

Lyme disease: a rare form of presentation

P. Ramos*, C. Fernandes**, J. Moreira***, A. Ventura****, M. Riscado****

Abstract

The authors present the case of a 32 year old male presenting classic features of Erythema Chronicum Migrans. Significant titers for *Borrelia burgdorferi* were then obtained. Following antibiotic treatment with tetracycline, a good response was obtained, with rapid disappearance of all the skin lesions. After two months, new secondary skin lesions of Lyme disease were observed, together with intense, bilateral and periorbital edema and purulent conjunctivitis. Once again, a good response was obtained with

doxycycline, with rapid disappearance of all the symptoms. In the following months, at almost monthly intervals, the patient suffered multiple episodes of recurrence with widespread secondary cutaneous lesions characteristic of Lyme disease. Definitive remission was then obtained with the use of ceftriaxone IV.

Key words: Lyme disease, periorbital edema, purulent conjunctivitis.

Introduction

Lyme disease is an infection with varied clinical manifestations and it is currently well defined. The ocular compromise that is characteristic of the secondary phase of the disease is an infrequent form of manifestation, occurring in only around 3% of cases¹.

Resistance to antibiotics, as well as certain clinical manifestations, appears to be associated with particular immunopathogenic characteristics, in relation to H.L.A², as we shall see in the case described here.

Clinical Case

A 32 year old male, white, a soldier, residing in Amadora, healthy, examined in August 1992 due to the appearance, about six weeks previously, of pruriginous red pimples on the left knee and trunk, of about 10 cm in diameter, confluent, migratory, and developing in about 3-5 days (figures 1 & 2). The patient had already been treated with ampicillin and amoxicillin at the outpatient clinic, without any clinical improvement.

A clinical suspicion of the Erythema Chronicum

Migrans stage of Lyme Disease was subsequently confirmed through positive serology with significant titers against *Borrelia burgdorferi* (IgM:1/320). Therapy with tetracycline (500 mg. t.i.d., P.O.) was then administered for twelve days, with complete remission of the condition and a clear reduction in serum titers in the subsequent weeks.

Approximately two months afterwards, the progressive appearance was observed of multiple round skin lesions, pruriginous, with a clearly-defined, round border, affecting both limbs, the trunk, face and scalp, resulting in hospitalization.

Twenty four hours after the observation, unexpected rapid, bilateral onset of intense periorbital edema and purulent conjunctivitis was observed, with inability to open the eyelids, fever (37.9°C) and arthralgias, particularly involving the metacarpus-phalangeal joints, elbows and knees.

Subsequent tests carried out in the laboratory showed unaltered hemogram, ESR 10 mm in the first hour, biochemistry unaltered (including P.F.H., P.E.R. and urine type II), coagulation within the normal limits, negative VDRL, immunoglobulin levels without alterations, CPR. = (4+), negative Widal, Huddleson and Wright, cryoglobulin (2+), C.I.C. 7ug/ml, and HLA (+) for Drw2 and Drw4.

Given the evidence of multiple secondary impairment (ocular, periorbital and cutaneous involvement) characteristic of Lyme disease (stage II), tetracycline therapy was initiated (500 mg, q.i.d, P.O. over 15 days), with excellent results; the symptomatology was completely reduced on the 5th day of therapy.

Over the nine subsequent months, seven episodes

*Resident to the Internal Medicine Supplementary Internship

**Resident to the Dermatology Supplementary Internship

***Internal Medicine Hospital Assistant

****Internal Medicine Hospital Senior Assistant

Medicine Service 2 and Dermatology Service.

Hospital Curry Cabral.



FIG. 1



FIG. 2

of recurrence of secondary cutaneous lesions were observed, with similar characteristics and evolution to those diagnosed previously, and which were not accompanied by any other symptomatology. The interval of respite between recurrences was, on average, 35 days (± 4 days), on which the patient remained asymptomatic. The serum titers remained high, with significant values (1/200, 1/156, 1/225), during the nine months in which recurrences were observed. The various episodes were treated in the same way, with tetracycline (500 mg q.i.d for 15 days) as well as doxycycline (100mg b.i.d. for 15 days) and the symptoms rapidly decreased after the start of treatment.

Given the clear evidence of resistance to the antibiotic therapy, and following the appearance of new secondary lesions similar to those previously described, ceftriaxone (3 gr/day/I.V. for 10 days) was prescribed, under hospitalization, with rapid resolution and remission of the symptoms. Since then there have been no other recurrences, and after several months, the serum tests were negative.

Discussion

The case described above shows the unpredictable variability classically associated with Lyme disease. Initiating, in this case, with the characteristic cutaneous lesion of Erythema Chronicum Migrans and confirmed by serology in significant titer², it was not, however, accompanied by the general manifestations usually associated with it, such as fever, arthralgia, myalgia, malaise or fatigue.^{1,3,4,5,6,7}

The appearance of multiple round cutaneous lesions is a frequent finding, occurring in around half

of all cases (48%).^{3,4,7} These were also found in the case described, demonstrating the ineffectiveness of the treatment and progression of the disease to a further level.⁶ Neurological and cardiac impairment are also common (present in 15% and 8% of cases), characterized by meningeal irritation, peripheral facial paralysis, neuritis or cardiac blockage to varying degrees, respectively.^{5,7} Ocular impairment, manifested in this case by purulent bilateral conjunctivitis and periorbital edema, is a rare form of presentation, occurring in only 11% and 3% of cases.^{1,7,8}

The uncharacteristic evolution, marked by successive episodes of recurrence after therapy, and the rare visceral compromise (ocular involvement) observed may be related to the particular immunopathogenic characteristics of the individual patient. Indeed, a particular association has been found between HLA-Drw2/Drw4 and the evolution to chronic disease,^{1,7,9} in which resistance to classic antibiotic treatment is not uncommon.

The therapy, which generally involves tetracycline or doxycycline^{1,2}, was clearly ineffective in this case. Therefore, in apparently resistant forms, or those associated with a typical visceral compromise in advanced stage, intravenous use of penicillin or a 3rd generation cephalosporin of (ceftriaxone) is prescribed.¹⁰ ■

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