

Letters to the Editor

Mesalazine-induced Churg-Strauss syndrome in a patient with Crohn's disease and sclerosing cholangitis

Sirs,

Churg-Strauss syndrome (CSS) is a rare disorder characterized by asthma, eosinophilia and systemic vasculitis (1). Three different phases can usually be recognized in CSS: asthma and atopic allergies such as rhinitis may precede of months, and sometimes of several years, the development of an eosinophilic infiltrative disease with eosinophilic pneumonia or gastro-enteritis followed by the vasculitic phase (1). The etiology of CSS is unknown but putative triggering factors have been identified, including desensitization, vaccination, rapid discontinuation of oral corticosteroid and, more recently, some drugs such as macrolide antibiotics and leukotriene receptor antagonists (2, 3).

We report a case of Churg-Strauss syndrome in a patient with Crohn's disease associated with use of mesalazine, a compound known to induce rarely eosinophilic pneumonia (4).

A 39-year-old man with a 10-year history of atopic rhinitis, nasal polyposis, and asthma presented with weakness, fever, weight loss, arthralgias, vertigo, and paresthesias. He was on salbutamol, mesalazine and ursodeoxycholic acid. Mesalazine had been started 10 months before presentation for Crohn's disease and sclerosing cholangitis, both histologically proven.

At presentation, his eosinophil count was $12,124 \times \text{mm}^3$ (56% of total), erythrocyte sedimentation rate 41 and C-reactive protein 7 mg/L (nv < 5). A urine specimen revealed haematuria (1 plus).

Antineutrophil cytoplasmic antibody (ANCA), antinuclear antibodies (ANA), and rheumatoid factor were negative. A nuclear magnetic resonance of the skull revealed pansinusitis, whereas a computed tomography of the lung showed initial centrilobular emphysema. Pulmonary function tests disclosed a mild obstructive disease. Electromyography of peripheral nerves was indicative of mononeuritis multiplex. A bone-marrow biopsy specimen showed extensive eosinophilic infiltration. Since a stool specimen was positive for *Blastocystis hominis*, the patient was discarded with metronidazole treatment.

One month later, the patient was admitted again because of worsening of vertigo. Laboratory tests confirmed the presence of eosinophilia (46.4%), and increased inflammatory markers.

An ENT doctor examination revealed vestibular decompensation and right perceptible hypacusia. Subsequently, there was the occurrence of spontaneous nystagmus and a left third cranial nerve palsy. A repeat nuclear

magnetic resonance of the skull showed new multiple small ischemic lesions.

Because of the presence of asthma, marked eosinophilia, sinusitis, and neuropathy a diagnosis of Churg-Strauss syndrome was made and treatment with prednisone 1 mg/Kg was started with prompt clinical improvement, normalization of inflammatory indices and of eosinophilia.

Since no reports on the association of Churg-Strauss syndrome with Crohn's disease were found in a literature search, and based on the well-known recognition that drugs can act as triggering factors in Churg-Strauss vasculitis (2, 3), mesalazine was stopped.

Eight months after diagnosis the patient is well under treatment with prednisone 15 mg/day. His eosinophil count is normal as well as his erythrocyte sedimentation rate and C reactive protein.

The cause(s) of Churg-Strauss syndrome is usually not known, but among putative triggering factors vaccinations, desensitisation, antibiotics and, more recently, leukotriene receptor antagonists have been described (2-3, 5). Mesalazine is known to induce eosinophilic pneumonia and can cause activation of eosinophil (4, 6). More rarely, it has been associated with vasculitis-like syndromes and, in a single case, with Churg-Strauss syndrome in a patient with ulcerative colitis (4, 6-10). In most cases blood eosinophilia was present (6-8).

To our knowledge this is the first report of a case of Churg-Strauss syndrome in a patient with Crohn's disease and sclerosing cholangitis. Even though we could not demonstrate a cause-effect relationship between mesalazine and Churg-Strauss syndrome, we believe that our patient's illness was probably caused by this drug because of its capacity to induce eosinophil activation and hypersensitivity reactions.

We suggest that mesalazine should be used with caution in patient with asthma.

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A case report of Takayasu arteritis with history of retroperitoneal fibrosis and coeliac disease: An unusual association

Sirs,

Takayasu arteritis (TA) is an idiopathic, inflammatory granulomatous vasculopathy of the aorta and its main branches. The pulmonary arteries can also be involved. The inflammation is caused by infiltration of lymphocytes and occasional giant cells in three vessel layers which leads to wall thickening and fibrosis with destruction of elastic tissue in the large vessel (1).

A retrospective review of medical records of 36 patients with TA has shown an association with other inflammatory autoimmune diseases such as, rheumatoid arthritis, systemic lupus erythematosus, spondyloarthritis, Crohn's disease, ulcerative colitis, coeliac disease and chronic thyroiditis (2). We described an unusual association of TA in a young woman with a medical history of coeliac disease and idiopathic retroperitoneal fibrosis.

In May 2005 a 34 year-old woman was admitted to the Vascular Surgical Unit of our Hospital with sub-nail necrotic lesions on her right hand. The patient reported episodes of claudicatio intermittens in the last six months, fatigue and discomfort in the muscles of her upper right arm, after minor efforts.

Remote anamnesis showed recurrent episodes of mono-oligoarthritis of knee and ankles since the age of six.

In 2000 as a result of persistent episodes of diarrhoea, progressive weight loss and iron deficiency anemia she was admitted to the Internal Medicine Unit where positivity of Antigliadin antibodies IgA, Antidendomyial antibodies IgA and Anti-Tissue transglutaminase IgA was demonstrated. A jejunal