Eosinophilic fasciitis in an African—possible benefit of chloroquine treatment

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Summary
A 58-year-old black woman with eosinophilic fasciitis is reported, this being the first report to our knowledge of the condition from Africa. The patient showed a subjective and objective improvement in symptoms on two occasions when treated with chloroquine.

KEY WORDS: eosinophilic fasciitis, Africa, chloroquine.

Introduction
The recently described scleroderma-like syndrome known as eosinophilic fasciitis is characterized by pain, swelling and stiffness of the hands, forearms, feet and legs, in association with a marked blood eosinophilia and, in some cases, hypergammaglobulinaemia (Rodnan et al., 1975; Shulman, 1977). The aetiology and pathogenesis of the condition are not understood, though a tendency for the illness to begin after strenuous exercise has been observed (Rodnan, Jablonska and Medsger, 1979). Treatment is symptomatic and appears to have no influence on the outcome of the disease, which usually results in stiff, though painless, extremities (Shulman, 1977). The patient reported here is, to the author's knowledge, the first to be described in Africa suffering from eosinophilic fasciitis, previous observations on the condition having been made mainly in North America.

Case report
A 58-year-old black Zambian woman was admitted to hospital complaining of painful swelling of the hands, wrists and feet. The symptoms had begun abruptly 6 days before admission and no similar symptoms had occurred in the past. There were no symptoms of Raynaud's phenomenon or the carpal tunnel syndrome. The patient was a subsistence farmer and was accustomed to regular exercise, though the symptoms started after an unusually long walk (55 km) through the bush. Physical examination revealed symmetrical non-pitting swelling and tenderness of the skin and subcutaneous tissues of the hands, wrists and the lower third of the forearms; movement of the wrist joints was limited by pain and stiffness to about 45 degrees of flexion and extension, and the patient was barely able to form a fist. The feet were slightly swollen and painful on pressure over the plantar surfaces. There were no other abnormal physical signs. Investigations revealed: haemoglobin 10·7 g/dl; leucocyte count $8.3 \times 10^9$/l, (29% eosinophils); ESR 88/hr; radiographs of the chest, hands, wrists and ankles, blood films for malaria parasites, urine and stool microscopy, sheep cell agglutination test, venereal diseases reference laboratory test for syphilis all normal or negative; deep wedge biopsy from the dorsum of the left hand showed marked thickening of the deep fascia and infiltration of all layers with eosinophils, lymphocytes and plasma cells.

Treatment was started with bedrest and aspirin, 600 mg 6-hourly. The clinical picture and blood eosinophilia suggested the diagnosis of eosinophilic fasciitis and prednisolone 5 mg 6-hourly was added, with a resulting fall in the blood eosinophilia to 8% of $7.2 \times 10^9$ after 7 days, though no significant improvement in the pain or swelling was evident. An attack of malaria (blood film positive for Plasmodium falciparum) occurred 18 days after admission and was treated with chloroquine 800 mg as a single dose followed by 400 mg daily for 3 days; the other treatment remained unchanged. The patient's joint symptoms began to improve about 3 days after chloroquine was started and, even though it was unlikely that a therapeutic effect of chloroquine on the fasciitis would occur so rapidly, chloroquine was continued at a dose of 200 mg daily in view of the failure of aspirin and prednisolone to relieve the symptoms. Prednisolone was withdrawn by a reducing regimen of doses over 10 days, during which time the patient became almost pain free. The patient was
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discharged on aspirin 600 mg 8-hourly and chloroquine 200 mg daily. She remained well but unilaterally decided to stop all medication about 6 weeks after discharge, and about 3 weeks later was readmitted with a relapse of symptoms. Treatment was started with chloroquine 200 mg daily, and the pain in her hands and feet began to subside about 3 weeks later. After discharge she continued to take chloroquine 200 mg daily with 3 monthly checks of her visual fields and acuity. She remained pain free, though induration and stiffness of the hands and feet progressed gradually. Chloroquine was stopped after a further 9 months and no relapse of pain occurred.

Discussion

The clinical presentation, blood eosinophilia and histological appearance secured the diagnosis of eosinophilic fasciitis in this patient. It appears that the condition can occur in central Africa and the case described was clinically and pathologically similar to those cases described in North America (Barnes et al., 1979).

Whether the apparent benefit of chloroquine in this patient was real or coincidental is open to conjecture as the condition may wax and wane spontaneously, and symptomatic benefit from treatment with low doses of corticosteroids has been observed (Rodnan et al., 1979). However, the improvement in pain which occurred on both occasions when chloroquine was given, the second time without concomitant therapy with aspirin and prednisolone, suggests that the effect might have been real, though there was no apparent influence of chloroquine treatment upon the eventual outcome of the disease, which left the patient with stiff, scleroderma-like hands, forearms and feet. The therapeutic value of chloroquine in rheumatoid arthritis and systemic lupus erythematosus is well known, though in those conditions the response to chloroquine is gradual and is usually not apparent for several weeks. Nevertheless, further appraisal of the role of chloroquine in the symptomatic treatment of eosinophilic fasciitis would probably be worthwhile.

References


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