REV BRAS REUMATOL. 2014;xxx(xx):xxx–xxx

REVISTA BRASILEIRA DE REUMATOLOGIA

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Case report

Presence of riziform bodies in a patient with juvenile idiopathic arthritis: case report and literature review☆

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ARTICLE INFO

Article history:
Received 17 April 2014
Accepted 14 September 2014
Available online xxx

Keywords:
Riziform bodies
Juvenile idiopathic arthritis
Tuberculosis
Chronic monoarthritis

ABSTRACT

Riziform bodies are structures formed by fibrin and cells that can be found in the synovial fluid or attached to the synovium, and have this denomination due to its rice grain-like appearance. They have already been described in several diseases such as tuberculous arthritis, rheumatoid arthritis, and rarely in juvenile idiopathic arthritis (JIA). This is the case of a boy with a 4-month course of chronic monoarthritis of the left knee, with family history of sarcoidosis in which diagnostic investigation showed the presence of these riziform bodies in the synovial biopsy. Diagnostic investigation ruled out sarcoidosis, tuberculosis and malignancies, establishing the diagnosis of JIA. Our objective was to describe what we believe is the 9th case reported on the presence of riziform bodies in JIA, which are probably underdiagnosed, and should be considered mainly in cases of severe arthritis of difficult medical treatment.

Presença de corpos riziformes em paciente com artrite idiopática juvenil: relato de caso e revisão de literatura

Palavras-chave:
Corpos riziformes
Artrite idiopática juvenil
Tuberculose
Monoartrite crônica

RESUMO

Corpos riziformes são estruturas constituídas por fibrina e células e que podem ser encontradas no líquido sinovial ou aderidos à sinóvia. Recebem essa denominação por sua

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http://dx.doi.org/10.1016/j.rbre.2014.09.004

Please cite this article in press as: Campos LR, et al. Presence of riziform bodies in a patient with juvenile idiopathic arthritis: case report and literature review. Rev Bras Reumatol. 2014. http://dx.doi.org/10.1016/j.rbre.2014.09.004

aparência semelhante a grãos de arroz. Já foram descritos em várias enfermidades como a artrite tuberculosa, a artrite reumatoide e, raramente, a artrite idiopática juvenil (AIJ). Trata-se de um caso de um menino com monoartrite crônica de joelho esquerdo de quatro meses de evolução, com história familiar de sarcoidose em cuja investigação diagnóstica evidenciou-se a presença desses corpos riziformes na biópsia sinovial. A investigação diagnóstica afastou sarcoidose, tuberculose e malignidades e estabeleceu-se o diagnóstico de AIJ. Nosso objetivo foi descrever o que acreditamos ser o nono caso relatado sobre a presença de corpos riziformes em AIJ, que devem ser subdiagnosticados e deveriam ser pensados principalmente em casos de grandes artrites de difícil tratamento clínico.

Introduction

Juvenile idiopathic arthritis (JIA) is the most common form of chronic arthritis in pediatric patients, and its diagnosis is made following exclusion of several other conditions that may present with prolonged musculoskeletal manifestations. The most common form of JIA is the oligoarticular one, which may present as a chronic monoarthritis.[1] In these cases, there are multiple diagnoses to be investigated: tuberculosis, sarcoidosis, villonodular synovitis, hemarthrosis, hemangioma, synovial osteochondromatosis, arborescent lipoma, malignancies and some autoinflammatory diseases.[2–4] Our goal is to report the case of a child with chronic monoarthritis whose biopsy showed a great amount of riziform bodies that are seldom described in this age group. This is the 9th case reported in literature about the presence of riziform bodies in patients with JIA and, to our knowledge, the first case reported in Brazil.

Case report

A male 8-year-old child, 2nd twin, born in the city of Rio de Janeiro, had a history of difficulty practicing exercises, with limited range of motion of the left knee, approximately four months before the 1st consultation. After a month, the parents noticed a great increase in volume that remained until the day of consultation. Within this period, there was no fever, skin lesions or any signs or symptoms of involvement of other organs. There was no previous history of trauma or infection in the three months before the onset of symptoms. He has a healthy twin brother, there is no family history of spondyloarthritis, and the mother had a facial skin lesion about 25 years ago, diagnosed as sarcoidosis, and treated with intralesional corticosteroids. Physical examination in the 1st consultation was normal except for the presence of large swelling of the left knee, with heat and slight hyperemia, piano key sign and motion limitation (flexion at 60° and extension at 150°).

With the syndromic diagnosis of chronic monoarthritis, tests were ordered, the results of which showed blood count, C-reactive protein, erythrocyte sedimentation rate, lipid profile, blood glucose, calcium, urea, creatinine, complement, muscle enzymes, protein electrophoresis, angiotensin converting enzyme (ACE) and urinary sediment within normal ranges. The rheumatoid factor, antinuclear antibodies and HLA B-27 were negative. Serologic markers for hepatitis B and C, toxoplasmosis, HIV and HTLV were also negative. Serology for rubella and cytomegalovirus showed negative IgM. The tuberculin test was negative and chest radiograph was normal. Also, his electrocardiogram, echocardiogram, and ophthalmologic evaluation were normal. Left knee X-ray showed only soft tissue swelling, and ultrasonography (USG) showed joint effusion with debris and synovial thickening. Magnetic resonance imaging showed massive joint effusion with heterogeneous signal in the presence of multiple small elongated images with hypotensive signal intensity on all sequences, suggesting riziform bodies, and pronounced enhancement of the synovium (Fig. 1). Left knee synovial biopsy was required and, in surgery, there was output of minimal amount of synovial fluid (SF) and a moderate amount of white mass, similar to rice grains (Fig. 2). Anatomic pathology showed, on macroscopic examination, the presence of numerous oval, friable, white and firm structures consistent with the diagnosis of riziform bodies. Microscopy showed synovium fragments with hyperplasia of lining cells, and extensive deposits of fibrin, with edema, vascular ectasia and neogenesis seen in the stroma, and lymphocytic inflammatory infiltrate occasionally aggregated in follicles, and some granulocytes (Fig. 2). The material represented by riziform bodies consisted of organized fibrin and had pervading mononuclear cells, and some polymorphonuclear cells. The histological report was suggestive of chronic inflammatory synovitis, or JIA. The patient was initially treated, before surgery, with non-steroidal anti-inflammatory drugs, with no clinical response. After surgery, methotrexate was added to the treatment and, in about two months, the patient was asymptomatic.

Discussion

Although imaging and laboratory studies suggest the diagnosis of JIA, synovial biopsy was requested in order to rule out sarcoidosis, not confirmed by histopathology. At surgery, a small amount of synovial fluid was found, along with the presence of a moderate amount of whitish mass that histopathology showed to be riziform bodies.

Riziform bodies are structures which can be found in synovial fluid or adhered to synovium, and have this denomination for its similar appearance to the rice grains. They consist of fibrin involving mononuclear cells, polymorphonuclear cells and red blood cells, and represent a nonspecific

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