Case Report

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

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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
L-TRYPTOPHAN
HYPOPROTEINEMIA
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 identified[1]. 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 inflammation[2].

Of the 1345 EMS cases reported, 191 (14%) were not related to LT. Symptoms of peripheral edema, rash, scleroderma-like 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 prognosis[1].

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 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/mm^3), 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), scleroderma-like 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 EMS[5]. 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.

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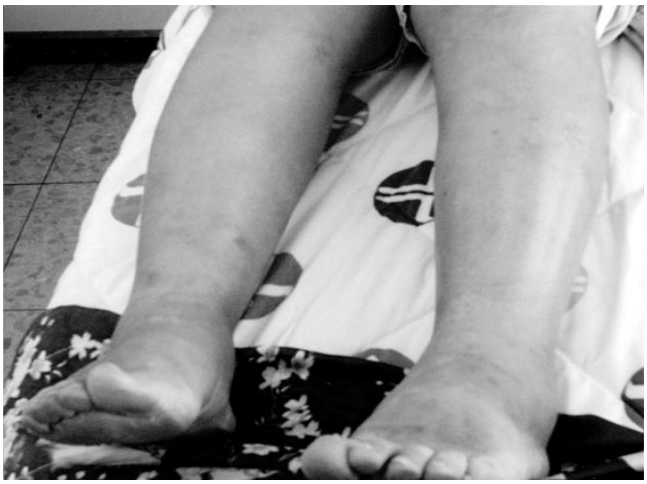

Figure 1. Tender nonpitting edema over the patient's shins and ankles.

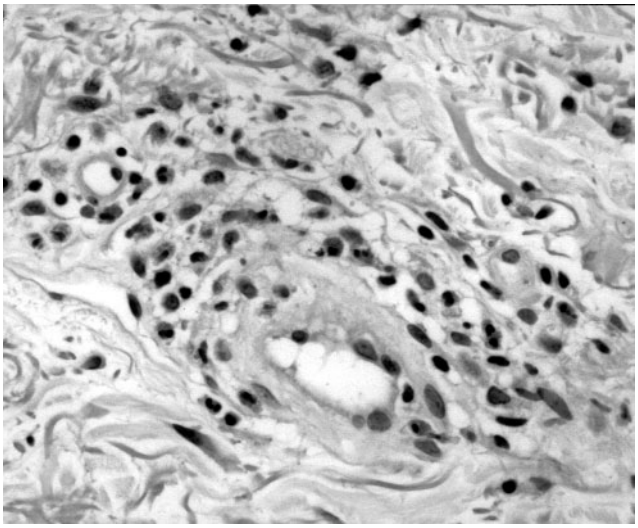

Figure 3. Histologic studies of the involved skin showed an intact epidermis and perivascular infiltrates of eosinophlils (H&E, ×200).

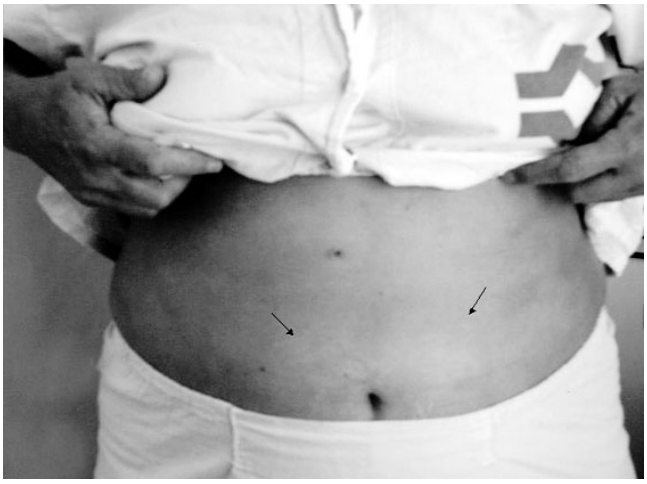

Figure 2. An erythemic induration over the lower abdomen.

There was no pulmonary or other systemic involvement characteristic for non-LT related cases of EMS[2]. Symptoms of peripheral edema, rash, and scleroderma-like skin change are also more prevalent in tryptophan cases[2]. 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

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