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A case and medical treatment of actinomycosis

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Abstract

Actinomycosis is a chronic bacterial infection characterized by “sulfur granules” in the discharge. Actinomyces species are susceptible to many antibiotics *in vitro*. Actinomycosis infections are uncommon, and the diagnosis is often missed or delayed because of general unfamiliarity with the disease; so it is still poses a great diagnostic challenge, especially in atypical cases, because of its insidious course and non-specific symptoms. A case of cervicofacial actinomycosis which presents as a persistent extraoral lesion is described.

Keywords: Actinomycosis, Sulfur granules, Antibiotic therapy

Introduction

Actinomycosis is a chronic suppurative bacterial infection characterized by fistulous tracts that contain “sulfur granules” in the discharge. Cervicofacial, pulmonary - thoracic, abdominal - pelvic and central nervous system involving clinical forms of actinomycosis have been described [1].

Actinomyces are closely related to Nocardia species, and both were once considered fungi because of their branching filaments, but are currently classified as bacteria. Actinomyces are a group of nonacid fast gram-positive strict or facultative anaerobic Gram-positive bacteria that normally colonize the mouth, colon, and urogenital tract, therefore mucosal disruption is necessary to proceed to infection. Occasionally bone tissue is involved due to direct inoculation or hematogenous spread. There is no documented person-to-person transmission of the disease [2].

The infection typically invades surrounding tissues without tracing fascial planes, and tends to form masses and mimic various infectious and noninfectious entities including malignancy. Regional lymphadenopathy is not common in initial representation [1].

The authors describe a case of cervicofacial actinomycosis which presents as a persistent extraoral lesion.

2. Clinical Report

26 year old male patient referred to our clinic with a 2 month history of a slowly-growing right buccal mass. He had a history of mandibular right third molar extraction 15 months before development of the mass.

On clinical examination, an indurated, nontender 4x1 cm swelling of buccal skin with inflamed overlying tissues was observed (Fig 1). Patient had a decent oral hygiene and there were no inflammatory signs seen intraorally (Fig 2). No lymphadenopathy was present. On radiographic examination, there were no specific findings encountered (Fig 3). The teeth that might be related were vital, and there were no signs of pain, trismus, infection of teeth or history of trauma.

With a presumptive diagnosis of actinomycosis, incision, drainage, curettage of tunnel-forming tracts and administration of a plastic drain were performed (Fig 4). A needle aspiration biopsy was obtained from the lesion and typical sulfur granules with solitarily present. Actinomyces bacteria has been identified by histopathologic examination. Microbiological examination of specimen revealed gram-positive branching, filamentous hyphae suggesting actinomycosis.

Patient has received a course of intramuscular Penicillin G 2 MIU daily for one month. Oral therapy of 100 mg Doxycycline and 1 g Ampicillin were continued for one month daily and

this was followed by an oral therapy of 1 g Ampicillin daily for two months. Significant regression of the lesion has been achieved in the 4th month of follow-up (Fig 5-7).

3. Discussion

Cervicofacial actinomycosis commonly occurs following a dental infection or trauma accompanying poor hygiene. Mandibular region is more frequently involved than maxillary region [3]. Even our patient had a decent oral hygiene and no history of trauma, actinomycosis infection had emerged.

Classically, Actinomycotic infection presents as a slowly enlarging dusky blue or reddish slightly tender swelling, which may become indurated due to scar formation. Sinuses may or may not be present and are often multiple. As in our case, purulent discharge is not necessarily a presenting sign [4].

A key characteristic of actinomycosis is described to be the "sulfur granules" from specimens. These granules resemble sulfur granules clinically though in fact are a conglomerate of bacteria. Sulfur granules can be identified by histopathologic examination or gram staining. Other granule-forming bacterial and fungal species (e.g. *Nocardia* spp and *Streptomyces madurae*) can be differentiated by the absence of peripheral clubs that are specific to *Actinomyces* subspecies [5]. Detection of sulfur granules and culture of the *Actinomyces* organisms are very difficult. Due to simultaneous growing of aerobic and anaerobic bacteria and slow growing characteristic of *A. israelii*, only 10-26 % of cultured cases can be shown to be positive. It is also possible that no sulfur granules be found in the specimen such as an *A. naeslundii* infection reported by Kimura H *et al.* [3].

Due to asymptomatic course of the infection, patients are generally seen in the chronic phase. Infection has a localized but infiltrating, slow-growing pattern, and focal abscesses are linked to each other by a tunnel-forming pattern. In case of involvement of masticatory muscles, trismus may be seen [2]. In our case, no signs of trismus has been noted due to detection before invasion to the muscular tissues.

Bacterial source in these infections is generally a decayed tooth which results in formation of an infectious granuloma that gets larger with eventual bone damage, possible suppuration, and formation of abscesses or fistulas. Bone involvement is uncommon but in recent years, reports of actinomycosis associated with bony sequestre, mandibular bone loss, osteomyelitis and post-operative infection of a dental cyst have been reported [6].

It is suggested that one may only speculate whether actinomycosis, which may be of low virulence, can remain in the tissues for 10 years and therefore be related to the extraction of the mandibular third molar. One cannot assume the presence of an actinomycotic lesion without classical clinical features or a positive culture or histology [4]. It is suggested that actinomycotic infection might be more common than previously thought, but often remaining undiagnosed [7]. It is reported a case of actinomycosis of the mandible that lasted for more than 3 years without displaying typical signs such as board-like swelling and multiple abscesses and sinuses [8]. Thus we cannot ignore the possibility that the infection entered the tissues through the extraction socket and remained there for a long time of 15 months before a clinically obvious lesion presented.

The reported case might have occurred by a previous periapical actinomycotic infection of the third molar; but extraoral inoculation of *Actinomyces* through skin is a more

possible pathway hence there is no evidence of any relationship of the lesion with intraoral tissues.

Though being still controversial, combining surgical and pharmacological treatment is generally accepted modality of treatment, with consensus on using surgery only in cases resistant to long term aggressive antibiotic therapy. Surgical treatment may include incision and drainage of abscesses, resection of necrotic tissue, and curettage of bone [2].

Actinomyces species are susceptible to many antibiotics *in vitro*. Clinical experience supports the use of penicillin G as the drug of choice, and in order to avoid recurrence, prolonged treatment is advisable.² Therapy should be individualized, but high doses (up to 18-24 million units/day) of penicillin over a long period of time (2-6 weeks) followed by oral therapy with amoxicillin to complete 6 to 12 months seems reasonable / is conventionally preferred regimen. For penicillin-allergic patients, doxycycline, minocycline, tetracycline, clindamycin and erythromycin have been proven to be effective. Metronidazole, TMP-SMX, ceftazidime, aminoglycosides, fluoroquinolones, penicillinase-resistant penicillins (eg, methicillin, nafcillin, oxacillin, cloxacillin), and cephalexin have been shown to be non-curative against *Actinomyces* species [9]. But most of other authors report no recurrence of cervicofacial actinomycosis after 3 months of IM antibiotic therapy [10].

Our patient's profession has limited us on our antibiotic protocol for being an active alpinist. We were forced to modify the antibiotic regimen protocol and could not be able to give IM injections more than a month time. Our antibiotic protocol (one month of intramuscular Penicillin G 2 MIU daily + one month of oral therapy of 100 mg Doxycycline and 1 g Ampicillin daily + two months of oral therapy of 1 g Ampicillin daily) is effective and could be useful in situations like ours.

In the literature, prolonged periods of (6-12 months) antibiotic treatment has been declared to be indispensable for an effective eradication of disease. This is particularly true for late-stage large lesions and patients who received intermittent antibiotic therapy. Nevertheless, there are several reports that depict success of short-term courses ranging from 10 to 104 days, especially in cervicofacial actinomycosis [10]. Close monitoring of clinical and radiological response is necessary, especially if a shorter regimen is considered.

3. Figures



Fig 1: Infectious Actinomycotic tunnel



Fig 2: Intraoral view



Fig 3: Radiologic view



Fig 4: Drainage of infectious tunnel



Fig 5: 15th day



Fig 6: 45th day



Fig 7: 120th day

4. Conclusions

Cervicofacial Actinomycosis infections are uncommon, and the diagnosis is often missed or delayed because of general unfamiliarity with the disease; so it still poses a great diagnostic challenge, especially in atypical cases, because of its insidious course and non-specific symptoms. Therefore, biopsies for histopathologic evaluation of sulfur granules and identification of organisms, and cultures are essential for definitive treatment. Therapy should be individualized according to initial presentation and response. The traditional 6-12 months of antibiotic course may not be necessary in all cases, as evidenced by successful short-term treatments in the literature.

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