CALCIFYING ODONTOGENIC CYST: A CASE OF CLINICAL MISDIAGNOSIS
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ABSTRACT

The article reports the successful management of a 43-year-old patient with facial swelling, which was initially misdiagnosed as periapical pathology and was later identified as Calcifying odontogenic cyst.

Keywords: Calcifying odontogenic Cyst; Cyst; Periapical Pathology

Introduction

The calcifying odontogenic cyst, (COC) first identified as a separate and distinct lesion by Gorlin’s et al in 1962, is an uncommon benign lesion, consisting of a proliferation of odontogenic epithelium and a scattered nests of ghost cells and calcification that may form the lining of a cyst or present as a solid mass.1-3 The lesion is a mixed odontogenic benign tumor, and although most of the cases present cystic characteristics, a few are of the solid type (15%) and its rare malignant transformation is well documented.4 The COC usually manifests itself as a painless slow growing swelling, anterior to the first molar.4-6 Radiographically, COC usually appears as a unilocular, well defined radiolucency, although it may well be multilocular.6 In about a third of cases radiopaque structures are present within the lesion.6 Review of literature shows that COC can occur at any age and usually it appear before the fourth decade of life.5,6,8 COC reported equally in the maxilla and mandible.5,6 The COC may be associated with other recognized odontogenic tumors like odontomas.5-11 Neville et al have also linked adenomatoid odontogenic tumors and ameloblastomas with this lesion. The diagnosis and therapeutic approach to periapical cysts in general is an extremely controversial concern for dentists according to Romero.12 For example, a case of a calcifying cystic odontogenic tumor mimicking a residual cyst was reported in the literature.13,14 Other authors have also reported a case where a persistent periapical periodontitis with unsuccessful attempts at root canal treatment (RCT) turned out to be a calcifying cystic odontogenic tumor upon histological examination.15

Case Report

A 43-year-old African American was referred to the Howard University Hospital Oral Surgery clinic in February 2011 with history of a non-healing extraction site. History reveals that the patient was initially diagnosed with periodontal abscess in relation to tooth #4 and had undergone incision and drainage under antibiotic coverage. The general dentist recalled the patient after two week for extraction of the tooth. During the three weeks follow up, a non-healing extraction socket was noticed and the patient was referred to the oral surgery department with pre operative radiographs (Figure 1,2). Patient admits the problem had been on going for over a year but the tissue out growth became very noticeable after the extraction of his mobile tooth. On clinical examination the patient was normocephalic. A small swelling was noted around the right malar region with no regional lymphadenopathy. Intra-orally, the oral soft tissue associated with the lips, buccal mucosa, floor of the mouth appeared normal. On the upper right jaw an exophytic lesion with reddish color and tender on palpation with moderate bleeding was noted. The lesion was in relation to teeth numbers 2-4 and measured 4 cm in diameter (Figure 3).

Tooth number 19 was also found to be grossly carious and tender to percussion. Radiographically, the periapical and the panoramic views show a well-defined unilocular radiolucencies and what appears to be calcified structure within the lumen. An initial diagnosis of odontogenic keratocyst was made and patient had tooth number 19 extracted along with an incisal biopsy of the lesion. Subsequent histology report confirmed a case of calcifying odontogenic cyst, solid type (calcifying cystic odontogenic tumor) as seen in the photomicrograph (6). Following the biopsy report surgical excision and extraction of tooth #3 was done under general anesthesia and the postoperative healing was uneventful.

Discussion

The WHO classification of odontogenic tumors groups the calcifying odontogenic cyst with all its variants as an odontogenic tumor rather than an odontogenic cyst.6,10,11 According to Neville et al,10 some calcifying odontogenic cysts appear to represent non neoplastic cyst; other members of this group, variously designated as dentinogenic ghost cell tumor or epithelial odontogenic ghost cell tumors, have no cystic features, may be infiltrative or even malignant, and are regarded as neoplasm. In this report, the neoplastic or what is known as a solid entity of COC is under discussion. Other names employed for this tumor according to literature will include calcifying ghost cell odontogenic tumor, cystic calcifying odontogenic tumor and Gorlin’s cyst. In the diagnosis and management of
odontogenic cysts, it is very essential to understand the clinical and radiographic features. Literature, have shown that misdiagnosis is very possible based on the clinical presentation alone.\textsuperscript{12,14,16} According to Romero\textsuperscript{17} there is a particular difficulty in establishing the differential diagnosis of periapical cyst compared to another very common maxillary pathology like apical periodontitis. In a case report of calcifying cystic odontogenic tumor mimicking as a residual cyst Manveen et al,\textsuperscript{18} concluded that a correlation of clinical and radiographic information with histological features is important in the diagnosis of odontogenic tumor and cyst. There are variety of lesions besides the typical granulomas and cysts that can appear at the apices of teeth. These other lesions must receive considera-
tion in the diagnosis of periapical disease because of their potential impact on patient treatment and outcome. Unless the clinician is thinking in broad rather than narrow terms, serious conditions may go undiagnosed and untreated for an inappropriate period of time.\textsuperscript{19}

Odontogenic keratocyst is a case in point and according to Ja-
son A. Garlock et al\textsuperscript{17} this lesion can manifest themselves as ra-
diolucencies that can appear anywhere in the maxilla or man-
dible, including periapical areas; and may thus masquerade as lesions of endodontic origin. Furthermore, the fact that the radicular cyst and peri-radicular granuloma occur so frequent-
ly, few clinicians consider other possibilities; hence, misdi-
agnosis and inappropriate treatment may result. Similarly, in lit-
erature several neoplastic lesions were presented as periapical radiolu-
cencies.\textsuperscript{16} These were all initially treated for presumed periapical infection. These studies concluded that the atypical features that should alert dentists to the possibility of a tumor presenting in this manner are a vital tooth with minimal car-
ries, root resorption and an irregular radiolucent outline, tooth mobility in the absence of generalized periodontal disease, regional nerve anaesthesia, and failure to respond to good endodontic therapy. The best way for any practitioner to avoid misdiagnosis of COC or odontogenic cysts in general is to rely on the basic fundamentals of clinical diagnosis which in this instance will include a through history, detailed clinical exam-
ination, and appropriate radiography. It is well documented that the most common clinical sign for COC is a raised or ex-
pansile alveolar process, which may or may not be symptom-
atic.\textsuperscript{18,19,20} It is predominantly an intraosseous lesion, although 13% to 30% of cases in reported series have appeared as pe-
ipheral (extraosseous) lesions.\textsuperscript{16} Occasionally, it can cause root resorption and displacement of teeth.

When evaluating or interpreting a radiograph one should re-
member that an ill-defined (diffuse, irregular) periphery is sug-
gestive of a lesion enlarging by invading the surrounding bone. A well-defined (circumscribed) periphery is suggestive of a self-contained lesion enlarging by expansion. A well-de-
finite periphery with a hyperostotic (sclerotic) radiopaque pe-
riphery is suggestive of an extremely slow-growing self-con-
tained lesion enlarging by expansion. If the bone surrounding a lesion does not show any changes, the clinical interpretation is that of a static lesion. COC generally appears as a unilocular lesion with well-defined margins, contains calcification and may occasionally appear multilocular.\textsuperscript{6,10,19} Possible differential diagnosis will include periapical cyst, dentigerous cyst, odontogenic keratocyst, ameloblastoma, incisive canal cyst and simple bone cyst.\textsuperscript{21} Because many other lesions can mimic COC as previously mentioned a definitive diagnosis can be reliably made on the basis of a histological examination. COC exhibits a diverse range of histological appearances which encompass both cystic and solid neoplastic forms,\textsuperscript{21} but the defining microscopic feature of this lesion is the presence of variable numbers of altered epithelial cells without nuclei.\textsuperscript{21} They tend to be lightly eosinophilic and retain the basic cell outline (ghost cells).\textsuperscript{21} The treatment of choice for COC is enu-
ucleation with long term follow-up\textsuperscript{5,10,21} Recurrence depends on completeness of cyst removal.\textsuperscript{21}

Conclusion
In conclusion, an understanding of the clinical course and ra-
diographic presentation of periapical lesions and an early re-
feral to a specialist will ultimately be beneficial to the patient and the primary care dentist alike.

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Figure 1. Pre-operative PA X-ray, Figure 2. Pre-operative Panoramic X-ray, Figure 3. Exophytic lesion of the right maxilla, Figure 4. Im-
mediate post excision view showing a well define basal bone, Figure 5. Gross specimen of the lesion following excision, showing solid mass, Figure 6. Low power (x100 H&E) histology shows a combined cyst and benign odontogenic tumor. There is fibrous connective tissue wall lined by ameloblastomous epithelium and supra basal stellate reticulum-like cells with interspersed clusters of ghost cells.
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References


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