A Management Approach for Periapical Actinomycosis: A Rare Case Report

Periapikal Aktinomikoz için Tedavi Yaklaşımı: Nadir Bir Olgu Sunumu

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ABSTRACT Actinomycosis is a rare, chronic, and slowly progressive bacterial disease caused by the anaerobic bacteria *Actinomyces*. It is usually diagnosed by identifying typical actinomycotic colonies in a surgical specimen. Frequently, a definitive diagnosis is only made with a cytological study and is based on the identification and/or isolation of the organism and presence of sulfur granules. Once the diagnosis is established, treatment of actinomycosis consists of prolonged administration of antibiotics with surgical excision or incision. Penicillin is still the drug of choice in the treatment of nonallergic patients. This article presents a management approach for periapical actinomycosis after radicular cyst enucleation in the posterior maxilla.

Key Words: Actinomycosis; maxillary sinus; radicular cyst

ÖZET Aktinomikoz, anaerobik *Actinomyces* bakterilerinin neden olduğu, nadir, kronik ve yavaş gelişen bir bakteriyel hastalıktır. Genellikle cerrahi örneğinde tipik olarak aktinomikotik kolonilerin varlığı ile teşhis edilirler. Kesin tanı için sitolojik bir çalışma ile yapılır ve sülfür granüllerinin varlığına veya aktinomiçeşlerin belirlenmesine ve/veya soyutlanmasına dayanır. Aktinomikozun tedavisi, cerrahi eksizyon ya da insizyonla birlikte uzun sureli antibiyotik kullanımını içerir. Penisiline karşı alerjisi olmayan hastalarda, tedavisinde penisilin hala tercih edilen bir ilaçtır. Bu makalede, üst çene arka bölgede radiküler kist enükleasyonundan sonra periapikal aktinomikoz için bir tedavi yaklaşımı sunulmuştur.

Anahtar Kelimeler: Aktinomikoz; maksiller sinüs, radiküler kist

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Actinomycosis is a rare, chronic, slowly progressive bacterial disease caused by anaerobic or facultative, gram-positive bacteria of the *Actinomyces* family with filamentous appearance, erroneously classified in the past as part of the mycetes families. This bacterium normally colonizes in the mouth, colon, and vagina.[1-4] Infection is the most common form of actinomycosis and is generally odontogenic in origin, and evolves as a chronic or subacute painless or painful soft-tissue swelling or mass involving the submandibular or paramandibular region. The submental and retromandibular spaces, temporomandibular joint, and cheek may also be involved. Infection involving the cervicofacial area is the most common clinical presentation, followed by pelvic, abdominal, and thoracic involvement.[5]

Actinomycosis was first described in 1877 and its pathogenesis in humans was reported in 1885.6 The incidence of the disease is not completely known.[3] Infection can take place in all age groups; however, it rarely develops in children or in patients older than 60. Most cases occur in individuals in the middle decades of life5 such as the fourth, fifth, and sixth decades, with a male-female ratio of 3:1.[4]

It is most often caused by the gram-positive bacterium *Actinomyces israelii* and less often by *Actinomyces naeslundii/viscosus* complex, *Actinomyces odontolyticus, Actinomyces meyeri,* and *Actinomyces gerencseriae*. It is an uncommon, both suppurative and granulomatous inflammation. This disease is characterized by localized swelling with suppuration, abscess formation, tissue fibrosis, and sinus drainage.[5] Its diagnosis is usually made by identifying the typical actinomycotic colonies in a surgical specimen.[2-5] Early diagnosis of actinomicosis is rare since it is usually difficult to diagnose on clinical features alone. Frequently, definitive diagnosis is only made with a cytological study and is based on the identification and/or isolation of the organism and the presence of sulfur granules.[3] Once the diagnosis is established, treatment of actinomycosis consisting of prolonged administration of antibiotics with surgical excision or incision, and drainage should be started as soon as possible. Penicillin is still the drug of choice in the treatment of nonallergic patients.[6]

This article presents a management approach for periapical actinomycosis after radicular cyst enucleation in the posterior maxilla and review of the literature.

CASE REPORT

A 26-year-old female was referred to the Oral and Maxillofacial Surgery Department of our faculty complaining of painful swelling for 3 months in the left posterior maxillary area. The patient indicated that the left first and second molars were the painful teeth. Radiographic examination revealed that both maxillary permanent first molar and second molar had previously undergone endodontic therapy by another dentist. There was a cystic radiolucent lesion in the periapical area of the left first and second molars related to the maxillary sinus (Figure 1). The patient's medical history showed no previous systemic disease or prior trauma to the area. The cystic lesion was enucleated and curetted. An apical resection procedure of the molars was then carried out (Figure 2-4). Inflammation in the tissue was observed during the surgery. The pathologic tissue was fixed in 10% buffered formalin and was submitted for histologic examination. Histopathologically, the tissue consisted of several fragments of granulation tissue containing various types of inflammatory cells with a colony of *Actinomyces* on the radicular cyst epithelium, and typical sulfur granules (Figure 5). After the surgery, the patient used intravenous ampicillin 1000 mg 4 times daily for 1 month. The radiographs at 3, 6, and 12 months after the operation showed complete regeneration of the bone in the periapical region and no sign of recurrence (Figure 6).

DISCUSSION

Actinomycosis is an uncommon chronic granulomatous inflammatory reaction, surrounded by polymorphonuclear neutrophils, usually caused by *Actinomyces israelii* and is characterized by abscess formation, draining sinus tracts, fistulae, and tissue fibrosis.[3,7,8] It mainly affects the soft tissue, but sometimes spreads to salivary glands, bone, or even the skin of the face and neck. Swelling and induration of the soft tissue are the most common symp-

FIGURE 1: Panoramic radiograph showing the persistent periapical lesion following endodontic therapy associated with molars in the left maxilla.

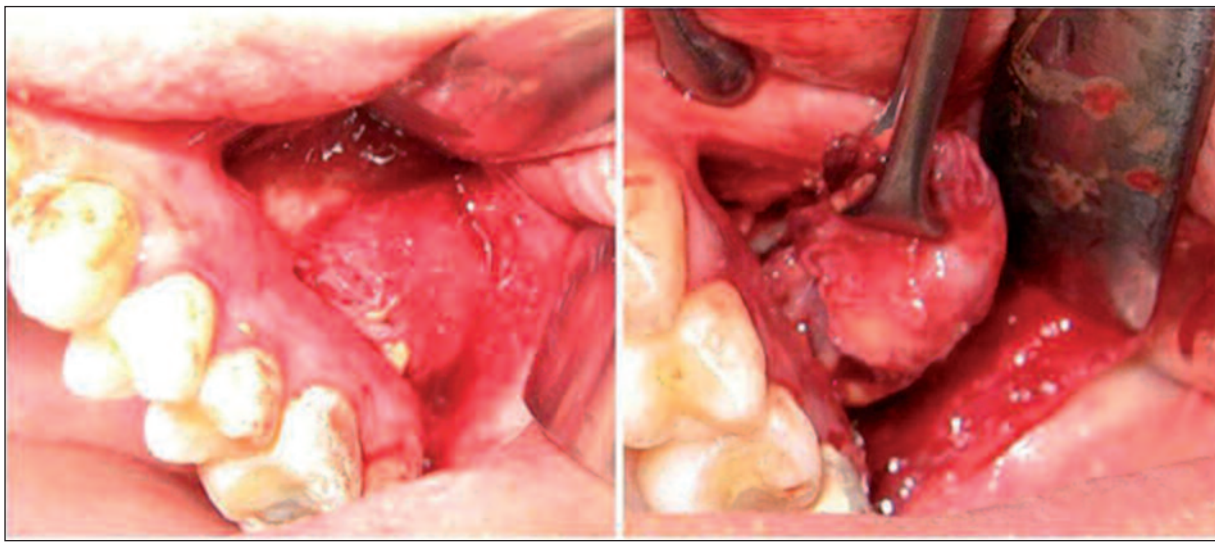

FIGURE 2: A picture of the pathologic tissue, during surgery.

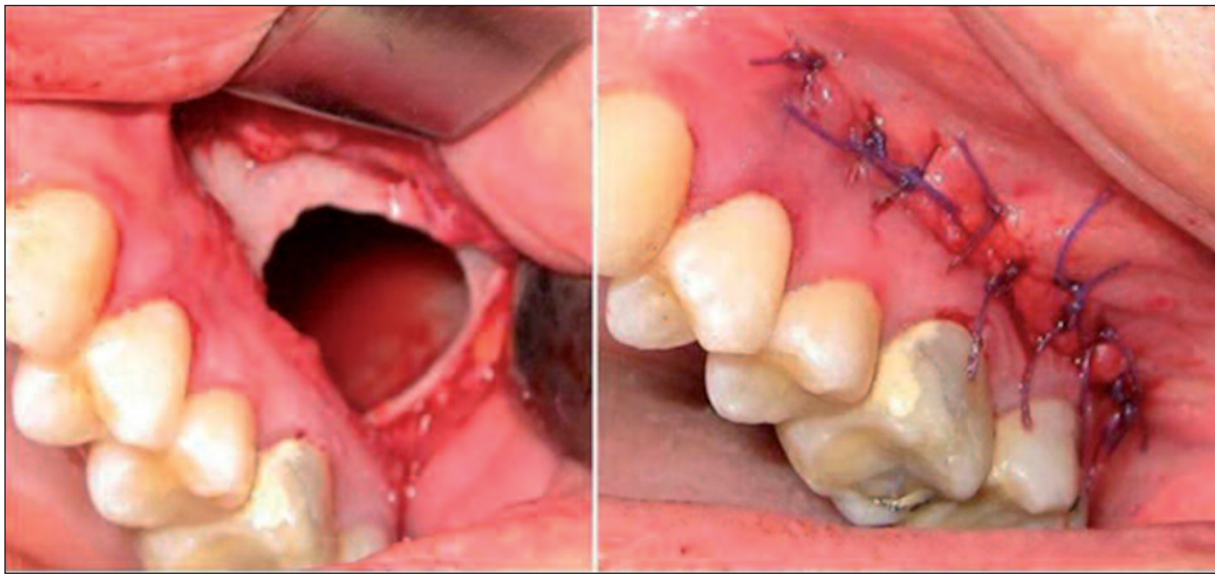

FIGURE 3: After removal of the lesion and closure of the operative area.

toms.[7] Four clinical forms of actinomycosis account for most of these human infections: cervicofacial, thoracic, abdominopelvic, and cerebral. Cervicofacial actinomycosis presents as a chronic, slowly evolving induration in the mandibular preauricular region, often accompanied by fistular tracts to the skin that discharge typical sulfur granules.[2] Cervicofacial infection is the most common manifestation of the disease, involving 50-70% of the cases reported in the literature.[3,8] Periapical actinomycosis is uncommon.2 A retrospective study of 317 patients treated at the Department of Dental Surgery of the University of Cologne showed that the mandibular region was the primary site of cervicofacial actinomycosis in 53.6%, the cheek in 16.4%, the chin in 13.3%, the submaxillary ramus and angle in 10.7%, the upper jaw in 5.7% and the mandibular joint in 0.3%.[9]

Actinomycosis is considered a rare disease in the oral region. The oral cavity is the principal habitat of the *Actinomyces* species.[10] It resides as a commensal organism in periodontal pockets and

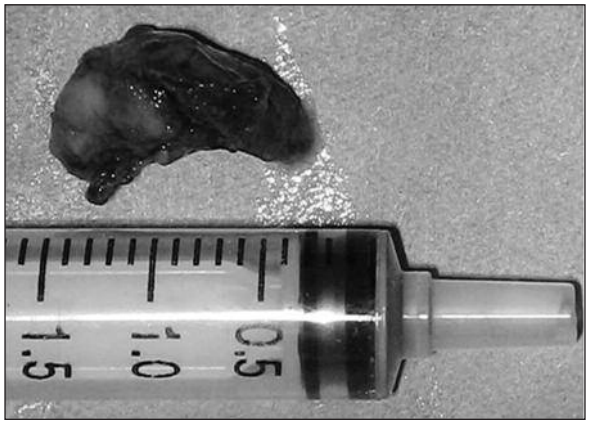

FIGURE 4: A picture of the pathologic tissue (size around 2 cm).

gingival crevices, in carious teeth, dental plaques, tonsillar crypts or in the periodontium. Penetration is facilitated by dental extraction and trauma. Oral and cervicofacial diseases are commonly associated with dental caries and extractions, gingivitis and gingival trauma, infection in erupting secondary teeth, chronic tonsillitis, otitis or mastoiditis, diabetes mellitus, immunosuppression, malnutrition, and local tissue damage caused by surgery, neoplastic disease, or irradiation.[5] Due to its inability to penetrate the mucosa, it exhibits low potential for pathogenicity in the absence of preceding infection or trauma.[10] In the majority of reported cases, there was either a history of trauma to the area or of previous endodontic therapy.[11] Most investigators agree that an actinomyces infection is a relatively rare occurrence.[10] Actinomycosis involving the maxilla usually is seen as a localized intraoral infection, in contrast to classical cervicofacial actinomycosis. Formation of draining sinuses, local swelling and pain are the most common presenting symptoms.[12]

The pathogenesis of oral actinomycosis is not clear; 6 however, in cases of persistent oral infection, the diagnosis of actinomycosis should be actively considered through microbiological and histological examination. Histopathological diagnosis of actinomycosis is important because clinical and microbiological studies cannot always identify the causative microorganism and primary infection source.[12] Most of the imaging modalities do not provide specific information about actinomycosis, but help to identify its inflammatory nature and differentiate it from neoplasms.[7] The laboratory diagnosis is based on a positive culture or detecting "sulfur granules" in the discharge material with typical branching pleomorphic rods. Major considerations in the differential diagnosis include carcinoma, tuberculosis, arthritis, osteomyelitis, cellulites, fungal infections, and pyogenic abscess.[4] The granulomatous reaction remains in the organ long after the pathogenic agent disappears from the tissue, decreasing slowly.[3] The disease is characterized by spread to contiguous tissues because of disruption of anatomical barriers. The disease presents clinically as single or multiple indurations with

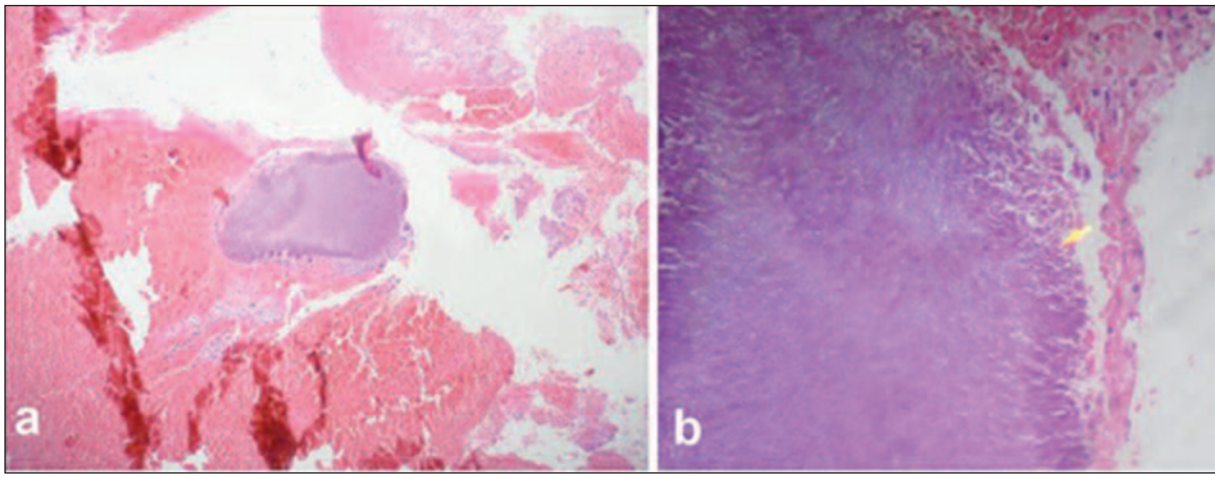

FIGURE 5: Photomicrograph showing the sulfur granule that was removed from the radicular cyst epithelial (Hematoxylin and eosin stain. Magnification, a. X 100 and b. X 400).

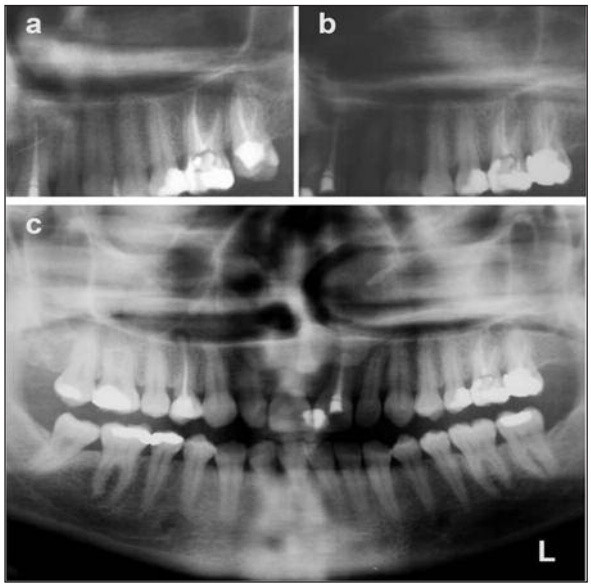

FIGURE 6: Radiographs taken after operation (a: 3. b: 6. c: 12. months).

central softening resulting in formation of fistulae and/or sinus tracts with discharging pus and sulfur granules, which is a classical feature of this disease.[4]

It appears from the literature, including previous case reports, that periapical lesions contaminated with *Actinomyces* are unlikely to heal after isolated nonsurgical root canal treatment.[13] When the actinomycosis infection is circumscribed, diagnosed early, and is not associated with draining fistulas, medical treatment is preferred to surgery. The surgical approach is indicated for excision or drainage of empyemas or when the outcome of medical treatment is not satisfactory.[3] The current recommended therapy for all clinical forms of actinomyces infection is administered at high doses over a prolonged period, because the infection has a tendency to recur.[3,5] Ampicillin, amoxicillin, or penicillin associated with surgical excision is the best therapy, but high doses must be used for a long time.[12] Penicillin is the drug of choice to be given in injectable form for 4 to 6 weeks.[4] Tetracycline, erythromycin, chloramphenicol, clindamycin, doxycicline, and lincomycin are suitable alternatives in patients allergic to penicillin.[4,6,12] First-generation cephalosporins, ceftriaxone, and imipenem also have been employed successfully.5 Sakallıoğlu et al.[6] suggested administration of doxycycline for those with gingival and periodontal involvement. In our case, we preferred to use IV ampicillin 1000 mg 4 times daily for 1 month.

In conclusion, periapical actinomycosis is an uncommon disease in the oral region. Such cases can be treated with conservative surgical curettage and prescription antibiotic after the surgery.

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