Case Report

Non-L-Tryptophan Related Eosinophilia-Myalgia Syndrome with Hypoproteinemia and Hypoalbuminemia

LEON MARGOLIN

ABSTRACT. Eosinophilia-myalgia syndrome (EMS) has been associated with L-tryptophan use since 1989; of the 1345 EMS cases reported, 191 (14%) were not related to the use of L-tryptophan. A case is described of non-L-tryptophan related EMS associated with transient hypoproteinemia and hypoalbuminemia that resolved with prednisone treatment. (J Rheumatol 2003;30:628–9)

> Key Indexing Terms: EOSINOPHILIA-MYALGIA SYNDROME HYPOPROTEINEMIA

L-TRYPTOPHAN HYPOALBUMINEMIA

The epidemia of the eosinophilia-myalgia syndrome (EMS) that started in the USA after use of L-tryptophan (LT) in the food industry in 1989 is well known. However, since the early 1990s after LT was eliminated from the market few if any cases have been reported.

EMS has been associated with LT use since 1989, but as yet no etiologic agent has been identified¹. It has been suggested that many different etiologic agents alone or together may initiate the common final pathways of tissue pathologic response resulting in the clinical syndrome of eosinophilia, myalgias, and fasciitis. LT and one or more of the impurities cause the characteristic features of the illness. The altered tryptophan metabolism in EMS is secondary to inflammation².

Of the 1345 EMS cases reported, 191 (14%) were not related to LT. Symptoms of peripheral edema, rash, sclerodermalike skin change, alopecia, and neuropathy were more prevalent in non-LT related patients. Mean eosinophil count was significantly higher for epidemic patients than for non-LT related patients. Non-LT related EMS cases were more likely to report the presence of scleroderma-like skin change, but not pulmonary symptoms, and have a better prognosis¹.

A case of non-LT related EMS associated with transient hypoproteinemia and hypoalbuminemia is described.

CASE REPORT

A 61-year-old female housekeeper of Jewish origin presented with tender nonpitting edema over her shins and ankles (Figure 1) and an erythemic induration over the lower abdomen (Figure 2). Her symptoms had developed

From the Department of Medicine, Rabin Medical Center, Petah Tiqva, Israel.

L. Margolin, MD, PhD.

Address reprint requests to Dr. L. Margolin, Rotenshraih 18/27, Tel Aviv, Israel. E-mail: leon3087@yahoo.com

Submitted March 5, 2002; revision accepted August 23, 2002.

rapidly over the preceding 2.5 months accompanied by severe generalized myalgia and poor mobility. She had no gastrointestinal, pulmonary, or other complaints. She denied any unusual exercise or exposure to environmental hazards (including mineral oils). She was not taking any medication or using any homeopathic remedies. Her history was unremarkable. Findings on physical examination were otherwise normal. The laboratory tests were remarkable for eosinophilia (950 cells/mm³), antinuclear antibody titer 1:80, hypoproteinemia 55 g/l, and hypoalbuminemia 25 g/l. Thyroid stimulatory hormone concentration was within normal limits. Extensive investigation ruled out an infectious or neoplastic etiology. Histologic studies of a biopsy from the involved skin showed an intact epidermis and perivascular infiltrates of eosinophlils (Figure 3), lymphocytes, and a few granulocytes in the upper dermis without thickening of collagen layers, with no involvement of the low dermis and hypodermis, and no mucin precipitates.

She was treated with prednisone 40 mg and PUVA (psoralen plus ultraviolet A) therapy that resulted in gradual involution of the edema and the plaques and improvement of her well being over 3 weeks. Her hypoproteinemia and hypoalbuminemia disappeared with treatment.

DISCUSSION

EMS is unrelated to L-tryphtophan in 14% of cases, and can be distinguished from morphea by the presence of severe myalgia and eosinophilia, lack of involvement of the low dermis, and good response to steroids.

This patient presented with generalized myalgia, nonpitting edema (mostly over the lower extremities), sclerodermalike skin changes, and eosinophilia that fulfilled the US Centers for Disease Control surveillance criteria for definition of EMS^{3,4}. Her histologic findings were consistent with the common histologic findings of EMS5. No other disorders could explain the findings in this case.

No exposure to products containing LT or mineral oils was documented. Eosinophilic fasciitis (Shulman syndrome) was ruled out by the results of the skin biopsy, which showed no involvement of the deeper to upper layers of the dermis. The age of the patient and the lack of grooving over the involved skin also were not characteristic for eosinophilic fasciitis.

Personal, non-commercial use only. The Journal of Rheumatology Copyright © 2003. All rights reserved.



Figure 1. Tender nonpitting edema over the patient's shins and ankles.



Figure 2. An erythemic induration over the lower abdomen.

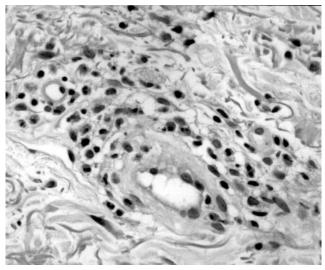


Figure 3. Histologic studies of the involved skin showed an intact epidermis and perivascular infiltrates of eosinophlils (H&E, ×200).

There was no pulmonary or other systemic involvement characteristic for non-LT related cases of EMS². Symptoms of peripheral edema, rash, and scleroderma-like skin change are also more prevalent in tryptophan cases². The transient hypoproteinemia and hypoalbuminemia had not been previously described in the constellation of symptoms of EMS. This case illustrates the possibility that cases of EMS not related to LT may appear long after the epidemic resolved. Clinical awareness of this syndrome unrelated to tryptophan is advocated.

REFERENCES

- Sullivan EA, Staehling N, Philen RM. Eosinophilia-myalgia syndrome among the non-L-tryptophan users and pre-epidemic cases. J Rheumatol 1996;23:1784-7.
- Sternberg EM. Pathogenesis of L-tryptophan eosinophilia myalgia syndrome. Adv Exp Med Biol 1996;398:325-30.
- Centers for Disease Control and Prevention. Eosinophilia myalgia syndrome – New Mexico. MMWR Morb Mortal Wkly Rep 1989;38:765-7.
- Centers for Disease Control and Prevention. Eosinophilia myalgia syndrome and L-tryptophan-containing products – New Mexico, Minnesota, Oregon, and New York. MMWR Morb Mortal Wkly Rep 1989;38:785-8.
- 5. Hertzman PA. Criteria for the definition of the eosinophilia-myalgia syndrome. J Rheumatol 1996;23 Suppl 46:7-12.