CASE REPORT

Borrelial pseudolymphoma of the nose

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SUMMARY

A 52-year-old Colombian woman, a patient with psoriasis, undergoing phototherapy with (ultraviolet B narrowband) UVBnb, presented with a symptomless solitary diffuse erythematous plaque on her nose for 3 months. Initially, she was treated with pimecrolimus 1% cream for 8 weeks, which was then combined with metronidazole cream for 4 weeks, with the initial diagnosis of UV-triggered rosacea, without improvement. A punch biopsy was performed and the histology showed a pseudolymphomatous reaction. The diagnosis of nasal pseudolymphoma of borreliosis was confirmed with PCR. The lesion completely resolved following oral doxycycline therapy.

BACKGROUND

Cutaneous pseudolymphomas are reactive lymphocytic proliferations that appear in the skin and resemble a malignant lymphoma. Pseudolymphoma is not a specific disease but rather an inflammatory response to known or unknown stimuli. The term pseudolymphoma without modification should be reserved for idiopathic cases. It is important to identify those pseudolymphomas in which the aetiology is clear. In endemic areas a common cause of pseudolymphoma is *Borrelia burgdorferi*, transmitted by tick bites.

CASE PRESENTATION

A 52-year-old Colombian woman presented with a symptomless solitary diffuse erythematous plaque on her nose for 3 months (figure 1). She had been undergoing phototherapy for psoriasis. During (ultraviolet B narrowband) UVBnb treatment, she developed erythema of the nose. Initially

diagnosed as UV-triggered rosacea, she was treated with pimecrolimus 1% cream for 8 weeks, which was then combined with metronidazole cream for 4 weeks, without improvement.

INVESTIGATIONS

Owing to persistent erythema of the nose, a punch biopsy was performed. Routine histology showed a dense superficial and deep dermal infiltration of lymphocytes, histiocytes and plasma cells (figure 2). Around 60% of the cells in the infiltrate were CD3-positive T-cells (figure 3), the other 40% were CD20-positive B-cells (figure 4). The λ and κ stains provided no evidence for a light-chain restriction. The histology was interpreted as a pseudolymphomatous reaction. An immunoblot of serum antibodies was reactive for six IgG antigens and one IgM antigen of *B. burgdorferi*, while PCR amplified *B. burgdorferi*-specific sequences from the biopsy material.

DIFFERENTIAL DIAGNOSIS

- ▶ Rosacea
- ▶ Lupus pernio, sarcoidosis
- ▶ Photo-allergic reaction
- ▶ Lymphoma
- ▶ Eczema
- ▶ Amyloidosis
- ▶ Malar rash in systemic lupus erythematosus

TREATMENT

With the diagnosis of nasal pseudolymphoma of borreliosis, treatment with oral doxycycline 100 mg twice daily was given for 21 days.

Figure 1 Diffuse erythematous plaque on the center of the nose.

Figure 2 Pseudolymphomatous reaction of lymphocytes, histiocytes and plasma cells within the dermis (H&E stain).

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Figure 3 Sixty per cent of lymphocytes in the infiltrate express T-cell markers (CD3 stain).

OUTCOME

Complete remission at 5 weeks.

DISCUSSION

B. burgdorferi is the major causative agent of cutaneous pseudolymphoma in endemic regions; it is also known as borrelial lymphocytoma (BL).¹ It is caused by tick bites and transmission of bacterium of the *Borrelia* genus.² The clinical presentation is usually a solitary, diffusely erythematous, painless, non-scaly plaque or nodule with rare association of extracutaneous symptoms³ and with an average interval of 5.5 weeks between the tick bite and the clinical cutaneous manifestation.⁴

It typically develops on the ear in children and the nipple or areola in adults. It develops also on the face, trunk or scrotum in almost 84.9% of patients. It has been suggested that *Borrelia* spirochetes may have a tropism for cooler body sites.⁵ The clinical diagnosis of BL is highly suggestive considering the typical location and the history of the illness, but confirmation through laboratory testing is recommended.² Histological criteria for diagnosis of BL

Figure 4 Forty per cent of lymphocytes in the infiltrate express B-cell markers (CD20 stain).

include presence of a top-heavy, wedge-shaped mixed-cell lymphoid infiltrate, characterised by nodular clusters of B-lymphocytes (CD20+) with presence of (CD21+) follicular dendritic cells. A large number of T-lymphocytes (CD3+) can sometimes be found around the B-cell areas.⁵ Lymphoproliferative lesions of the skin include pseudolymphomas, atypical lymphoid proliferations and lymphomas. Sometimes it is difficult to separate these lesions from each other.⁶

Serology is the gold standard for the diagnosis of borreliosis. Usually, western blotting for several antigens is performed to increase the sensitivity and specificity of the serological test. PCR is sometimes used for ambiguous cases and can be applied to a tissue specimen directly. The sensitivity reported is at 67.5% for the detection of *Borrelia* sequences in reported series.⁵ Culture is not routinely used, because its performance and outcome are inferior to the methods discussed above.⁷ The recommended first-line treatment for BL is oral amoxicillin (500 mg 3×/day for adults, 50 mg/kg per day for children) or oral doxycycline (200 mg per day) for 14–21 days.^{2,7}

Learning points

- ▶ Atypical presentations of borrelial lymphocytoma are not uncommon and should be included in the differential diagnosis and work-up with biopsy and serology for erythematous lesions of the face.
- ▶ Biopsy is necessary to establish a diagnosis of pseudolymphoma.
- ▶ The response to treatment with doxycycline 100 mg twice daily for 3 weeks in our case was as expected with a complete remission, underlining the need for a broad work-up of such clinical presentations to quickly initiate an appropriate treatment.
- ▶ Patients may not give a history of tick bite and physicians should be aware of this in endemic areas.

Competing interests None.

Patient consent Obtained.

Provenance and peer review Not commissioned; externally peer reviewed.

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