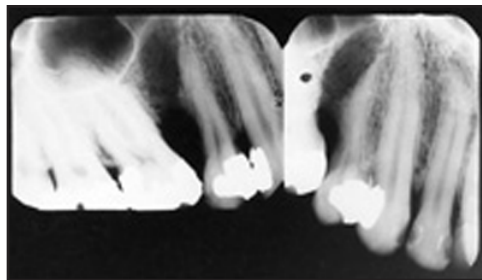


## Chronic Inflammation on the Right Side of the Maxilla

**Karl-Åke Omnell, DDS, Ph.D; Madeleine Rohlin, DDS, Ph.D**



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

### Case Summary

This case challenge presents a patient with a problem that has been unresolved for 18 years.

A 30-year old male was referred by a dental practitioner to the Department of Oral Radiology at the University of Lund, Sweden, for a radiological evaluation of chronic symptoms of inflammation on the right side of the maxilla. According to the patient, at age 12 he had surgery to remove a non-erupted maxillary right second premolar. Postoperatively, a draining sinus tract appeared on the buccal aspect of the alveolar process. Thirteen years later, at age 25, surgery was again performed and the sinus tract reappeared. Intermittent discharge of exudate from the sinus tract occurred since the second surgery.

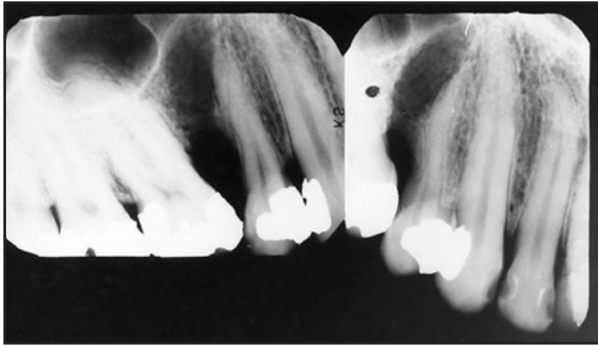
Clinical and radiological examinations were conducted prior to a third surgery. The clinical examination revealed pus draining from the tract located on the buccal aspect of the alveolar process between the right maxillary first molar and first premolar. The radiological examination included periapical radiographs of the right maxillary first premolar and canine, a panoramic radiograph, and frontal tomograms of the maxillary right premolar area.

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

## Diagnostic Information

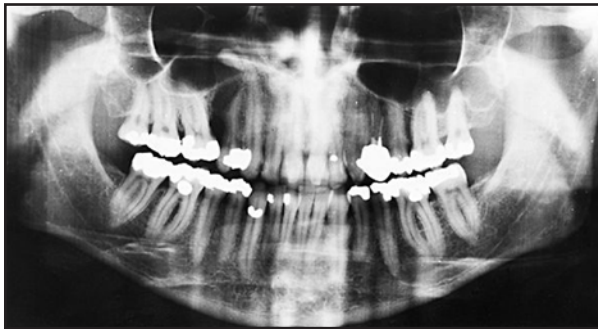
### Preoperative Periapical Radiographs

These periapical radiographs were taken prior to the third surgery. The second premolar is missing and there is an osteolytic lesion between the first molar and the first premolar. The lesion seems to be well demarcated with a partial radiopaque outline.



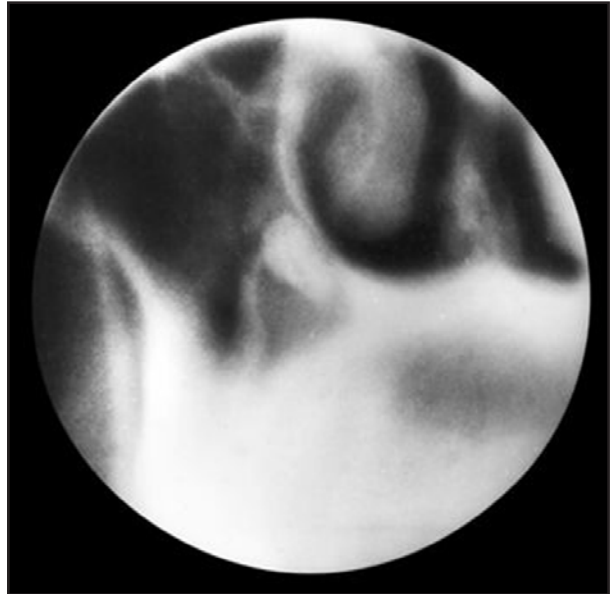
### Preoperative Panoramic Radiograph

This panoramic radiograph was taken prior to the third surgery. The osteolytic lesion between the maxillary right first molar and first premolar is not confined to the alveolar process but encroaches upon the maxillary sinus. A homogeneously radiopaque structure seems to be associated with the lesion.



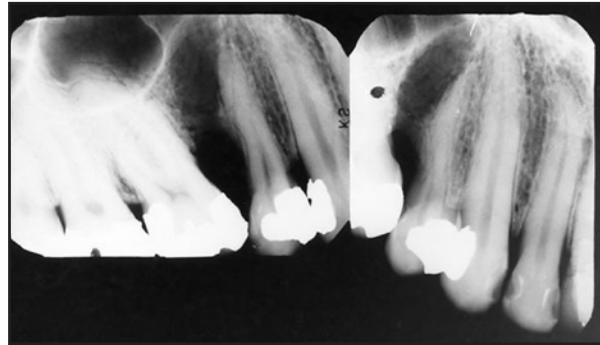
### Preoperative Tomogram of the Affected Area

This frontal tomogram through the center of the lesion was one of several tomograms taken prior to the third surgery. It shows a pear-shaped radiolucent lesion located in the infero-medial part of the right maxillary sinus. A bony wall separates the lesion from the sinus and the nasal cavity. The radiopaque structure observed in the panoramic radiograph looks like a tooth or part of a tooth. It seems to be totally enveloped by the borders of the radiolucent lesion.



### Can you make the diagnosis?

This case challenge presents a patient with a problem that has been unresolved for 18 years.



#### Select the Correct Diagnosis

- A. Adenomatoid Odontogenic Tumor
- B. Calcifying Odontogenic Cyst
- C. Dentigerous Cyst
- D. Unicystic Ameloblastoma

## Adenomatoid Odontogenic Tumor

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

The adenomatoid odontogenic tumor is rare. Less than 200 cases have been reported.<sup>1</sup> There seems to be a first radiolucent stage and a

second stage where calcifications occur within the lesion. In this case, AOT is an unlikely diagnosis since it is very rare and would have grown to a much larger size in the 18 years in which the patient's chronic problems remained unresolved.

Please re-evaluate the information about this case.

## Calcifying Odontogenic Cyst

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

Like the dentigerous cyst, the calcifying odontogenic cyst is often diagnosed in the second decade of life.

The classical paper by Praetorius et al.<sup>2</sup> presented a reevaluation and a classification of the COC based upon a detailed analysis of 16 cases. They concluded that the group of pathoses called calcifying odontogenic cyst contains two entities, three variants of a cyst, and a neoplasm for which they suggested the name “dentinogenic ghost cell tumor.” The cyst

variants grow slowly, mostly without symptoms. In the early stages they can mimic dentigerous cysts. As the name indicates, there may be calcifications. The calcified bodies vary in size, and they appear close to the occlusal/incisive surfaces of nonerupted or impacted teeth. In one cystic subtype the calcifications take on the shape of compound or complex odontomas. Although the diagnosis calcifying odontogenic cyst should be considered for the case at hand, the shape of the radiopaque structure associated with the radiolucency does not look like the calcifications usually observed in connection with this disease.

Please re-evaluate the information about this case.

## Dentigerous Cyst

Choice C. Congratulations! You are correct.

### Discussion

The diagnosis is “infected dentigerous cyst.” This diagnosis is the result of the sketchy patient history and radiographic findings combined with the sign of infection/inflammation expressed by the persistent discharge through a sinus tract. At the subsequent surgery, a cyst-like sac containing pus and part of a root was easily removed without perforation of the walls of the maxillary sinus and the nasal cavity. The pathology report confirmed the radiological and surgical diagnosis.

One can only speculate why the cyst and part of a root remained after the two previous surgeries. One reason could be a combination of surgical incompetence and incomplete radiological work-up. The radiographic information in a case like this should give the full extent of the cyst, its position in regard to the maxillary sinus and the nasal cavity, and its association with the remainder of the non-erupted second premolar.

In the two preoperative periapical radiographs, the maxillary sinus contour and the distal part of the root of the first premolar create the false impression that the radiolucent area between these two teeth is well demarcated. However, the panoramic radiograph clearly showed that the lesion extended into an area normally occupied by the lower anterior part of the maxillary sinus. The frontal tomograms, finally, yielded the information necessary for an accurate diagnosis and for the planning of the third surgery.

### Synopsis on Dentigerous Cysts

Dentigerous means bearing teeth and stems from Denti plus Latin *gerere*, to bear. Dentigerous cysts are also called follicular cysts, from Latin, *folliculus*, little bag.

The dentigerous cyst is believed to originate in the reduced enamel epithelium of the dental follicle when the crown is fully formed but before the root starts to develop. In most cases, the cyst is attached to the tooth at the cemento-enamel junction and envelopes the crown. It is the second most common jaw cyst, with the radicular cyst being the most prevalent.<sup>5</sup>

The majority of patients diagnosed with a dentigerous cyst are in their second decade of life. The cysts are twice as common in males as in females.<sup>6</sup>

Typically, dentigerous cysts are located in the premolar- and molar areas of both jaws. They grow slowly and are generally asymptomatic. In the absence of infection or expansion beyond the normal limits of the jaws, they may remain unrecognized for many years. They are often discovered serendipitously when a clinically missing tooth triggers a radiographic examination. In the maxilla they can be quite destructive and occupy the maxillary sinus and even encroach upon the orbit.<sup>7</sup> Once removed, dentigerous cysts recur very rarely.



A panoramic view showing the presence of a massive dentigerous cyst in the right mandible.

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A variant of the dentigerous cyst is the so-called “eruption cyst.” It acquires its typical clinical appearance when a dentigerous cyst penetrates the crest of the alveolar bone giving rise to an easily palpable, typical, bluish swelling. Usually eruption cysts do not require treatment since they gradually disappear as the associated tooth erupts.



An intraoral view of an eruption cyst in the area of the maxillary left lateral incisor.

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Excellent accounts of the radiological features of dentigerous cysts are given by Goaz and White<sup>8</sup> and Langlais, Langland and Nortje.<sup>1</sup>

From a differential diagnostic standpoint, calcifying epithelial odontogenic tumor, adenomatoid odontogenic tumor, and cystic ameloblastoma should be considered. However, odontogenic fibroma, central giant cell granuloma,

and ameloblastic fibroma may mimic dentigerous cysts and should not be overlooked as possibilities.

In rare instances other pathoses have been associated with dentigerous cysts, such as aneurysmal bone cysts,<sup>9</sup> squamous cell carcinomas,<sup>10</sup> and hemangiomas.<sup>7</sup>

## Unicyclic Ameloblastoma

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

The unicyclic ameloblastoma is also called mural ameloblastoma since it originates in a cyst wall. It most commonly occurs in the wall of a dentigerous cyst.<sup>3</sup> Unicyclic ameloblastoma is a rare, slow growing tumor with a clear propensity for the mandible. The first report of a maxillary

UCA seems to have been that of Gardner et al.<sup>4</sup> A detailed account of the radiological features of the unicyclic ameloblastoma was given by Langlais, Langland, & Nortje.<sup>1</sup> The diagnosis of unicyclic ameloblastoma is very far-fetched for the same reason as calcifying epithelial odontogenic tumor.

Please re-evaluate the information about this case.



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## About the Authors

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

### Karl-Åke Omnell, DDS, PhD



Dr. Karl-Åke Omnell is a Professor and Dean Emeritus of the Department of Oral Medicine, Division of Oral Radiology at the:

University of Washington  
Box 356370  
Seattle, WA 98195-6370  
USA

E-mail: [omnell@u.washington.edu](mailto:omnell@u.washington.edu)

### Madeleine Rohlin, DDS, PhD



Dr. Madeleine Rohlin is a Professor and Chair of the Department of Oral Radiology at the Centre for Oral Health Sciences:

Carl Gustafs väg 34  
SE-214 21 Malmö  
SWEDEN

E-mail: [madeleine.rohlin@od.mah.se](mailto:madeleine.rohlin@od.mah.se)