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CASE REPORT HAND

Mycetoma of the hand: an unusual case of *Actinomadura* infection in Australia

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Introduction

Mycetoma is a rare infective disease caused by fungi or bacteria that leads to chronic, destructive, granulomatous inflammation causing tumefaction. Prevalence in developed countries is exceptionally low, with mycetoma most commonly reported in Asia, South America and Africa. Mycetoma predominantly affects the lower limb. This report describes an unusual case of a 57-year-old male presenting with mycetoma of his hand in Australia. Current treatment of mycetoma is with antibiotics. However, the hand surgeon must be aware of mycetoma because patients will be referred for primary surgical review since infection is rarely considered the cause of chronic subcutaneous nodules.

Case

A 57-year-old right-hand-dominant male of Lebanese heritage, with a background of hypertension and hypercholesterolaemia, was referred to our hand surgery unit with a two-year history of multiple slow-growing, rubbery lumps on the volar surface of his right hand. He had developed central palmar swelling nine years prior (2013) for which he had undergone surgical excision and a six-month course of trimethoprim/sulfamethoxazole. On completion there was a clinical resolution of infection, however, a relapse of symptoms occurred in 2020. The patient has been living in Australia for over 20 years, with travel only to Lebanon. He has worked as a painter throughout his life and denies having a traumatic or penetrating injury to his hand or exposure to soil and occupational materials.

Examination revealed generalised swelling of the entire right hand with multiple small lumps on the volar surface and one sinus on the palm discharging small red grains. There was no cellulitis or tenderness present in the hand (**Figure 1**). He had a full range of motion and normal sensation in the hand with no lymphadenopathy. Imaging with a CT scan found cortical erosions and cystic changes at the base of the third metacarpal suggestive of osteomyelitis (**Figure 2**). These soft tissue lesions were shown to be multiple rim-enhancing collections within the intrinsic musculature of the hand, in keeping with infective abscesses on MRI. Imaging also showed erosion of the third metacarpal base, loculated collections around multiple carpometacarpal joints and bony inflammation.

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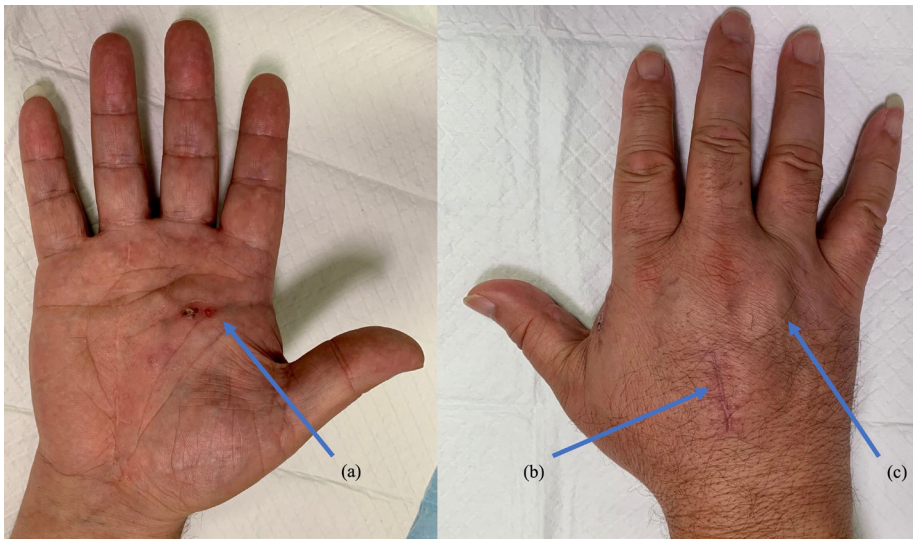

Fig 1. Examination findings: (a) sinus with discharging bacterial grains; (b) surgical scar from excisional biopsy; (c) subcutaneous nodule

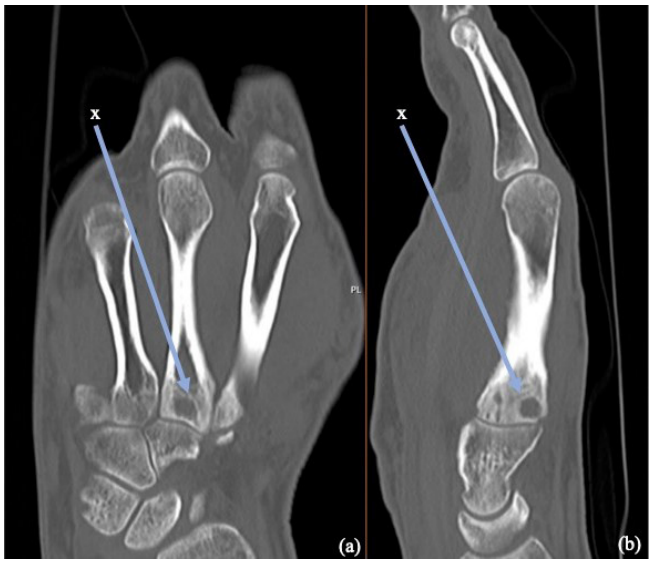

Fig 2. Right hand CT scan: (a) coronal view; (b) sagittal view; (x) cystic erosion of third metacarpal base

Based on the imaging findings, an excisional biopsy was performed and demonstrated widespread inflammatory changes in the right hand. The granulomatous inflammation communicated from volar to dorsal plate, invading the intrinsic musculature and metacarpals, with macroscopic osteomyelitis of the third metacarpal base. A single discrete lesion that was heavily adherent to the surrounding inflammatory tissue was amenable to excision (**Figure 3**). Histopathological analysis showed filamentous bacteria which was confirmed to be *Actinomadura* species on subsequent 16S rRNA testing, but multiple specimens (tissue and bone) sent for culture were all negative.

Following confirmation of relapse of infection and the presence of mycetoma, the patient was referred to the infectious diseases department for targeted antimicrobial therapy. A regimen of trimethoprim/sulfamethoxazole and amoxicillin/clavulanic acid was prescribed. The patient completed a total course of 27 months of antimicrobial therapy and has achieved disease control with resolution of swelling and no palpable subcutaneous nodules on examination (**Figure 4**). Disease regression was confirmed on MRI with resolution of rim-enhancing collections and only limited bony and soft tissue changes remaining, likely indicative of scarring (**Figure 5**).

Discussion

Mycetoma is a rare infection that causes a chronic and destructive granulomatous inflammatory process.[1,2] Mycetoma was first described in 1842 by John Gill in Madurai, India, where he referred to the condition as 'Madura foot' due to its podal predominance.[3] Since then, mycetoma has been found to affect not only the foot but also the entire lower limb, hand and upper limb, face, chest and abdomen. The hand is affected by mycetoma in only around 4 per cent of cases, with the foot still comprising the vast majority of cases.[2,4,5]

Mycetoma can be categorised according to causative organism. Mycetoma caused by fungi is referred to as eumycetoma, while mycetoma caused by bacteria is called actinomycetoma. Common causative organisms include the fungi *Madurella mycetomatis* and *Scedosporium boydii*, and the aerobic bacteria *Streptomyces somaliensis*, *Nocardia* species and *Actinomadura* species.[6] In this particular case, our patient was afflicted by actinomycetoma.

The recorded global prevalence of mycetoma is only approximately 127 cases per year, although this is thought to be an underestimation.

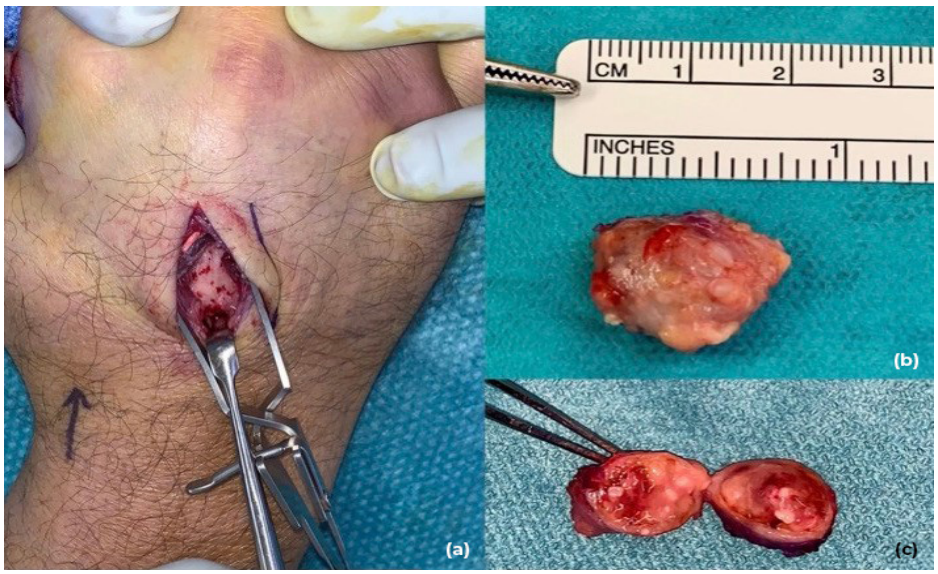

Fig 3. Intraoperative findings: (a) cystic erosion of third metacarpal base; (b) infective lesion; (c) infective lesion cut open to reveal bacterial grains

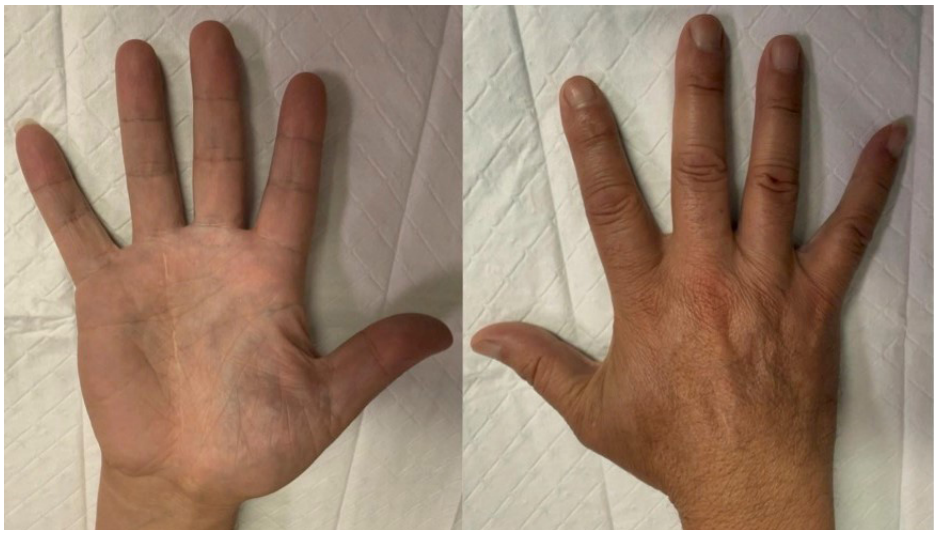

Fig 4. Examination findings following adequate antibiotic therapy, demonstrating resolution of subcutaneous nodules and swelling, and closure of discharging sinuses

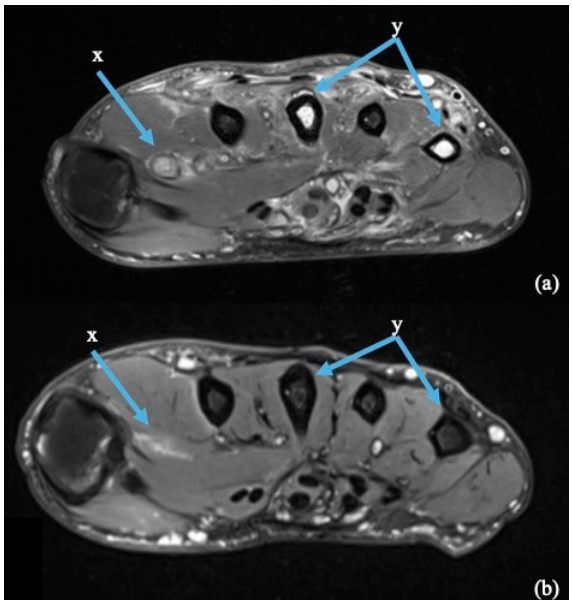

Fig 5. T2-weighted right hand MRI scan: (a) axial view at the time of presentation demonstrating rim-enhancing collection (x) and bony inflammation with marrow oedema (y); (b) axial view following 27 months of antibiotic therapy, demonstrating residual scarring (x) and resolution of bony inflammation (y)

Mycetoma is most commonly reported in areas near the equator between latitudes 15°S and 30°N, especially in Asia, South America and sub-Saharan Africa, with cases in developed countries being remarkably rare.[1,6] To our knowledge, only one other case of mycetoma of the hand has been reported in Australia.[4]

While a rare condition, especially in Australia, and even more so when considering infections of the hand, mycetoma of the hand can be potentially devastating.[7] Traumatic inoculation of the tissue with the offending organism leads to granulomatous inflammation which causes subcutaneous tumefaction. The lesion progresses insidiously until a sinus erupts through the skin and begins draining grains containing clumps of fungi or bacteria.[1] Mycetoma can have severe consequences because the inflammatory process diffuses across tissue planes and extensively invades the native tissue affected. This makes disease control very difficult and, in the hand,

can lead to significant disability as anatomical structures have such specific roles that small changes have large consequences.

Mycetoma is most commonly a subcutaneous condition.[8] This particular case demonstrates an unusual progression of disease with bony involvement present in addition to soft tissue disease. We suspect that bony disease is likely to have occurred as a consequence of inadequate disease control on initial presentation nine years ago because deeper structural involvement (eg, bone and even viscera) has only been described in severe, late-stage cases.[8]

Current management of mycetoma is largely with medical supervision and appropriate antibiotic treatment, with surgical intervention having a limited role. Surgical treatment alone results in recurrence in 20–90 per cent of cases, however, surgery may be indicated in treatment-resistant cases or in conjunction with antibiotic treatment.[1,5] In this case, excisional biopsy plus antimicrobial management was the only therapeutic option as debridement of the infected tissue would strip all of the intrinsic musculature of the hand, resulting in severe disability. Therefore, in cases of mycetoma of the hand, the ordinary surgical principles related to debridement of infected tissue do not apply, with surgery being limited to excisional biopsy for diagnostic intent rather than debridement for curative intent. Nevertheless, the hand surgeon must be aware of such conditions as infection is rarely considered the primary cause of chronic subcutaneous nodules and, therefore, patients may be referred for surgical review without knowledge of the aetiology of their condition.

Conclusion

Mycetoma of the hand is a rare but potentially destructive condition owing to the diffuse invasion of infection without respect for tissue planes. This case demonstrates late-stage disease and shows the limited but imperative role of surgery.

Patient consent

Patients/guardians have given informed consent to the publication of images and/or data with the Western Sydney Local Health District Research and Education Network providing case report approval (reference 2302-04).

Conflict of interest

The authors have no conflicts of interest to disclose.

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