

FIBROBLASTIC RHEUMATISM TREATED WITH METHOTREXATE AND COLCHICINE

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ABSTRACT – Objective: Fibroblastic rheumatism (FR) is a rare disease first described by Chaouat in 1980 which is characterized by a combination of rheumatologic and dermatological manifestations. Rheumatologic features are symmetrical polyarthralgia with joint stiffness, associated with cutaneous nodules and sclerodactyly. Histology shows an increased number of fibroblasts and marked dermal fibrosis. In this report, we described a rare cutaneous disease-fibroblastic rheumatism, in which the patient had an excellent response to methotrexate associated with colchicine.

Case Presentation: A 13 years old boy who was previously healthy started in 2013 deformities over his fingers without pain or any inflammatory signs associated with joint stiffness. This is the second Brazilian case of FR treated with methotrexate and colchicine.

Conclusions: The patient observed a slight improvement in cutaneous nodules and joint stiffness. Though FR is a rare disease, early diagnosis is mandatory. This case reinforces that MTX associated with colchicine may be an additional therapeutic tool for these patients.

KEYWORDS: Fibroblastic rheumatism, Cutaneous nodules, Fibrosis, Peyronie's disease, Methotrexate, Colchicine.

INTRODUCTION

Fibroblastic rheumatism (FR) is a rare disease characterized by joint manifestations associated with cutaneous nodules and sclerodactyly¹. The critical mark is the histologic feature of the nodules that are represented by the proliferation of fibroblasts in the dermis.

With the present case included, 43 case reports on RF were already described in the literature. An excellent response to methotrexate is usually observed, mainly in the nodule's size reduction. We have previously described a case with a good response to methotrexate².

Therefore, this article describes an additional Brazilian case of fibroblastic rheumatism treated with methotrexate and colchicine.

CASE REPORT

A 13 years old boy who was previously healthy started in 2013 deformities over his fingers without pain or any inflammatory signs associated with joint stiffness. He denied fever, weight loss, skin alterations, or any other clinical manifestation. His physical examination revealed nodules over the proximal interphalangeal joints of the hands and flexion contractures of these proximal interphalangeal





Figure 1. Subcutaneous nodules on the extensor face of the metacarpophalangeal joints of the hands and flexion of the interphalangeal joints.

joints. Sclerodactyly and contracture of the fingers and toes were also observed (Figure 1). The active motion was limited. Laboratory tests demonstrated positive antinuclear antibodies in a low titer of 1:80, speckled pattern, with negative rheumatoid factor, ANCA, anti-CCP, antinuclear antibodies, anti-dsDNA, anti-Sm, anti-U1RNP, anti-Ro/SS-A, anti-La/SS-B, anti-Scl-70, anticentromere, anti-Jo-1 and anticardiolipin antibodies. The erythrocyte sedimentation rate was 8 mm in the first hour, and the C-reactive protein level was less than 5 mg/L. X-rays of the hands did not show erosions or joints involvements. Lung function test, chest x-ray, Doppler echocardiogram, and abdominal ultrasounds were all normal. Magnetic resonance imaging (MRI) of the right hand showed solid nodular formation in the dorsal subcutaneous tissue, tenosynovitis of the flexors, and synovitis of the second, third, fourth, and fifth metacarpophalangeal joints (MCPs). Biopsy specimens of one nodule revealed the proliferation of spindle-shaped cells focally arranged in a storiform pattern, thickened collagen fibers, and dermal fibrosis. These alterations were compatible with the diagnosis of FR. Methotrexate was then initiated at 10 mg per week associated with folic acid 5 mg/week, and the dose increased to 20 mg/week; in the subsequent visit, colchicine 1 mg/day was added. The patient felt better, slightly reducing the nodules and stiffness.

DISCUSSION

In this report, the authors describe a rare cutaneous disease-fibroblastic rheumatism, in which the patient had an excellent response to methotrexate associated with colchicine.

FR is a rare disease, first described in 1980¹, and until now, there are only 37 cases in the medical literature¹⁻²⁰. The typical clinical presentation is cutaneous nodules, polyarthralgia or polyarthritis, contracture of the fingers, and sclerodactyly associated with characteristic histologic features.

The pathogenesis of this disease is unknown. However, histologic findings show dermal and synovial fibrosis may be due to fibroblastic proliferation rather than increased collagen synthesis.

A large number of therapeutic options have been tried for FR, including hydroxychloroquine¹⁰, non-steroidal anti-inflammatory drugs (NSAIDs)³, glucocorticosteroids¹¹, interferon-alpha¹¹, colchicine¹², D-penicillamine¹³, and methotrexate (MTX)¹⁶⁻²⁰.

Regarding MTX, this drug was successfully used in the treatment of FR for the first time in 1996. There are at least 12 cases of RF treated with MTX, and most subjects responded well. Notably, at least one patient with complete disease resolution was described with this immunosuppressive drug^{4,7,14,15-20}.

CONCLUSIONS

Here, we described a Brazilian FR case with an excellent response to MTX and the colchicine association. The patient observed a slight improvement in cutaneous nodules and joint stiffness. Though FR is a rare disease, early diagnosis is mandatory. This case reinforces that MTX associated with colchicine may be an additional therapeutic tool for these patients.

CONFLICTS OF INTEREST:

The Authors have no conflict of interest to declare.

INFORMED CONSENT:

An informed and signed consent was collected from the patient's parents.

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