REV BRAS REUMATOL. 2015;xxx(xx):xxx–xxx

REVISTA BRASILEIRA DE REUMATOLOGIA

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Case report

Lyme disease and juvenile idiopathic arthritis – a pediatric case report☆

Mário Correia de Sá[a,*], *Catarina Moreira*[b], *Cláudia Melo*[c], *Álvaro Sousa*[c], *Sónia Carvalho*[c]

[a] *Serviço de Pediatria, Centro Hospitalar de Vila Nova de Gaia/Espinho, Vila Nova de Gaia, Portugal*
[b] *Unidade de Saúde Familiar Ribeirão, Vila Nova de Famalicão, Portugal*
[c] *Serviço de Pediatria, Centro Hospitalar do Médio Ave, Vila Nova de Famalicão, Portugal*

ARTICLE INFO

Article history:
Received 2 March 2015
Accepted 14 August 2015
Available online xxx

Keywords:
Juvenile idiopathic arthritis
Lyme disease
Eritema migrans

ABSTRACT

Introduction: Lyme disease, a pathology secondary to infection by bacteria of the genus *Borrelia*, can present with erythema migrans and monoarticular or oligoarticular arthritis. The relationship between infection with this agent and Juvenile Idiopathic Arthritis has been already mentioned.
Case report: In this paper, the case of a 6-year-old female child, seen at our outpatient clinic because of symptoms compatible with Lyme disease confirmed by a serological work-up, is described. Erythema migrans lesions disappeared with the appropriate antibiotic treatment. The resolution of joint symptoms was only transitory; chronic anti-inflammatory and immunosuppressant treatment was needed.
Discussion: In this case, the resolution of cutaneous symptoms and the persistence of chronic arthritis suggest that the strong likelihood of infection by *Borrelia* has triggered Juvenile Idiopathic Arthritis.

Doença de Lyme e artrite idiopática juvenil – Relato de caso clínico pediátrico

Palavras-chave:
Artrite idiopática juvenil
Doença de Lyme
Eritema migrans

RESUMO

Introdução: A doença de Lyme, patologia secundária à infeção por bactérias do género *Borrelia*, cursa com *eritema migrans* e artrite monoarticular ou oligoarticular. Foi já apontada a relação entre a infeção por este agente e a Artrite Idiopática Juvenil.
Caso clínico: Descreve-se o caso de uma criança de 6 anos, sexo feminino, avaliada em consulta externa por sintomatologia compatível com Doença de Lyme confirmada por estudo

☆ Study conducted at Serviço de Pediatria, Centro Hospitalar do Médio Ave, Vila Nova de Famalicão, Portugal.

* *Corresponding author.*
E-mail: mario.s.sa@gmail.com (M.C. de Sá).

http://dx.doi.org/10.1016/j.rbre.2015.09.006

Please cite this article in press as: de Sá MC, et al. Lyme disease and juvenile idiopathic arthritis – a pediatric case report. Rev Bras Reumatol. 2015. http://dx.doi.org/10.1016/j.rbre.2015.09.006

serológico. As lesões de *eritema migrans* resolveram com o tratamento antibiótico adequado. A resolução da sintomatologia articular foi apenas transitória necessitando a criança de tratamento anti-inflamatório e imunossupressor crónico.

Discussão: A resolução da sintomatologia cutânea e a persistência de artrite crónica aponta para uma forte probabilidade de neste caso a infeção por *Borrelia* ter desencadeado Artrite Idiopática Juvenil.

Introduction

Lyme disease (LD) is an infectious disease caused by spirochetes of the genus *Borrelia* and transmitted by tick bite.

The clinical presentation of LD is divided into three separated phases: an early localized disease, characterized by erythema migrans (EM), an early disseminated disease with potential involvement of the central nervous system and heart, and a late stage of disease with monoarticular or oligoarticular arthritis of large joints. During the early phase, oral antibiotics and, in the case of disseminated disease with neurological or cardiac damage, intravenous antibiotics, are indicated. The arthritis of LD (late stage) should be initially treated with oral antibiotics for a month; intravenous treatment should be limited to patients with severe or persistent disease.

Several studies have suggested a possible influence of several infectious agents, including *Borrelia*, in the etiopathogenesis of Juvenile Idiopathic Arthritis (JIA).[1]

Case report

A 6-year-old female patient with no relevant past medical history was referred to Pediatric Rheumatology consultation, with complaints of pain and swelling of the proximal interphalangeal joints (PIP) of hands and wrist and tibiotarsal joints bilaterally with several months of development and progressive worsening. There was no fever or history of trauma. She resided in an urban environment in northern Portugal, but with regular visits to relatives living in a rural area, where she had contact with dogs. The family history was irrelevant.

The girl showed multiple circinate erythematous lesions of 2–5 cm diameter, with a 5-month progression, and was refractory to oral and topical antifungal treatment. At physical examination, the patient had signs of inflammation and limitation on active and passive mobilization of all PIP joints of her hands (Fig. 1) and of the knee, tibiotarsal joints and wrists bilaterally; and also multiple circinate erythematous lesions with irregular shapes, scattered on the trunk, upper and lower limbs and neck (Fig. 2).

Given the clinical picture suggestive of LD, antibiotic therapy was instituted with oral amoxicillin 1.5 g/day and ibuprofen 30 mg/kg/day for 21 days.

The laboratory investigation showed normal complete blood count (CBC) and general biochemistry (renal function, ALT, AST, alkaline phosphatase, thyroid function, ionogram); erythrocyte sedimentation rate 24 mm/1st hour, C-reactive protein 1 mg/dL, immunological studies with ANA, ANCA and rheumatoid factor negative, C3 and C4 slightly increased (184 mg/dL and 46 mg/dL, respectively), and serology (HIV, CMV, EBV, toxoplasma, VDRL, Weil-Felix reaction and Wright reaction) negative. Serological studies for *Borrelia burgdorferi* were positive (indirect immunofluorescence, IgG 53.30 AU/mL [positive, >10 AU/mL]; IgM: 1.37 [positive, >1.09]). In the face of such results, the diagnosis of EM and arthritis in a context of Lyme disease has been confirmed. The child showed no changes in her heart and eye examination.

In spite of the treatment, the child related the same complaints of arthralgia, with the appearance of new lesions of EM. A cycle of 28 days of intravenous ceftriaxone 2 g/day was then introduced, with complete disappearance of skin lesions.

The resolution of joint symptoms was only temporary; about two months later, re-aggravation of arthralgia complaints and mobility limitation of wrist and PIP joints occurred. There was no recurrence of other signs of arthritis and even of EM.

Taking into account the persistence of the signs and symptoms of chronic arthritis, anti-inflammatory treatment with oral deflazacort (7.5 mg/day) and naproxen (500 mg/day) and also immunosuppression with oral methotrexate (14.5 mg/m^2/week) were initiated. Then the child presented a progressive improvement of her pain complaints, but with persistence of a slight limitation to wrist extension. Analytically, there were no new changes. Currently, the patient remains in remission, depending on this therapy; her behavior and response to therapy are absolutely identical to

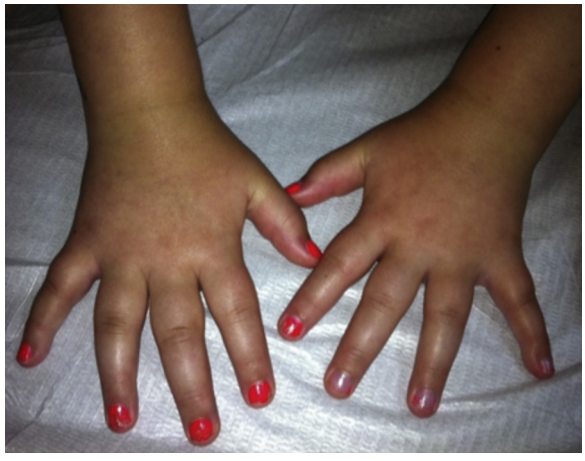

Fig. 1 – Proximal interphalangeal joints with inflammatory arthritis observed in a Pediatric Rheumatology consultation.

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