

Unusual Presentation of Actinomycosis Misdiagnosed as Severe Periodontal Destruction

Rania Rodan BDS*

ABSTRACT

Actinomycosis is an infiltrative disease caused by *Actinomyces* species that frequently has chronic granulomatous and suppurative lesions. The present case reports an adult female patient with a diffuse and atypical actinomycotic lesion which had a large desquamation and subsequent exposure of the alveolar bone in the region of the maxillary left first and second molars. Diagnosis was based on histopathological examination. Due to the opportunistic characteristics of the actinomycotic infection, early diagnosis of the lesion, together with the adequate therapy and management are of great importance to prevent the spread of the disease. Proper knowledge of the different types of periodontitis is essential to distinguish any atypical presentation of tissue destruction.

Key words: Actinomycosis, Cervicofacial, Differential Diagnosis.

JRMS March 2012; 19(1): 53-56

Introduction

Oral mucosa and skin can be affected by many infectious diseases that range from subacute, chronic or cellulitic invasion of the soft tissues. It was estimated that about 500 species of bacteria inhabit the human oral cavity.⁽¹⁻³⁾ While most of these organisms are commensals, some of them are likely to become opportunistic pathogens that might cause systemic disease.⁽⁴⁾

Actinomycosis is one of the suppurative and chronic infectious diseases that invade the tissues of the face and neck and causes the formation of external sinus tracts that discharge sulfur granules and causes extensive destruction and sequestration of the maxillary and nasal bone. Actinomycosis may also spread to the pulmonary and gastrointestinal systems.⁽⁵⁾

Clinical manifestation occurs in one of three forms: cervicofacial, abdominal-pelvic or pulmonary actinomycosis. Cervicofacial actinomycosis accounts for about 50%-60% of the cases.⁽⁶⁾ The most frequently affected cervicofacial sites are the parotids, submandibular glands, and the

mandible.^(7,8) It is also known as lumpy jaw and involves both the soft and hard tissue of the head and neck region.⁽⁹⁾

Clinical presentation of cervicofacial actinomycosis is characterized by the presence of an indurative mass with discharging sinus. The multiple discharging sinuses that subsequently develop and the sulfur granules that are often present in the pus are almost pathognomic and diagnostic of the disease.⁽¹⁰⁾

Actinomycosis is caused by many various bacterial species of the actinomycete group, it is usually caused by *Actinomyces israelii*, which is an anaerobic gram-positive organism that enters the tissue through a break in the mucosa.⁽¹¹⁾

Actinomycosis is sometimes difficult to diagnose and it should be considered in the differential diagnosis of numerous infectious and non-infectious diseases.⁽¹²⁾

Case Report

A 38years old female was referred to periodontal clinic at Princes Aysheh Bint Al-Hussein Medical

* From the Department of Dentistry, Princes Aysheh Bint Al-Hussein Medical Complex, Marka-Jordan

Correspondence should be addressed to Dr. R. Rodan, P.O. Box 391, Al-Salt, 19110, Jordan. E-mail: rrodan@hotmail.com

Manuscript received March 9, 2010. Accepted July 8, 2010



Fig. 1: A palatal gray-black necrotic lesion, between upper first and second molar teeth.



Fig.2: The sulfur granules that are often present in the pus are almost pathognomic and diagnostic of the disease.



Fig.3: Periapical radiographs of the area of the lesion revealing advanced bone loss around the upper left second molar.

Complex from a general dental practice clinic for evaluation and management of periodontitis in the region of the maxillary left first and second molars. The patient complained of severe pain in the hard palate, and tenderness of the upper left first and second molars with salty taste in the mouth. There was no complaint related to the maxillary sinus involvement, such as heaviness or nasal obstruction.

The patient was depressed, uncooperative, seeking only relieving of the pain in the hard palate. The patient was suffering from social problems due to her husband death and was treated in the psychiatric clinic and used (promazen 1gm/day) as an antidepressant for the last year.

Extraoral examination revealed no facial swelling on the left side of the face. Neither asymmetry nor lymph node involvement were detected.

Clinical intraoral examination revealed a gray-black necrotic palatal lesion between the upper left first and second molar teeth with loss of interdental papillae buccally and palatally (reverse architecture) superimposed by yellowish discharge. Grade III mobility was detected in the second molar, and grade I in the first molar. There was a 7mm loss of attachment distal to the upper left first molar and 9mm attachment loss mesiopalatal to the upper left second molar. Oral hygiene was fair, with no gingival inflammation except in the area of the lesion. No deep pockets were detected in other

sites (Fig. 1 & 2).

Periapical radiographs of the area of the lesion revealed advanced bone loss around upper second molar and moderate bone loss distal to the first molar. The maxillary first and second molars had large and deep restorations. The upper left third molar was impacted (Fig. 3).

Adequate surgical debridement under local anaesthesia was carried out interproximally between upper left first and second molars as an emergency treatment. A biopsy was performed with considerable amount of granulation tissue curetted from areas with the lesion from the alveolus and areas with absence of frank tissue necrosis. The specimen was sent for histopathological evaluation. Oral penicillin therapy was started as 500 mg Amoxicillin three times daily.

One week later, the patient came with the result of the histopathological report as Actinomycosis. The pain decreased but the tenderness of the upper left second molar persisted. The lesion was improved as there was no pus discharge. Extraction of upper left second molar was carried out due to advanced bone loss and grade III mobility. Amoxicillin was continued for four weeks after the diagnosis of actinomycosis was established.

Six weeks after, the patient came with no pain, no tenderness of the teeth, improved tissue condition, with no complaints in the upper 1st molar.

Discussion

Actinomycosis is considered as “the most misdiagnosed disease” even by experienced clinicians and is listed as a “rare disease” by the Office of Rare Disease (ORD) of the National Institute of Health (NIH).⁽¹³⁾ The first case of actinomycosis in humans was, described by Von Lnagebeck, in 1845 and it was attributed to a fungus.⁽¹⁴⁾ Aktino referred to the radiating organism in the sulfur granules as ray fungus that exhibits a number of simple fungus like characteristics, such as tendency to grow as mass of rounded bodies (clubs) and filaments in tissue, low virulence, and the property of eliciting suppuration, necrosis, and a chronic granulomatous tissue response. The unique nature of the organism is the absence of a nuclear membrane, which places *Actinomyces* species among the higher prokaryotic bacteria.⁽¹³⁾

Five species of *Actinomyces* have been identified: *A israelii*, *A bovis*, *A naeslundii*, *A viscosus* and *A odontolyticus*. *Actinomyces* are either strict or facultative anaerobic gram positive rods. Morphologically they are filamentous and branching in nature, except for *A bovis*, all the species are normal inhabitants of the human oral cavity. Actinomycosis is caused primarily by *Actinomyces israelii*, residing as commensal in periodontal pockets and gingival crevices, in carious teeth, dental plaques, tonsillar crypts or in the periodontium.⁽¹²⁾

One of the characteristics of Actinomycosis is the lack of immediate tissue reaction after implantation of the organism. It usually requires 6 weeks or longer for an actinomycotic swelling to break down and discharge pus.

Precipitating factors that are believed to lead to disease in the cervicofacial region include carious teeth, dental manipulations and maxillofacial trauma. The pathogenesis is related to the microorganism ability to act as an intracellular parasite therefore resisting phagocytosis as well as its tendency to spread regardless of the established tissue plains or anatomic barriers.

The presentation is facilitated by dental extraction and trauma to the tissues that facilitate the invasion of the microorganism, so it is considered as one of many post extraction complications. The common site of presentation following extraction is the region around the angle of the mandible.⁽¹⁵⁾

This patient was under treatment by antipsychotic drug (promazen). The most common side effect of psychiatric medication is a dry mouth (xerostomia)

caused by reduced salivary flow. This has a significant impact on oral health, increasing the risk of dental caries, periodontal disease and oral infections.⁽¹⁶⁾

Actinomycosis is relatively rare; most reports cite an incidence of one case per year per institution. There is a slight male predominance from 1.5:1 to 3:1. Actinomycosis is seen more in patients in the 4th to 6th decade of life, it is very rare in infants and children.⁽¹⁷⁾ There are no known predisposing racial, environmental or geographic factors.⁽¹⁸⁾

The microorganism that cause actinomycosis does not spread via the lymphatic system because of the size of the bacterium, that is why regional lymphadenopathy is uncommon or it develops late.^(19,20)

The lesions of the nasal, oral and pharyngeal cavities contiguously extended to the adjacent neck spaces, crossing facial planes. This infiltrative nature is also reported in the abdominopelvic and thoracic actinomycosis, and it may be attributed to the actions of the proteolytic enzymes of the organism.^(21,22)

Actinomycosis is often difficult to diagnose and most of the cases are misdiagnosed.⁽²³⁾ Cervicofacial actinomycosis may mimic either a malignant neoplasm of the head and neck or a chronic granulomatous lesion such as that due to tuberculosis or fungal infection.⁽²⁴⁾

At present the actinomycosis of the oral cavity and face occurs rarely with relatively small inflammatory reaction and the formation of small periodontal abscesses, together with progressing destruction of the maxillary bone.⁽²⁵⁾

Actinomyces israelii is sensitive to most commonly used antibiotics, Penicillin being a narrow-spectrum antibiotic, is considered a good choice for treatment in most cases of actinomycosis, antimicrobial therapy is the only treatment required, although surgery can be adjunctive in selected cases. The traditional treatment of extensive actinomycosis is usually a long term course, generally with up to one month intravenous penicillin G, followed by weeks to months of oral penicillin.⁽²⁶⁾

Cervicofacial actinomycosis is especially responsive to brief courses of antibiotic treatment. In recent series, 3-6 weeks of oral antibiotic therapy combined with surgical drainage, has been curative.^(27,28)

In the microscopic picture the presence of actinomycosis colonies are evident. Colonies are surrounded by numerous neutrophilic granulocytes

and macrophages containing fat bodies (Xanthoma cells). Outside the lesion there is new connective tissue (granulation tissue).⁽²⁹⁾

Conclusion

Cervicofacial actinomycosis is a relatively infrequent infection, despite the fact that this organism is part of the normal oral flora. Predisposing factors include debilitating states such as diabetes, malignancy and immunodepression. There can be also local predisposing conditions such as trauma, surgical intervention, poor oral hygiene or dental caries.

Actinomycosis should be included in the differential diagnosis of a relatively homogeneously enhancing soft tissue masses associated with substantial inflammatory reaction and infiltration of the cervicofacial area. Even in the absence of typical clinical presentation, i.e., discharging sinus tract on the skin surface, Actinomycosis infection should be considered in the case of unusual destruction in the oral tissues.

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