

Primary Actinomycosis of the Knee and Ankle Joint: An Unusual Manifestation

Recep Tekin^{1*}, Mehmet Gem², Yeliz Arman Karakaya³, Vuslat Boşnak⁴ and Ahmet Kapukkaya²

¹Department of Infectious Disease and Clinical Microbiology, Faculty of Medicine, Dicle University, Diyarbakir, Turkey

²Department of Orthopedics and Traumatology, Faculty of Medicine, Dicle University, Diyarbakir, Turkey

³Department of Pathology, Faculty of Medicine, Dicle University, Diyarbakir, Turkey

⁴Department of Infectious Disease and Clinical Microbiology, Faculty of Medicine, Gaziantep University, Gaziantep, Turkey

Abstract

Actinomycosis caused by *Actinomyces* spp. is a chronic, progressive, suppurative and granulomatous bacterial infection. The general sites of infection are the head and neck, thorax and abdomen. Primary actinomycosis infection both on the knee and the ankle is uncommon. We report a very rare case of actinomycosis of the knee and ankle joint that was diagnosed and successfully treated with surgical resection and long-term antibiotic therapy.

Keywords: Actinomycosis; Knee; Ankle; Treatment

Introduction

Actinomycosis caused by *Actinomyces* spp. is a chronic, progressive, suppurative and granulomatous bacterial infection which is characterized clinically by multiple abscesses formation, recurrent draining sinuses, chronic fistulae and fibrous tissue formation. A variety of Gram-positive, non-spore-forming anaerobic or microaerophilic rods, primarily from the genus *Actinomycetes*, are responsible agents, with *Actinomyces israelii* being the most common one [1-3]. The major clinical forms of actinomycosis are cervicofacial, thoracic, abdominal, pelvic and systemic forms [4,5]. Primary bone and joint actinomycosis are very rare and usually associated with external trauma, punk injury and local ischemia [1,6,7]. In this article we report a very rare case of primary extremity actinomycosis presenting as osteomyelitis on both the knee and on the ankle of the left leg.

Case Report

A 90-year-old female presented with a seven month history of progressive, severe pain, swelling with erythema and intermittent discharging sinuses around the left knee and ankle joint. Physical examination revealed a stiff, moderately swollen and tender, left knee and ankle joint with multiple sinuses adherent to around the left knee and ankle joint (Figure 1). Eight years ago she had undergone an operation for her left femur fraction. Results of laboratory investigations revealed a white blood cell count of 5380/mm³, erythrocyte sedimentation rate of 56mm/h and a C-reactive protein of 101 mg/L (reference: 0 to 5mg/L). No other laboratory parameters were prominent. Radiographic studies of the left knee and ankle joint showed changes consistent with osteomyelitis, multiple lytic areas with surrounding sclerosis in the peri-articular bones, with reduction of the joint space and soft tissue swelling (Figure 2). Ampicillin/sulbactam was administered intravenously (4g/d) for 2 weeks. The patient was discharged on an outpatient basis, maintaining oral treatment with amoxicillin-clavulanate for 6 months. Considering the severe pain and inability to eradicate the disease lower thigh amputation was performed. The pathological examination was consistent with chronic osteomyelitis. (Figure 3). Patient was discharged on completion of treatment. One-year follow-up was done and there was no recurrence of disease.

Discussion

Actinomycosis is a suppurative and granulomatous chronic infectious disease, late diagnosis and less common that comes to mind. Ac-



Figure 1: Left knee with ankle joint showing a swelling and multiple discharging sinuses.

tinomycosis is believed to be acquired by endogenous implantation into deep tissues where anaerobic conditions prevail. The disease develops in tissue adjacent to the mucosal surfaces that harbour the microorganisms and human actinomycosis most frequently affects the face and neck, but may also be encountered in thoracic and abdominal sites [1,3,5,7,8]. *Actinomyces* osteomyelitis is a rare, recurrent disease of the skeletal system that most commonly effects chin. Extremity involvement is rare and usually occurs after trauma or open fractures. Initially it is 75% of the soft tissue of the disease, because of the trauma that is around 19%. In literature, cases involving finger, metacarpal and leg have been reported [4,9]. The majority of the cases reported have a clear history of trauma, either a human bite or a perforating injury with contamination from outside [4]. Primary actinomycosis of the knee and

***Corresponding author:** Dr. Recep Tekin, Department of Infectious Diseases, Faculty of Medicine, Dicle University, Yenişehir 21280 Diyarbakir, Turkey, Tel: +90 412 248 80 01- 4858; Fax: +90 412 248 84 40; E-mail: rectek21@hotmail.com

Received April 02, 2012; Accepted April 18, 2012; Published April 24, 2012

Citation: Tekin R, Gem M, Karakaya YA, Boşnak V, Kapukkaya A (2012) Primary Actinomycosis of the Knee and Ankle Joint: An Unusual Manifestation. J Bacteriol Parasitol 3:137. doi:10.4172/2155-9597.1000137

Copyright: © 2012 Tekin R, et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.



Figure 2: Radiograph of the left knee and ankle joint showing radiolucent areas and lytic lesion with sclerosis.

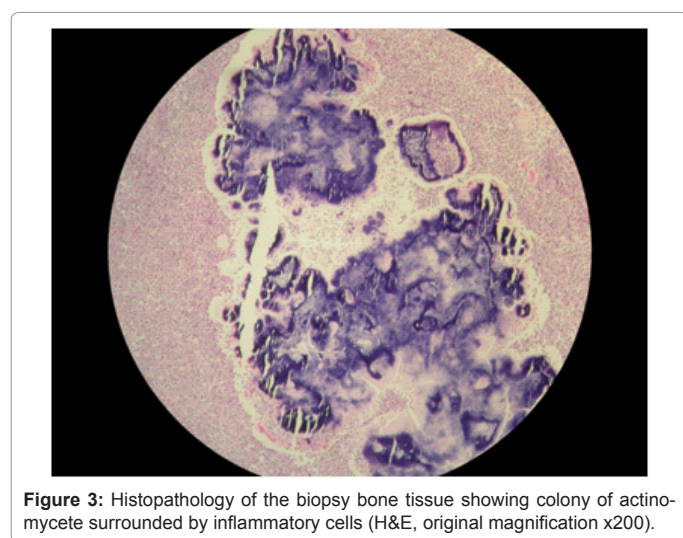


Figure 3: Histopathology of the biopsy bone tissue showing colony of actinomycete surrounded by inflammatory cells (H&E, original magnification x200).

ankle is very rare because of the exclusively endogenous habitat of the etiologic organism. Actinomycosis of the joint is usually due to adjacent spread of soft tissue infection but the infection may occur following after punch injury and trauma or it can be hematogenous spread. Infection of bones and soft tissue usually develops and may be due to the hematogenous spread in some cases [4,7,10].

In our case it had no systemic symptoms, catarrhal purulent wound in the knee and ankle but had a large fistula with reference to an amount higher than the white blood cell erosion. Multiple lytic areas with surrounding sclerosis in the peri-articular bones are observed. The presence of direct radiography helped our assessment of lesion. In our case the clinical, radiological and laboratory findings suggest chronic osteomyelitis and diagnosis was confirmed by pathological examination. Our patient was operated for hip replacement six years ago. Actinomycosis recurred six years after the operation and is extremely unlike present.

Skeletal actinomycosis has chronic indolent course and presents

with abscess and sinus tracts draining pus mixed with sulphur granules. Clinical diagnosis of actinomycosis is generally supported by the Gram-positive bacilli on Gram stain and confirmed by showing a clump of filamentous bacilli surrounded by neutrophils in crushed sulphur granules and granulomatous reaction on histopathology [1,2,4].

The histopathological examination of the lesions is significant to support the pathology by demonstrating the showing of the organisms and attendant inflammatory change [1]. Discharge of sulphur granule is a characteristic feature helping the clinical diagnosis and its conformation by demonstrating the filamentous bacilli. According to some authors, the characteristic sulphur granules, in the specimens, are present in only 35-55% of cases. In these cases, the diagnosis is definitive [4,7].

Radiographs are helpful to determine the extent of the infection, although they are not diagnostic. Radiological features of actinomycosis include both destruction and formation of bone, manifesting as multiple lytic lesion with sclerosis and periosteal reaction [4,5,9]. As the disease spreads to bone, the earliest changes include periosteal reaction with loss of cortical margins, often followed by erosion and adjacent sclerosis. Computed Tomography (CT) or Magnetic Resonance Imaging (MRI) can be helpful to determine the full extent of bony involvement and to delineate soft-tissue involvement [2].

The main treatment principle in skeletal actinomycosis is to use long-duration intensive antibiotic treatment. The optimal duration of total antibiotic therapy for thoracic actinomycosis, as well as the optimal duration of intravenous antibiotic therapy, has not been thoroughly evaluated, although a total duration of 6-12 months is generally recommended [1,2,4,7,10]. A combined medical-surgical approach is frequently required for actinomycosis. In the majority of cases and also in the present case, the therapy consisted of surgical debridement combined with prolonged antibiotic therapy. With this approach, a successful outcome was attained in every case [6,7,10].

In the pre-antibiotic period, surgical removal of infected tissue was the only treatment. Despite the advent of efficacious antimicrobial therapy, combined surgical therapy is still advocated. Although recommendations for incision and drainage of abscesses, fistulotomy, sinus tract excision, and more extensive debulking of infected tissue are made, it is unclear how often these procedures are necessary. Furthermore, percutaneous drainage of well-localized abscesses has become an additional option [3,7]. Surgical treatment is often indicated for curettage of bone, resection of necrotic tissue, excision of sinus tracts, drainage of soft tissue abscesses. Surgery plays a significant role both in the treatment and diagnosis of actinomycosis [5].

The clinical experience with actinomycosis has been extensive and supports the use of penicillin G and ampicillin as the drugs of choice for all clinical forms of the disease. For penicillin-allergic patients, tetracycline, erythromycin, and clindamycin are suitable alternatives. Owing to the potential for relapse of actinomycosis, prolonged antibiotic treatment is prudent. Penicillin should be administered parenterally in high doses initially. Oral therapy should be continued for an additional 6 to 12 months depending on the original site of infection and the clinical response [7].

Actinomycetes rarely effects the knee and ankle. In these cases if there is no response to antibiotics, amputation may be required to control the infection. Awareness of clinical symptoms of this disease, early diagnosis and treatment, reduces the rate of surgical treatment, mortality and morbidity.

References

1. Metgud SC (2008) Primary cutaneous actinomycosis: A rare soft tissue infection. *Indian J Med Microbiol* 26: 184-186.
2. Foltz KD, Fallat LM (2004) Madura foot: atypical finding and case presentation. *J Foot Ankle Surg* 43: 327-331.
3. Kargi E, Akduman D, Güngör E, Deren O, Albayrak L, et al. (2003) Primary extremity actinomycosis causing osteomyelitis of the hand. *Plast Reconstr Surg* 112: 1495-1497.
4. Kumar A, Varshney MK, Trikha V, Khan SA, Yadav CS, et al. (2008) A rare actinomycosis of humerus: an unusual location and a diagnostic dilemma. A case report. *Arch Orthop Trauma Surg* 128: 121-124.
5. Robinson JL, Vaudry WL, Dobrovolsky W (2005) Actinomycosis presenting as osteomyelitis in the pediatric population. *Pediatr Infect Dis J* 24: 365-369.
6. Espina B, Fariñas MC, Matorras P (2004) Primary actinomycosis of the humerus: a quite unusual form. *Am J Med* 116: 785-786.
7. Smego RA Jr, Foglia G (1998) Actinomycosis. *Clin Infect Dis* 26: 1255-1261.
8. Song JU, Park HY, Jeon K, Um SW, Kwon OJ, et al. (2010) Treatment of thoracic actinomycosis: A retrospective analysis of 40 patients. *Ann Thorac Med* 5: 80-85.
9. Kundu ZS, Bhardwaj G, Sangwan SS, Arora B (2003) Actinomycosis of the knee: a case report. *J Orthopaed Traumatol* 4: 133-135.
10. Mert A, Bilir M, Bahar H, Torun M, Tabak F, et al. (2001) Primary actinomycosis of the hand: a case report and literature review. *Int J Infect Dis* 5: 112-114.