Granulomatous cheilitis, originally described by Miescher [1], is clinically characterized by intermittent lip swelling (which may become long-standing) and is accepted by many authors as an oligosymptomatic form of Melkersson-Rosenthal syndrome [2, 3].

The cause of this condition is unknown. A genetic predisposition is suspected. Relationships with Crohn’s disease, sarcoidosis, vaso-motor instability, infections and allergies have been suggested [4–8].

Various therapeutic regimens for granulomatous cheilitis have produced disappointing or variable results [4, 9, 10], e.g. systemic or intraleisonal steroids, clofazimine, hydroxychloroquine sulfate or cheiloplasty.

On reviewing the literature, we found only little mention of antibiotic treatments [2, 11, 12]. Fisher [13] reported a good response using a combination of tetracycline (500 mg daily) and prednisone (10 mg every other day) for 2.5 years.

We have used minocycline in 6 patients with granulomatous cheilitis.

Report of Cases

In the last 3 years we have studied 6 patients with granulomatous cheilitis, 4 women and 2 men, median age 33.4 years, median age of onset 29.2 years.

All 6 patients had persistent swelling lasting for 4–48 months.

Histologically the tissue involved showed in all cases nonnecrotizing granulomas, edema, lymphangiectasia and perivascular lymphocytic infiltration.

Special stains showed no evidence of fungal organisms or acid-fast bacilli.

There was no evidence of odontogenic infections.

Clinical and laboratory studies were normal in 5 of the 6 patients.

The sixth case, previously reported [14], was a 29-year-old female presenting a 4-month history of persistent macrocheilitis. She was a chronic carrier of Salmonella and had suffered from recurrent cystitis and vulvovaginitis.

Several investigations (generation of H2O2 and O2 by neutrophils and monocytes, nitroblue tetrazolium test, activation of NADPH oxidase in cell-free systems, measurement of cytochrome b558) performed on the patient’s and her mother’s phygocytes led to the diagnosis of chronic granulomatous disease, type I, X-linked form.

A 4- to 6-month trial with systemic steroids and a 5- to 7-month trial with clofazimine (Lamprene) produced no effect in any of the 6 patients.
Minocycline was subsequently used in doses of 100 mg daily for 4–6 months in all patients. The swelling receded by about 80% only in the case with chronic granulomatous disease. The success of treatment with minocycline only in the patient with granulomatous cheilitis and chronic granulomatous disease could be due to the antibacterial action of the drug against an infectious agent that we failed to detect, despite a thorough search. Alternatively, the positive response could be due to the action of tetracyclines as biological response modifiers [15] in the only patient with abnormal immunologic function.

References


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