for an explanation by studying the patient's habits. In fact, after several talks he told us that when he went hunting in Sardinia, where the disease has focal endemic areas, he was often bitten by *Phlebotomus* during defecation in the open.

In the diagnosis of leishmaniasis we often observe small nodules such as the "oriental sore". The atypical clinical pattern in our case may be due to the anatomical site of the lesion, a particularly favourable region for infiltration, and/or to pathomorphism as a consequence of the numerous and inappropriate topical treatments previously applied.

The therapeutic procedure normally applied by us in the cure of CL consists of intralesional infiltration with melamine antimoniate and has allowed us to achieve excellent results without significant side-effects (3, 4). However, in this case, because of the site and extension of the lesion, the protozoal density and the disease duration, our preference was given to systemic therapy. Itraconazole, which has been reported by other authors as an active drug (5), was totally ineffective in our case. On the other hand, meglumine antimoniate triggered a severe urticarial rash, necessitating the interruption of treatment. The excellent results obtained with rifampicin without any clinical or laboratory adverse effects confirm the therapeutic value of this molecule (6). We therefore believe that it may represent a valuable alternative in the cure of extensive multifocal CL or in case of intolerance to meglumine antimoniate.

REFERENCES


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Nikola Aste M.D., Giuseppe Fumo M.D., Monica Pau M.D. and Pietro Biggio M.D.
Clinic of Dermatology, University of Cagliari, Via Ospedale, 54, I-09124 Cagliari, Italy.

Histiocytic Necrotizing Lymphadenitis (Kikuchi-Fujimoto's Disease) with Cutaneous Involvement

Sir,

Histiocytic necrotizing lymphadenitis (Kikuchi-Fujimoto's disease) was first described in 1972 as a benign lymphadenopathy with a self-limited clinical course. Later, this disease was frequently reported in Japan but rarely in Europe and in the United States. Its aetiology is unknown, possibly caused by a virus. The disease primarily affects cervical lymph nodes of young adult women, with fever and leucopenia. The histopathological features in the lymph nodes are peculiar: nodular areas with necrotizing foci in the cortex and paracortical areas of lymph node composed of foamy histiocytes and lymphoid cells, neutrophils being absent (1).

Cutaneous involvement is not frequent; when present, it is usually characterised by short and transient skin rashes (2). Some authors described similar skin manifestations as papules (3, 4) or nodules (5) which showed similar histopathological features as the affected lesion in the lymph node.

An association with or a possible evolution of Kikuchi's disease to systemic lupus erythematosus has been previously described (6). This kind of possible evolution in the skin of Kikuchi's disease should be considered in differential diagnosis with cutaneous lymphonephosis, such as large cell lymphomas or Hodgkin's disease (2, 7).

A 21-year-old Italian woman came to our observation for a red-brown erythematous plaque measuring 2×1 cm, developed on the left cheek, 1 year before with slow and progressive enlargement. In the past 6 years she had been admitted three times to the Medicine Department of our hospital for recurrent episodes of lymphadenopathy associated with systemic symptoms such as fever and leucopenia. Each episode lasted for 1 month; the previous two had been characterised by right cervical lymphadenopathy and interpreted as due to viral infection, according to serological positivity of Epstein-Barr virus antibodies. She went to the third admission because of left cervical lymphadenopathy, and a diagnosis of Kikuchi's disease was made, based on histopathological and immunohistochemical features of the lymph node biopsy.

Laboratory examinations, including serum titers for infective diseases, autoantibodies and bone marrow biopsy, showed leucopenia (white cell count of 2,510/mm³) but no other pathological alterations. The skin lesion which addressed the patient to our Department appeared shortly after the last episode of left cervical lymphadenopathy. No lymphadenomegaly and no systemic symptoms were present at the time of our observation, and haematological data were normal.

A total excision of the cutaneous lesion was performed, and the histological examination of the skin biopsy showed heavy lymphoid infiltration of the dermis and subcutaneous fat (Fig. 1). This lymphoid population did not show any epidermotropism and was mainly constituted by small round lymphocytes with scarce cytoplasm; scattered large mononuclear cells, with an irregularly shaped nucleus, could easily be seen. Among the lymphoid nodules, necrotic foci were present, surrounded by histiocytes with no granulocytes.

The immunohistochemical analysis of the lymphoid infiltrate failed to demonstrate any monoclonal populations. The monoclonal cells were positive for UCHL1/CD45 RO and MT1/CD43. In view of the morphological and immunophenotypical features, we consider this case as cutaneous involvement of Kikuchi's disease.
Successful Treatment of Pemphigus Vulgaris with Prednisolone and Tranilast

Sir,

Tranilast, N-(3,4-dimethoxyanilinomethyl) anthranilic acid, has been used clinically as an anti-allergy drug (1). Recently it has been reported from Japan that it can be an effective treatment in combination with other forms of treatment for several bullous diseases such as dermatitis herpetiformis Duhring (2). Here we report the successful effects in a patient with pemphigus vulgaris.

CASE REPORT

A 37-year-old Japanese man was referred to our clinic in April 1990 for investigation and management of erosions affecting the palate, gingiva, face, scalp, chest and back. He gave a 2-month history of gingival erosions with bleeding, and erosions on his palate, face, and scalp, and a 1-month history of bullae on his chest and back. A skin biopsy taken from a bulla on his chest showed hyperkeratosis and an intraepidermal blister with a mononuclear cell infiltrate in the papillary dermis. Some of these cells infiltrated into the epidermis. An examination using direct immunofluorescence showed deposit of IgG and C3 in the intercellular space of the epidermis. Anti-epidermal antibody was detected in the serum (1/240), and deposits of intercellular substance were observed with indirect immunofluorescence. The diagnosis was made on the basis of the multiple erosions in the oral cavity and bullae on the head and trunk and the findings on histopathology and immunofluorescence. At first, the patient was only treated with oral prednisolone (40 mg/day) from April 1990. This was gradually decreased and dapsone (75 mg/day) was started in July 1990. As multiple erosions recurred on his scalp and face in October 1990 despite the combination therapy of prednisolone (15 mg/day) and dapsone (75 mg/day), intramuscular sodium aurothiomalate was started immediately at a dose of 25 mg once a fortnight. However, erosions recurred in July 1991 on his face, and so azathioprine (50 mg/day) was started in September 1991. Although erosions recurred on the gingiva in July 1993, and on the face, upper arm and back in August 1993, most erosions had regressed by November 1993 as the result of increasing the doses of prednisolone, azathioprine and dapsone, but there was still a walnut-sized erosion with crusts on the scalp and there were several residual rice-grain-sized erosions on the face. The medication at this time consisted of oral prednisolone (15 mg/day), dapsone (100 mg/day), azathioprine (100 mg/day) and intramuscular sodium aurothiomalate (25 mg once a fortnight). Tranilast (300 mg/day) was therefore added in February 1994. The erosions on the patient’s scalp and face gradually disappeared; erosions on his face regressed in November 1994, and the erosions and crusts on the scalp in March 1995. Azathioprine was stopped in January 1995, intramuscular sodium aurothiomalate in February 1995, and dapsone in April 1995. The patient has since been treated with prednisolone (7.5 mg/day) and tranilast (300 mg/day) and erosions have not recurred. Minocycline 100 mg once daily was administered from November 1991 to November 1993 but was almost ineffective. Anti-epidermal antibody was detected in the serum (1/120) in September 1990 but was not detectable in August 1995.

DISCUSSION

Although we used prednisolone, azathioprine, dapsone and sodium aurothiomalate in the treatment of our patient, he had three recurrences and at no point did his eruptions disappear. However, after starting tranilast, all of his lesions regressed.

REFERENCES


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Rosina P, D’Onghia FS, Barba A, Colombari R and Chiericato C. Institutes of Dermatology and Venerology and Anatomic Pathology, University of Verona, Piazza Stefani I, 1-37126 Verona, Italy.