Cutaneous actinomycosis and long-term management through using oral and topical antibiotics: A case report

Ali Hassan Najmi,[1] Ibrahim Hassan Najmi,[2] Mosa Mohmmed Hassan Tawhari,[3] Khadija Hafed Sawadi,[1] Khaled Ahamed Hassan Khbrani,[3] Fawaz Hadi Tawhari,[1] Mohammed Abdu Tawhari,[3] Majed Hassan Mathkur,[3] Khalid Mohammed Al-attas[4]

[1]College of Pharmacy, Jazan University, Jazan; [2]Department of Dermatology, Khamis Mushayt General Hospital, Khamis Mushayt; [3]Pharmaceutical Care Services, King Fahad Central Hospital, Jazan; [4]Department of Dermatology, King Fahad Central Hospital, Jazan, Saudi Arabia

Abstract

Actinomycosis is a subacute or chronic suppurative bacterial infection which caused because of filamentous gram-positive, anaerobic to microaerophilic nonacid fast bacilli primarily of the genus *Actinomyces* that normally colonize the mouth, colon, and vagina. Primary cutaneous actinomycosis is a rare entity and is generally associated with trauma. A 61-year-old Yemeni male firstly presented to the surgical department in King Fahd Central Hospital with multiple swelling, petted lesions and boring of sensation in the right foot. Local examination showed multiple erythematous nodules and plaques with discharge in the right leg, and active sinus was not determined.

Introduction

The term actinomycosis was derived from the Greek terms *aktino*, that refers to the radiating appearance of a sulfur granule, and *mykos*, which labels the condition a mycotic disease. Actinomycosis is a rare subacute or chronic bacterial infection caused by Gram-positive, anaerobic or microaerophilic bacilli, *Actinomyces* spp. These are higher prokaryotic bacteria belonging to the family *Actinomyceataceae*.[1] Primary cutaneous actinomycosis is a rare entity and is generally associated with trauma.[2] Actinomycosis israelii as the causative organism was isolated in 1891.[1] The clinical findings, diagnostic steps, and management options are shortly discussed as well.

Case Report

A 61-year-old Yemeni male firstly presented to the surgical department in King Fahd Central Hospital with multiple swelling, petted lesions and boring of sensation in the right foot. Local examination showed multiple erythematous nodules and plaques with discharge in the right leg, and active sinus wasn't determined. The patient was firstly advised for complete antifungal treatment and follow up, and also he was recommended for X-ray and computed tomography (CT) scan and referred to the dermatology department. The following up photos were shown in Figure 1. The results of an X-ray on the left and right legs were illustrated in Figures 2 and 3.

The dermatology specialist advised the patient for pathological and microbiological examination of the skin plaques for a definite diagnosis of our case. The pathological report of skin punch biopsy from the right leg stated that, the growth description was three pieces of tissue; the first one measuring 0.5×0.5×1.2 cm, the second one measuring 0.8×0.8×0.4 cm while the third piece measuring 0.5×0.5×0.7 cm. All tissues were used in three blocks. The pathological microscopic description stated that examined sections show grains in the center of suppuration zones. The grains show relative basophilia alternating lobules of granulation tissue and chronic active inflammation, foamy histiocytes, and focal multinucleated grain cells noted. The pathological diagnosis of the examined skin biopsy showed consistent with mycetoma most probably actinomycetoma. The pathologist recommended the patient for clinical-pathological correlation with microbiology studies.

Special studies for Periodic acid-Schiff and Grocott methenamine-silver stains were negative for fungal elements. Also, give a negative gram stain and Ziehl-Neelsen. The microbiological examination report of a wound swab stated that the culture was contaminated.

The dermatologist diagnosed the case as actinomyosis and advised the patient to: i) Itraconazole; ii) Chlorphenamine maleate 4 mg; iii) Diaminodiphenyl sulfone 100 mg; iv) Betamethasone valerate ointment BID; v) Fusidic acid cream BID.

After one month of treatment, the patient is feeling better, and he was recommended to continue the same treatment scheme until the next visit.

After three months of treatment, the patient was well improved, no discharge and mild itching and he was advised to: i) Diaminodiphenyl sulfone 100 mg; ii) Betamethasone valerate ointment BID; iii) Miconazole BID; iv) Cetirizine 10 mg tablets O.D.

The effect of the treatment on hemoglobin level, blood indices and white blood cell counts was in concern where the results of laboratory complete blood count (CBC) along the five years of following up (Hb%: 12.9, 12.4, 13.0, HCT: 39.9, 38.7, 42.6, RBCs: 4.75, 4.74, 5.19 ($\times 10^{12}$ /L), MCH: 27.2, 26.2, 25.0, MCHC: 32.3, 32.0, 30.5, MCV: 84.0, 81.6, 82.1, MPV: 12.2, 11.4, 11.1, RDW: 13.1, 13.5, 13.0, WBCs: 4.54, 5.43, 5.66 ($\times 10^{9}$ /L) and platelets: 224, 268, 274 ($\times 10^{9}$ /L)). By revising the CBC normal ranges, the dermatological specialist found that the results were normal during the receiving of treatment over the 5 years of the following up.

After five years of continuous treatment and follow-up the physical examination indicate the presence of multiple cysts on

Correspondence: Ali Hassan Najmi, College of Pharmacy, Jazan University, Jazan, Saudi Arabia.
Tel.: +966541400256.
E-mail: Ph.ddali@gmail.com

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Contributions: AHN conceived of the presented idea, directed the project, proofed the outlines, finalized the data, extracted form patient medical record and got the consent form from the patient; MMHT, KHS, KAHK, MHM, FHT, MAT, involved in the patient care provision, collected the patient data including the medication charts and correlated to the patient past and current condition; KMA, attending physician of the case, supervised the project, in cooperation with IHN, the dermatology consultant who provided critical feedback on the final version of the manuscript.

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the right foot and the laboratory investigations showed normal renal function tests and normal lipid profile, while the treatment has a side effects on liver function test where the level of serum bilirubin and alkaline phosphatase were elevated, and he was advised to: i) Keplexin 750 BID; ii) Terbinafine 250 O.D; iii) Vasline daily; iv) Fusidic acid BID.

After one year of receiving this treatment, the patient was completely cured where the physical examination indicated no oblique, no granules and no patches. At this point, the follow-up and treatment were finally completed.

Discussion

Actinomycosis is a rare, slowly progressive infectious bacterial disease,[3] caused by anaerobic or microaerophilic bacteria, primarily of the genus *Actinomyces*, which colonize the mouth, colon, and vagina.[4] Actinomycosis has been called *the most misdiagnosed disease* and listed as a *rare disease* by the Office of Rare Diseases of the National Institutes of Health.[5]

Disease incidence is greater in males between the ages of 20 and 60 years than in females. Before the discovery of antibiotics, the incidence in the Netherlands and Germany was one per 100,000 people/year. The incidence in the U.S. in the 1970s was one per 300,000 people/year, while in Germany in 1984, it was estimated to be one per 40,000 people/year in females the incidence of genitourinary actinomycosis has increased by increasing the use of intrauterine devices.[6]

The most common symptoms of cutaneous actinomyosis are having progressive skin and soft-tissue inflammation, which can become an abscess or cold mass, or nodular lesions with fistulas that need to be differentiated from chronic inflammatory skin disease, cutaneous mycobacterial infections, and sporotrichosis.[7] Basically the diagnosis of Cutaneous Actinomycosis is made by the following tests and exams:[8] i) A thorough physical examination and assessment of symptoms. ii) Evaluation of the affected individual's medical history. iii) Microscopic observation of pus or tissue samples, to check for the presence of sulfur granules (which are round and yellow and are named for their characteristic appearance; but, they do not contain sulfur). The tissue samples may have to be obtained surgically. iv) The culture of fluid or tissue from the infected area: These bacteria are slow to grow in culture, and it may take over 3 weeks to obtain a (positive) culture result. v) X-ray, CT, or magnetic resonance imaging scans of the affected area, to ascertain location and number of abscesses, as well as differentiate inflammatory masses due to infection from the tumors.

The treatment of cutaneous actinomycosis of the lower extremity is primarily based on antimicrobial therapy. Drug of choice is Penicillin G. High-dose Penicillin G – 12–24 million U/d intravenous by continuous infusion or in divided doses is given followed by oral amoxicillin, ampicillin, or penicillin V which is administered over a prolonged period (6 months to 1 year). An alternative to penicillins, if the patient is hypersensitive to penicillin or the causative organisms are resistant to penicillin, are Ceftriaxone, Imipenem/Cilastatin, Clindamycin, Amoxycillin/Clavulanic acid, Doxycycline, Tetracycline, Lincomycin and Macrolides (erythromycin, Carbomycin, Spiramycin, and Oleandomycin). Surgery is indicated mainly for taking a biopsy specimen, to drain abscesses, to extirpate a fibrotic sinus tract or a refractory fistulous tract which are not responding to the conservative medical treatment.[9] The follow-up should be adequate and meticulous to have a complete cure for the disease as the prognosis is usually excellent, especially when it is diagnosed early and treated with the appropriate antibiotic therapy.[10]

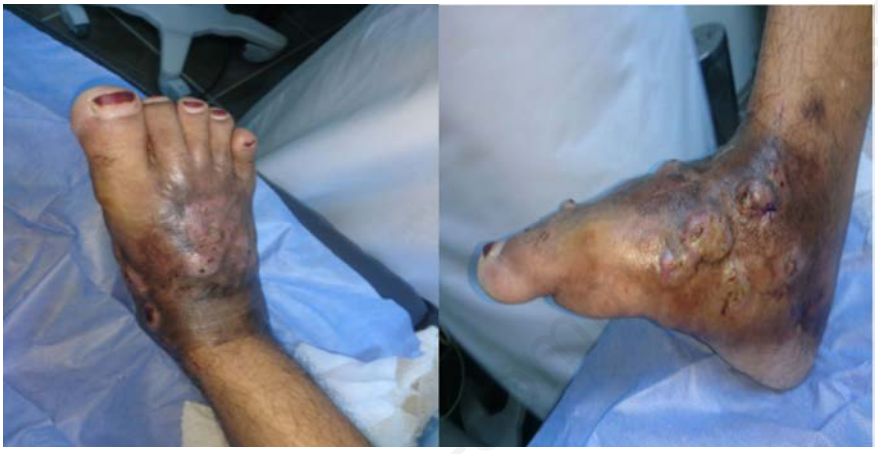

Figure 1. Follow up.

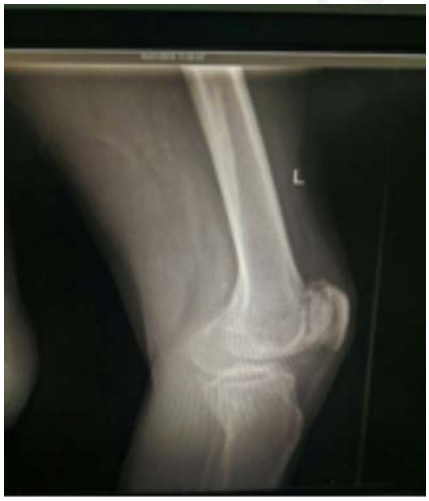

Figure 2. Left leg X-ray film.

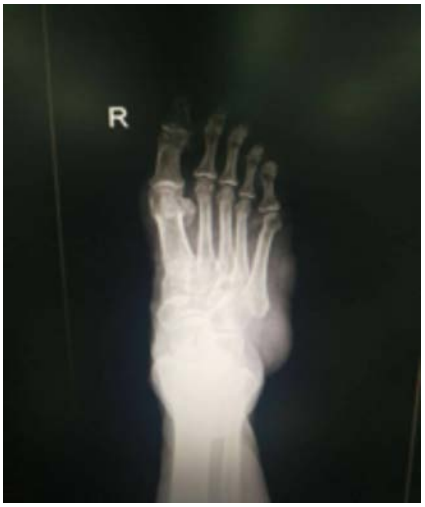

Figure 3. Right leg X-ray film.

Conclusions

Actinomyosis is a rare disease. The incidence of actinomyosis is higher in males than in females. It was characterized by swelling of the skin, nodules and soft tissue inflammations. X-ray and CT plays an important role in the diagnosis of actinomyosis. The treatment is mainly depending on the usage of antifungal and antibiotics. Continuous follow up is strongly requested.

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