DISCUSSION

Chronic relapsing inflammatory diseases of the face may be particularly disabling for the patient. Both GF and LGS belong to this type of skin disease. Their treatment may often be unsatisfying and frustrating. Corticosteroids and dapsone have been used with some benefit in some cases of GF (2). We observed a very good effect of clofazimine in widespread GF. To the best of our knowledge, there is only one additional report on a 36-year-old Nigerian man treated with 300 mg a day for 3 months (4).

Our second patient, a 40-year-old woman, suffered from a longstanding, mainly facial LGS. The cause is one of chronic relapsing and its treatment sometimes frustrating. Clofazimine was of outstanding benefit in this case. The initial dosage of 100 mg/day was well tolerated and without any side-effects.

Clofazimine is a phenothiazine drug with antibacterial and anti-inflammatory effects. Recently anti-proliferative activity has been observed for lymphocytes and carcinoma cells (5, 6). Unwanted side-effects have been reported in high-dosage and long-standing treatment regimens, as for leprosy, including skin hyperpigmentation, toxic erythema, gastrointestinal irritation, eosinophilic enteritis, renal infarction and pedal oedema (7–9).

In our first patient a mild hyperpigmentation of the skin was seen, but no other side-effects were noted. On the other hand, the clinical response was obviously very good and the dosage of potentially hazardous corticosteroids could be minimized.

We suggest that clofazimine seems to be a beneficial and reliable alternative to other therapeutic efforts in certain cases of disabling inflammatory skin diseases.

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Recurrent Aphthous Stomatitis: Treatment with Pentoxifylline

Sir,

We read with great interest the letter by Wahba-Yahav (1). The author reports 3 patients with severe recurrent aphthous stomatitis (RAS); successfully treated with a short course of oral pentoxifylline (PTX), 400 mg t.d., without side-effects and with a prolonged remission. Wahba-Yahav asserts that his study is the first attempt to treat idiopathic RAS with PTX (1). However, we have reported previously 6 cases of RAS successfully treated with PTX (2, 3). Oral therapy with PTX (400 mg twice to three times daily) suppressed recurrence of aphthae in 5 patients and led to a reduction in the number of ulcers, with symptomatic improvement, in one patient (3). Recently we have conducted an open trial on a new series of 22 patients with minor RAS (4). All the patients received oral therapy with PTX at a dose of 400 mg three times daily during a 6-month period. No relapses of aphthous ulcers during the course of treatment were observed in 11 patients (50%); 6 patients (27%) showed recurrence of the lesions with symptomatic improvement; 3 patients (14%) showed recurrence of aphthous ulcers without symptomatic improvement; finally, 2 patients (9%) noted gastrointestinal intolerance and the treatment was discontinued in the first month. Taken together, all these observations suggest that PTX may play a role in the treatment of most patients with idiopathic RAS. In addition, a beneficial effect of PTX on aphthous ulcers in patients with HIV-1 infection has been observed by us (5) and other authors (6). In agreement with Wahba-Yahav (1) we think that additional controlled studies on large numbers of patients are warranted in order to confirm the usefulness of PTX for the treatment of RAS, as well as to determine the optimal dose and duration of therapy.

REFERENCES


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Pretibial Epidermolysis Bullosa with Vulvar Involvement

Sir,

As venereologists are often confronted with patients with longstanding genital complaints, it is of importance to be aware of the possibility of a chronic bullous disease with manifestations in the ano-genital area. Hereditary epidermolysis bullosa is the name of a group of disorders characterized by the formation of blisters following minor trauma (1). A rare variant is pretibial epidermolysis bullosa, a probably dominantly inherited disorder with onset at a young age, usually between 11 and 24 years. Slowly healing pruritic papulo-nodular crural lesions are usually found (2, 3). A woman with pretibial epidermolysis bullosa developed severe macerative bullous lesions on the vulva and perigenital area as part of the clinical manifestations.

CASE REPORT

A 45-year-old otherwise healthy woman was referred to the clinic due to a painful blistering eruption of 3 months' duration, located on the vulva. The history was unremarkable except for a tendency since childhood to develop slowly healing bullous lesions on the pretibial areas after even minor trauma. None in the family had similar skin problems. Clinical examination revealed remnants of bullae, erosions and erythema in the vulval area (Fig. 1). Milia were noticed in the genital lesion. Milia, cicatrices and slight erythematous nodular lesions could be detected on the crura (Fig. 2). A single bulla was seen on the medial aspect of the left lower leg. The nails were dystrophic, but hair and teeth were uninvolved. Pathogenic bacteria, Candida albicans or herpes simplex virus could not be demonstrated in the genital lesions. A histopathological examination of biopsy specimens from the genital and crural lesions showed subepidermal bulla formation with no or only slight lymphohistiocytic inflammation. Direct immunofluorescence findings of perilesional skin were negative for immunoglobulins and complement. Electron microscopic examination showed that epidermis was separated from dermis. A large blister had formed under the epidermis (Fig. 3). The blister roof was epidermis with basal lamina. Remnants of the dermal tissue were attached to the basal lamina. Anchoring fibrils were not found. The blister floor was dermal connective tissue. Symptomatic treatment with mupirocin ointment to the ulcerated lesions was initiated, with good symptomatic effect. Varying vulvar lesions are still seen after 4 years.

DISCUSSION

The history and clinical findings in our patient are in accordance with the diagnosis of pretibial epidermolysis bullosa (1). None of the previously described cases have shown ano-genital bullous lesions. Pretibial epidermolysis bullosa is considered a minor or localized variant of dominant dystrophic epi-