Inflamed Odontogenic Cyst with Actinomyces Colonization: Management of an Atypical Case in a 16-Year-Old Patient

Inflamed odontogenic cysts, if not treated, may lead to progression of osteolytic activity, potential paresthesia, and loss of teeth. A 16-year-old female patient was referred by a pediatric dentist for asymptomatic abnormal radiolucency found interproximally to the mandibular left first and second premolars. Radiographic, clinical, and pathologic analyses led to a diagnosis of an inflamed odontogenic cyst (type K09.0) with Actinomyces colonization. The cyst was treated by periodontal regenerative therapy and resulted in successful osseous regeneration. This was a rare case because of the patient’s age, the location of the lesion, its association with vital teeth, and its presentation.


Odontogenic cysts arise from proliferation of remnants of the epithelial rests of Malassez, stimulated by inflammation that results from pulpal necrosis of an associated tooth that expanded by hydrostatic pressure/osmotic gradient, leading to fluid transport.\(^1\) Interleukin-1, interleukin-6, and tumor necrosis factor alpha are important proinflammatory cytokines released by macrophages,\(^2\) and this inflammatory response induces proliferation of fibroblasts that stimulate prostaglandins, causing the bone resorption seen in a cyst.\(^3\) This is the driving force of osmotic gradient differences that lead to increases in internal hydrostatic pressure, driving fluid into the cyst.

The World Health Organization 2017 classification of head and neck tumors recognizes two main types of inflammatory odontogenic cysts: Radicular and collateral.\(^4\) Radicular cysts, also known as periapical cysts or apical periodontal cysts, are the most common type of inflammatory odontogenic cysts associated with a nonvital tooth, occurring predominantly in the maxilla and at a mean age of 37.5 years in males.\(^5\) Conservative conventional therapy of radicular cysts includes root canal treatment followed by apical surgery if the radiolucency persists.\(^6\) The residual cyst remaining after extraction of the affected tooth

Ahmad Soolari, DMD, MS\(^1\)
Amin Soolari, BS, CRT\(^2\)
Christopher Fielding, DDS\(^3\)

Inflamed odontogenic cysts, if not treated, may lead to progression of osteolytic activity, potential paresthesia, and loss of teeth. A 16-year-old female patient was referred by a pediatric dentist for asymptomatic abnormal radiolucency found interproximally to the mandibular left first and second premolars. Radiographic, clinical, and pathologic analyses led to a diagnosis of an inflamed odontogenic cyst (type K09.0) with Actinomyces colonization. The cyst was treated by periodontal regenerative therapy and resulted in successful osseous regeneration. This was a rare case because of the patient’s age, the location of the lesion, its association with vital teeth, and its presentation.


Odontogenic cysts arise from proliferation of remnants of the epithelial rests of Malassez, stimulated by inflammation that results from pulpal necrosis of an associated tooth that expanded by hydrostatic pressure/osmotic gradient, leading to fluid transport.\(^1\) Interleukin-1, interleukin-6, and tumor necrosis factor alpha are important proinflammatory cytokines released by macrophages,\(^2\) and this inflammatory response induces proliferation of fibroblasts that stimulate prostaglandins, causing the bone resorption seen in a cyst.\(^3\) This is the driving force of osmotic gradient differences that lead to increases in internal hydrostatic pressure, driving fluid into the cyst.

The World Health Organization 2017 classification of head and neck tumors recognizes two main types of inflammatory odontogenic cysts: Radicular and collateral.\(^4\) Radicular cysts, also known as periapical cysts or apical periodontal cysts, are the most common type of inflammatory odontogenic cysts associated with a nonvital tooth, occurring predominantly in the maxilla and at a mean age of 37.5 years in males.\(^5\) Conservative conventional therapy of radicular cysts includes root canal treatment followed by apical surgery if the radiolucency persists.\(^6\) The residual cyst remaining after extraction of the affected tooth

Ahmad Soolari, DMD, MS\(^1\)
Amin Soolari, BS, CRT\(^2\)
Christopher Fielding, DDS\(^3\)

Inflamed odontogenic cysts, if not treated, may lead to progression of osteolytic activity, potential paresthesia, and loss of teeth. A 16-year-old female patient was referred by a pediatric dentist for asymptomatic abnormal radiolucency found interproximally to the mandibular left first and second premolars. Radiographic, clinical, and pathologic analyses led to a diagnosis of an inflamed odontogenic cyst (type K09.0) with Actinomyces colonization. The cyst was treated by periodontal regenerative therapy and resulted in successful osseous regeneration. This was a rare case because of the patient’s age, the location of the lesion, its association with vital teeth, and its presentation.


Odontogenic cysts arise from proliferation of remnants of the epithelial rests of Malassez, stimulated by inflammation that results from pulpal necrosis of an associated tooth that expanded by hydrostatic pressure/osmotic gradient, leading to fluid transport.\(^1\) Interleukin-1, interleukin-6, and tumor necrosis factor alpha are important proinflammatory cytokines released by macrophages,\(^2\) and this inflammatory response induces proliferation of fibroblasts that stimulate prostaglandins, causing the bone resorption seen in a cyst.\(^3\) This is the driving force of osmotic gradient differences that lead to increases in internal hydrostatic pressure, driving fluid into the cyst.

The World Health Organization 2017 classification of head and neck tumors recognizes two main types of inflammatory odontogenic cysts: Radicular and collateral.\(^4\) Radicular cysts, also known as periapical cysts or apical periodontal cysts, are the most common type of inflammatory odontogenic cysts associated with a nonvital tooth, occurring predominantly in the maxilla and at a mean age of 37.5 years in males.\(^5\) Conservative conventional therapy of radicular cysts includes root canal treatment followed by apical surgery if the radiolucency persists.\(^6\) The residual cyst remaining after extraction of the affected tooth

Ahmad Soolari, DMD, MS\(^1\)
Amin Soolari, BS, CRT\(^2\)
Christopher Fielding, DDS\(^3\)

Inflamed odontogenic cysts, if not treated, may lead to progression of osteolytic activity, potential paresthesia, and loss of teeth. A 16-year-old female patient was referred by a pediatric dentist for asymptomatic abnormal radiolucency found interproximally to the mandibular left first and second premolars. Radiographic, clinical, and pathologic analyses led to a diagnosis of an inflamed odontogenic cyst (type K09.0) with Actinomyces colonization. The cyst was treated by periodontal regenerative therapy and resulted in successful osseous regeneration. This was a rare case because of the patient’s age, the location of the lesion, its association with vital teeth, and its presentation.


Odontogenic cysts arise from proliferation of remnants of the epithelial rests of Malassez, stimulated by inflammation that results from pulpal necrosis of an associated tooth that expanded by hydrostatic pressure/osmotic gradient, leading to fluid transport.\(^1\) Interleukin-1, interleukin-6, and tumor necrosis factor alpha are important proinflammatory cytokines released by macrophages,\(^2\) and this inflammatory response induces proliferation of fibroblasts that stimulate prostaglandins, causing the bone resorption seen in a cyst.\(^3\) This is the driving force of osmotic gradient differences that lead to increases in internal hydrostatic pressure, driving fluid into the cyst.

The World Health Organization 2017 classification of head and neck tumors recognizes two main types of inflammatory odontogenic cysts: Radicular and collateral.\(^4\) Radicular cysts, also known as periapical cysts or apical periodontal cysts, are the most common type of inflammatory odontogenic cysts associated with a nonvital tooth, occurring predominantly in the maxilla and at a mean age of 37.5 years in males.\(^5\) Conservative conventional therapy of radicular cysts includes root canal treatment followed by apical surgery if the radiolucency persists.\(^6\) The residual cyst remaining after extraction of the affected tooth

Ahmad Soolari, DMD, MS\(^1\)
Amin Soolari, BS, CRT\(^2\)
Christopher Fielding, DDS\(^3\)

Inflamed odontogenic cysts, if not treated, may lead to progression of osteolytic activity, potential paresthesia, and loss of teeth. A 16-year-old female patient was referred by a pediatric dentist for asymptomatic abnormal radiolucency found interproximally to the mandibular left first and second premolars. Radiographic, clinical, and pathologic analyses led to a diagnosis of an inflamed odontogenic cyst (type K09.0) with Actinomyces colonization. The cyst was treated by periodontal regenerative therapy and resulted in successful osseous regeneration. This was a rare case because of the patient’s age, the location of the lesion, its association with vital teeth, and its presentation.


Odontogenic cysts arise from proliferation of remnants of the epithelial rests of Malassez, stimulated by inflammation that results from pulpal necrosis of an associated tooth that expanded by hydrostatic pressure/osmotic gradient, leading to fluid transport.\(^1\) Interleukin-1, interleukin-6, and tumor necrosis factor alpha are important proinflammatory cytokines released by macrophages,\(^2\) and this inflammatory response induces proliferation of fibroblasts that stimulate prostaglandins, causing the bone resorption seen in a cyst.\(^3\) This is the driving force of osmotic gradient differences that lead to increases in internal hydrostatic pressure, driving fluid into the cyst.

The World Health Organization 2017 classification of head and neck tumors recognizes two main types of inflammatory odontogenic cysts: Radicular and collateral.\(^4\) Radicular cysts, also known as periapical cysts or apical periodontal cysts, are the most common type of inflammatory odontogenic cysts associated with a nonvital tooth, occurring predominantly in the maxilla and at a mean age of 37.5 years in males.\(^5\) Conservative conventional therapy of radicular cysts includes root canal treatment followed by apical surgery if the radiolucency persists.\(^6\) The residual cyst remaining after extraction of the affected tooth

Ahmad Soolari, DMD, MS\(^1\)
Amin Soolari, BS, CRT\(^2\)
Christopher Fielding, DDS\(^3\)
and lateral radicular cysts associated with a lateral root canal also fall under the radicular cyst category. Included in the collateral inflammatory cyst category are paradental cysts, arising from third molars, and mandibular buccal bifurcation cysts on the buccal surface of first and second molars.\textsuperscript{5}

Actinomyces are opportunistic anaerobic, gram-positive, rod-shaped bacteria known to cause actinomycosis. Actinomyces species are highly susceptible to β-lactam antibiotics such as amoxicillin or penicillin G.\textsuperscript{7} Gomes et al reported Actinomyces israelii to be the most commonly identified microorganism in radicular cysts.\textsuperscript{8}

**Case Report**

A 16-year-old Caucasian girl was referred for periodontal evaluation of a bony lesion at the interproximal space of the mandibular left first and second premolars (Fig 1). The patient was asymptomatic, with an atypical large radiolucency detected by the referring pediatric dentist. The patient had received orthodontic treatment between 2014 and 2016. Clinical and conventional radiographic evaluation disclosed deep probing, bleeding on probing, purulent exudates, significant bone loss, and occlusal disharmony (Fig 2). Since the lesion was not typical of periodontal disease, a cone beam computed tomography (CBCT) image was taken, which disclosed a well-defined unilocular low-density lesion extending from the crest to the root apices, approaching the mental foramen. The intraosseous lesion in the axial cut (mesial-distal direction) was $8.40 \times 10$ mm, with 10.00-mm buccolingual dimensions (Figs 3 and 4). The sagittal cut showed close proximity of the lesion to the mental foramen with thinned cortical plates. No other pathology was detected in the oral cavity.

The lesion had a speck of linear high density within it (a possible odontogenic remnant). From the buccolingual dimensions, the lesion was estimated to extend between the cortical plates, with thinning of the lingual plate and interruption suspected in the buccal plate. The lamina dura adjacent to the lesion and in the interproximal area was missing along the mesial aspect of the second premolar and distal aspect of the first premolar, but there were no changes to the root apex and both teeth were alive on vitality testing.

Differential diagnosis included radicular cyst, odontogenic kerato-cyst, calcifying odontogenic cysts, and lateral radicular cyst. To confirm a diagnosis of the findings, the authors performed an incisional biopsy to rule out odontogenic tumors. The biopsy confirmed an inflamed odontogenic cyst with Actinomyces colonization. There was no evidence of malignant or neoplastic processes. Although cyst fluid samples and culturing were not performed, 150 mg of the antibiotic clindamycin was prescribed (three times a day for 10 days) along with the anti-inflammatory methylprednisolone, 800 mg of the analgesic ibuprofen, and a chlorhexidine rinse (Acclean 0.12% oral rinse USP, twice
Periodontal regenerative therapy was offered to rebuild the hard and soft tissue that had been compromised by the inflammatory response. The patient and her parents accepted the treatment plan.

Panoramic radiographs were taken on December 24, 2011; July 25, 2013; and September 1, 2017 (Fig 5); these radiographs show that the mandibular first premolar erupted in 2011 (Fig 5a) and that the mesial root of the second premolar caused a delay in eruption in 2013 (Fig 5b), but all permanent premolars were present and the crowding had been corrected by 2017 (Fig 5c). The premolar arrangement, however, differed between the mandibular left and right sides: A large space was observed between premolar roots on the left side but not the right side. Both sides presented symmetrically up until 2013, before orthodontic treatment.

**Treatment**

The CBCT images were read by a board-certified oral and maxillofacial radiologist whose report was reviewed with the patient and her parents; they agreed to proceed with the treatment plan, which was a biopsy of hard and soft tissues followed by periodontal therapy to regenerate lost tissues. Anesthesia was achieved with one carpule of lidocaine (2%) with epinephrine (1:100,000). The pretreatment photo (Fig 6a) showed the presence of a fistula at the interproximal area of the mandibular left first and second premolars. Two vertical incisions were made (Fig 6b) from the distal aspect of the second premolar and mesial aspect of the first premolar using a no. 15 blade (carbon steel, Benco Dental), and an intrasulcular incision connected the two vertical incisions. A full-thickness flap was raised and extended beyond the mucogingival junction to enable full access to the bony defects and to facilitate the ridge-augmentation procedure followed by coronal advancement. The full-thickness flap was used instead of a partial-thickness flap to assist in locating the mental nerve and avoiding injury to the neurovascular bundle near the surgical area. Reflection of the flap (Fig 6c) disclosed a large bony lesion (8.40 × 10 mm) in the vertical and horizontal directions at the interproximal area and a paper-thin lingual plate. In addition, the buccal

![Fig 3](image1) The CBCT scan disclosed an 8.40 × 10-mm intraosseous lesion in the axial cut (mesial-distal direction) and 10.00-mm buccolingual dimensions of bone resorption.

![Fig 4](image2) The lesion was in close proximity to the mental foramen.

![Fig 5](image3) Panoramic radiographs showing that (a) the first premolar erupted in 2011, and (b) the mesial root of the second premolar caused a delay in eruption in 2013. (c) By 2017, all permanent premolars were present and the crowding had been corrected.
The plate was partially missing on the first premolar. The inferior border of the bony lesion approached the mental foramen.

The debridement of the bony lesion was performed until bare bone was seen with no bleeding following removal of all necrotic tissues. The ridge augmentation procedure consisted of reconstruction of the lost tissue using freeze-dried bone allograft (Cortical DN025, Maxxeus Dental; Fig 6c) and a nonresorbable titanium-reinforced membrane (Cytoplast, Osteogenics Biomedical) that was trimmed and fitted to the interproximal area (Fig 6d). The facial flap was coronally advanced to cover the membrane and secured into position with simple interrupted 4.0 monofilament sutures (Cytoplast Non-Absorbable PTFE Sutures, Osteogenics Biomedical).

A surgical soft and hard tissue excisional biopsy specimen (1.3 × 1.1 × 0.2 cm) was removed and sent for histopathologic evaluation, which showed a reactive and hyperplastic cystic squamous epithelium, with background inflammatory cells, that was infected with Actinomyces (Fig 7).

The nonresorbable membrane was removed 2 months later due to premature exposure. The area was left to heal for closure and complete soft tissue remodeling. The area was clinically and radiographically evaluated at 6 and 12 months posttreatment (Fig 8). Conventional radiographs showed significant improvement but did not provide a comprehensive picture of the existing hard and soft tissues; the CBCT taken 17 months after treatment disclosed a successful ridge augmentation in both the horizontal and vertical dimensions (Fig 9). Bone...
gain was 10 mm in the buccolingual direction and 8 mm in the occluso-gingival dimension.  

The interdisciplinary approach in this case avoided potential paresthesia (lesion advancing to the mental foramen) and loss of two teeth (progression of osteolytic activity) and involved delicate management of a large bony lesion, which is rare.

**Discussion**

In this case, all common inflammatory odontogenic cysts were ruled out. Vitality testing ruled out radicu-
lar and lateral radicular cysts. Residual cysts were ruled out because the patient had no history of extractions associated with cysts. Paradental cysts could not be completely ruled out as they are histologically indistinguishable from other inflammatory odontogenic cysts, but they are clinically mostly associated with third molars in the mandible and a history of pericoronitis. Mandibular buccal furcation cysts were ruled out, as they are defined by their association with first or second molars. Because of the clinical and radiographic findings of this case, the authors hypothesized that this was a reactive inflammatory odontogenic cyst induced by some coronal remnants of the prior deciduous tooth infected by Actinomyces that gained access from the draining fistula. A retained root tip (of the same tooth) was also observed by CBCT just apical to the lesion.

An inflamed odontogenic cyst with Actinomyces colonization was
the final diagnosis; to the best of the authors’ knowledge, the patient’s age and the location of the lesion, its association with vital teeth, and its presentation classify this as a rare case. Inflamed odontogenic cysts with Actinomyces colonization are three times more prevalent in males (with a peak incidence in the second to fourth decades of life) and are typically associated with nonvital teeth.

Clinical, Histopathologic, and Radiographic Appearance

The inflamed odontogenic cyst with Actinomyces colonization manifested as an asymptomatic lesion, which was detected by a pediatric dentist on a routine dental visit. The lesion was central in the mandible and exhibited an asymptomatic osteolytic behavior, although it was painless. The Actinomyces colony demonstrated the Splendore-Hoeppli phenomenon, an in vivo formation of eosinophilic material around microorganisms leading to prevention of phagocytosis and chronicity of infection. This reaction is thought to represent the deposition of antigen-antibody complexes and debris from host inflammatory cells.11

Histologically, an inflamed odontogenic cyst is defined as a lesion that contains reactive odontogenic epithelium. The cyst may have been associated with the mandibular left second premolar. Cystic lesions in the maxilla are typically observed in the anterior region, while mandibular lesions tend to occur in the posterior region. Careful diagnosis was important because of the unusual histologic features. Scholl et al reviewed cysts and cystic lesions of the mandible, and noted that many non-odontogenic lesions can mimic odontogenic lesions.12 Marx and Stern recommended examination of the original pathologic specimen as well as examining a biopsy specimen if a recurrence is observed.13 Cases of inflamed odontogenic cysts should be monitored at least 5 years after surgical removal.

Microscopic examination of the biopsy material showed an inflammatory odontogenic cyst characterized by a reactive and non-keratinizing hyperplastic squamous epithelium set within markedly inflamed granulation tissue. The inflamed granulation tissue contained a mixture of inflammatory infiltrate composed of lymphocytes, plasma cells, neutrophils, and scattered histiocytes. Abundant filamentous bacterial colonies were present that were morphologically consistent with Actinomyces species.

Conclusions

A 16-year-old female patient with a large bony lesion in the premolar region of the mandible was diagnosed with an inflamed odontogenic cyst with Actinomyces colonization without developed complications. This case of an extremely rare osteolytic lesion required special management of diagnosis, treatment planning, treatment, evaluation of results, and follow-up.

Acknowledgments

The authors declare no conflicts of interest.

References