exquisitely painful upon palpation. Pulp-related dental diseases are associated with nonvital teeth. Therefore, while the initial clinical impression favored a dental or periodontal cause for the swelling, it soon became clear that the lesion was associated with some other bony pathology.

A 90-degree lateral mandibular occlusal radiograph was obtained to evaluate the relationship of the lesion to the bony cortex and rule out the presence of reactive subperiosteal bone formation (ie, proliferative periostitis). The occlusal radiograph (Fig 3) demonstrated a barely visible 5-H11003 5-mm faint radiopaque shadow buccal to the mandibular premolar teeth where the swelling was noted. It was not initially clear whether the radiopaque shadow was induced by subperiosteal bone formation or by thinning and expansion of the bone cortex. Nevertheless, the presence of a triangular-shaped radiopaque area adjacent to the edge of the swelling resembling a Codman triangle was disconcerting. Codman triangle is one of the radiographic features associated with osteogenic sarcoma.7

Because of the unusual radiographic findings, the differential diagnosis included malignancy. However, given the location, a lateral periodontal cyst was also considered. A central giant cell granuloma was also under consideration. For definitive diagnosis, an excisional biopsy was performed under local anesthesia.

Histopathologic examination revealed a fibrocollagenous cystic structure lined by a thin, uniform layer of parakeratinized stratified squamous epithelium (Fig 4). The epithelium exhibited surface corrugation and a prominent columnar basal cell layer. Based on these findings, a diagnosis of odontogenic keratocyst was rendered.

The patient healed uneventfully and was subsequently lost to follow-up.

DISCUSSION

Odontogenic keratocyst is a unique odontogenic lesion that is more commonly observed in the posterior mandible and ascending ramus but can occur in any portion of the
A 7 × 5-mm firm mass was identified on the buccal aspect of the mandibular left second premolar. The gingival margin adjacent to the swelling is slightly erythematous and thickened. Generalized calculus deposition and gingival swelling are evident around many teeth.

Periapical radiograph demonstrating a faint radiolucent shadow with ill-defined border (arrows) located between the mandibular premolar teeth. The crestal lamina dura between the 2 premolars is less clear than that of the adjacent teeth. The lamina dura and periodontal ligament space around the apical portion of the teeth in the mandibular left posterior area appear normal and intact.

Occlusal radiograph demonstrating a barely visible 5 × 5-mm faint radiopaque shadow (white arrows) in the buccal aspect of the mandibular premolars. Note the triangular-shaped radiopaque area adjacent to the edge of the swelling, resembling a Codman triangle (black arrow).

An enhanced view of the occlusal radiograph visualizing more details of the radiopaque shadow (white arrows). Codman triangle (black arrow).
tooth-bearing areas of both jaws.\textsuperscript{1,2} OKCs have a greater propensity to appear as a multilocular radiolucent lesion or a lesion with scalloped margins than do other jaw cysts.\textsuperscript{1,2} In addition, OKC is the sole jaw cyst that is characteristically associated with a systemic syndrome. The occurrence of multiple distinct OKCs is pathognomonically associated with nevoid basal cell carcinoma syndrome.\textsuperscript{8}

The nature and behavior of OKC contributes to a higher recurrence rate than do other odontogenic and nonodontogenic cysts.\textsuperscript{3} The higher recurrence rate of OKC is probably associated with the characteristics of the cyst epithelial lining.\textsuperscript{9,10} Different authors have reported that increased mitotic activity, as well as higher levels of expression of various cell proliferation markers, are indicative of a greater level of cellular proliferation compared with that seen in dentigerous or radicular cysts.\textsuperscript{9–13} These findings support the theory that in contrast to periapical and dentigerous cysts, the epithelial lining of OKC has neoplastic behavior.\textsuperscript{13} Thus, the World Health Organization has recently proposed changing the nomenclature of OKC to better reflect its putative neoplastic growth potential.\textsuperscript{14} The term \textit{keratocystic odontogenic tumor} has been proposed.

OKCs are usually asymptomatic and initially expand within the substance of the jawbone with little or no overt bony expansion. Consequently, an OKC may go undetected for months or occasionally years until it is large enough to produce a noticeable swelling or become symptomatic.\textsuperscript{2} Nevertheless, in the current case, a small OKC lesion appeared between the mandibular premolar teeth, where it produced a detectable bony, hard swelling in its very early stages of development.

A small OKC initially appearing as a radiolucent shadow in the tooth-bearing areas can be confused with more commonly occurring dental or periodontal pathologic processes. Furthermore, secondary infection of OKCs can generate pain and accelerate swelling.\textsuperscript{5} In such a case, more confusion may arise when the patient presents to a clinician complaining of pain and swelling similar to those induced by dental or periodontal infection. However, radiologic and clinical findings that do not appear to be classic features of regular dental and periodontal diseases should be considered as worrisome signs. In such cases, a more detailed clinical and radiologic evaluation or use of other advanced imaging modalities may be required to rule out the presence of a more aggressive lesion.

In the current case, the firmness of the swelling located adjacent to vital teeth was an important unexpected finding that influenced the decision to include lesions other than dental and periodontal diseases in the differential diagnosis and to perform a biopsy for histopathologic examination. It is thus recommended that for any lesion that does not respond or resolve following initial therapy, a biopsy should be performed and tissue submitted for microscopic examination.

**CONCLUSION**

Odontogenic keratocysts have a variable clinical and radiographic appearance. In some patients, these lesions may develop in such a manner that they may be mistaken for more commonly occurring endodontic and periodontal diseases. Early diagnosis of OKC and initiation of appropriate therapy may reduce the possibility of its recurrence. However, misinterpretation of such a condition as dental or periodontal disease may lead to incorrect treatment planning and unnecessary complications.
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REFERENCES