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Scedosporium prolificans osteomyelitis following penetrating injury: A case report

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ABSTRACT

Scedosporium prolificans are opportunistic moulds that can cause mycetoma following penetrating injuries. This fungus is more virulent than other species and treatment options are limited. Here we describe the first known case in the UK of *S. prolificans* osteomyelitis, in a 4 year old following penetrating injury. Successful outcome with limb salvage and foot function is achieved after repeated surgical debridement, and combination chemotherapy with voriconazole/terbinafine.

1. Introduction

Scedosporium prolificans is a ubiquitous fungi present in the soil, water and potted plants [1]. Since the first description of *S. prolificans* as an agent of human disease, several cases have been reported from across the developed world, in both immunocompetent and immunocompromised patients. Though septic arthritis and osteomyelitis after penetrating injury are recognised in immunocompetent patients [2–4], there are no cases reported from the United Kingdom. Here we report a case of osteomyelitis caused by *S. prolificans* in a 4-year-old immunocompetent child treated with surgery and combination antifungal agents.

2. Case

A previously well 4-year-old boy presented to the Accident & Emergency Department (day 0) accompanied by his mother in June 2013 with painful left ankle. There was a history of penetrating injury by a thorn 2 weeks previously. The thorn had been removed promptly by the parents. The child had been born in United Kingdom to Welsh father and Vietnamese mother who settled in UK. The family spent 6 weeks in Vietnam between February and March 2013 and apart from a diarrhoeal illness had been well during that time.

On examination, child was alert, apyrexial but walking with a limp. The medial malleolus was swollen and tender and the ankle joint was warm but there was no restriction of movement. A small healing wound from the thorn injury was noted on the midsole but there was no suggestion of any foreign body. Initial plain X-ray of the left foot was unremarkable (Fig. 1). He was sent home and reviewed 2 days later with little change in his condition. Mild swelling was noticed distal to medial malleolus which was tender but he remained apyrexial. Initial blood tests showed a white cell count of 12.8×10^9/L (normal limits) and CRP of 26 g/L (mildly raised). He was sent home on analgesic treatment with a plan to review in the orthopaedic clinic the following week.

Two days later, his mother brought him back to hospital with increased pain and swelling of the left ankle. An ultrasound scan of the foot showed synovial thickening and increased vascularity around the talonavicular joint, most marked laterally, suggesting septic arthritis. An ultrasound guided aspiration of the joint and blood cultures were taken and he was started on co-amoxiclav. All cultures were negative on microscopy and culture.

After 5 days of co-amoxiclav, the CRP continued to rise peaking at 150 g/L. Clinically he remained in pain and unable to weight bear. An MRI scan (day 9) of left foot (Fig. 2) showed a thick effusion and synovial proliferation in the talonavicular and anterior subtalar joints, marrow oedema in the medial aspect of the talar head but no evidence of any intraosseous collection.

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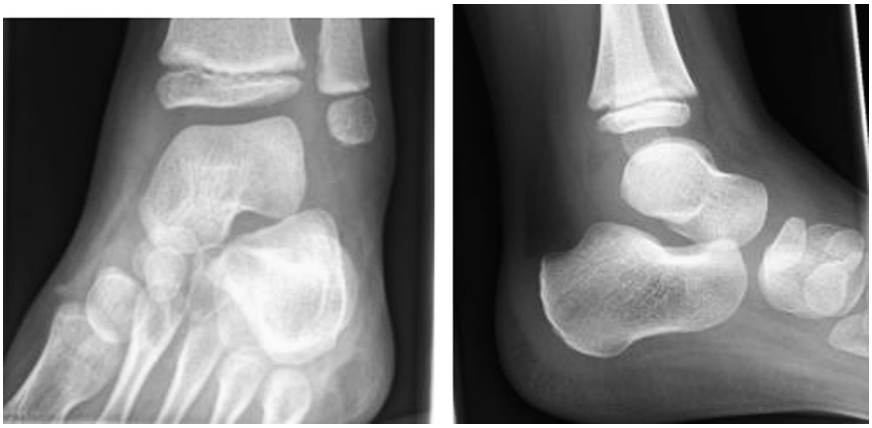

Fig. 1. AP and lateral views of the foot – no significant abnormality seen.

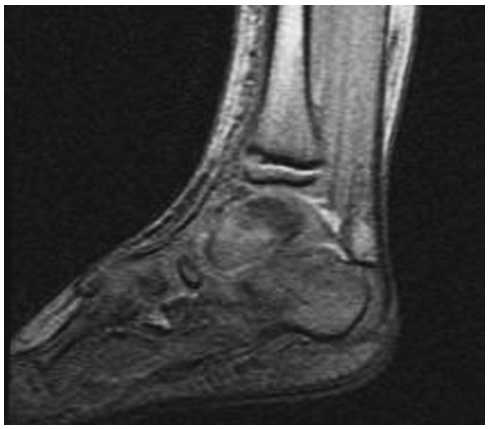

Fig. 2. MRI of the foot showing effusion and synovial proliferation in the talonavicular and anterior subtalar joints.

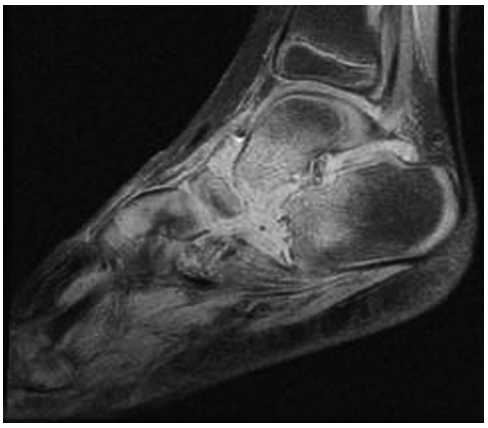

Fig. 3. Extensive synovial thickening within the talonavicular and calcaneocuboid joints associated with complete loss of articular cartilage at these two joints.

A washout of the subtalar and talonavicular joints was performed the same day and washout fluids and an interoperative swab were sent for culture. Culture from the swab grew a mycelial fungus after 3 days incubation although other specimens remained negative. Initial presumptive identification based on microscopic appearance suggested *Scedosporium spp*.

The orthopaedic team was advised to repeat the washout and send multiple tissue and fluid samples to exclude contamination (day 14). Antifungal treatment was not started at this stage but he was commenced on flucoxacillin and fucidic acid empirically. Synovial tissue and pus samples taken at the second washout again grew a mycelial mould and presumptive identification was modified to *Exophiala* species based on microscopic appearance and colonial morphology. Isolates were sent to the reference laboratory for further identification and susceptibility testing. Itraconazole with a loading dose of 10 mg/kg/day followed by maintenance dose 5 mg/kg/day in 2 divided doses was commenced on day 19.

Six days after starting itraconazole (day 25) the patient deteriorated clinically with pus oozing from the wound. A further washout was undertaken and samples taken at this stage again grew a mould.

Reports from the reference laboratory received day 28 identified the mould as *S. prolificans* resistant to amphotericin B, itraconazole, posaconazole, terbinafine; intermediate susceptibility to miconazole and susceptible to voriconazole. Itraconazole was discontinued and the patient was commenced on voriconazole and terbinafine.

Repeat MRI scan on day 29 (Fig. 3) showed extensive fluid and synovial thickening within the talonavicular and calcaneocuboid joints associated with significant periarticular oedema. Also, there is complete loss of articular cartilage at these two joints associated with erosions at the calcaneocuboid joint and inferior medial aspect of the talus. He was taken to theatre for radical debridement involving washout followed by synovectomy and removal of the navicular bone and 50% of the talar head articular surface; large erosions in the cuboid and calcaneum were curetted to healthy bone. Samples taken during debridement were sent for routine and mycobacterial cultures. All samples were negative at this stage. Full immunological review was undertaken and no defects were detected.

Alternate day washouts and theatre review were performed and a vacuum drain was inserted. Intravenous flucloxacillin and fusidic acid were discontinued. All further cultures were negative for all organisms. No further progression of the bony destruction was seen and the boy improved clinically. He underwent weekly change of vacuum dressings in theatre until the cavity was small enough to be treated comfortably in the outpatient clinic.

He was sent home 6 weeks after initial diagnosis on voriconazole and terbinafine with weekly review in clinic. At 3 months he had healed both lateral and medial wounds and was weight bearing in a functional boot. Whilst clinical progress is good, minor radiological abnormalities persist. (Fig. 4) At 4 months finger nail discoloration and skin rash were noted and were presumed to be related to terbinafine. The plan was to continue long-term antifungal treatment with therapeutic drug monitoring