

Colchicine as possible treatment of non-cryoglobulinaemic vasculitis in Sjögren's syndrome

Sirs,

We herewith report a case of recurrent cutaneous vasculitis in a patient suffering from Sjögren's syndrome (SS) and rheumatoid arthritis (RA), successfully treated with colchicine.

A 54-year-old woman was admitted at our outpatients clinic for the appearance of purpura at both lower limbs. She was diagnosed with SS in 2006 according to the following criteria: xerophthalmia, xerostomia, positive anti-Ro/SSA antibody, Schirmer's test documenting hypolacrimation, minor salivary glands histology showing a focus score of 4 (1).

Other relevant features were rheumatoid factor (RF) positivity, polyclonal hypergammaglobulinaemia and Raynaud's phenomenon.

Initially, she was treated with hydroxychloroquine, pilocarpine and lacrimal substitutes. Nine years later, erosive arthritis of hands and feet small joints occurred and low titre anti-cyclic citrullinated peptide antibodies were detected, therefore a diagnosis of overlapping RA was made and methotrexate therapy was introduced.

A year later, because of severe articular flares with concomitant intolerance to methotrexate, abatacept (CTLA4-Ig) was started, taking into account literature data indicating this drug as an effective therapeutic option for glandular involvement in SS (2).

A year ago, purpura in the lower limbs (Fig. 1) and general malaise occurred and the patient was admitted to the emergency room, where high-dose corticosteroid therapy was administered.

Laboratory examination revealed positive antinuclear antibody (ANA), positive anti-Ro/SSA antibody, mild increased erythrocyte sedimentation rate (ESR: 37 mm/h) and C-reactive protein (C-RP: 8 mg/l), high RF titre (232 UI/ml), hypocomplementaemia (C4: 2.1 mg/dl with normal serum C3) and hypergammaglobulinaemia (2.2 mg/dl) without a monoclonal component on immunofixation. No cryoglobulins were detected. Serological markers for B and C viral hepatitis were negative.

The patient refused to undergo a skin biopsy. Abatacept therapy was suspended and treatment with oral prednisone (50 mg/day) was started. However, purpura recurred when the tapering of steroid dosage was attempted. Then the patient was treated with two 1000 mg rituximab infusions within two weeks followed by maintenance therapy with azathioprine.

Despite rituximab, purpura recurred and intravenous immunoglobulins (IVIgs) coupled with intravenous methylprednisolone pulse doses were added in treatment.

Fig. 1. Purpura at both lower extremities at physical examination.



Laboratory tests showed persistent polyclonal hypergammaglobulinaemia and a slight increase in inflammatory markers. Therefore, colchicine 1 mg/day therapy was introduced, based on a case report, published in 2004, describing two patients with SS and hypergammaglobulinemic purpura who had promptly responded to colchicine treatment (3).

Six months after the introduction of colchicine and three months after the second cycle of rituximab, the patient showed a clear improvement in purpura, resolution of hypergammaglobulinaemia (1.12 mg/dl) and reduction of inflammatory markers (ESR: 10 mm/h; C-RP: 1.67 mg/l).

Cutaneous vasculitis is a common finding in patients with SS. In a study of 558 patients with pSS cutaneous involvement was found in 16% of patients, and cutaneous vasculitis was the main manifestation, excluding xeroderma (4).

Although the manifestation of purpura in our patient occurred during treatment with abatacept, we cannot define a correlation between the two events. Examining data in literature, there are some reports of skin adverse events during treatment with abatacept, including the appearance of medium-vessel vasculitis, rheumatoid vasculitis, cutaneous polyarteritis nodosa and pyoderma gangrenosum (5-7).

Colchicine is an ancient drug, with a wide use in dermatology (8). Collen demonstrated the efficacy of colchicine treatment in the control of cutaneous leukocytoclastic vasculitis in a group of 13 patients, in whom the resolution occurred within 7 to 10 days after colchicine introduction (9). Although these results have not been further confirmed (10), the prompt resolution of purpura in our patient and the concomi-

tant reduction of inflammatory markers suggest that colchicine could play a role not only in the resolution of symptoms, but also in the immunopathogenetic pathways beneath SS-associated cutaneous vasculitis.

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