Case Report

Actinomycosis of the ankle joint

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Keywords: *Actinomycosis, Ankle Joint, Chronic Synovial İnfection, Cotrimoxazole, Rifampicin.*

Abstract

We report a case of actinomycosis of the ankle joint with synovial involvement which resisted the usual treatment. Actinomycotic infection of the extremities involving the bone and joints is rare. After the initial diagnosis, our case was treated with ampicillin for six months with no benefit. Instead the condition worsened with the development of tiny pockets of fluid collections within the soft tissue and increased inflammation. Surgical debridement was done to drain the fluid collections and the patient was treated with combined regimen of Cotrimoxazole and Rifampicin with good response clinically and radiologically. Timely intervention and the correct diagnosis saved the limb.

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Date of Submission: May 29, 2014 **Date of Acceptance:** June 13, 2014 **Date of Publishing:** July 20, 2014

How to cite this paper:
Ramachandra S, Kuruvila S, John CK. Actinomycosis of the ankle joint. Annals of Pathology and Laboratory Medicine. 2014;1(1):38-40

1. Introduction

Actinomycosis is a chronic suppurative and granulomatous, slow growing infection.[1] Cases of synovial actinomycosis have been reported following arthroplasty, as involvement of musculoskeletal system is rare.[2] Synovial infection of the ankle joint is uncommon and our case resisted the usual treatment. The condition responded to cotrimoxazole, a drug stated as not active against actinomyces and rifampicin, an antitubercular drug administered in actinomyces of the central nervous system (CNS).

2. Case Report

A 31-year-old male patient presented with complaints of swelling and pain in the right ankle for the past six months. Swelling preceded the pain. Pain gradually increased, became intolerable disturbing his routine activities and was not able to walk without support. He had some relief with pain killers and was treated at various hospitals with no improvement in his condition before presenting at our hospital. CT and MRI done elsewhere showed extensive ankle joint synovitis and effusion with pannus formation, erosions of the talus and superior part of the calcaneus with features consistent with inflammatory arthritis. The signal intensity of the pannus was not typical of pigmented villonodular synovitis (PVNS) or gout. Fever, trauma or family histories of arthritis were negative and past history was nothing contributory. He is a soldier rendering his services in the armed forces. Hematological profile and ESR (erythrocyte sedimentation rate) were normal with mild increase in CRP (C-reactive protein). With this background he received three weeks of treatment in our hospital following which his symptoms reduced and subsequently synovial tissue was biopsied for histopathological assessment. The microscopic examination of the synovial tissue showed nodular aggregates of chronic inflammatory cells and micro abscess formation. Many colonies with eosinophilic matrix with radiating filaments at the periphery were noted (Fig. 1 & 2). These filaments were PAS (periodic acid schiff), GMS (gomori's methamine silver) (Fig. 3 & 4) and gram's stain positive. Modified Ziehl-Neelsen stain showed negative filaments which ruled out *nocardia* species (Fig. 5). Synovium showed neovascularisation and oedema. The histopatological diagnosis of synovitis with actinomycotic colonies was rendered.

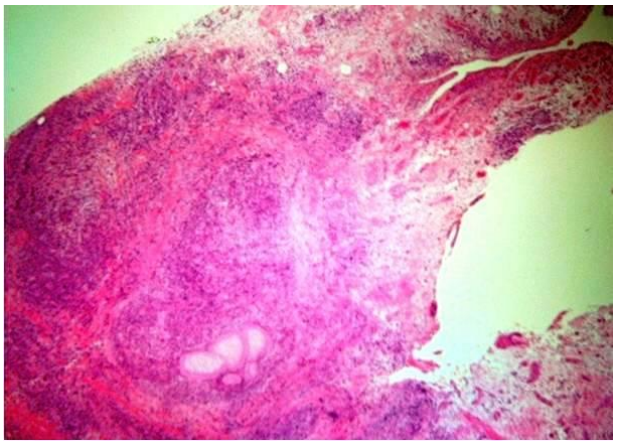

Figure 1: Actinomycotic colonies, abscess and inflammation (H&E, x4)

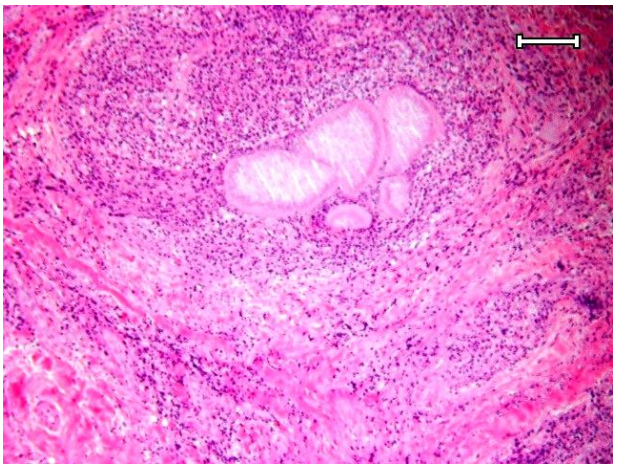

Figure 2: Actinomycotic colonies (H&E stain, x10)

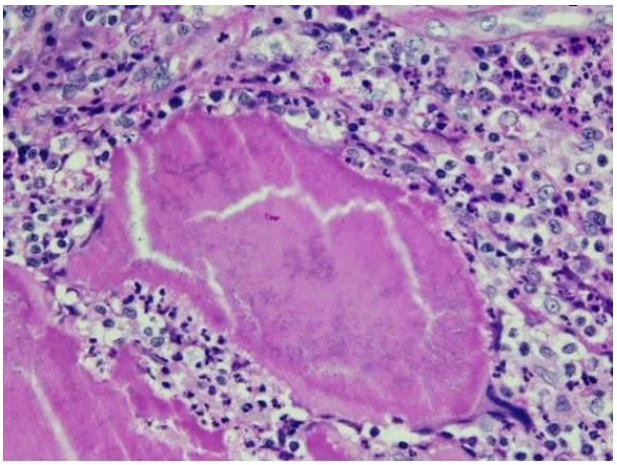

Figure 3 : Periodic acid Schiff (PAS) positive filaments of actinomyces (PAS, x40)

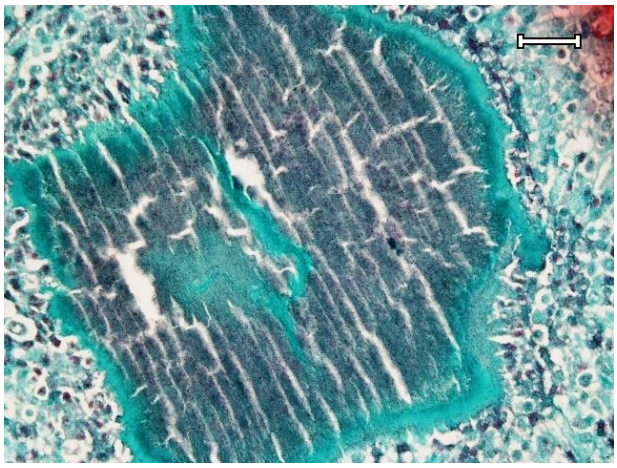

Figure 4 : Gomori's methamine silver (GMS) positive filaments (GMS, x40)

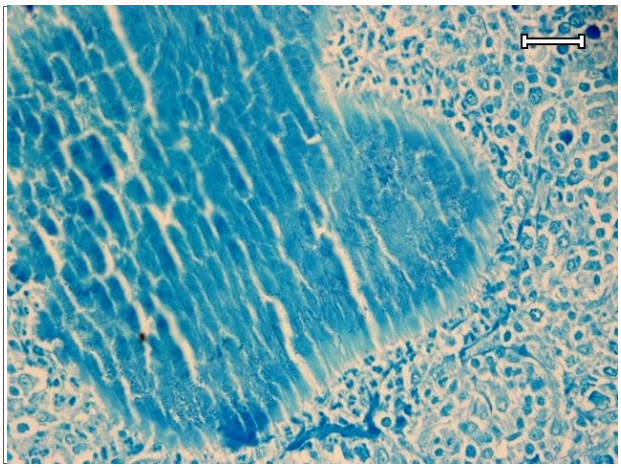

Figure 5 : Modified Ziehl Neelsen stain, negative acid fast filaments (ZN stain, x40)

With this diagnosis, he was treated initially with intravenous rocephin followed by oral ampicillin. Later on cotrimoxazole was combined with ampicillin. He improved clinically, but remained symptomatic with ankle swelling and pain. Ten months after treatment follow-up MRI showed soft tissue inflammation with multiple fluid collections and calcaneal osteomyelitis with no significant interval change of the infective/inflammatory process. With the help of dermatological consultation it was decided for a second surgical debridement of the inflamed tissue and drainage of the fluid collections. The histopathology showed similar colonies as observed in the first biopsy. The patient was now started with cotrimoxazole (800+160mg) and rifampicin (600mg). He has been on this combination drugs for the past one year with marked clinical improvement, healed sinuses and resolution of the bony and soft tissue edema as compared to his previous images in the repeat MRI.

3. Discussion

Actinomycosis is an indolent slowly progressive granulomatous lesion characterized by external sinuses and discharging granules.[2] They are an important endogenous constituent of the commensals found on the mucous membranes of the tracheobronchial tree, oral cavity and within the crevices of the carious teeth or tonsils.[3] They become pathogenic and cause infection after trauma, surgery or aspiration.[4]Their slow chronic course of the disease resembles fungal infection, tuberculosis or even malignancy.[4] Hence called as the most misdiagnosed infection.[5] The common sites of infection are cervicofacial, thoracic, abdominal, pelvic, primary cutaneous and CNS.[2,6] Involvement of the extremities with infection of bone and joint is uncommon.[2]Synovial actinomycosis and involvement of the ankle joint is rare. In our case, source of endogenous infection is unlikely suggesting an exogenous source. Cases of actinomycosis associated with knee and hip arthroplasty have been reported.[5,7] The infections that occur following arthroplasty are classified as early, delayed and late infections.[7] The early and delayed infections are those that are acquired during implantation and late infections present after a year or later. Bone and joint infections are usually involved by spread of adjacent soft tissue infection (75%), hematogenous spread (3%) or direct inoculation through trauma (19%).[8] A few cases have been reported in intravenous drug users.[9]

The genus *Actinomyces* consists of several species of anaerobic and microaerophilic gram positive non-spore forming organisms.[5] There are different species; *A. bovis*, *A. israelii*, *A. propionicus*, *A. naeslundii, A. eriksonnii and A. nevii*.[3] They have been classified on the basis of rRNA molecular and biochemical analysis.[7] A few preformed enzyme detection panel are available for identifying certain species of *Actinomyces*. The best growth is obtained by anaerobic culture on blood agar at 37^0C.[7] Atleast 72 hours are required to isolate the strain and several weeks to identify the colonies.[3] In our case the species was not identified.

Diagnosis can be established by microscopic examination of the purulent material or histopathological examination of the tissue. Purulent material or granule shows Gram positive branching fungal hyphae. The biopsy shows actinomycotic colonies with radiating filaments surrounded by inflammation. These filaments are Gram's, PAS and GMS positive but negative for acid fast staining thus differentiating it from its similar histomorphologic counterpart, nocardiosis whose filaments are acid fast positive. They are usually known to be associ-

ated with other gram negative organisms that have a synergistic role initiating actinomycotic infections and are penicillin resistant.[3,6] Granulomatous reaction with multinucleated foreign body giant cells along with lymphocytes and plasma cells are noted. Synovial tissue shows mild increase in mast cells.[8] Management includes both surgical and medical. Surgical drainage of the sinuses and curettage of the inflamed tissue is the mainstay of the treatment followed by antibiotics. This chronic infection elicits marked fibrotic reaction making difficult for drug penetration and unable to achieve the desired serum levels of the antibiotics delaying response to treatment.[6] Penicillin is the drug of choice. Oral amoxicillin and ampicillin are used. Tetracycline is used in patients allergic to penicillin and erythromycin is used in pregnancy. The above mentioned drugs are the first-line drugs which includes first-generation cephalosporins and clindamycin.[9] Cotrimoxazole is supposedly stated as a drug not active against *actinomyces*.[10] However, serum concentrations of sulphonamides (4-8mg/dL) inhibit some strains of *A. isrealii* therefore may occasionally respond to sulphonamides.[9] Rifampicin is used in CNS infections which helps in good penetration of the drug across the blood brain barrier.[1] It is an anti-tubercular drug also known to have antibacterial effects against anaerobic bacteria.[6] In combination with erythromycin, it is considered as the most active drug against *actinomyces* in vitro.[1] Our case resisted the usual treatment and responded to cotrimoxazole which suggests the infecting species belongs to one of the strains of *A. israelii* and rifampicin has probably acted against the associated anaerobic organisms that resisted the initial treatment with ampicillin and might have helped in penetration of the drug across the synovial membrane.

4. Conclusion

Actinomycosis of the synovium is rare, resisted the actual treatment course and responded to cotrimoxazole (a drug usually not active against *actinomyces*) and rifampicin (an antitubercular drug). Timely intervention and the two unusual antibiotics through their unique pharmacological properties saved the limb.

Acknowledgements

None.

Funding

None.

Competing Interests

None declared.

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