Vacuum sealing drainage: A novel treatment method for primary cutaneous *Mycobacterium intracellulare* infection

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Abstract

The incidence of primary cutaneous *Mycobacterium intracellulare* infection is very low. We report a case of primary cutaneous *M. intracellulare* infection which presented as painful erythematous swelling of the right upper limb without systemic involvement. A novel technique of vacuum sealing drainage was successfully implemented after antimycobacterial treatment proved ineffective at the end of 3 months. Our technique also revealed some additional practical advantages.

Key words: Cutaneous, *Mycobacterium intracellulare*, primary, vacuum sealing drainage

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Introduction

Nontuberculous mycobacteria are important opportunistic human pathogens, and the *Mycobacterium avium* complex is a prominent subgroup in most epidemiologic series.[1,2] *M. avium* complex is composed of several closely related slow-growing nonchromogens including *M. intracellulare,* which is widely prevalent, often causing pulmonary infection, lymphadenitis, disseminated infection, and involving the lungs, spleen, liver, lymph nodes, gastrointestinal tract, bone marrow and skin.[3] The skin is typically involved as a part of disseminated infection, while skin involvement alone is rare. Here we report a case of primary cutaneous *M. intracelluare* infection in a patient on prolonged methotrexate and methylprednisolone therapy. We have also described a novel treatment method for cases unresponsive to medical therapy.

Case Report

A 72-year-old Chinese woman, farm-worker by profession, presented with a solitary, painful, erythematous swelling on the right forearm and the back of right hand since last 2 months. The lesion started as a small erythematous, painful swelling on the back of right hand which gradually increased in size and involved the right forearm. Penicillin was first administered without any appreciable improvement. The lesion progressively enlarged in size along with formation of multiple pus-filled nodules, which prompted her referral to our hospital. No history of fever, night sweat, cough or antecedent trauma was obtained. The patient had been receiving oral methotrexate (10 mg/week) and methylprednisolone (6 mg/day) since 2 years for rheumatoid arthritis.

Physical examination revealed an erythematous, non-fluctuant swelling involving the right forearm and back of right hand with multiple linearly arranged cystic nodules discharging pus, along the ulnar margin of the same hand [Figure 1]. The lesions were warm and tender on palpation. Routine laboratory investigations were within normal limits. Human immunodeficiency virus screening was negative. Chest roentgenogram, B-ultrasonography and computed tomography revealed no abnormality. Lesional biopsy

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revealed a normal epidermis along with mild edema of the upper dermis and extensive lower dermal infiltration with histiocytes, neutrophils and lymphocytes. Few tuberculoid granulomas were also observed in the lower dermis [Figure 2]. The tuberculoid granulomas demonstrated histiocytes and multinucleate giant cells in the center, while plasma cells and lymphocytes were observed at the periphery [Figure 3]. Ziehl–Neelsen staining ruled out the presence of acid-fast bacilli. Periodic acid-Schiff and Gomori methenamine silver staining were also negative. Microscopic examination demonstrated acid-fast bacilli in the purulent discharge from the lesions without presence of any fungi [Figure 4]. Culture was positive for mycobacteria. Further identification of the isolated mycobacteriawas performed by polymerase chain reaction restriction fragment length polymorphism and gene sequencing of hsp65 and 16S rDNA. The sequencing analysis revealed 99 and 100% homology, respectively, with *M. intracellulare* strain ATCC 13950 (GenBank accession number AF126035.1 and GQ153276.1). Further *in vitro* drug susceptibility assay revealed sensitivity to clarithromycin, rifampicin and moxifloxacin.

A final diagnosis of primary cutaneous *M. intracellulare* infection was made, based on laboratory findings. The patient was prescribed oral clarithromycin (500 mg/day), rifampicin (450 mg/day) and moxifloxacin (400 mg/day) for 3months, based on drug susceptibility tests. The erythematous swelling improved but the purulent nodules and subcutaneous sinuses remained unchanged [Figure 5]. Subsequently, the patient was treated by surgical debridement to take care of the abscess, nodules, sinus, purulent secretion and necrotic tissue. Postoperative vacuum sealing drainage was carried out in the incised area to continuously drain out the pathogens and secretion from the infected area [Figure 6]. Repeated debridement and vacuum sealing drainage was done after assessing the healing response of the tissue. After 15 days of the application of vacuum sealing drainage, granulation tissue appeared, the abscesses disappeared and the incision was sutured. The patient continued to take the same oral medications for 10 months and achieved complete recovery [Figure 7]. There was no recurrence during long-term follow-up (6 months).

Discussion

M. intracellulare is a non-chromogenic, urease-positive nontuberculous mycobacterium belonging to Runyon group III. *M. intracellulare* cannot be differentiated from *M. avium* by conventional methods expect for DNA analysis. Although *M. avium* and *M. intracellulare* possess similar

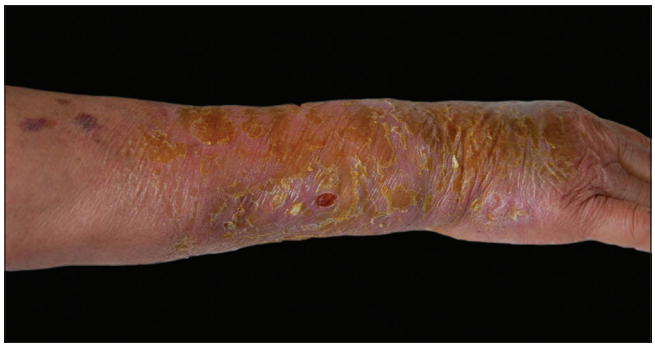

Figure 1: Erythematous swelling and nodules on the right forearm and the back of right hand

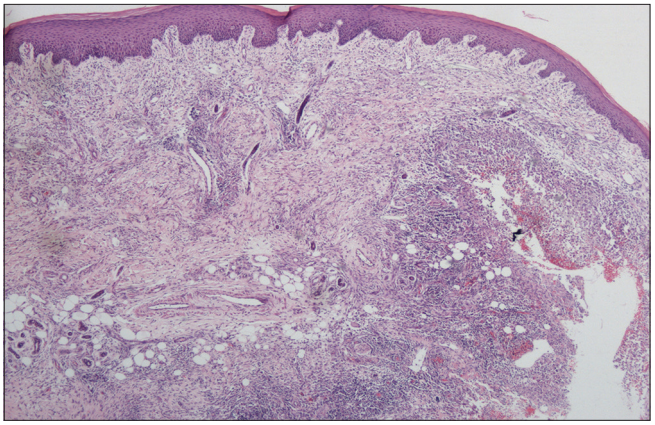

Figure 2: Histiocytes, neutrophils, lymphocytes and tuberculoid nodules in the lower dermis [hematoxylin and eosin (H and E), ×100]

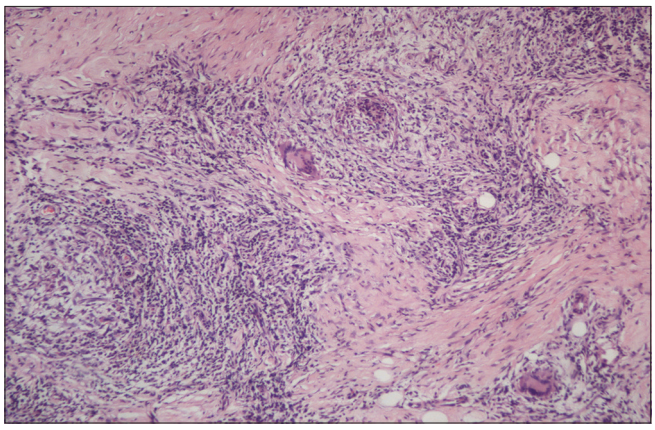

Figure 3: Histiocytes and multinucleate giant cells in the center, plasma cells and lymphocytes at the periphery (H and E, ×400)

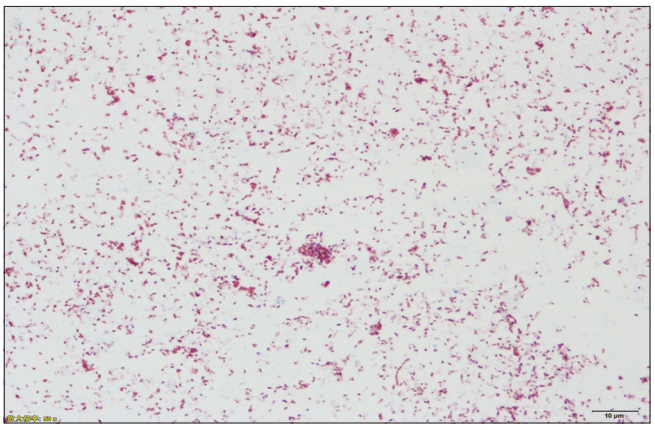

Figure 4: Direct microscopic examination of the purulent discharge was positive for Ziehl–Neelsen staining

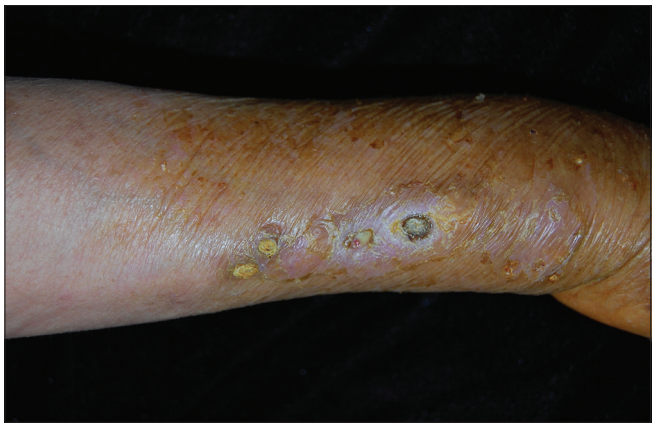

Figure 5: Nodules arranged in clusters, purulent secretions after extrusion

Figure 6: Postoperative vacuum sealing drainage was conducted on incision

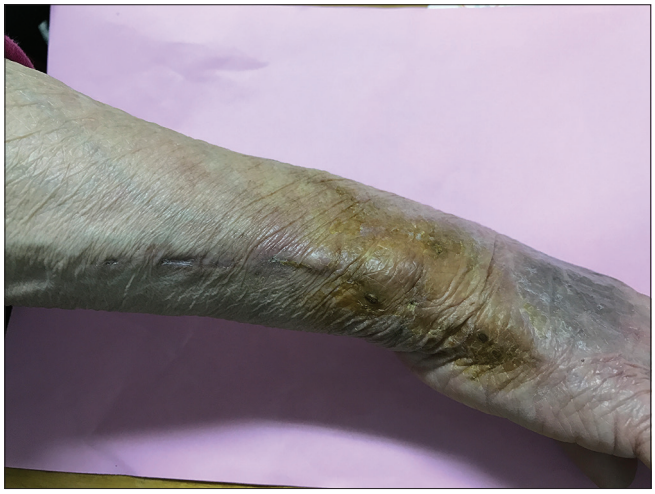

Figure 7: The patient continued to take oral medicine for 10 months with complete cure

biochemical characteristics, their virulence pattern is different. The vast majority of *M.avium* complex infections in AIDS patients were caused by *M. avium*, whereas *M. intracellulare* was the main cause of *M.avium* complex pulmonary infection in non-HIV patients.[4] An ecologic study revealed that *M. intracellulare* was more inclined to form biofilm than *M. avium*.[5] This ability might give an explanation for the higher pathogenic property of *M. intracellulare* in non-HIV patients. In the present case, *M. intracellulare* in a non-HIV patient corroborates the above view. *M. intracellulare* is one of the most common opportunistic pathogens in AIDS patients, and is also an important pathogen in non-HIV immunocompromised individuals, but rarely attacks immunocompetent patients.[6,7] Our patient had been receiving methotrexate and methylprednisolone for 2 years resulting in an immunocompromised state, thus becoming susceptible to *M. intracellulare* infection.

Cutaneous infection caused by *M.avium* complex shows a varied range of presentations including nodules, erythematous papules, ulcers, pustules, abscesses, folliculitis, panniculitis, infiltrated plaques, verrucous lesions, draining sinuses, sporotrichoid growth patterns or rosacea-like papulopustules.[8-12] Generally, skin manifestations associated with *M. avium* complex infection can be categorized into two types: direct lesion, where the microorganism appears by direct inoculation (via hematogenous dissemination, or through contiguous infection); and hypersensitivity vasculitis, where the bacillus is absent, but clinical features appear due to deposition of immune complexes in the walls of the small vessels. In the present case, *M. intracellulare* was detected in the skin lesion. However, there were no signs of contiguous or hematogenous dissemination. Thus, we believe the present case to be a primary cutaneous *M. intracellulare* infection by direct inoculation. Reed *et al.* reported that cumulative occupational exposure to soil was the most significant environmental risk factor for *M.avium* complex infections.[13] Regular farm work of our patient might have contributed to her *M. intracellulare* cutaneous infection.

Nowadays, the established treatment regime for *M.avium* complex pulmoray infection is a macrolide-based two- or three-drug combination for 6–12 months.[14] The proper drug regimen for cutaneous *M.avium* complex infection still remains unknown, so the prognosis varies widely depending on affected sites, underlying diseases, and early treatment. In this case, we prescribed oral clarithromycin, rifampicin and moxifloxacin based on drug susceptibility testing, resulting in initial remission of erythematous swelling, without affecting the abscesses and nodules. Complementary surgical debridement and vacuum sealing drainage was conducted and remission was obtained after 10 months with complete cure. No relapse was observed after a 6months follow-up.

The vacuum sealing drainage technique is a therapeutic concept to achieve rapid wound healing in traumatic soft wounds and chronic infections.[15] The wound cavity is filled with polyvinyl alcohol embedded foam, covered by a transparent vapor transmitting polyurethane film. Then a drainage tube is connected between the wound cavity and a vacuum bottle, to

generate the negative pressure. Secretions are continuously drained by this mechanism, removing potentially infectious secretions from the wound to accelerate its healing. An important advantage of this set-up is the formation of a protective barrier against microorganism translocation. We were unable to find any previous report of vacuum sealing drainage technology used in the treatment of primary cutaneous *M. avium* complex infections in the literature. Thus we opine that this therapy may be used in these kinds of cutaneous infections, which is a safe, convenient and effective strategy.

This case has been reported for its rarity and also to highlight a novel treatment mechanism.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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