Multiple cutaneous abscesses in a patient with Sjögren’s syndrome

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Case report

A 63-year-old woman presented with multiple tender, nonulcerating, erythematous nodules on her right hand and forearm for 10 days (Figure 1A). She was suffering from Sjögren’s syndrome for more than 1 year and had been treated continually with azathioprine and prednisolone. Under the tentative diagnosis of erythema nodosum, an incisional biopsy was performed on her right forearm. Histopathology showed septolobular panniculitis, heavy infiltrates of neutrophils and granulomatous inflammation (Figures 1B and 1C). Periodic acid-Schiff (PAS) and acid-fast stains were negative. She was then treated with prednisolone 20 mg per day for suspected autoimmune panniculitis. The patient returned to our clinic 2 months after initial presentation. Multiple erythematous to purpuric macules, papules, and punched out ulcers spread centripetally over the patient’s four extremities in a symmetrical distribution (Figure 2A). Tender erythematous nodular eruptions without ulcerations similar to her previous skin lesions at initial presentation were found over both hands and forearms (Figure 2B). There were no associated systemic symptoms, such as fever, chilling and myalgia. She was treated with higher dose of prednisolone of up to 60 mg per day for suspected autoimmune vasculitis by the rheumatologist. However, more papulonodules, ulcers and pustules developed over her extremities as we tapered the dose of systemic steroid. Another skin biopsy of a fresh pustule from her right hand was taken and submitted for both histopathology and culture. Acid-fast positive bacilli were found (Figure 3).
Diagnosis

*Mycobacterium abscessus* infection.

Discussion

Histologic examination of the biopsy specimen revealed numerous acid-fast bacilli in the abscesses and adjacent dermis. Identification of *Mycobacterium abscessus* in the mycobacterial culture from the same biopsy specimen clinched the diagnosis of *M. abscessus* in this case.

*M. abscessus* is a ubiquitous, rapidly growing mycobacterium that rarely causes significant clinical infection in immunocompetent hosts. However, in recent decades with the emergence of AIDS and the increased use of systemic immunosuppressants, the recognition of infections caused by rapidly growing mycobacterium has increased.1

Cutaneous *M. abscessus* infection usually presents as erythematous papules or nodules that become centrally purpuric and then develop ulcers or abscesses. A granulomatous tissue reaction is the most common pathologic change.2 The immunocompromised patients tend to have more prominent ulceration of and abscess formation within the cutaneous lesions. Histologically, these immunocompromised hosts show more involvement of the subcutaneous tissue with more prominent microabscess formation.1 Microbiological diagnosis is essential, with lesional biopsies offering the highest yield (55%) compared to debridement specimens (34%) and swabs of wound (11%).3 Polymerase chain reaction–based technology also provides a rapid, accurate system for the identification of clinically important species of rapidly growing mycobacterium.

In our case, there was a lack of granulomatous inflammation in the latter biopsy specimen, which is unusual for nontuberculous mycobacterial infection. However, in immunocompromised patients, a high index of suspicion for infection with acid-fast bacilli should be considered in the differential diagnosis of prominent acute inflammation and abscess formation in skin and soft tissue.

*M. abscessus* fails to respond to standard antituberculous agents. This emphasizes the need for tissue diagnosis and obtaining specimens for culture and drug susceptibility testing, with use of empirical therapy suggested until susceptibilities are known. *M. abscessus* is susceptible to amikacin, cefoxitin, imipenem, and clarithromycin.4 In disseminated disease, combination therapy for at least 6 months is recommended to prevent resistance.5

Following the diagnosis of cutaneous *M. abscessus* infection, the patient was started on a regimen of intravenous amikacin (500 mg once a day) and imipenem (500 mg four times a day) and oral clarithromycin (500 mg twice a day) for 2 months with a dramatic improvement in her skin lesions. She was then discharged and regularly received periodic parenteral antibiotics (amikacin 500 mg once a week, and imipenem 1000 mg twice a week) and oral azithromycin (250 mg once a day) for another 4 months. After 6 months of treatment and surgical debridement, the nonulcerative lesions cleared almost completely with postinflammatory hyperpigmentation, and the ulcers healed with dry crust.

This case highlights a number of important points regarding the management of a patient with autoimmune disease under immunosuppressive treatment and nontuberculous mycobacterial infections. Early consideration of atypical mycobacterial infection should be made in any patient in whom there is an unexplained persistent suppurative cutaneous lesion with relatively mild symptoms and poor response to standard treatment. In such cases, skin biopsy for histology and microbiological studies are crucial in the diagnosis.

References