

*Médecine Interne
Immunologie Clinique et Allergologie
Hôpital Central
54035 Nancy
France
Tel: 0383852863
Fax: 0383852864
E-mail: arschull@yahoo.fr

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Severe venous thromboembolic disease in Churg–Strauss syndrome

G. Garcia*, L. Achouh, D. Cobarzan, D. Fichet, M. Humbert

Key words: Churg–Strauss syndrome; deep vein thrombosis; eosinophilia; pulmonary embolism; vasculitis.

Churg–Strauss syndrome (CSS) is a systemic vasculitis, characterized by severe asthma, blood and tissue eosinophilia and diffuse organ involvement (1). Venous thromboembolic disease (VTED) has exceptionally been reported in CSS.

We report three cases of severe VTED in a cohort of 10 CSS patients.

A 58-year-old man had a diagnosis of CSS based on the association of steroid-dependent asthma, blood eosinophilia (21% of WBC, $3.3 \times 10^9/l$), transient pulmonary infiltrates, sinusitis, profound asthenia, and weight loss. No tissue biopsy was performed. Antineutrophil cytoplasmic antibodies (ANCA) were negative. Cyclophosphamide pulses and increased steroid therapy improved markedly his condition. One month later, the patient presented right calf pain and an episode of syncope. Ultrasonography revealed right femoro-popliteal venous thrombosis and spiral computed tomography (CT) of the chest demonstrated bilateral proximal pulmonary embolism (Fig. 1). There was no blood hypereosinophilia at that time. No coagulation abnormalities were found.

A 37-year-old woman was admitted in intensive care unit for dyspnea, fever, and congestive heart failure confirmed by echocardiography and right-heart catheterization. She had a 1-year history of difficult-to-control asthma and sinusitis with recent worsening of her respiratory symptoms with profound asthenia.

Three patient boxes of severe venous thrombo-embolic diseases are described in a cohort of ten patients with Churg–Strauss syndrome.

Blood counts showed 19% of eosinophils ($1.94 \times 10^9/l$). The ANCA were negative. A clinical diagnosis of CSS was established, but no tissue biopsy was performed because of the patient's critical condition. Spiral CT of the chest showed a right posterobasal pulmonary embolism confirmed by pulmonary angiography. Doppler ultrasonography revealed bilateral deep vein thrombosis.

A 72-year-old man with a history of steroid-dependent asthma, chronic sinusitis, marked blood eosinophilia (28% of WBC, $3.2 \times 10^9/l$), pericardial effusion, abdominal pain, arthralgia and asthenia had a high clinical suspicion of CSS. No tissue biopsy was performed. The ANCA were negative. During clinical follow up, he experienced an episode of idiopathic lower limb thrombosis (left common femoral vein). Eosinophil blood count was normal at that time. No coagulation abnormalities were found.

Coronary involvement leading to myocardial infarcts and heart failure is a known complication of CSS (2). A report of coronary artery stenosis repermeabilization after corticosteroid therapy in CSS highlights that vasculitis presumably plays a significant role in these patients (2). VTED is believed to be a rare occurrence in the course of CSS. There are only a few cases in the literature, including a report of deep vein thrombosis and pulmonary infarction in a 13-year-old boy, and a case of thrombosis of the left subclavian vein (3). In



Figure 1. Spiral computed tomography of the chest showing proximal pulmonary embolism in a patient with Churg–Strauss syndrome.

addition, Guillevin et al. describe a single episode of VTED in their follow up of 96 patients with CSS (4).

Several factors may explain the association between CSS and VTED. First, VTED may be the mere consequence of classical thromboembolic risk factors such as heart failure, immobilization or hospitalization, as evidenced in our first and second cases. Secondly, inflammatory diseases may predispose to vascular thrombosis. For instance, it has been demonstrated that patients displaying inflammatory bowel disease have a threefold increased risk of developing acute VTED (5). More specifically, eosinophilic inflammation may be a risk factor for thrombotic events. Follow up of 15 patients with hypereosinophilic syndrome led to the description of 10 thromboembolic complications including two VTED (6). A possible explanation of this association could be the ability of eosinophil cationic protein to bind, *in vitro*, to Hageman factor (XII) and activate the intrinsic pathway of coagulation.

In conclusion, we describe three episodes of severe VTED in a cohort of 10 CSS patients. It remains unclear whether VTED was the consequence of specific vascular lesions or the mere result of a combination of classical VTED risk factors.

*Department of Pulmonary Diseases and Intensive Care

Antoine Bécèle Hospital
157 Rue de la Porte de Trivaux
92141, Clamart
France
Tel: 01 45 37 48 10
Fax: 01 45 37 48 28
E-mail: gilles.garcia@abc.ap-hop-paris.fr

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Atopic myelitis: first case report outside Japan?

A. Zoli, M. Mariano, A. Fusari, F. Bonifazi, L. Antonicelli*

Key words: atopic dermatitis; atopic myelitis; atopy; myelitis.

Atopic myelitis (AM) is a syndrome which was thought to be a peculiarly Japanese phenomenon.

A recent Japanese nationwide survey identified 79 patients with AM (1).

This condition is characterized by the association between allergic and central nervous system (CNS) diseases, although the pathogenetic mechanism is unknown.

The disease affects a young to middle-aged adult population with atopic diathesis (atopic dermatitis plus rhinitis and asthma).

The main features of AM are: paresthesia/dysesthesia are the initial and predominant symptoms, the stepwise progression and fluctuation of the symptoms are frequent.

Hyperreflexia is frequent, but severe motor weakness is rare.

Cerebrospinal fluid (CSF) abnormalities, if any, are infrequent and mild.

Very high total immunoglobulin E (IgE) serum level and mite antigen-specific IgE are present in most cases.

Study of the motor evoked potentials revealed abnormalities in the upper and lower limbs.

High signal intensity lesions at the cervical or at the thoracic spinal cord on the T2-weighted magnetic resonance imaging (MRI) were documented (1–5).

The biopsy specimens of the CNS lesions showed perivascular mononuclear cell cuffs accompanied by eosinophils around the small veins and capillaries as well as in the parenchyma; myelin and axons were equally damaged. Immunohistochemical analysis revealed memory T cells infiltrating the lesions with only CD8+ T subset in the parenchyma and dominating over CD4+ cells in the perivascular area (6, 7).

Peripheral blood mononuclear cell flow cytometry revealing an increase in the CD3+CD8+ and in CD4+CD45RO+CD30+ T-cell subsets was reported in a study performed on 13 patients with AM (8). The flow cytometry used to analyse the intracellular interferon (IFN)γ/interleukin (IL)-4 ratio in peripheral blood CD4+ T cells showed a significantly lower value than in the controls (6).

A 34-year-old man (N.B.), with allergic rhinitis and mild bronchial asthma since childhood, developed atopic dermatitis in adulthood (22 years). At the age of 25 years, slight, intermittent neurological symptoms appeared: muscle weakness, migrant paresthesia/dysesthesia in the limbs, anisocoria and right eyelid ptosis. A brain and spinal cord MRI scan revealed an area of abnormal intensity located at the bulbar medulla junction to C1 segment (i.e. low intensity in T1 images and high in T2) consistent with myelitis-like lesions (Fig. 1 – circle). Examination of the motor provoked potentials showed abnormalities in the lower limbs. Neurological examination excluded other CNS diseases and in particular multiple sclerosis. Skin prick tests with commercial extracts (Stallergenes France) were positive for several inhalant allergens (house dust mite, cat, grass and cypress pollen) with the mite sensitization playing the

Neurological symptoms in atopic disease may be related to an immunological impairment of CNS.