We present a rare case of fungal osteomyelitis of tibia and fibula with extensive involvement of ankle, intertarsal and tarsometatarsal joints leading to bony ankylosis of all joints over a four years duration. Diagnosis was confirmed with histological and microbiological examinations, which showed Aspergillosis osteomyelitis. The patient was treated with Amphotericin B for 6 weeks. The outcome after the therapy was good after adequate follow up. The clinical, pathologic, and therapeutic features of aspergillosis osteomyelitis are described and compared with previously published cases.

Introduction

Fungal arthritis and osteomyelitis are uncommon diseases and generally present in an indolent fashion. The incidence of fungal bone and joint infections is increasing with an increase in the prevalence of factors predisposing to invasive fungal disease, such as the use of central venous catheters, broad spectrum antibiotics, immunosuppression, and abdominal surgery. Definitive diagnosis relies on bone or synovial culture or biopsy. Successful management has traditionally consisted of amphotericin B in combination with surgical debridement. Given the rarity of this disease, treatment is not well defined, but reports of success with the use of azole antifungal agents, including itraconazole, fluconazole, voriconazole, and posaconazole, are promising.

Case Presentation

A 42 year old male farmer from Homtang-1, Bhojpur an Eastern remote village of Nepal presented to the Orthopaedics OPD of B P Koirala Institute of Health Sciences, Dharan, with multiple discharging sinuses and swelling of the left leg and foot of 4 years duration. The swelling of the left leg was followed by the multiple discharging sinuses. These symptoms were not associated with constitutional symptoms like fever, cough, or chest pain. Definitive treatment was not taken for the disease till he visited this hospital. On examination, there were multiple discharging sinuses on the distal part of left leg with ulceration and multiple scabs. (Fig 1) There was no movement at the ankle, subtalar and tarsometatarsal joints. Radiographs of the leg, ankle and foot showed features of osteomyelitis of tibia and fibula along with bony ankylosis of ankle, intertarsal and tarsometatarsal joints. (Fig. 2, 3,) An incisional biopsy was done and culture confirmed fungal infection with Aspergillus flavus. With this Amphotericin B was started according to body weight, for 6 weeks. After follow up of 6 months, the discharging sinuses and skin condition was improved with painless plantigrade foot and sound ankylosed ankle and subtalar joints.

Discussion

Holtom et al described mucormycosis, which is an uncommon but highly aggressive fungal infection most commonly occurring in hosts who are immunologically depressed. Holtom et al reported an unusual case of mucormycosis osteomyelitis...
developing in a patient who was immunocompromised, after routine tibial Steinmann pin placement for the application of traction. In his case, surgical debridement and amphotericin B were not sufficient to control the infection, and the patient subsequently underwent above-knee amputation. This is the first description of mucormycosis causing osteomyelitis as a result of Steinmann pin tract infection.

Alvarez et al. presented an unusual case of osteomyelitis of the tibia and septic arthritis of the knee caused by *Aspergillus fumigatus* in a renal transplant recipient. Although the characteristic joint involvement was present, the synovial fluid was initially sterile and contained numerous pyrophosphate crystals.

De Vuyst D et al. have reported a case of osteomyelitis involving the proximal epiphysis of the left tibia in a heart transplant patient, caused by *Aspergillus flavus*. History revealed a previous pretibial wound due to a fall in the street as the consequence of a sudden cardiac arrest. Surgical debridement combined with fungostatic treatment including amphotericin B and itraconazole was followed by clinical improvement, although the fungus could still be recovered by culture on subsequent samples.

Corrall CJ et al. have treated a black male adolescent with intact cellular and humoral immunity who had developed *Aspergillus flavus* caused osteomyelitis involving the right tibial epiphysis following penetrating injury to that area. Following amphotericin B therapy for six weeks, the patient was apparently cured. This case report represents the first well documented case of Aspergillus osteomyelitis in an immunocompetent host. The increasing incidence of invasive disease due to Aspergillus species and the increased awareness of the incidence of mycotic bone infections, particularly in pediatric patients, may allow further definition of pathogenesis and appropriate therapy.

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In our case, the patient presented at the late stage of osteomyelitis and arthritis of joints affected by *Aspergillus flavus*, but the outcome of treatment was satisfactory. As prompt diagnosis and start of treatment was done, the morbidity of the disease was much decreased.

**Reference**


**Fig. 2** Ankylosis of ankle, subtalar and metatarsotarsal joints of patient with *aspergillus flavus* osteomyelitis

**Fig. 3** Diaphyseal osteomyelitis from *aspergillus flavus*