MACROPHAGIC MYOFASCIITIS: A CASE REPORT OF AUTOIMMUNE/INFLAMMATORY SYNDROME INDUCED BY ADJUVANTS [ASIA]

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Introduction

Macrophagic myofasciitis is an immune mediated disease known at least since 1993^{1,2}. This disease has unclear etiology, although it is often related with aluminium hydroxide adjuvant used in vaccines, as depicted in an electron microscopy study³. It manifests with myalgia, arthralgia, marked asthenia, muscle weakness, chronic fatigue, and low grade fever. Some authors postulate that this disease might be a feature of a common syndrome: ASIA – autoimmune/inflammatory syndrome induced by adjuvants. It is estimated that 30 % of the patients have elevations of creatinine kinase (CK) and less than 30 % have a myopathic electromyogram^{1,4}.

Case Report

This case report refers to a 47 years-old female, observed due to diffuse mechanical arthralgia, low back pain, asthenia and fatigue that lasted for more than 4 years. The observation was normal, except for the presence of two Heberden nodes, and 12 fibromyalgia tender points, fulfilling the classical ACR diagnostic criteria for this disease. The laboratory evaluation showed slightly increased (399 U/L; N 33-211 U/L) creatinine kinase and ESR (41 mm/1st hour), she was HLA-B27 positive and anti-nuclear antibodies (including anti-Jo-1) were negative. The radiographs were compatible with osteoarthritis affecting the cervical and dorsal spine, as well as the hips, shoulders and hands. Sacroiliac joints were normal. Treatment with glucosamine sulphate, paracetamol,

*Rheumatology Research Unit, Instituto de Medicina Molecular, Faculdade de Medicina da Universidade de Lisboa **Rheumatology Department, Centro Hospitalar de Lisboa Norte, EPE, Hospital de Santa Maria, Lisbon, Portugal ***Neurology Department, Centro Hospitalar de Lisboa Norte, EPE, Hospital de Santa Maria, Lisbon, Portugal NSAIDs and cyclobenzaprine was ineffective. Due to slight diminished proximal strength a muscle biopsy was performed and showed features compatible with macrophagic myofasciitis. As shown in Figure 1, there is a centripetal infiltration of the endomysium by sheets of large cells of the monocyte/macrophage lineage, absence of necrosis, of both epithelioid and giant cells, and of mitotic figures. These features might also be found in the inflammatory myopathy with abundant macrophages (IMAN), usually associated with dermatomyositis features that were absent in this patient⁵. No vaccine correlation could be established, although there was a Tetanus-pertussis vaccination three years before. The electromyography was normal. Prednisolone 0,5 mg//kg/day was started with slight improvement.

Conclusion

Macrophagic myofasciitis is a rare entity within the context of inflammatory myopathies and fascii-

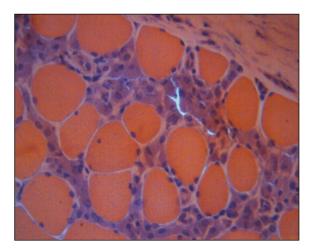


Figure I. Muscle biopsy, 400x, Hematoxilin & Eosin, macrophagic perifascicular infiltrate

tides. It does not correspond to any of the previously described histiocytoses or any known macrophage-overload disease. The clinical manifestations are unspecific and the diagnosis can only be established on a muscular biopsy with fascia. In this particular case, the abundant macrophagic infiltrate of the endomysium might also suggest IMAN, although the absence of dermatomyositis features is against this hypothesis⁵. No precise treatment recommendations have been established, but the disease tends to respond to steroids and immunossupressants, although antibiotics have also been used^{2,6,7}.

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