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Case Report

Clinico-radiological improvement in an immunocompetent patient presented with scedosporium apiospermum osteomyelitis

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ABSTRACT

Scedosporium apiospermum is frequently found as a saprophyte in soil, sewage and contaminated water. Its manifestations in immunocompetent patients are usually localised and in immunodeficient patients, it causes invasive systemic diseases. We are reporting the case of a 40-year-old male, who presented with pain, oedema and multiple discharging sinuses over the lateral aspect of the left foot with history of thorn prick. On examination, there were multiple active sinuses with tenderness and local rise in temperature. Calcaneum on palpation showed a thickened and irregular surface with limitation of ankle and subtalar movements. Blood investigations showed a moderate rise in ESR and CRP. X-rays showed typical features of chronic osteomyelitis with sclerosis, cavities and irregular bone contour. CT report showed features of osteomyelitis involving calcaneum, talus, cuneiforms and navicular bone with periarticular soft tissue involvement and mild focal collection in the calcaneum. On repeated culture, it was found to be scedosporium apiospermum fungus. We managed the case with voriconazole therapy and it responded with excellent clinical and radiological improvement by 9 months.

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1. Introduction

Scedosporium apiospermum is frequently found as a saprophyte in soil, sewage, manure and contaminated water.¹ Its manifestations in immunocompetent patients are usually localised. Local manifestations usually follow minor trauma with splinters or thorns and cause invasive systemic diseases in immunodeficient patients.¹ We are reporting a case of scedosporium apiospermum osteomyelitis of the calcaneum.

2. Case report

A 40-year-old male presented with multiple discharging sinuses over the lateral aspect of the left hind foot. He was a manual labourer without any past history suggestive of immunodeficiency. He had a history of thorn prick over the plantar aspect of left foot 6 months back. Following this, he developed pain and oedema of the left foot. It was a pricking type of pain, gradually increasing in character without

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Fig. 1 – Image of left foot showing diffuse oedema with two discharging sinuses over the lateral aspect of the left calcaneum.

radiation of pain. Oedema was pitting in type, which gradually aggravated. Three months back, a sinus appeared over the lateral aspect of the left hind foot with serous discharge. Discharge aggravated gradually with a yellowish fluid.

On examination, mild local rise in temperature and tenderness were present. There was a diffuse oedema of the left foot with two discharging sinuses over the lateral aspect of the calcaneum [Fig. 1]. One was 3×2 cm and the other 1×1 cm size with induration of the surrounding skin. Margins were indurated with a reddish brown discoloration. Sinus base was not fixed to the bone. On applying pressure, there was increased discharge with associated tenderness. Calcaneum was found to be irregular and thickened on palpation. Range of ankle movement was 15° dorsiflexion and 20° plantar flexion actively and 20° dorsiflexion and 25° plantar flexion passively. Subtalar movements were severely limited both actively and passively. Midfoot movements were normal. He had inguinal lymphadenopathy but no limb length discrepancy or distal neurovascular deficit. So we suspected bacterial osteomyelitis and managed accordingly.

Fig. 2 – X-ray (lateral view of ankle) showing typical features of chronic osteomyelitis of calcaneum with sclerosis, cavities and irregular bone contours.

Blood investigations showed haemoglobin to be 12.1, total count 12,000 and a differential count of 60% polymorphs and 25% lymphocytes. ESR was 42 mm per hour and CRP was 12. Liver and renal function tests were normal. RBS normal, HIV and other screening tests were negative. Investigations showed normal immune status.

On radiological evaluation, X-ray (Fig. 2) showed typical features of chronic osteomyelitis with sclerosis, cavities and irregular bone contours. The CT images (Fig. 3a and b) showed features of osteomyelitis like osteopenia, lytic areas and cortical irregularities involving calcaneum, talus, cuneiforms and navicular bone with enlarged peri-articular soft tissue shadow and lytic hypodense areas in calcaneum.

Culture from the sinus track showed mixed growth and we embarked on the antibiotics. We planned curettage and biopsy, whose culture reported as *scedosporium apiospermum* osteomyelitis. Through the microscope (Fig. 4a and b) it is commonly seen as cylindrical conidiogenous cells arising from branching septate hyphae ($2-4 \mu\text{m}$ in diameter) producing slimy masses of conidia¹ [unicellular, smooth, and oval, which may be found in clusters-graphium state].

We treated him by intravenous injection of voriconazole 6 mg/kg (360 mg) 12th hourly for a day followed by 4 mg/kg (240 mg) 12th hourly for 20 days.² After starting voriconazole pain, oedema and discharge decreased in two weeks and sinus started healing. After the initial intravenous management, voriconazole was maintained as oral dose 200 mg 12th hourly for 2 months. By 3rd month [Fig. 5a and b] sinuses healed, and cavities in the calcaneum as well as sclerosis started improving radiologically. By 9th month [Fig. 6a and b] we found excellent outcome with nearly normal soft tissue and completely healed sinus, while radiologically, cavities were obliterated with regular bone contours and normal bone density of calcaneum. The patient started his work by 10th month.

3. Discussion

Scedosporium infections are usually opportunistic, ranging from colonisation in cystic fibrosis patients to neurological affections in drowning cases and disseminated diseases in

Fig. 3 – (a) Sagittal CT image, with soft tissue window width 400. (b) Axial CT image, with bone window width 1500: showed features of osteomyelitis like osteopenia, lytic areas and cortical irregularities involving calcaneum, talus, cuneiforms and navicular bone with enlarged peri-articular soft tissue shadow and lytic hypodense areas in calcaneum.

Fig. 4 – (a, b) Microscopic images showing scedosporium apiospermum in graphium state.

Fig. 5 – (a, b) By the 3rd month clinical image showed healed sinus. Lytic hypodense areas and sclerosis started improving radiologically (lateral view of the ankle).

immunocompromised individuals.³ The genus *scedosporium* contains two clinically significant species *scedosporium apiospermum* and *scedosporium prolificans*. *Scedosporium apiospermum* is the asexual state of its teleomorph, *Pseudallescheria boydii*. Local manifestations in immunocompetent patients include subcutaneous infections, septic arthritis, osteomyelitis and sinusitis. Invasive systemic manifestations in immunodeficient patients include pulmonary, brain abscess, endocarditis,

peritonitis, etc.¹ An extensive literature search revealed very few cases of *scedosporium apiospermum* osteomyelitis affecting immunocompetent patients.

Mycetoma is a clinical condition affecting the skin and subcutaneous tissues, fascia, bone and joints of the foot and is caused by soil-inhabiting bacteria (*actinomycetoma*) or fungi (*eumycetoma*).³ *Scedosporium mycetoma* is common in temperate zones and are responsible for approximately 10% of the

Fig. 6 – (a, b) Clinical and radiological images (lateral view of the ankle) by the 9th month showing excellent outcome with nearly normal soft tissue and completely healed sinus clinically, while obliterated cavities, regular bone contours and normal density of calcaneum radiologically.

cases.³ *Scedosporium mycetoma* is more common in males, because of the greater outdoor activities as in our case report. The proportion of males to females affected varies from 3 to 5:1. The disease is more common in the age group of 20–45 years.³ Our case is of a 40-year-old male involved in outdoor activities.

As it is a rare infection, its diagnosis can only be made with good anticipation during clinical examination and culture [in Sabouraud glucose agar, 30°, 14 days] for its typical microscopic picture.¹ A time lag has been noticed between the investigations and *scedosporium* isolation.⁴ In our case, we found the organism in the second culture. *Scedosporium* antigen detection can be done using counter immune electrophoresis, but cross-reactions with other fungal antigens may occur.⁵ Pinto et al. isolated a peptidorhamnomannan antigen from mycelial forms of *scedosporium*. It may become the diagnostic antigen test in the future.⁶ Use of radiology in its diagnosis is limited to measuring the extent of the disease, bone involvement and follow-up of the disease. In our case, X-rays helped a great deal, not only for diagnosing osteomyelitis but also for assessing prognosis with treatment.

The treatment of *scedosporium* osteomyelitis is a challenge. Antifungal therapy combined with surgical debridement is the choice.¹ *Scedosporium* is resistant to the broad spectrum antifungal drug amphotericin B, indeed *scedosporium* infections can occur with amphotericin therapy.¹ Several subjects have demonstrated successful use of voriconazole in *scedosporium* infections;² hence we used voriconazole following surgical debridement and curettage. Many side effects have been reported, of which arrhythmia is the most dangerous complication, hence in consultation with the cardiologist, an echo and ECG was done prior to starting voriconazole. Neutrophils appear to exercise a more destructive effect on *scedosporium*. It may be possible that the addition of G-CSF to the antifungal therapy may be a useful adjunctive therapy.³

Our case responded well with surgical debridement and voriconazole monotherapy clinically and radiologically with a follow-up of 9 months.

Conflicts of interest

The authors have none to declare.

Author contributions

Author Rahul Mohan contributed to Concepts, Design, Definition of intellectual content, Literature search, Clinical studies, Data acquisition, Data analysis, Manuscript preparation, Manuscript editing and Manuscript review; Author T.S. Gopakumar contributed to Concepts, Definition of intellectual content, Literature search, Clinical studies, Manuscript editing and Manuscript review.

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