A Multilocular Radiolucency Presenting at the Apex of a Tooth: Lessons to be Learned

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Abstract

An accurate diagnosis and treatment plan can increase the chances for a timely and effective treatment and better outcomes of our patients. Clinicians can use clinical and radiographic examinations to help them in their everyday practice for making a correct differential diagnosis.
Traditionally, a periapical lesion that is treated by non-surgical endodontic therapy is not biopsied. As a result, no histological diagnosis is available prior to endodontic treatment. While this approach is effective in the vast majority of cases, some cases are more complex and may be deceptive, resulting in failed treatment. One such case is presented in this case report. Interdisciplinary consultation and collaboration as a team of the endodontist, radiologist, oral surgeon and histopathologist is very important to be able to make a correct diagnose, treatment plan and to give the best treatment to our patients.

Keywords: Multilocular, Cyst, Radicular Cyst, Glandular Odontogenic Cyst, Radiolucency, Periapical, Endodontics.

Introduction

The prevalence of periapical radiolucencies in a systematic review of cross-sectional studies evaluating the periapical status of over 300,000 teeth was 5%1, emphasizing the importance of being able to accurately make a differential diagnosis and treatment plan for the timely and effective treatment of the patients. Radiographic examinations can help in better visualization of the inner structures in the oral and maxillo-facial area. However there are some limitations that should be taken into consideration, such as the limitation of 2D radiographs to identify periapical lesions with mineral loss less than 1 to 7 mm located in cancellous bone (2,3), the appearance of the defect smaller than in clinical evaluation4 and the influence of the thickness of the cortical plate and volume of bone in the visualization of the lesion2. Three-dimensional cone-beam computed tomography (CBCT) can be a very helpful tool in diagnosing and determining the size and extend of a lesion, through accurate identification of periapical inflammatory disease, and measurements (5-10). In the Joint Position Statement published in 2015, the American Association of Endodontists and the American Academy of Oral and Maxillofacial Radiology11 provide scientifically based guidelines for the use of CBCT in endodontic treatment. They recommend that CBCT can be used for the differential diagnosis of complex pathoses, such as lesions of endodontic or non-endodontic origin (11). The importance of an accurate differential diagnosis reflects in a timely effective treatment of the patient (12). Periapical inflammatory disease have unilocular radiographical presentation and do not respond to pulp testing. In cases of presence of a multilocular radiolucency on a periapical image, it is recommended to take a panoramic radiograph or CBCT to confirm the multilocular growth pattern and extension of the lesion. The extensive differential diagnosis would include common multilocular radioluciences such as odontogenic ameloblastoma, keratocyst (OKC), central giant cell lesion (CGCL) and uncommon multilocular radioluciences such as am-

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How to Cite
Deeba Kashtwari, Almira Isufi, Zachary S. Peacock, Bernard Friedland.
A Multilocular Radiolucency Presenting at the Apex of a Tooth: Lessons to be Learned
Annali Di Stomatologia, 15(2),57-.
https://doi.org/10.59987/ads/2024.2.57-63

eloblastic fibroma, odontogenic myxoma, central odontogenic fibroma, calcifying epithelial odontogenic tumor, orthokeratinized odontogenic cyst, lateral periodontal cyst, calcifying odontogenic cyst, glandular odontogenic cyst, central hemangioma/arteriovenous malformation, aneurysmal bone cyst, cherubism, hyperparathyroidism, intraosseous mucoepidermoid carcinoma (13).

The glandular odontogenic cyst (GOC) can present radiographically as unilocular or multilocular (14-22), with a well-defined border19, might occur in both maxilla and mandible, with a predilection for the anterior mandible (18,19,21), might be present in both men and women, with a predilection for men (15,16,18), in a wide range of age, with an average age of 50 years old (15,16). Studies show a recurrence of more than 30% of cases (23) and may have an extensive and aggressive clinical aspect (17-19,24). The histopathologic appearance of the cyst differs from that of other cysts of the jaws (24). The GOC may be connected to the lateral periodontal cyst (25) and to the botryoid odontogenic cyst (26).

This paper also highlights the importance of interdisciplinary consultation and work as a team of the endodontist, radiologist, oral surgeon and histopathologist, to be able to give the best treatment to our patients.

Case Report

This case report has been written according to Preferred Reporting Items for Case reports in Endodontics (PRICE) 2020 guidelines (27). All patient identifiers have been removed.

We present a case of an apical multilocular lesion in the maxilla in the manner and sequence in which it was brought to our attention.

A 34-year-old Hispanic female was referred to the Endodontic Department of Harvard School of Dental Medicine in October 2015 regarding a failed endodontic treatment. After first examination and checking the intraoral radiographs, (Figure 1), and cone-beam CT (CBCT), a consult with the radiologist was requested. All periapical images in this study were acquired with a Planmeca Promax at 63 kvP, 7mA & 0.1 sec. The scan was acquired with a Kodak 9000 unit with exposure parameters of 90 KvP, 8 mA, and 10.8 Seconds, and a field of view of 4x4cm.

The periapical images showed a well-defined, partially corticated unilocular radiolucency with an epicenter between tooth 13 and tooth 12 and at the level of approximately the apical fifth of the root of the canine. Coronally, the radiolucency extended to the midpoint of the root of the canine. On the mesial of 13 the lamina dura was absent where radiolucency overlay the tooth. Mesiodistally, the radiolucency extended from 12 to the distal of 14. Teeth 13 and 12 had undergone endodontic therapy. The root of 12 was short. The appearance of 12 was consistent with external resorption and root end surgery. Based on these images, the presence of a lateral radicular cyst was considered, especially if 13 had tested non-vital at the initial presentation. There was no independent information concerning the vitality of the teeth prior to endodontic treatment. The CBCT, apart from showing loss of both the buccal and palatal cortices and an intact floor of the nose, did not contribute much more towards arriving at a definitive radiographic interpretation.

The review of the patient's chart showed the following history: In May 2011, the patient had been referred from a community health center for the evaluation and retreatment of teeth 13 and 12, both of which had undergone endodontic therapy approximately two years earlier. Upon presentation in 2011, the patient was on antibiotic medication. The record further indicates that on examination, she was "symptomatic to palpation over teeth 13/12 area. Also, on teeth 13 and 12, there was probing <3mm. Teeth showed no mobility or sensitivity to percussion. PFM margins were intact. There was also no

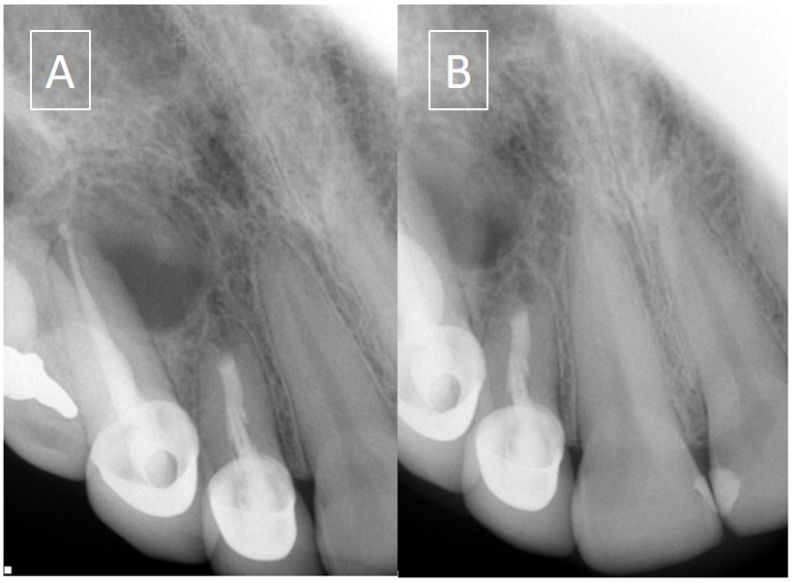

Figure 1. Periapical radiographs taken in October 2015, following endodontic therapy on teeth 13 and 12, as well as an apicoectomy on tooth 12. The images show a well-defined, partially corticated unilocular radiolucency with an epicenter between tooth 13 and tooth 12 and at the level of approximately the apical fifth of the root of the canine. Coronally, the radiolucency extended to the midpoint of the root of the canine. On the mesial of tooth 13 the lamina dura was absent where the radiolucency overlay the tooth. Mesiodistally, the radiolucency extended from tooth 12 to the distal of tooth 14. Teeth 13 and 12 had undergone endodontic therapy. The root of tooth 12 was short.

10.59987/ads/2024.2. 57-63

response to cold test. In February 2012, an apicoectomy was performed on tooth 12. The lesion was excavated, the tissue was removed and the area was filled with mineral trioxide aggregate (MTA). The biopsy report was received with a diagnosis of a radicular cyst.

In April 2015, the patient was diagnosed with a "failing endodontic treatment." Tooth 13 was retreated. In October 2015, the patient presented again. On intra-oral examination, the apical region of 13 was slightly tender to palpation. The radiology consult evaluated the periapical radiographs and CBCT and the interpretation showed that the epicenter of the lesion was between two teeth rather than at the apex, that endodontic therapy and retreatment, as well as an apicoectomy on 12 had failed to resolve the lesion, which all called into question the diagnosis of a radicular or lateral radicular cyst. At this time, it was decided to review the case from the beginning, with an emphasis on reviewing the radiographs (Figures 2,3). The periapical image of 05/09/11 showed a similar picture to that seen on the October 2015 image, namely, endodontically treated teeth 13 & 12, a short root on 12 and a relatively radiolucent, circular area with its epicenter between tooth 13 and tooth 12 and at the level of approximately the apical fifth of the root of the canine (Figure 4). The lamina dura was absent on the mesial of 13 where the radiolucency overlay the tooth. On the

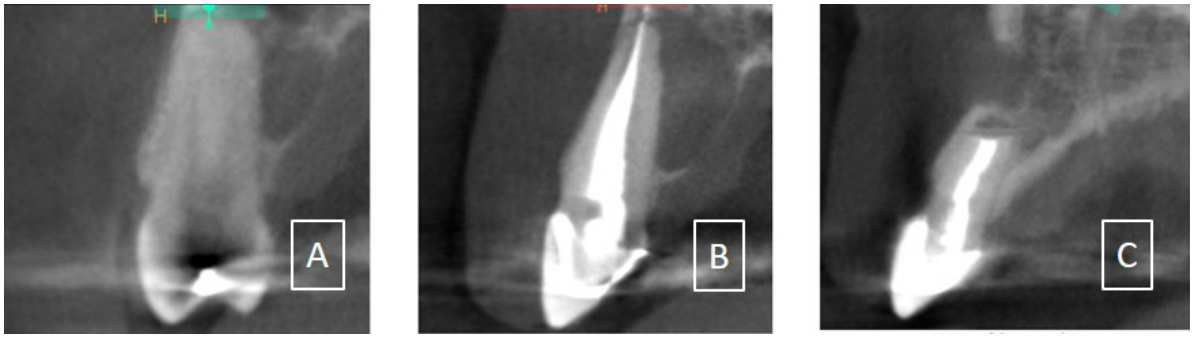

Figure 2. Coronal views of teeth numbers 12-14 sites from a CBCT taken after endodontic therapy on tooth 13 and tooth 12, as well as an apicoectomy on tooth 12. The images show loss of the buccal cortex, consistent with previous root end surgery, in tooth 12 site, as well as palatal expansion in tooth 13 site.

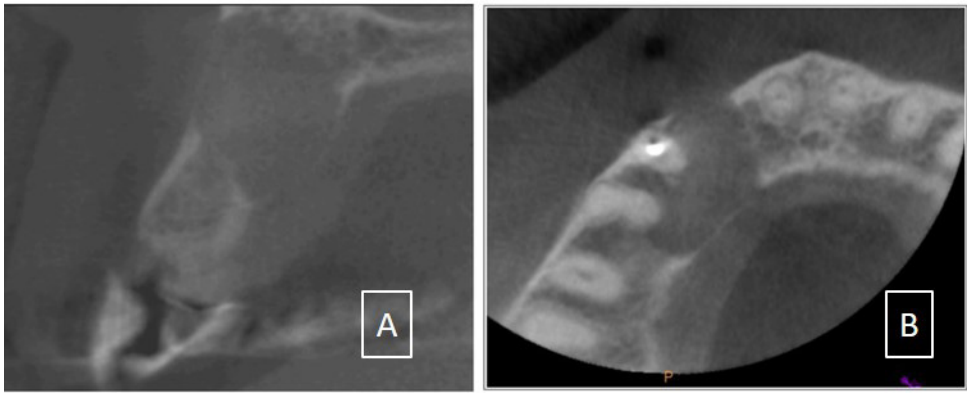

Figure 3. Coronal view between teeth 13 and 12 and axial view of the October 2015 CBCT showing loss of the buccal and palatal cortices.

Figure 4. Periapical image taken in December 2011 (left) and in February 2012 (right) showing a multilocular radiolucency in teeth 13 and 12 sites. The left image was taken after endodontic therapy on teeth 13 and 12. The right image was taken following an apicoectomy on tooth 12.

periapical image taken on 12/09/11, the lesion is clearly multilocular, rather than unilocular, with well-corticated locules. The multilocular appearance persists on the periapical image of 02/15/12.

Based on the multilocular appearance it was decided that the lesion was unlikely to be a radicular cyst. The appearance was more consistent with an odontogenic keratocyst and an ameloblastoma. Upon consultation of an oral pathologist, a histopathologic diagnosis of a glandular odontogenic cyst was established (Figure 5). The patient was referred to an oral surgeon for definitive treatment, following which the second pathologist provided a diagnosis of a glandular odontogenic cyst.

A 3D reconstruction of the DICOM files from the CBCT scan data was used to help in the visualization of the radiolucent lesion, to better understand in 3D its extend and size, as shown in Figure 6. The different tissues were segmented according their greyscale values and

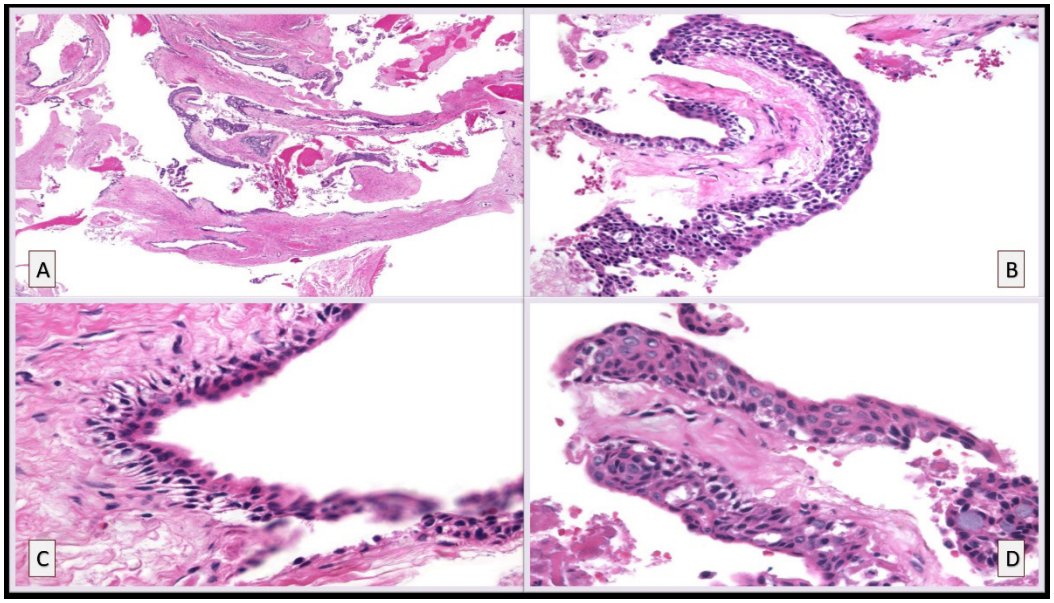

Figure 5. Histopathological examination. A. Fragmented cyst wall with epithelial lining. The epithelial lining is thin and not proliferative. B. The epithelial lining is composed of squamoid cells, 5-7 cells in thickness, and is flat. C. Focally, the squamoid cells appear more spindle with clear cytoplasm, mimicking ameloblastic epithelium (ameloblastoma), however, definitive ameloblastic differentiation (reverse nuclear polarity) is not seen. D. Scattered goblet cell-typed mucous cells are present. The combination of mucous cells and squamoid cells with the possibility of any cells in between (intermediate cells) raises a possibility of intraosseous low-grade mucoepidermoid carcinoma.

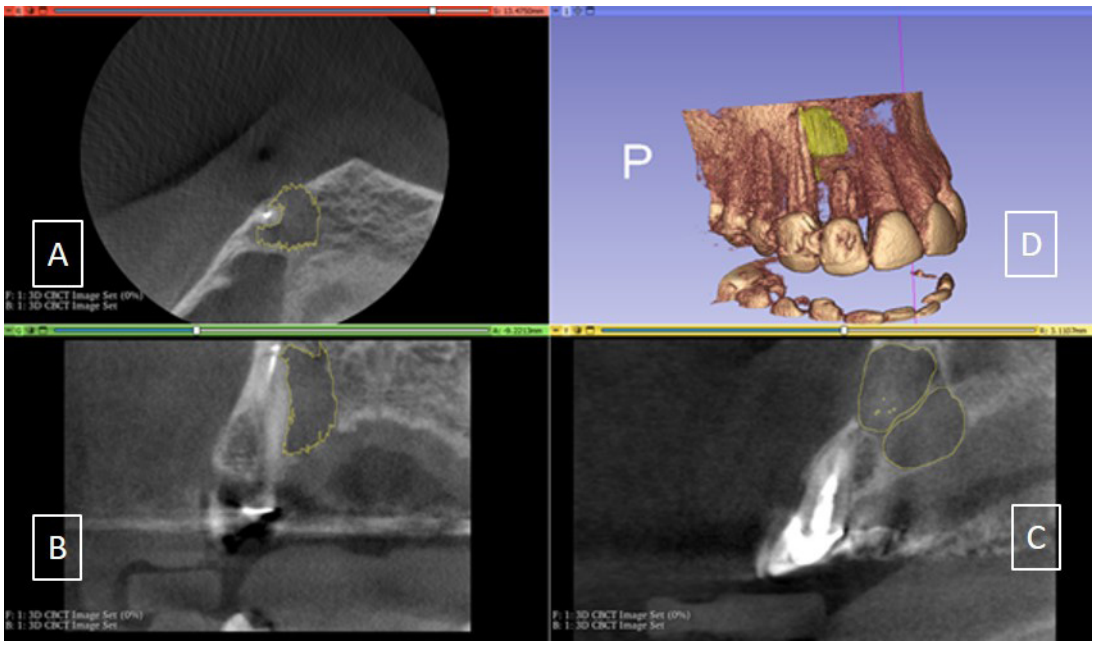

Figure 6. (A-D) Axial, coronal, sagittal and 3D visualization, segmentation and reconstruction of the radiolucent multilocular lesion, with a histologically confirmed diagnosis as GOC after surgical treatment (2015).

10.59987/ads/2024.2. 57-63

visualized in 3D with the use of 3DSlicer software platform28.

The 8-year follow-up control and data of the patient show healing and no recurrence as shown in Figure 7, in the panoramic and CBCT examinations.

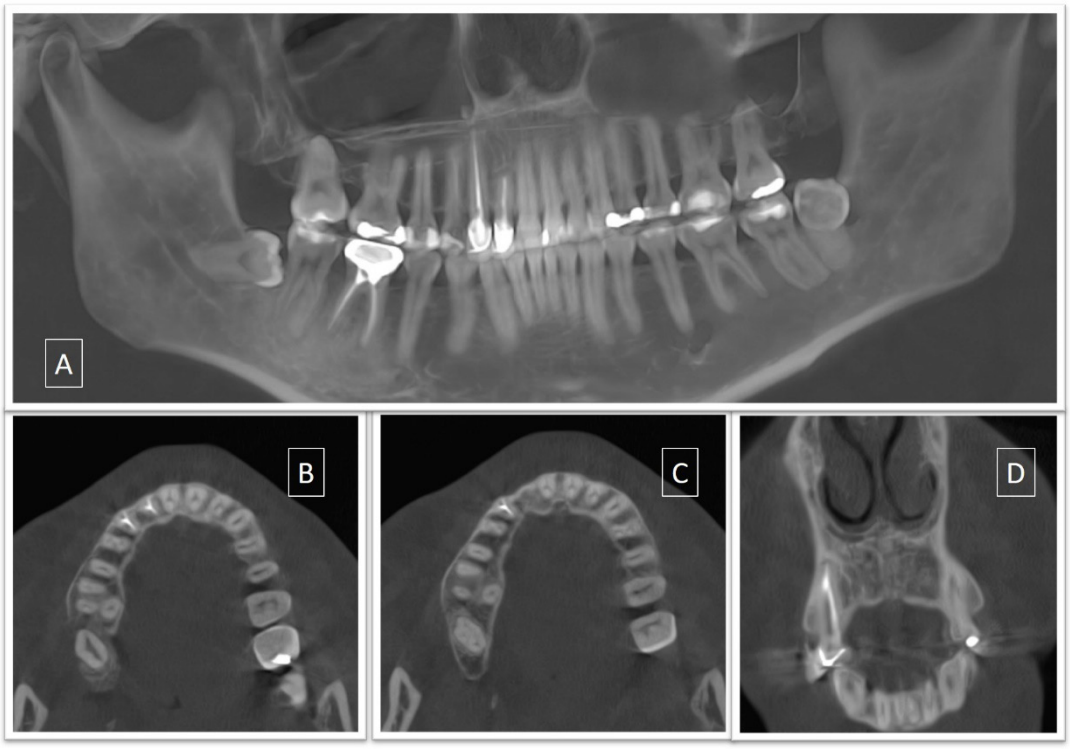

Figure 7. A. Panoramic radiograph at 8-year follow-up visit. B.C.D. CBCT scan views (axial and coronal) at 8-year follow-up showing healing and no recurrence

Discussion

The glandular odontogenic cyst is a rare developmental cyst of the jaws24, with an aggressive potential, a high incidence of cortical perforation, and a relatively high rate of recurrence, with size ranges from 0.5 to 12 cm, with a mean of 4.9 cm (29). GOC have been reported in literature in patients older than 30 years old, with a mean age of 50 years old, both in male and female (29). They have been reported in both jaws, 70% in the lower jaw and 30% in the upper jaw (29). Radiographically, the glandular odontogenic cyst can be unilocular or multilocular radiolucent lesions, with well-defined borders, however, without any pathognomonic features that can make it possible to distinguish these lesions from other with similar characteristics (29-3). GOC can present radiographically with root resorptions, displaced or impacted teeth, thinning, erosion or perforation of the cortical plates30. The differential diagnosis must be evaluated carefully with other radiolucent lesions such as ameloblastoma, odontogenic keratocyst, central giant cell lesion, central mucoepidermoid tumor, lateral periodontal cyst, and botryoid odontogenic cyst. (32,34) Because of the higher incidence of ameloblastoma and the radiographic features very similar to GOC, there should be paid particular attention in the differential diagnosis when evaluating multilocular radiolucencies. Features present in GOC that could help us in the diagnosis are irregular loculation and sclerotic borders with foci of perforations.35 In the literature, it is suggested that more investigation is needed to explore the differential diagnosis of GOC and mucoepidermoid carcinoma and botryoid odontogenic cyst of the jaws because of the very similar radiological and histopathological features they present (31,34).

Depending on the size and locularity of the lesion, conservative or aggressive treatment methods have been described in literature (29,36-39). However, because of the aggressiveness of GOC, more aggressive approaches have been recommended for its treatment, as literature shows that more recurrence was found associated with more conservative methods of treatment, in large multilocular lesions with cortical perforations, while more aggressive approaches such as major surgical procedures have had no reported recurrence (29,30,39) and the sooner the lesion is treated, the smaller may be the area needing to be resected (38). The same is true for other radiolucent lesions taken into consideration for the differential diagnosis of GOC, such as odontogenic keratocysts and ameloblastomas. Nonetheless, the general clinical situations of the patients and their preference about treatment should be taken into consideration, trying to follow-up the patients, to timely diagnose possible recurrences (35).

The case presented raises and highlights a number of important points. Since multilocular radiolucent lesions are rare, the question arises of what endodontists should do when confronted with a multilocular lesion at the apex of a tooth.

Recommendations in literature regarding histopathological examination of tissue collected from endodontic surgery are controversial, with authors suggesting it to be not beneficial and expensive for patients and careful clinical diagnosis would differentiate endodontic and non-endodontic lesions (40-42), while, on the other

hand, other studies have recommend histopathological examination for the exact diagnosis especially in large periapical lesions or cases of failure of endodontic treatments needing endodontic surgery43-50. Many clinicians do not submit tissue in cases where they believe they made a correct diagnosis or the tissue recovered is considered 'limited' (43,51,52).

The case underscores the importance for all practitioners to be familiar, at least in a general way, with the radiographic features of lesions. In this case, based on the imaging alone – and without benefit of knowing the vitality of the teeth when the patient presented to the community health center – the decision to initially undertake endodontic therapy may have been reasonable. However, once the lesion did not regress and especially when the multilocularity of the lesion became apparent, surgical intervention was indicated.

The correct use of CBCT to help in diagnosing a lesion, accurately measure its size and determine it is extend, and moreover, in diagnosing complex pathoses, such as lesions of endodontic or non-endodontic origin, has been recommended in the Joint Position Statement of the AAE and AAOMR (11). Different new software platforms for radiological 3D image visualization and analysis can be used to aid in the diagnosis and treatment planning and to teach dental students and residents about challenging clinical cases.

Finally, from a practice perspective, the case points to the value of establishing and maintaining a mutually trustworthy and respectful relationship with fellow practitioners, in this case, endodontist, radiologist, oral surgeon and histopathologist. The limitation of this case report is that it is a single case involving one patient. The result may thus not be generalizable to all multilocular lesions that present at the apices of teeth.

This case highlights the difficulty faced by endodontists when an apparent "endodontic" lesion presents as a periapical radiolucency. Two reasonable courses of action can be adopted when a clinician is confronted with this scenario. These are more frequent follow up with periodic imaging and/or a surgical intervention followed by a biopsy. Studies aimed at the histopathological diagnosis of multilocular lesions that present as apical radiolucencies should aid in determining how frequently radicular cysts present as multilocular lesions.

Acknowledgement

The authors have no conflicts of interest and all authors contributed equally in the design, writing and reviewing of the manuscript.

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