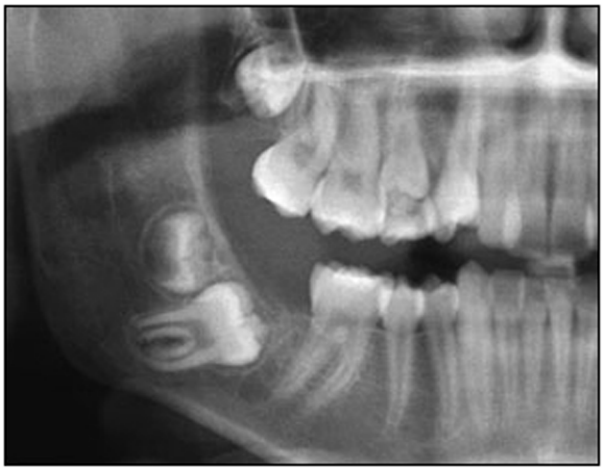
dentalcare.com
Case Challenge
Brought to you by Crest • Oral-B

A Non-expansile Radiolucency of the Posterior Mandible

James W. Rokos, DDS; Ricardo J. Padilla, DDS



The following Case Challenge is provided in conjunction with the American Academy of Oral and Maxillofacial Pathology.

Case Summary

A 14-year-old Caucasian female was referred by her orthodontist with a non-expansile radiolucent lesion associated with impacted tooth #31.

After you have finished reviewing the available diagnostic information, make the diagnosis.

Diagnostic Information

History of Present Illness

The patient is asymptomatic and in good general health, has no known drug allergies, and does not take any medications daily. The lesion was an incidental finding on the radiographic survey at the initial orthodontic consultation.

Clinical and Radiographic Findings

The patient was evaluated for a radiolucent lesion at the right mandibular second molar area associated with impacted tooth #31. No bony expansion was noted clinically. The rest of her intraoral and head and neck examination was unremarkable.

A panoramic radiograph revealed a unilocular radiolucency adjacent to but not associated with the impacted #31 (Figure 1).

A cone-beam CT examination revealed a 1.5-cm well-defined radiolucent area located lingual and distal to the roots of tooth #30 and mesial to the crown of the impacted tooth #31 (Figure 2). There was some expansion and thinning of the lingual cortical plate but no root resorption.

Histopathological Findings

An incisional biopsy was performed revealing an odontogenic neoplasm composed of a myxomatous connective tissue stroma containing nests and cords of cuboidal to columnar epithelium arranged in a pattern suggestive of early enamel organ development (Figures 3 and 4).

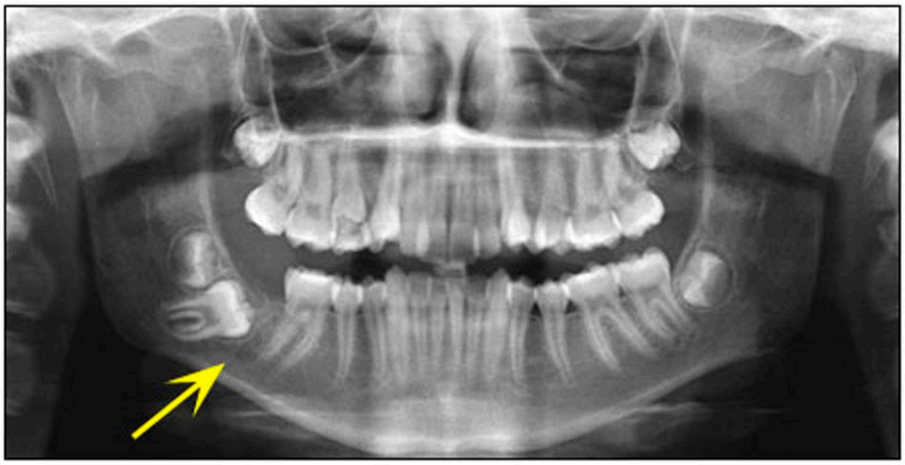


Figure 1. Note the radiolucency distal to roots of tooth #30 (arrow).

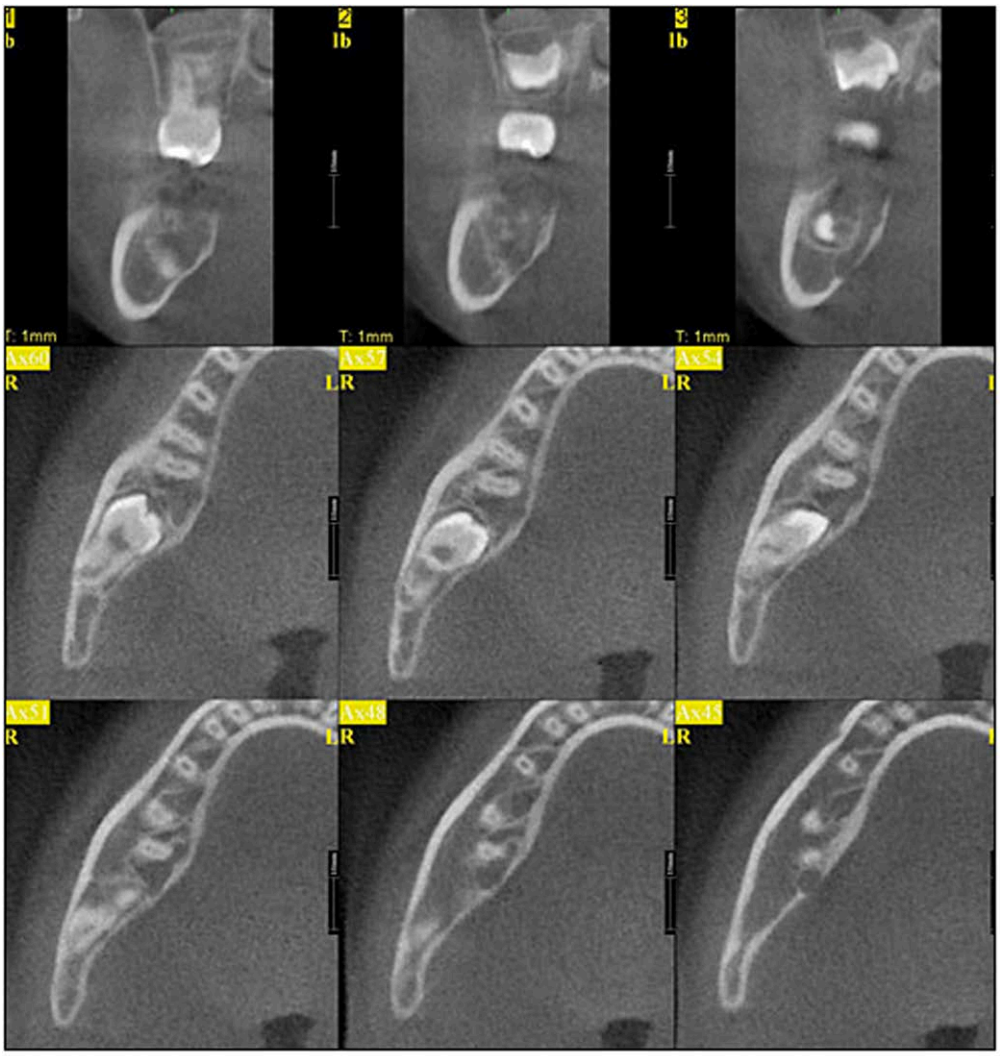


Figure 2. Cone beam CT showing expansion adjacent to impacted tooth #31.

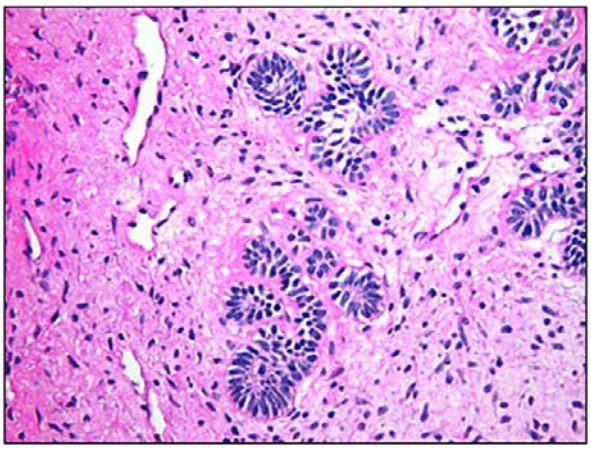


Figure 3. Numerous nests of odontogenic epithelium in a myxomatous stroma.

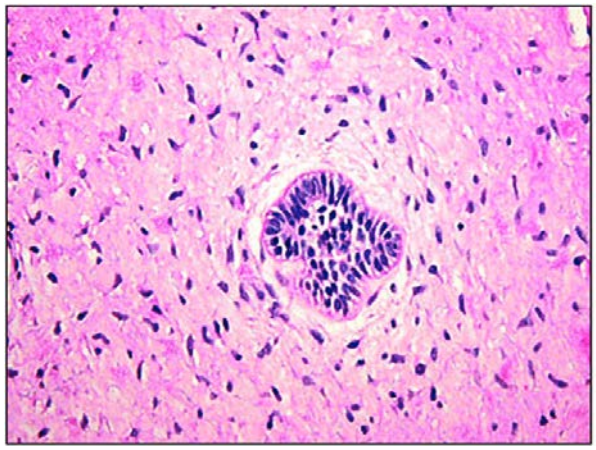
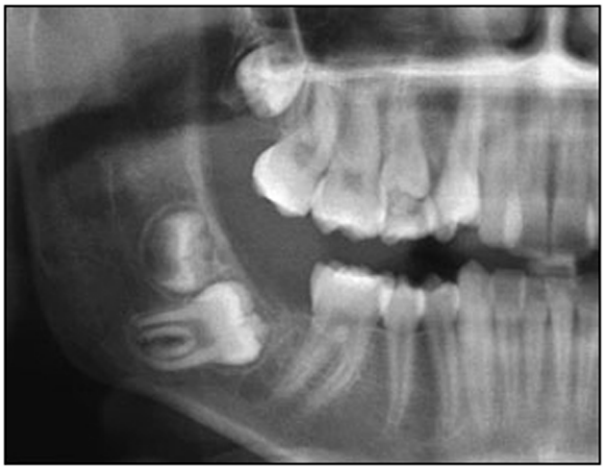


Figure 4. The appearance of the epithelium is reminiscent of the developing enamel organ.

Can you make the diagnosis?

A 14-year-old Caucasian female was referred by her orthodontist with a non-expansile radiolucent lesion associated with impacted tooth #31.



Select the Correct Diagnosis

A. Unicystic Amelblastoma
B. Keratocystic Odontogenic Tumor (Odontogenic Keratocyst)
C. Ameloblastic Fibroma (AF)
D. Odontogenic Myxoma (OM)

Unicystic Amelblastoma

Choice A. Sorry, this is not the correct diagnosis.

Unicystic ameloblastoma is a benign, epithelial, odontogenic neoplasm found most often in the posterior mandible but can also be found in the maxilla and premolar regions. It represents 5% of all ameloblastomas. The neoplasm can arise de novo or in the wall of a dentigerous cyst. Although the age range for other variants of ameloblastomas is predominantly in the third to fifth decade of life, the unicystic variant is seen mainly in younger age groups (10-29 years old).[1,2] The radiographic presentation is typically a unilocular radiolucency, many times associated with an impacted tooth. Oftentimes it presents as an asymptomatic expansion of the affected jaw. Occasionally a developing malocclusion, caused by the displacement of teeth by the lesion, is the first sign of its existence.[3] Histologically, the unicystic ameloblastoma is a cystic neoplasm in which the lining epithelium is composed of a basal layer of palisaded, polarized columnar cells demonstrating hyper-chromatic nuclei and sub-nuclear vacuolization. Adjacent to the basal cells are variably spaced stellate-shaped cells reminiscent of the stellate reticulum of the enamel organ. The current case exhibits a dental papilla-like stroma which is inconsistent with ameloblastoma.

Please re-evaluate the information about this case.

Keratocystic Odontogenic Tumor (Odontogenic Keratocyst)

Choice B. Sorry, this is not the correct diagnosis.

Keratocystic odontogenic tumor is a newly suggested name for the odontogenic keratocyst proposed by a World Health Organization panel of experts because they believe the lesion is actually a neoplasm that grows with a cystic architecture. Many oral and maxillofacial pathologists, however, still classify this entity as a developmental odontogenic cyst. Histologically it is characterized by a parakeratinized, stratified, squamous epithelial lining. It represents about 4-10% of all odontogenic cysts. Patients in their second to third decade of life are most often affected, and the posterior mandible is its favored location. The odontogenic keratocyst can be associated with an unerupted tooth or it may occur in an area where a tooth never formed.

Radiographically, small lesions typically present as a unilocular radiolucency, although larger lesions can become multilocular. Microscopically the keratocystic odontogenic tumor (odontogenic keratocyst) is lined by epithelium demonstrating specific histopathologic features. The luminal surface of the epithelium is parakeratinized and shows marked corrugation of the parakeratin layer. The epithelial lining maintains a uniform thickness of six to eight cells and has a flat interface with the underlying connective tissue of the cyst wall without rete ridge formation. The basal cell layer is composed of cuboidal to low columnar cells that are palisaded and hyperchromatic. Despite its benign nature, this lesion has a significant recurrence rate.[2,4,5] At surgery, the current case was found to be a solid neoplasm without any cystic component, arguing against odontogenic keratocyst. The microscopic features of the current case are also inconsistent with that diagnosis.

Please re-evaluate the information about this case.

Ameloblastic Fibroma (AF)

Choice C. Congratulations! You are correct

Ameloblastic fibroma (AF) is a benign odontogenic neoplasm composed of tissues of both epithelial and mesenchymal origin. The AF has a mean age of occurrence of 14 years. It is found in the posterior mandible 80 to 88% of the time. There is a slight (1.6: 1) predilection for this lesion to be found in males. Oftentimes, it produces a swelling of the affected segment of the jaw. Radiographically, it presents as a well defined radiolucency that may be uni- or multilocular with a sclerotic border. It can be associated with an unerupted tooth or with an area where a tooth failed to develop.[3] Microscopically the AF is "dominated by an immature myxoid connective tissue background reminiscent of the dental papilla. Within the background are islands and cords of cuboidal to columnar epithelial cells. The islands are reminiscent of the early stages of enamel organ development, including a central component with stellate reticulum-type morphology".[3] In this lesion both the epithelial and the connective tissue components are neoplastic. A well defined, narrow, cell-free zone borders the epithelium, and sometimes hyalinization of the adjacent connective tissue occurs. This area of acellular hyalinization is suggestive of induction of the mesenchyme by the epithelium.[6] The mesenchyme is composed of uniformly dispersed bipolar to stellate-shaped cells with a delicate intercellular fibrillar and granular amorphous matrix. No calcified material is seen.

Controversy surrounds the true nature of the AF in terms of whether it is a neoplastic process or whether it represents the earliest stage of a hamartomatous development of an odontoma. Some believe it is a neoplasm. Others contend it is the earliest stage in the development of an odontoma, and an untreated AF will mature into an ameloblastic fibro-odontoma (AFO) which then continues the maturation process to eventuate in an odontoma. If this latter situation were the case, one would predict the AF to be seen in a young age group, the AFO in a slightly older age group, and the odontoma in an even older range. In reality, this is not the case. The average age for an AFO is 9.6 years, at least 5 years younger than the average for the AF.[7] Furthermore, one can see AFs in age groups well beyond the expected completion of odontogenesis. Lastly, data from a study by Chen et al.[8] is enlightening. They examined 13 cases of AF which had an average age at diagnosis of 22 years. This is much higher than the average age seen in other studies, most likely owing to geographic, cultural, and racial differences in the Chinese population studied. Despite these patients being older and their tumors clinically more advanced, none of the lesions had "matured" further into an AFO as the hamartomatous theory would predict.[8]

The treatment for AF is conservative surgical removal either by enucleation, curettage, or simple excision. In a review of the English literature a recurrence rate of 33.3% is noted.[8] When AF's recur, surgical resection to include sound bony margins is the treatment of choice.

Of utmost concern with the AF is the potential for malignant transformation into an ameloblastic fibrosarcoma. In this malignant counterpart to the benign AF, the mesenchymal component transforms into a sarcoma, while the epithelial component shows no change or a progressive reduction in its prominence in the lesion. The average age of occurrence of the ameloblastic fibrosarcoma is 27 years, suggesting a step-wise transformation from the AF rather than de novo genesis of the malignancy. The incidence of this transformation can be as high as 11.4%.[8] Moreover, multiple surgical interventions of recurrent lesions seem to be an important factor in the malignant transformation. Of note is the fact patients whose lesions were discovered before the age of 22 were unlikely to develop malignant transformation.

Odontogenic Myxoma (OM)

Choice D. Sorry, this is not the correct diagnosis.

Odontogenic myxoma (OM) is an uncommon odontogenic neoplasm considered to originate exclusively from undifferentiated odontogenic mesenchyme of the dental papilla. It has a relatively high recurrence rate and mainly affects patients in the second to third decades of life. Young children and patients older than 60 are rarely affected. It too produces as an asymptomatic swelling of the posterior, tooth-bearing area of the jaws, and radiographically presents as a radiolucency. In contrast to the AF, OMs tend to be multilocular, displacing roots adjacent to it. Microscopically, it exhibits loosely arranged bipolar and stellate-shaped ectomesenchymal cells in a myxoid to slightly collagenized stroma. Although rare, nests of inactive odontogenic epithelium may be present but are considered an incidental finding.[9-12] In contrast, the histopathology of the current case shows neoplastic nests of odontogenic epithelium within a myxomatous stroma.

Please re-evaluate the information about this case.

References

1. Jing W, Xuan M, Lin Y, Wu L, Liu L, Zheng X, Tang, W, Qiao J, Tian W. Odontogenic tumours: a retrospective study of 1642 cases in a Chinese population. Int J Oral Maxillofac Surg 2007; 36:20–5.
2. Ledesma-Montes C, Mosqueda-Taylor A, Carlos-Bregni R, Romero de León E, JM Palma-Guzmán JM, Páez-Valencia C, Meneses-García A. Ameloblastomas: a regional Latin-American multicentric study. Oral Dis 2007; 13:303–7.
3. Sciubba JJ, Fantasia JE, Kahn LB. Tumors and Cysts of the Jaw. Washington, D.C.: AFIP, 2001.
4. Habibi A, Saghravanian N, Habibi M, Mellati E, Habibi M. Keratocystic odontogenic tumor: a 10-year retrospective study of 83 cases in an Iranian population. J Oral Sci 2007; 49:229–35.
5. Okada H, Yamamoto H, Tilakaratne WM. Odontogenic tumors in Sri Lanka: analysis of 226 cases. J Oral Maxillofac Surg 2007; 65:875–82.
6. Takeda Y. Ameloblastic fibroma and related lesions: current pathologic concept. Oral Oncol 1999; 35:535–40.
7. Chen Y, Li TJ, Gao Y, Yu SF. Ameloblastic fibroma and related lesions: a clinicopathologic study with reference to their nature and interrelationship. J Oral Pathol Med 2005; 34:588–95.
8. Chen Y, Wang JM, Li TJ. Ameloblastic fibroma: a review of published studies with special reference to its nature and biological behavior. Oral Oncol 2007; 43:960–9.
9. Boleaga-Duran B, Palacios E. Odontogenic myxoma of the maxilla. Ear Nose Throat J 2007; 86:444, 446.
10. Butt FM, Chindia ML, Wakoli KA. Problems in diagnosing odontogenic myxoma: case report. East Afr Med J 2007; 84:141–5.
11. Fellegara G, Mody K, Kuhn E, Rosai J. Normal dental papilla simulating odontogenic myxoma. Int J Surg Pathol 2007; 15:282–5.
12. Noffke CE, Raubenheimer EJ, Chabikuli NJ, Bouckaert MM. Odontogenic myxoma: review of the literature and report of 30 cases from South Africa. Oral Surg Oral Med Oral Pathol Oral Radiol Endod 2007; 104:101–9.

About the Authors

Note: Bio information was provided at the time the case challenge was developed.

James W. Rokos, DDS

Dr. Rokos is a second year resident in the Advanced Education Program in Oral and Maxillofacial Pathology at the University of North Carolina School of Dentistry. Prior to entering the residency training program, he enjoyed a 16 year career in the private practice of general dentistry.

Ricardo J. Padilla, DDS

Dr. Padilla is a Clinical Assistant Professor in the Department of Diagnostic Sciences and General Dentistry at the University of North Carolina School of Dentistry. He is a Diplomat of the American Board of Oral and Maxillofacial Pathology and the director of the Advanced Education Program in Oral and Maxillofacial Pathology.

Email: padilla@dentistry.unc.edu