中文題目：發生在氣喘病人的雙下肢麻與無力

英文題目：Progressive lower legs numbness and weakness in an asthma patient

作　　者：鐘堉緁、楊志仁、黃明賢、黃吉志

服務單位：高雄醫學大學附設中和紀念醫院 內科部胸腔內科

摘　　要：

Case Presentation

A 69-year-old woman with a history of asthma had taken inhalating salmeterol/fluticasone for years. She started to take montelukast 10mg daily due to poor control of her asthma. About 2 weeks after starting use of montelukast, she suffered from progressive numbness and weakness in bilateral legs. She was soon admitted for further evaluation. The initial laboratory analyses showed leukocytosis ($29100*10^6$/L) eosinophilia (absolute eosinophil count of $21534 *10^6$/L (74%)), high total IgE level (2768 IU/mL), and high rheumatoid factor, while the serum were negative for antineutrophil cytoplasmic antibody (ANCA) or antinuclear antibody (ANA). Chest radiograph revealed right lower lung pneumonia. Computed tomography of the chest revealed pneumonia in the both lungs, particularly in the right lung with bilateral pleural effusion. Nerve conduction velocity (NCV) showed axonal degenerative polyneuropathy. Water's view revealed bilateral maxillary sinusitis. Based on the findings, including asthma, eosinophilia, polyneuropathy, sinusitis, and non-fixed radiographic pulmonary infiltrates, Churg-Strauss syndrome (CSS) was diagnosed.

Tracing back her previous laboratory data, the level of eosinophil count increased notably after the start of using montelukast 10mg daily. Therefore, montelukast was discontinued and the weakness and numbness in her legs dramatically improved within days. Due to recurrent pneumonitis with eosinophilia and high rheumatoid factor, pulse steroid therapy with intravenous methylprednisolone 1g daily for 3 days, followed by methylprednisolone 40 mg 3 times a day, were given as suggestion from the rheumatologist. Her symptoms improved rapidly and the chest radiograph showed resolution of the pneumonia. After starting on a rehabilitation program, she was discharged home with stable clinical condition.

Discussion

Churg-Strauss syndrome (CSS) is a rare syndrome that affects small- to medium-sized arteries and veins. The American College of Rheumatology (ACR) has proposed 6 criteria for the diagnosis. The presence of 4 or more criteria yields a sensitivity of 85% and a specificity of 99.7%. These criteria include (1) asthma (wheezing, expiratory rhonchi), (2) eosinophilia of more than 10% in peripheral blood, (3) paranasal sinusitis, (4) pulmonary infiltrates (may be transient), (5) histological proof of vasculitis with extravascular eosinophils, and (6) mononeuritis multiplex or polyneuropathy.

CSS related to leukotriene receptor antagonist has been previously reported. Different mechanisms may be involved in antileukotriene-induced CSS. One possibility is that leukotriene receptor antagonist may unmask underlying CSS by facilitating steroid-tapering.

Some cases of antileukotriene-associated CSS, however, have occurred without concomitant steroid treatment, as in our case. Therefore, other mechanisms may be involved. Our patient received inhaled bronchodilator and corticosteroid and had never received systemic corticosteroid. She developed CSS soon after starting montelukast. How the antileukotriene involved in the pathogenesis of CSS needs further study.

The clinical and electrophysiologic signs of neuropathy in CSS usually include asymmetric abnormal motor and sensory responses that are suggestive of mononeuritis multiplex. Lower limbs are more likely to be affected and the sciatic nerve is most commonly affected followed in frequency by the tibial and peroneal nerves. Through literature review, neuropathy as the initial manifestation of montelukast-induced CSS remains rare. This case report demonstrated that leukotriene receptor antagonist may trigger CSS, which may be treated successfully with discontinuation of the leukotriene receptor antagonist and with the use of intravenous steroid. In conclusion, CSS should be considered in patients with asthma who develops marked eosinophilia following the introduction of leukotriene receptor antagonist.