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Case Report

Actinomycosis May Be Presented In Unusual Organs: A Report Of Two Cases

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ABSTRACT

Actinomycosis is a chronic granulomatous *suppurative* disease characterized by direct extension to the contagious tissue with the formation of multiple drainage sinus tracts through which tiny colonies of organisms called sulfur granules are discharged. Here, we report 2 cases of actinomycosis from Iran. One of them had actinomycosis on the hand and the other on the foot. Samples of tissue biopsy showed sulfur granules associated with colonies of actinomyces, thus confirming the diagnosis in both cases. The response to curettage and penicillin therapy was satisfactory in our patients. The chronic and indolent course of actinomycosis resembles tuberculosis, fungal infection and malignancy. So, increasing awareness among the clinicians and clinical microbiologists will help in the early diagnosis of the disease and in the initiation of early and proper treatment.

Key Word: Actinomycosis, hand, foot.

skin, where skin sinuses form, with the drainage of the groin [4], and in later stages can cause fracture and osteomyelitis [5]. Although it is not an opportunistic infectious disease, it has been associated with the use of corticosteroids, leukemia and children with congenital immunodeficiencies and HIV infection. The 3 major forms of actinomycosis are cervicofacial (65%), thoracic (15%) and abdominal/pelvic (20%) [6]. A severe form of periodontitis may be associated with actinomyces. Reddy et al reported that actinomyces had the propensity to infect the heart valve and thus cause the insidious presentation of endocarditis with fever in less than half of the cases [16]. Hand and foot are the rare sites of involvement and it is reported in other sites also, in various studies. Also, our diagnosis progressed from suspicion about clinical signs to confirmation by biopsy, but other studies reported initial suspicion that it might be another disease but after the biopsy, actinomycosis were confirmed.

Case Report

The first case was a 44 years old man who presented with multiple fistulas, sinus tracts

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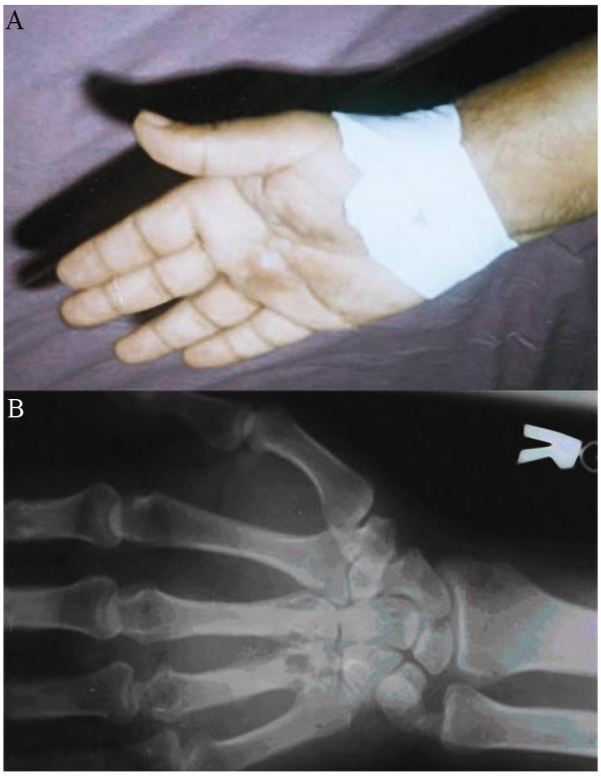
Introduction

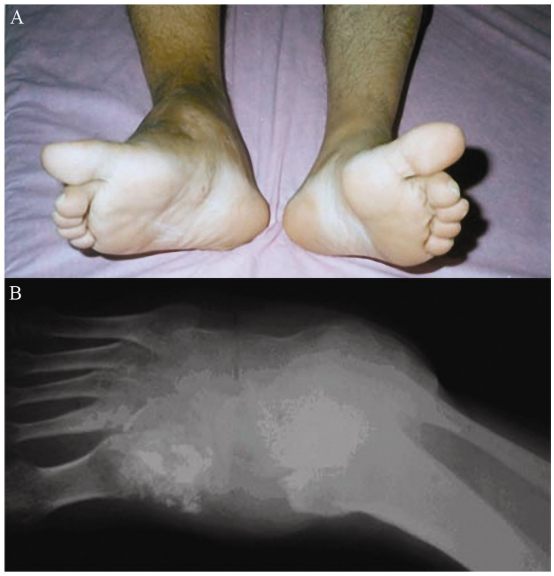
Actinomycosis is a chronic indolent infection caused by anaerobic or micro aerophilic bacteria, primarily of the genus actinomyces that colonize the mouth, colon and vagina [1]. It is an endemic in tropical and subtropical countries [2]. It may occur in all age groups. The causative agent gets implanted into the body from soil or plants through traumatic injuries or superficial lesions [3]. After the minor injury, a silent period takes place when the lesion grows slowly and extends to the



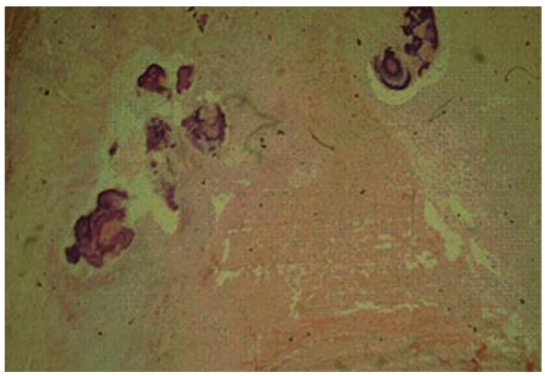
(Table/Fig 1) A-Actinomycosis in right hand; B- AP x-ray of right hand that revealed lytic areas surrounded by sclorosis in the base of 4th metacarpal bone.

The second case was a 39 years old man who presented to the outpatient with a chronic swelling on the right foot since 2 years. He was from Minoodasht city in the Golestan province in the North of Iran. He remembered a blunt foot trauma 6 weeks before the development of painless soft tissue swelling on the foot. After that, the foot became indurate, followed by a period of apparent inactivity after which draining sinuses appeared. He received different antibiotics over a 2-years period which yielded only a partial response or no response at all. There were no systemic symptoms or underlying disease. The clinical examination showed a non tender swollen area in the right ankle and forefoot and the overlying skin was adherent, with multiple draining sinuses. Due to the endemicity of tuberculosis in this area, an initial clinical diagnosis of tuberculosis with less possibility of actinomycosis was proposed. The routine hematological and biochemical tests including ESR were normal. Aerobic and anaerobic tissue cultures for usual bacteria were negative [Table/Fig 2]. The microscopic examination of curettage and the biopsy of the draining sinuses revealed multiple sulfur granules [Table/Fig 3]. Among histochemical stains, periodic Acid-schiff and and pain and swelling on the right hand. He was from an urban area and had the problem since 10 years. He underwent surgery 7 times and took multiple antibiotic therapies, but was left without cure. He was a toll worker and remembered a history of occupational blunt trauma to his hand 12 years ago. There was no history of immune deficiency or underlying disease. Physical examination revealed normal vital signs, but had a tender swollen right hand with multiple discharging sinuses at the volar and palmar areas and also significant movement restriction of the fingers [Table/Fig 1]. The laboratory results were unremarkable and the first hour erythrocyte sedimentation rate (ESR) was 12mm.The microbiological cultures of soft tissue for aerobic and anaerobic organisms showed no growth. Our suspicion about tuberculosis was ruled out by smear and tissue culture. Finally, the curettage and biopsy from discharging sinuses revealed a large amount of inflammatory granulation tissue [Table/Fig 3]. Gram staining showed colonies of actinomyces, confirming the diagnosis of actinomycosis. Treatment with high doses of penicillin (24 million units intravenously per day) was started and continued for six weeks. During intravenous antibiotic therapy, the infection gradually improved and so we also recommended oral penicillin for the next 8-12 months.

findings. In Iran, the first case of actinomycosis was identified in 1962 and about 34 cases from different anatomical sites have been reported so far [10]. In 1972, Barokhion and Bahadory from Iran found 28 cases of actinomycosis in a retrospective investigation of 149872 biopsy specimens. The infection was found to be located on the foot or hand (11 cases), abdomen (8 cases), thorax (1 case) and head and neck (8 cases) [11]. But our observations were in contrast to those from the above mentioned study, as in our area, actinomycosis of the upper and lower limbs was rare. Blinkhorn et al reported a patient with osteomyelitis of the distal right first metacarpal bone due to actinomyces israelii, following a punch injury during fisticuffs [12]. Mendelsohn reported a case of actinomycosis of the metacarpal bone in a sea-man. He also reviewed case reports of actinomycosis of the hand in six studies in which all patients were male, with an age range of 20-55 years [13]. Kumar et al reported actinomycosis of the thigh which mimicked a neoplasm, but on surgical removal, histopathological examination confirmed the diagnosis [14]. Vundeveld has also reported actinomycosis of the lower leg in a young man for whom amputation below the knee was done [15]. Both of our patients had noticed prior trauma to their extremities and had received multiple treatments with no response for a long period of time because of the undiagnosed disease, but responded well to penicillin after diagnosis. In a study by Smith et al (2005), the actinomyces species appeared to be susceptible to a wide range of beta-lactam agents and these, when combined with beta-lactamase inhibitors, should be regarded as agents of first choice. Ciprofloxacin and Tetracyclines demonstrated poor performance. There are a number of species which are different in their susceptibility profiles, which may have an impact on clinical outcome [17]. Some strains of *A. israeli* have been reported to be resistant to penicillin. Both of these reported cases of actinomycosis were negative for actinomyces and other bacteria, which could have added to the species identification. The chronic and indolent course of actinomycosis resembles tuberculosis, fungal infections and malignancy. Because actinomycosis is often difficult to diagnosis, it has been referred to as a forgotten or a misdiagnosed disease. So, a heightened gram staining were positive and staining for acid fast bacilli was negative and so the diagnosis of actinomycosis was made. The patient was treated twice with surgical debridment and also intravenous and oral penicillin for 6 months. The response to this treatment was excellent, but the foot deformity was permanent.



(Table/Fig 2) A- Actinomycosis in right hand; B- Radiograph of right foot that shows erosions of first metatarsal bone.



(Table/Fig 3) Tissue section shows prominent sulfur granules composed of colonization of filamentous bacteria surrounded by supportive inflammatory reaction in connective tissue.

Discussion

Actinomyces are branching (except *A. meyeri*), filamentous, gram positive rods that are normal commensals of the mouth and tonisillar crypts [7]. Primary actinomycosis of the extremities [8] is rare, because of the exclusively endogenous habitat of the aetiological organism . Since actinomycosis is usually, if not always, polymicrobial in nature involving mixed bacteria and the cultures are positive in only 24% of cases [9], the diagnosis is often based on histopathological

awareness among the clinicians and clinical microbiologists will help in the early diagnosis of the disease and the initiation of early and proper treatment.

Reference

[1] Russo TA , Kasper DL , Fauci AS, et al. Harrison's principles of internal medicine. 16th ed. New York: McGraw-Hill, 2005: 937-39.

[2] Rippon JH. Medical mycology: the pathogenic Fungi and the pathogenic Actinomycetes, lst ed. Philadelphia: WB Saunders, 1988; 97-103.

[3] Zaini F, Mehbod ASA, Emami M. Comprehensive medical mycology. 1st ed. Tehran: Tehran University press, 1999; 147-61.

[4] Fahal AH, Hassan MA. Mycetoma. British Journal of surgery 1992; 79: 1138–41.

[5] Baril L, Boiron P , Manceron V, et al. Refractory craniofacial actinomycetoma due to streptomyces somaliensis that required salvage therapy with Amikacin and Imipenem. Clinical Infectious Diseases 1999; 29: 460-61.

[6] Wilson DC, Redmond AOB. An unusual cause of thoracic mass. Arch Dis Child 1990; 65: 991-92.

[7] Moses J, Bonomo GF, Wenlund DE, Halldorddon TS ,Connelly JP. Actinomyces in childhood. Clin Pediatr 1967; 6: 221-26.

[8] Reiner SL, Harrelson JM , Miller SE , Hill GB, Gallis HA . Primary actinomycosis of an extremity: a case report and review. Rev Infect Dis 1987; 9: 581-9.

[9] Golden N, Cohen H, Weissbort J, Silverman S. Thoracic actinomycosis in childhood . Clin Pediatr 1985; 24: 646-50.

[10] Moghaddami M, Shidfar MR, Omidi K. Study of the etiologic agent of mycetoma and sporotrichosis in the North of Iran (Gilan and Mazandaran provinces). Med J I.R. Iran 1991; 5: 139-43.

[11] Barokhian E, Bahadory M. 28 cases of human actinomycosis: investigation of observed cases in pathology laboratories of Medical school and Pahlavi Medical center. Medical Journal of Tehran University of Medical Sciences 1972; 9: 346-53.

[12] Blinkhorn JR, Strimbu V, Effron D, Spugnuolo J. Punch actinomycosis causing osteomyelitis of the hand. Archives of Internal Med 1988; 12: 148.

[13] Mendelsohn B.G. Actinomycosis of a metacarpal bone: Report of a case .The journal of bone and joint surgery 1965; 47: 739-42.

[14] Kumar A, Detrisac DA, Krecke DF, Jimenez MC. Actinomycosis of the thigh presenting as a soft tissue neoplasm. J Infect 1991; 23: 187-90.

[15] Vandevelde AG, Jenkins SG, Hardy PR. Sclerosing Osteomyelitis and actinomyces naeslundii infection of surrounding tissue. Clin Infect Dis 1995; 20: 1037-9.

[16] Reddy I, D.A. Ferguson and F.A. Sarubbi, Endocarditis due to Actinomyces pyogenes. Clin Infect Dis 1997; 25: 1476–77.

[17] Smith AJ, Hall V, Thakker B, Gemmell CG. Antimicrobial susceptibility testing of Actinomyces species with 12 antimicrobial agents. J Antimicrob Chemother. 2005 Aug;56(2):407-9. Epub 2005 Jun 21.