Rapidly Progressing Generalized Morphea with High Lyme Disease Titer

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Sir,

A 51-year-old male presented with erythematous to brownish ill-defined indurated enlarging plaques on the right leg, left flank, and left anterior chest over 10 days with hardness. All of the lesions were exceeding 5 cm in diameter (Figures 1 and 2). He did not give tick bite history and never had traveled to the endemic areas of Lyme borreliosis. Biopsy from the right leg showed lymphoplasmacytic infiltrate and fibrosis in the dermis and subcutis (Figures 3 and 4). Complete blood count, chemistry, HIV testing were normal, and antinuclear antibody, anti-Ro/La, anti-RNP, anti-Scl70, anti-centromere antibody were all negative. Lyme disease immunoglobulin G titer was 1:512 and IgM titer was <1:16 in immunofluorescence assay (IFA), but negative in Western blot test. Warthin Stary stain was negative. DNA was not detected in polymerase chain reaction.

Transthoracic echocardiography, electrocardiography, esophagogastroduodenoscopy, colonofibroscopy, chest and abdominopelvic computed tomography, and pulmonary function test were done and all were normal. Oral doxycycline (200 mg/day) for 3 weeks was started due to the possibility of Lyme borreliosis but without response. After systemic corticosteroid administration, the progression was slowed. After discontinuing the medication for 3 months, new skin lesions were found on the left buttock and thigh. Corticosteroid was started again, and the progression was stopped. There was no progression after cessation of medication until reporting.

Follow-up IFA and western blot determinations were performed 6 months after the onset of symptom, but the titer did not decrease, and western blot was still negative.

Lyme disease, also known as Lyme borreliosis, is a multi-organ infection caused by spirochetes of the *Borrelia burgdorferi sensu lato* group which are transmitted by ticks of the species *Ixodes*.¹ Common skin manifestations include erythema migrans, lymphocytoma, and acrodermatitis chronica atrophicans.¹ Recently, there have been some reports linking morphea or lichen sclerosus to *B. burgdorferi* infection.¹

The presence of antibodies to *B. burgdorferi* in 50% of patients with morphea was found with enzyme-linked immunosorbent assay (ELISA),¹ but contradictory data have been proposed.² Most reports of the association between positive antibody test to *B. burgdorferi* and morphea come from Europe, and have been rarely found in the USA.³ Differences between *Borrelia* strains and prevalence in the USA and in Europe have been raised to explain these conflicting results.³ *B. garinii*, *B. afzelii*, and *B. burgdorferi sensu stricto* are present in Europe, only *B. burgdorferi sensu stricto* is identified in the USA. It has been suggested that only certain types of infection of *B. burgdorferi* may lead to morphea. *B. burgdorferi sensu stricto*, has never been found in late dermatologic manifestations of Lyme borreliosis, this...
Figure 1: Erythematous to brown colored, irregularly circumscribed indurated plaques are found from upper thigh to calf

Figure 2: Slightly whitish to erythematous irregular shaped indurated patch on chest, around nipple

Figure 3: Histopathology of specimen from right calf shows perivascular and interstitial infiltrates of lymphocytes and plasma cells in dermis (H and E, ×40)

Figure 4: Patchy fibrosis in the subcutaneous layer, and lymphoplasmacytic infiltrates are found. Trabeculae are thickened in subcutaneous layer (H and E, ×100)

may explain the reason of conflicting data from different regions.\[3\]

The overall false-positive rate of Lyme borreliosis testing is approximately 5%.\[4\] When patients have other viral or spirochetal infections or autoimmune diseases, false-positive findings with ELISA or IFA may occur.\[4\] There has been only one case of morphea with false positivity to Lyme titer since 1993.\[5\] We experienced this interesting case of rapidly progressing generalized morphea with high antibody titer to \textit{B. burgdorferi}.

**Declaration of patient consent**

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand 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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A male patient presented with complaints of abdominal pain, nausea, and fatigue that started 4 years ago, and an abscess was found in the liver, and skin, who was treated with steroids and report a case of multiple abscesses involving the spleen, which included fever, fatigue, weight loss, and multiple wounds with discharge on the extremities. His history was followed up for malignancy, and he was managed with symptomatic treatment. He had severe abdominal pain, nausea, and fatigue that was first reported in 1995. It manifests as round abscesses composed of neutrophils, and it involves the liver, abdominal lymph nodes, lungs, skin, and sterile abscesses. Elevated erythrocyte sedimentation rate, and elevated reactive protein; however, it does not respond to antibiotics. Its response to steroids is excellent.

Considering the differential diagnosis, polyarteritis nodosa, disseminated tuberculous abscess, ecthyma gangrenosum, mycosis, and syphilitic gumma, multiple biopsies were carried out. The histopathology of the epidermis revealed spongiosis, slight acanthosis, and exocytosis abscesses carried out. The histopathology of the epidermis revealed spongiosis, slight acanthosis, and exocytosis abscesses in the epidermis, focal abscesses in the epiderm and deep dermis, with multinuclear giant cells. The patient was examined with a chest X-ray, thoracic tomography, sacral magnetic resonance imaging, and computed tomography of the abdomen. He developed abscesses on the distal parts of the upper and lower limbs, with purulent discharge that rapidly increased 2 months after splenectomy; therefore, he underwent a splenectomy.

The laboratory workup was as follows: C-reactive protein was 110 mg/L (<55); alkaline phosphatase level was 265 U/L (30–120); gamma-glutamyl transferase level was 235 U/L (<50); alanine transaminase level was 142 U/L (<50); aspartate aminotransferase level was 18,000 (3.98–10.2); and procalcitonin level was 0.02 ng/mL. No growth was seen in nonspecific bacteria, aerobes, anaerobes, and tuberculosis, as well as a mycotic examination. No growth was seen in tuberculosis complex polymerase chain reaction. QuantiFERON test was negative, and chronic granulomatous disease titer. Indian J Dermatol 2020;65:432-4

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