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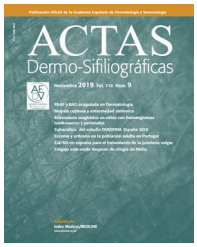

CASE AND RESEARCH LETTER

[Translated article] Sporotrichoid Nodules in a Woman With Sarcoidosis

Nódulos de distribución esporotricoide en una paciente con sarcoidosis

To the Editor,

Mycobacterium chelonae is an atypical mycobacterium classified as a rapidly growing nonchromogenic mycobacterium.[1] It is universally distributed and is normally found in the environment (e.g., in water and soil).[2–5] It is one of the most common mycobacteria responsible for skin infections in immunocompromised patients in whom lesions may be deeper and/or more disseminated. Infections can manifest as abscesses, painful erythematous nodules,[2] folliculitis, cellulitis, and sporotrichoid lesions.[3] Most cases are nosocomial and are generally associated with trauma or surgical or cosmetic procedures, although these events are often not evident.[2–5]

A 70-year-old woman with stage III sarcoidosis under treatment with salmeterol/fluticasone propionate, terbutaline, and inhaled prednisone (10 mg daily) presented at the dermatology clinic with lesions of 1 month's duration on her left forearm. She did not recall any previous trauma and reported no fever or associated systemic symptoms. Physical examination showed 2 erythematous nodules, firm to palpation, with sporotrichoid spread: one on the dorsum of the left hand and the other on the dorsum of the left forearm, (Fig. 1A). Suspecting deep mycosis or cutaneous sarcoidosis, we performed skin biopsy, which showed an intense inflammatory infiltrate in the deep dermis composed of lymphocytes, histiocytes, and clusters of polymorphonuclear leukocytes with cell debris. Ziehl–Neelsen staining showed long pink structures (Fig. 2). With a histopathologic diagnosis of suppurative granulomatous nodular dermatitis of probable infectious origin, DNA was extracted for mycobacterial species identification by polymerase chain reaction (PCR), which, together with the culture findings, confirmed a diagnosis of skin infection due to *M. chelonae*. The patient was prescribed clarithromycin 500 mg/12 h for 4 months. She responded well initially, but on completion of treatment, she developed a recurrent infection. Susceptibility testing at this point showed susceptibility to clarithromycin, ethionamide, and tobramycin. Follow-up tests revealed hypogammaglobulinemia with an immunoglobulin (Ig) G level of 380 mg/dL (normal, > 650 mg/dL) and B-cell lymphopenia (30 cells/mL; normal, >100). On reviewing the patient's clinical records, we detected a history of respiratory infections and bronchiectasias and established a diagnosis of a primary immunodeficiency disorder (PID) with predominantly deficient antibody production. The patient was started on intravenous IG replacement therapy at a dose of 0.4 mg/kg every 3 weeks, which, together with clarithromycin for 2 months, led to definitive resolution of the lesions (Fig. 1B).

The sporotrichoid pattern observed in *M. chelonae* infection is due to the ascending spread of the mycobacteria along the lymphatic channels.[6] It is an unusual pattern, and just 15 cases have been reported in the literature (Table 1), none of them in a patient with sarcoidosis. The main entities to include in the differential diagnosis are infections due to other pathogens that present with a similar distribution, such as *Sporothrix schenckii*, *Mycobacterium marinum*, *Nocardia* species, and *Leishmania* species.

Immune system alterations should be ruled out in patients with atypical mycobacterial infections, especially in the presence of an uncommon pattern, such as sporotrichoid spread. Skin infections are the most common dermatologic manifestations of PIDs. Susceptibility may be specific to certain pathogens, depending on which part of the immune system is compromised.[7] Patients with PIDs caused by mutations in interferon γ genes, which are characterized by phagocyte defects without altered humoral immunity, are prone to severe disseminated infections caused by atypical mycobacteria.[8] In our case, we observed IgG deficiency and B-cell lymphopenia. Antibody deficiencies are commonly associated with respiratory bacterial infections, which were also present in our patient's history.

Biopsy is key to diagnosis. Histopathologic patterns include a diffuse histiocytic infiltrate, microabscesses, panniculitis, tuberculoid or sarcoid granulomas, and/or reactive vasculopathy.[9] Acid-fast bacilli are demonstrated by specific stains such as Ziehl-Neelsen, although a negative test does not rule out a mycobacterial infection.[5,10] Diagnosis is

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Table 1 Cutaneous *Mycobacterium chelonae* Infections with a Sporotrichoid Distribution Reported in the Literature.

Case	Age, y/sex	Location	Underlying disease	Immunosuppression	Treatment	Recurrence	Treatment after recurrence
Greer, 1979[12]	76/F	Leg		No	Isoniazid + amithiozone	No	
Higgins, 1988[13]	65/F	Forearm	Chronic active hepatitis	Yes	Erythromycin + amikacin	No	
Murdoch, 1989[14]	61/F	Leg	Kidney transplant	Yes	Pyrazinamide + rifampicin 6 mo	Yes	Erythromycin
Jopp-McKay, 1990[15]	52/F	Leg	Kidney transplant	Yes	Minocycline 2 mo	Yes	TMP-SMX + surgery
Zahid, 1994[16]	70/M	Hand	COPD	Yes	Ciprofloxacin + clarithromycin 6 mo	No	
Endzweig, 2001[17]	59/M	Leg	Kidney transplant	Yes	Surgery + ciprofloxacin + TMP-SMX + imipenem	Yes	Surgery + amikacin + cefoxitin + clarithromycin
Haas, 2001[18]	66/F	Forearm	Rheumatoid arthritis	Yes	TMP-SMX + clarithromycin	Yes	Azithromycin + ciprofloxacin + surgery
Demitsu, 2001[19]	46/M	Forearms	Congestive heart failure Diabetes	No	Minocycline 2 mo	Yes	Surgery
Rosón, 2002[20]	42/F	Forearm		No	Minocycline	No	
Phillips, 2008[21]	43/F	Forearm	Bilateral panuveitis	Yes	Imipenem + piperacillin-tazobactam + amoxicillin-clavulanic acid 5 mo	No	
de Vasconcelos, 2015[22]	60/M	Forearm	Rheumatoid arthritis	Yes	Clarithromycin 6 mo	No	
Orrin, 2016[23]	65/F	Leg	Cryptogenic organized pneumonia	Yes	Clarithromycin 9 mo	No	
Boulavsky, 2017[24]	75/F	Leg and foot	Lupus nephritis	Yes	Clarithromycin + amikacin + levofloxacin	No	
Kemp, 2017[3]	54/F	Forearm	Systemic lupus erythematosus	Yes	Linezolid + clarithromycin 4 mo	No	
DuBow, 2019[6]	31/F	Leg	Systemic lupus erythematosus	Yes	Linezolid + clarithromycin 8 mo	Yes	Linezolid + clarithromycin 3 mo
Current case	70/F	Forearm	Sarcoidosis Primary immunodeficiency	Yes	Clarithromycin 4 mo	Yes	Clarithromycin 2 mo + IVIG

Abbreviations: COPD, chronic obstructive pulmonary disease; F, female; IVIG, intravenous immunoglobulin; M, male; TMP-SMX: trimethoprim-sulfamethoxazole.

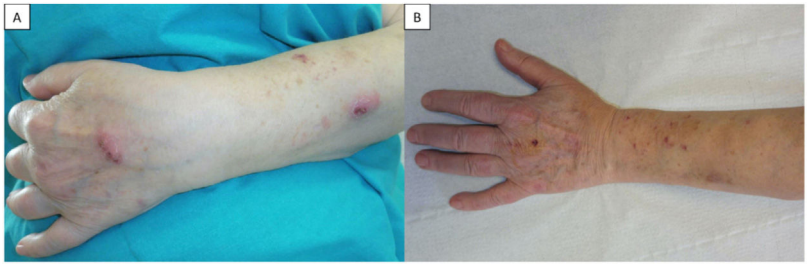

Figure 1 A, Two indurated erythematous nodules with a sporotrichoid distribution on the back of the hand and on the left forearm. B, Resolved lesions after treatment.

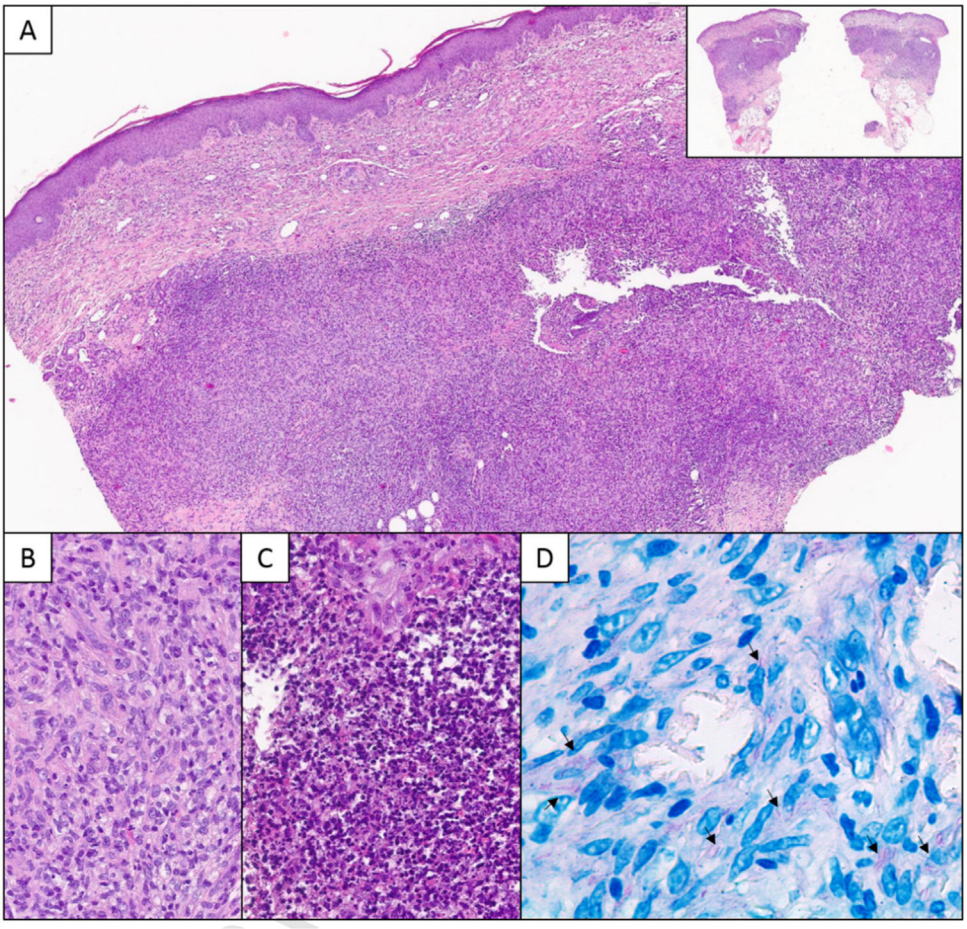

Figure 2 A, Histologic section showing a deep granulomatous dermal infiltrate with a nodular pattern (panoramic view in top-right corner) (hematoxylin–eosin, original magnification ×40). B, Detailed view showing a lymphocytic and histiocytic infiltrate (hematoxylin–eosin, original magnification ×200). C, Suppurative areas with abundant neutrophils and cell debris (hematoxylin–eosin, original magnification ×400). D, Ziehl–Neelsen staining. Note the long pink structures (arrows) (original magnification ×630).

confirmed by culture or molecular techniques such as PCR restriction fragment length polymorphism analysis.[2,10]

M. chelonae infections tend to have an unpredictable resistance profile, hence the importance of susceptibility testing. Although clarithromycin monotherapy is sufficient in most cases, combined therapy is recommended due to the risk of resistance developing during treatment, which is frequently administered for long periods.[6,11] Adjuvant surgical treatment may be required in certain cases.[5]

In conclusion, when dealing with a patient with sporotrichoid cutaneous lesions, it is important to rule out an atypical mycobacteria infection, especially in immunosuppressed patients.

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A. Navarro-Bielsa[a,*], A. Bielsa[b], M.C. Gomez-Mateo[c], I. Abadías-Granado[a]

[a] *Servicio de Dermatología, Hospital Universitario Miguel Servet, Paseo Isabel la Católica, Zaragoza, Spain*
[b] *Servicio de Medicina Interna, Hospital Universitario Miguel Servet, Paseo Isabel la Católica, Zaragoza, Spain*
[c] *Servicio de Anatomía Patológica, Hospital Universitario Miguel Servet, Paseo Isabel la Católica, Zaragoza, Spain*

* Corresponding author.
E-mail address: albanavarrobielsa@hotmail.com (A. Navarro-Bielsa).