CASE REPORT

# Fungal Osteomyelitis of Cuboid Bone Case Report with Literature Review

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## Abstract

**Background:** Chronic osteomyelitis with sinus formation of right cuboid bone has often been missed, which leads to the morbidity of the patient.

**Case description:** Here, we present a case of a 29-year-old male with fungal involvement of the cuboid bone. Fungal osteomyelitis of cuboid bone is of an unusual occurrence. Delayed diagnosis is one of the prime concerns leading to the progression of the disease and further bone involvement. In our study, we did a prompt diagnosis which changed the progression and treatment of the disease.

**Conclusion:** Early diagnosis and prompt treatment with surgical intervention, antifungal medications, and a multidirectional approach is the key in treating cases of fungal osteomyelitis for immunocompetent and immunocompromised fungal infection.

**Keywords:** Case control study, Cuboid bone, Fungal infection, Fungal osteomyelitis, Immunocompetent fungal infection.

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## Background

Osteomyelitis and avascular necrosis of bone are opportunistic infections that may result from mucormycosis.[1] The course of the disease is initially similar to any other infective etiology making the diagnosis unlikely. In cases where the diagnosis is promptly delayed, as seen in most of the cases, the natural course of the disease varies positively, leading to further damage of the bone architecture and eventually leading to necrosis of the involved bone.[1] If a broad mindset with early diagnosis is followed by prompt treatment, favorable prognosis is seen.[2]

The main difficulty in such cases are lack of awareness, delayed patient presentation, improper diagnosis, delayed diagnosis, lack of treatment protocols in such unusual cases, which all together affect the prognosis of the disease.

Our case study presents such an unusual case report with delayed presentation to the tertiary institute, but with proper investigations and regular follow-up, we came to a proper diagnosis, followed by a staged plan of care, and we came on with a favorable outcome.

A 29-year-old male patient presented to orthopedics outpatient department at Sri Aurobindo Medical College and PG Institute with chief complaints of pus discharging sinus over the lateral side of midfoot for the past 6 months (Fig. 1). Patient had a history of trauma due to a prick by a nail 1 year back, following which he was managed conservatively elsewhere.

Patient was alright for 6 months, then he started complaining of pus discharge, for which he consulted in various private hospitals and serial debridement were done elsewhere, but the patient was not relieved from the symptoms. Preoperative magnetic resonance imaging (MRI) (Fig. 2) is suggestive of osteomyelitis changes of the right cuboid bone.

Keeping in mind the MRI report, we planned for debridement with curettage with antibiotic cement beads insertion (Fig. 3) under spinal anesthesia to overcome the infection. Initially, we also concluded a pyogenic etiology for which a broad-spectrum of intravenous antibiotics was used.

But in spite of intravenous antibiotic and antibiotic beads insertion sinus didn't heal and discharge continued, so we suspected tubercular or fungal pathology as the infection was not spreading aggressively like in cases of pyogenic infection.

So, we planned for antibiotic beads removal (Fig. 4) and sample were sent for culture sensitivity and cartridge-based nucleic acid amplification test, which was suggestive of fungal hyphae (*Mucor* species).

Patient was started on antifungal as per the culture sensitivity report and an alternate day dressing of the wound was done.

Delayed suture removal was done on postoperative day 15 and patient has been walking full weight bearing since then with no active discharge (Fig. 5).

On 1 ½ month follow-up (Fig. 6) the patient has no complaints of pain or pus discharge and is walking full weight bearing without the support and is able to perform his daily routine activity (Table 1).

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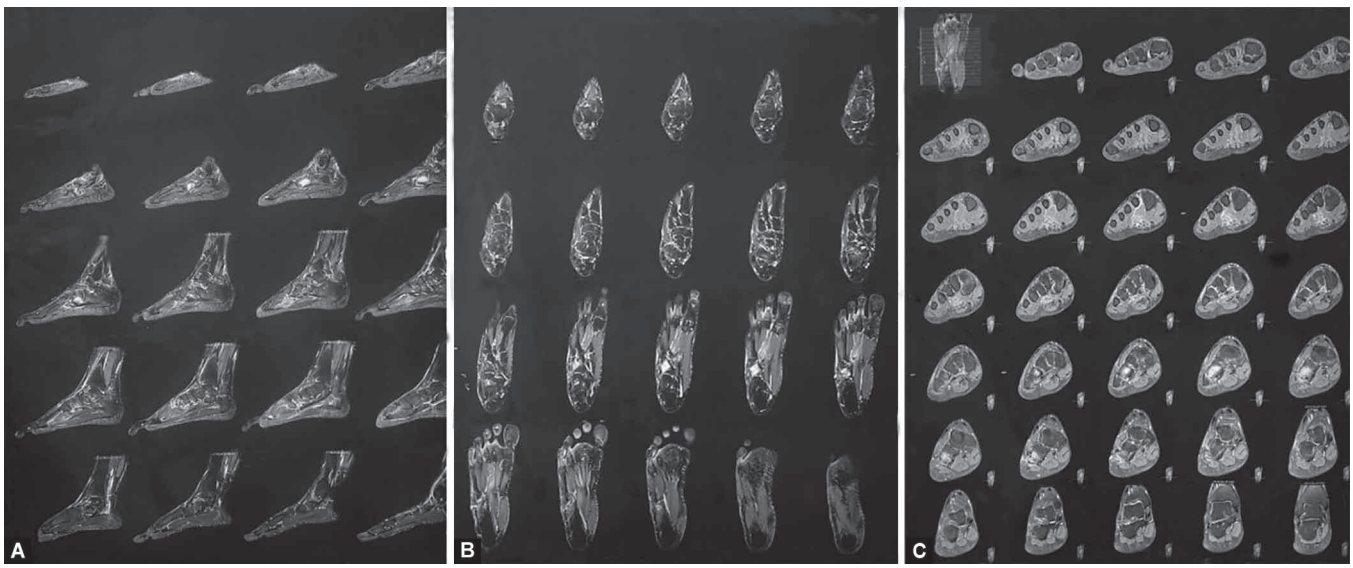
**Patient consent statement:** The author(s) have obtained written informed consent from the patient for publication of the case report details and related images.


**Figs 1A and B:** Preoperative clinical Image and X-ray




**Figs 2A to C:** Preoperative MRI


**Fig. 3:** Postbead insertion X-ray

## Discussion

Bone mycotic infection is uncommon. Many fungi can damage bones, but *Coccidioides* and *Blastomyces* species are the most frequent offenders.[1] These fungi are found in the soil and are spread through inhalation. A minor respiratory infection is the first symptom of the disease. A tiny fraction of these infections develops into a systemic illness, which can cause granulomatous lesions in cancellous bone and occasionally even necessitate amputating the diseased limb.[1]

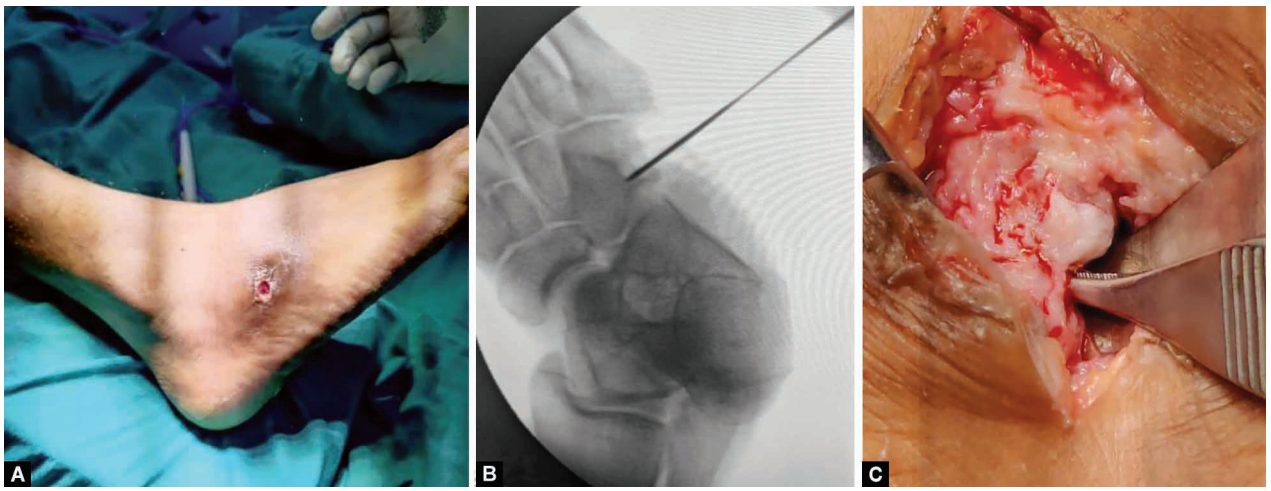
As they affect both immune-normal and immune-impaired hosts, cryptococcal skeletal infections are more common in men and afflict people of all ages.[3] From an asymptomatic patient with an aberrant finding on a routine radiograph to a patient exhibiting signs and symptoms of a systemic infection, the disease spectrum at the time of presentation is wide. The usual patient complains of soft tissue pain and swelling, and the gram stain of a straightforward closed aspiration is frequently used to make a diagnosis.[3] Surgery alone, systemic antifungal medication alone, or a combination of both methods can all be used to cure a patient. Some patients are reportedly treated by surgical excision alone, while other individuals are reportedly diagnosed with meningitis after being thought to be cured by surgery.[3] It can be challenging to distinguish between a patient with cryptococcal skeletal illness and clinically undetectable spreading. Radiographic information may help with diagnosis, but

because resolution takes time, it is difficult to determine how long therapy should last. Serologic markers have not been demonstrated to be effective in either monitoring of therapy or stratification of disease in cryptococcal skeletal infections, in stark contrast to their utility in cryptococcal meningitis.[3]

Maduromycosis is a type of fungal osteomyelitis common in tropical and subtropical regions and may be brought on by many fungi species. The most prevalent infection site (Madura foot) mainly affects unsheathed agricultural laborers.[1] After an often-minor injury, the fungus can develop, and its early stages are sometimes overlooked. The foot may already have a large number of discharge sinuses and severe underlying necrosis at the time of initial presentation.[1] Amputation is necessary for severe damage and early lesions.

Although the rhinocerebral region is the most frequently affected area by mucormycosis, pulmonary, gastrointestinal, and cutaneous variants have also been reported. In the past 30 years, there have been very few occurrences of osteomyelitis of an

**Fig. 5:** Postbead removal X-ray




**Figs 4A to C:** Intraoperative images

**Table 1:** Literature review of similar studies

| *S. no* | *Author* | *Year of study* | *Sample size* | *Location* | *Conclusion* |
|---|---|---|---|---|---|
| 1 | Chaudhari et al.[2] | 1992 | 1 | Cuboid bone | Rapid diagnosis, therapy and appropriate surgery therapy could be offered to patient. Nonetheless, the exemplary and rapid response to treatment in this case is unusual in this disease. |
| 2 | Shaw et al.[1] | 1994 | 1 | Cuboid bone | Mucormycosis very occasionally causes osteomyelitis and avascular necrosis of bone. If early treatment is instituted the prognosis is good. |
| 3 | Peene et al.[5] | 1998 | 2 | Cuboid bone | MRI diagnosed osteomyelitis of the cuboid bone when conventional X-rays were negative. Osteomyelitis of the cuboid bone has been reported in adults with diabetes and in mucormycosis. |
| 4 | Dinasarapu et al.[6] | 2010 | 1 | Calcaneum | Mucormycosis should be considered as one of the pathogens in patients with diabetes and ketoacidosis and extensive soft tissue infections. Diagnosis, prompt surgical treatment, and inclusion of appropriate antifungal therapy. |
| 5 | Pattanashetty et al.[7] | 2013 | 1 | Great toe | Isolated Aspergillus osteomyelitis of the bone are very rare. Treatment with wound debridement and with a course of amphotericin B for 6 weeks provided good results. Fungi should be kept in mind for differential diagnosis of osteomyelitis and culture should be appropriately ordered. |


**Figs 6A to C:** A1.5-month follow-up

extremity reported. Each of these patients had a predisposing condition, such as burns or renal failure, and the disease developed as a result of mild skin injuries. Avascular necrosis develops because local infiltration and vascular invasion are easily caused by the fungus, which causes little tissue reaction.[4]

Such cases of infection that were not properly treated highlight the value of an early diagnosis, which is typically accomplished through the presence of hyphae in a biopsy specimen. Even from infected tissue, the Mucoraceae is highly challenging to culture, and it may take weeks to get a result.[4] In our case, MRI was helpful in determining the soft tissue condition, and culture was necessary for a certain diagnosis. The preferred treatment today is amphotericin B therapy, followed by extensive surgical resection. Because of its negative side effects, amphotericin B must be used with caution.

We anticipate seeing more instances of fungal osteomyelitis recognized as a concurrent or late manifestation of hematogenous spread as patients with a high risk of severe fungal infections become more prevalent. The pathophysiology, clinical signs and symptoms, radiographic findings, and therapeutic outcomes resemble those of the stealthily progressing bacterial osteomyelitis quite a little.

The above table shows different case reports of cuboid osteomyelitis, which enlightens the unusual occurrence of the disease of tarsal bones with limited cases all over the world in the past few decades.

In conclusion, early diagnosis and prompt treatment with surgical intervention and antifungal medications is the key in treating such cases. Also, the surgeon should have a multidirectional approach for diagnosing such unusual pathologies.

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