SHORT COMMUNICATION

Borrelia-associated Fasciitis: Two Cases

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Late manifestation of infection with *Borrelia species* can induce several cutaneous symptoms due to the ability of *Borrelia spp*. to colonize and induce structural changes in the collagen fibres (1). The classical form is acrodermatitis chronica atrophicans, but infection with *Borrelia spp.* has also been related to morphoea (2), lichen sclerosus et atrophicus (3) and, rarely, eosinophilic fasciitis (EF) (4–7). Diagnosis is mainly clinical, and a deep skin biopsy can reveal a typical lymphoplasmacytic infiltrate. Serology is very sensitive (95–99%), but does not provide any information about the activity of the disease. PCR on skin biopsies is a useful tool for the diagnosis of early stage skin borreliosis when there is a high load of *Borrelia* present in the skin, but its sensitivity decreases during the course of the infection (8). A relatively new test, using antibodies targeting VlsE, a lipoprotein expressed by *Borrelia spp*. implicated in escaping the immune system, correlates with the disease activity and is a useful tool for follow-up of the efficiency of the treatment. The test is highly specific, but not very sensitive (9–11). We report here 2 cases of fasciitis associated with *Borrelia spp*. infection.

CASE REPORTS

Case 1

A 70-year-old man presented with widespread deep cutaneous sclerosis of the limbs (**Fig. 1**) associated with fatigue, arthralgia and night sweats. He remembered having had a tick bite in Sweden several months earlier. A computed tomography (CT) scan revealed a bilateral infiltration of the superficial and deep fascia of both thighs. A deep biopsy of the skin and subcutaneous tissue including the fascia and some superficial muscle tissue showed a moderate perivascular and interstitial inflammatory infiltrate composed of lymphocytes and plasma cells throughout the dermis, extending into the fascia and skeletal muscle. There was no blood eosinophilia. Western blot was positive for *Borrelia spp*. IgG and anti-VlsE were strongly positive. PCR for *Borrelia spp*. on deep skin biopsy, synovial liquid and cerebrospinal fluid were negative. Fasciitis associated with *Borrelia spp*. infection was diagnosed; intravenous ceftriaxone, 2 g once daily, was administered for one month, with a positive biological response showing a decreasing anti-VlsE level. Clinically, there was a significant improvement, but the persistence of sclerosis, oedema of the limbs and arthralgia required additional treatment with prednisone and methotrexate.

Case 2

A 79-year-old woman presented with non-pruriginous erythematous lesions with sclerotic changes of the skin involving the thighs, groin, abdomen, axillary regions and breasts (**Fig. 2**) She reported having had a tick bite on the left groin one year earlier. Laboratory tests revealed an eosinophilia, a slightly elevated C-reactive

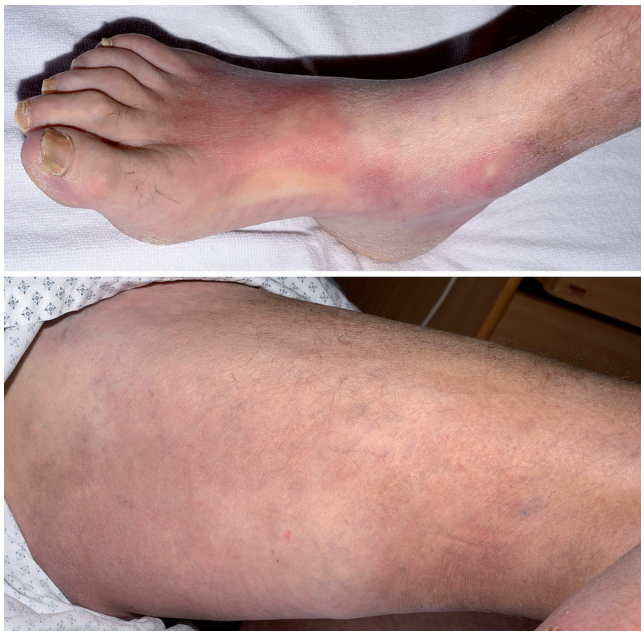

Fig. 1. Diffuse cutaneous fibrosis of the right foot and right thigh (Case 1)**.**

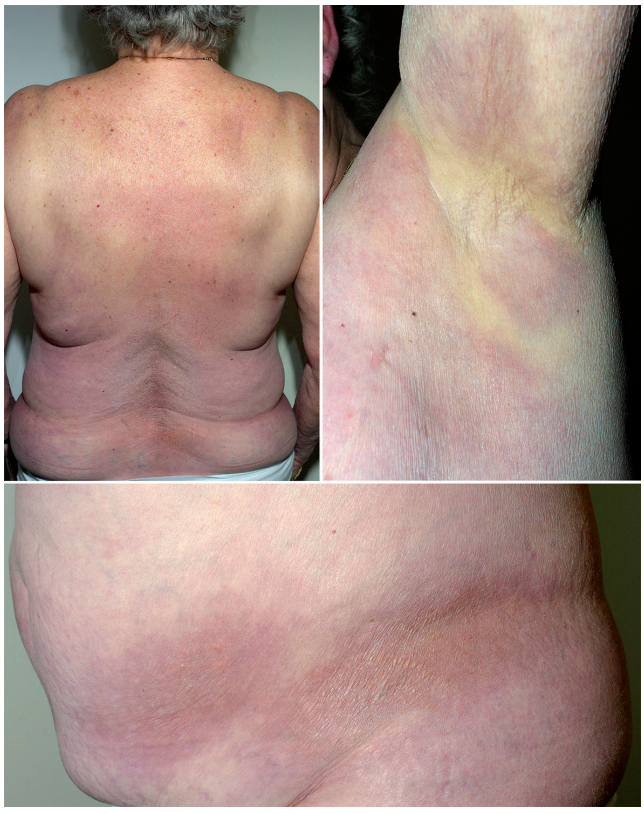

Fig. 2. Sclerotic erythematous lesions involving the flanks, the lumbosacral area, the abdomen and the axillary regions (Case 2)**.**

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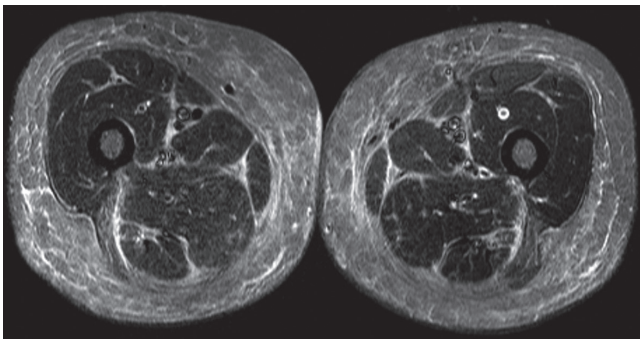

Fig. 3. Magnetic resonance imaging showing deep infiltration of subcutaneous fat and fascia in both thighs.

protein (CRP) and erythrocyte sedimentation rate (ESR), and a discrete hypergammaglobulinaemia. Magnetic resonance imaging (MRI) showed a deep infiltration of subcutaneous fat and fascia of the abdomen and thighs (**Fig. 3**). Deep biopsy of the skin and subcutaneous fat including the fascia (**Fig. 4**a) revealed fibrosis and a moderate lymphoplasmacytic infiltrate in the deep dermis and dermal-subcutaneous fat junction (Fig. 4b). Diffuse oedema and a lymphoplasmacytic infiltrate with scarce eosinophils were observed in the fascia (Fig. 4c). Western blotting was repeatedly positive for *Borrelia spp*. IgM, whereas anti-VlsE was negative, as was PCR of the deep tissue. EF was diagnosed, possibly associated with a chronic infection with *Borrelia spp*. Following a one-month course of intravenous ceftriaxone 2 g once a day with no other therapy, complete remission was observed.

DISCUSSION

EF is a rare disorder characterized by scleroderma-like skin induration and fascia thickening, with or without eosinophilic infiltration, depending on the stage of the disease. The aetiology of EF is uncertain; *Borrelia spp*. has been proposed as a possible triggering factor (4–7). Treatment with oral corticosteroids remains the standard therapy for EF, taken alone or in association with an immunosuppressant drug (12).

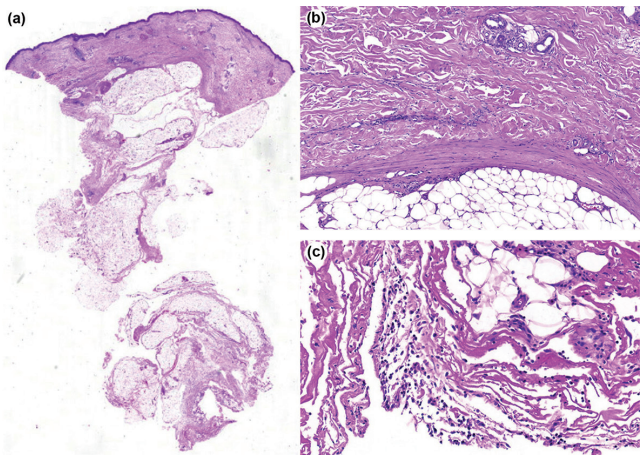

Fig. 4. Deep cutaneous biopsy showing fibrosis and moderate lymphoplasmacytic infiltrate in the deep dermis and dermal subcutaneous fat junction, and diffuse oedema and lymphoplasmacytic infiltrate with scarce eosinophils in the fascia (original magnification: a ×2; b, c ×20).

In our first case, positive Western blot for *Borrelia spp*. IgG, highly positive anti-VlsE and partial clinical response to intravenous ceftriaxone associated with a decreased level of anti-VlsE were strongly suggestive of a causative role of *Borrelia*. New studies explore whether *Borrelia* infection can induce an autoimmune reaction, which may persist even after the eradication of the pathogen and explain the partial response to antibiotics (13, 14). Meanwhile, we cannot exclude a spontaneous remission in that case.

In the second case, the clinical presentation, Western blotting repeatedly positive for *Borrelia spp*. IgM, and the complete resolution of symptoms after intravenous ceftriaxone was strongly in favour of the causative role of *Borrelia* in EF. It had been suggested that *Borrelia spp*. infection drives the humoral response away from protective, high-affinity, and long-lived antibody responses and toward the rapid induction of strongly induced, short-lived antibodies of limited efficacy, explaining the presence of IgM and the absence of IgG in our case (15).

We could not precisely determine the exact species of Borrelia implicated in these cases, as Western blot used in our laboratory detects antigens of 4 species of *Borrelia*: *B. afzelii, B. garinii, B. burgdorferi* and *B. spielmanii*, but cannot distinguish between the species due to cross-reactivity of the antigens. Anti-VlsE also recognizes *Borrelia spp*. antigens that are common to the different species, and currently the precise identification of *Borrelia* species can only be done by PCR analysis.

In conclusion, *Borrelia spp.* should be screened in EF, and laboratory studies should be repeated in cases of strong clinical suspicion of *Borrelia spp.* as a causative agent.

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