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Case Report

Churg Strauss Syndrome after Leech Therapy: A Case Report

Sülük Tedavisi Sonrası Gelişen Churg Strauss Sendromu: Olgu Sunumu

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Abstract

Churg Strauss Syndrome (CSS) is a necrotizing vasculitis affecting small vessels. Although there are many accompanying findings, this syndrome has distinctive clinical findings such as asthma, nasal polyposis, and hypereosinophilia. Here, we describe a case report of a 48-year-old male patient with known asthma and nasal polyposis, who applied to the emergency department with a sudden worsening of consciousness and bullous lesions on the ankle after undergoing leech therapy (hirudotherapy), accompanied by mononeuritis multiplex and other clinical findings. Clinical findings, laboratory, electromyography, lumbar puncture, radiological imaging methods, histopathological examination and transthoracic echocardiography were used to diagnose CSS. In addition, it was desired to draw attention to the fact that leech therapy may trigger hyperosinophilia in vasculitis group diseases and cause serious complications in the individual.

Keywords: Leech therapy, hypereosinophilia, Churg Strauss Syndrome

Öz

Churg Strauss Sendromu (CSS); küçük çaplı damarları tutan nekrotizan bir vaskülittir. Eşlik edebilen pek çok bulgusu olmakla birlikte bu sendrom, astım, nasal polipozis, hipereozinofili gibi belirgin klinik bulgulara sahiptir. Burada bilinen astım ve nasal polipozisi olan 48 yaşında bir erkek hastanın sülük terapisine gittikten sonra ani gelişen bilinçte kötüleşme ve ayak bileğinde yine ani gelişen büllöz lezyonlar ile acil servise başvurduğu ve beraberinde mononöritis multisipleks ve başka klinik bulguların da eşlik ettiği bir CSS olgu sunumu anlatılmaktadır. CSS tanısının konulmasında klinik bulgular, laboratuar, elektromiyografi, lomber ponksiyon, radyolojik görüntüleme yöntemleri, histopatolojik inceleme ve transtorasik ekokardiyografiden yararlanıldı. Ayrıca sülük terapisinin vaskülit grubu hastalıklarda hiperozinofiliyi tetikleyip kişide ciddi komplikasyonlara yol açabileceğine dikkat çekilmek istendi.

Anahtar Kelimeler : Sülük tedavisi, hipereozinofili, Churg Strauss Sendromu

INTRODUCTION

Churg Strauss Syndrome (CSS) is a necrotizing vasculitis affecting small vessels. The prominent clinical features are asthma, eosinophilia, allergic rhinitis, pulmonary infiltrates and extravascular granulomas. It is in the ANCA-associated vasculitis group. Asthma, eosinophilia, paranasal sinusitis, pulmonary infiltration, histological detection of vasculitis and mononeuritis multiplex are the 6 American College of Rheumatology (ACR) criteria used in the diagnosis of CSS. In addition, intestinal bleeding due to gastrointestinal involvement, intestinal perforation, cholecystitis, pancreatitis or unexplained abdominal pain, less commonly glomerulonephritis and interstitial nephritis due to renal involvement, pericarditis due to cardiac involvement, cardiomyopathy and myocardial infarction are also seen in this syndrome (1).

Leech therapy, also known as Hirudotherapy, is a traditional and complementary treatment method applied with leeches. Its therapeutic feature is realized by the enzymes that the leech, which is placed on the problematic area, secretes into the body during blood sucking. This secretion has anticoagulant, blood pressure stabilizer, antidepressant, antibacterial and antioxidant effects.

Here, we present a case report of a patient with a history of asthma and nasal polyposis, who developed bullous

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lesions on his skin after leech therapy and was diagnosed with CSS. The diagnosis of CSS was made by laboratory and clinical findings accompanied by eosinophilia and mononeuritis multiplex.

CASE REPORT

A 48-year-old patient with known asthma and nasal polyposis was admitted to the emergency department with complaints of worsening consciousness and drowsiness. The patient stated that the bullous lesions noticed on his right ankle appeared after the leech treatment he had done 2 days ago. He said that he had numbness and widespread joint pain in the extremities. There was also loss of muscle strength in the extremities, and the muscle strength was evaluated as 3/5 on the right side and 4/5 on the left side, more prominently in the right arm and leg. The patient applied to the emergency department 1 month ago with the complaints of numbness in the extremities and tongue and weakness on the right side. Her complaints were thought to be due to a transischemic attack, but no pathology was observed in cranial CT, and her vertebral and carotid artery Doppler results were also normal. Bile sludge and acalculous cholecystitis were also observed in abdominal USG. In his family history, the patient's mother had angioneurotic edema and patient's siblings had asthma and nasal polyposis. In laboratory tests, leukocytosis (57.700/uL), eosinophilia (75.8-43.770/uL), urea 25mg/dL, creatinine 1.21mg/dL, CRP 171.96mg/L, sodium 120mmol/L, potassium 4.9mmol/L, chloride 83 mmol/L, troponin 220, creatine kinase 3001U/L, brain natriuretic peptide (BNP) 1959ng/L, antinuclear antibody (ANA) and proteinase 3 antineutrophilic cytoplasmic antibody (c-ANCA) negative but myeloperoxidase antineutrophilic cytoplasmic antibody (MPO-ANCA) was positive (76.63RU/ml). Sinus tachycardia was observed in his EKG. Echocardiography of the patient showed 65% EF, normal left ventricular systolic function, and mild mitral valve regurgitation. Since the troponin elevation continued in the follow-ups, the patient was consulted with the cardiology, but myocardial infarction was not considered in the patient. Significant eosinophilia was observed in the peripheral smear (figure 1). With peripheral smear, the patient was consulted to hematology, but hypereosinophilic syndromes or other hematological malignancies were not considered.

Contrast-enhanced lower abdominal MRI revealed 1-2 reactive lymph nodes in both inguinal areas and 1-2 lymph nodes in the paraaortic region in contrast-enhanced upper abdomen MRI. In the thorax CT, bronchovascular prominences in both lungs, interlobular septal thickening, sequela fibrosis in both upper lobes, pleural thickening and irregularities in the right upper lobe were observed. The patient was consulted to dermatology because of bullous lesions on the ankle. The lesions were not evaluated as lesions of a specific pathology.

He was consulted with neurology because he had mononeuritis multiplex-like symptoms describing numbness in his extremities and an arachnoid cyst in his cranial MRI taken in another hospital. EMG was taken. In his EMG, 'polyneuropathy which includes the signs of demyelination, axonal involvement and damaged subacute motor fibers' was observed. Considering Guillain-Barre, the patient underwent lumbar puncture (LP). Cerebrospinal fluid (CSF) examination was normal in LP, there was no growth in the empty fluid culture, viral and bacterial meningitis panel was normal, serum and CSF IgG index were normal.

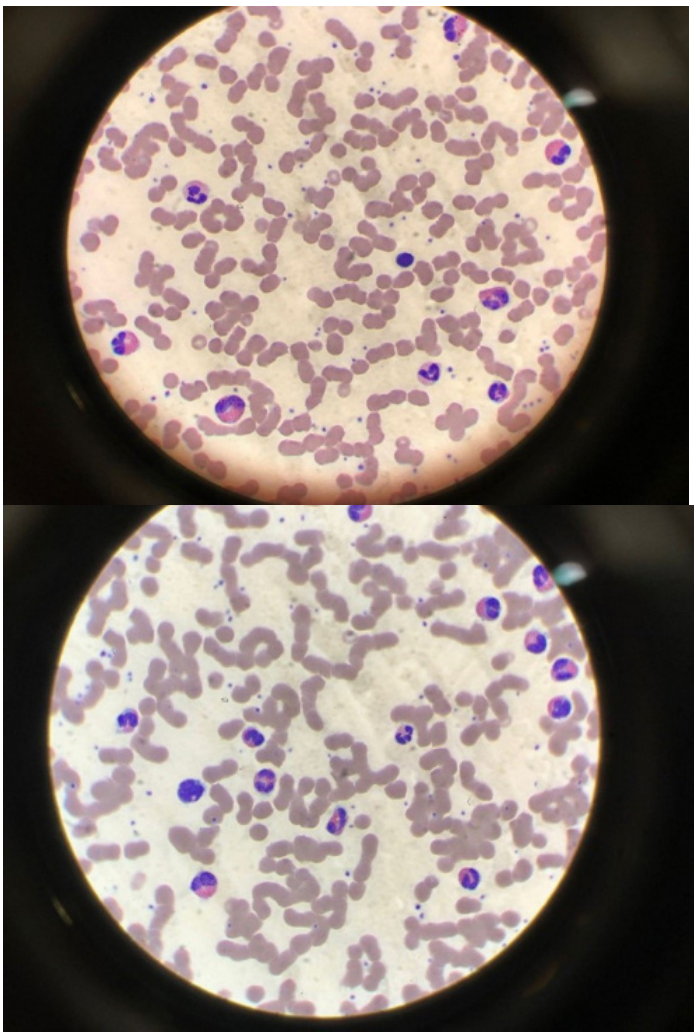

Figure 1. Eosinophilia

The patient's EMG result was re-evaluated by the neurologist and it was decided that he had vasculitic type neuropathy. Fundoscopic examination of the patient was performed, no vasculitic finding was detected. The patient's paranasal sinus CT was taken, multiple polyps were observed (figure 2). Biopsy was taken from the polyps and the pathology result was evaluated as normal. Renal USG showed grade 2 renal parenchymal damage in the right kidney.

There was 1.28g proteinuria in spot urine. In his 24-hour urine, 3.73g proteinuria was detected. The patient was consulted with rheumatology and nephrology because of proteinuria, eosinophilia and pANCA positivity. Renal biopsy was recommended for the histopathological diagnosis of nephropathies and rheumatological diseases.

However, the patient refused.

His hyponatremia resolved with fluid restriction. With the recommendation of neurology and rheumatology, the patient was given pulse steroid (1000mg-5 days) and Ivlg (0.4g/kg/day) for 3 days). Improvement in muscle strength and dramatic decrease in eosinophilia were observed. Afterwards, Prednol (40mg in the morning - 20 mg in the evening) was continued. The patient was discharged after his eosinophil level decreased to 0.02 10^3/uL. He did not have any active complaints and improvement was observed. He started to be followed from rheumatology.

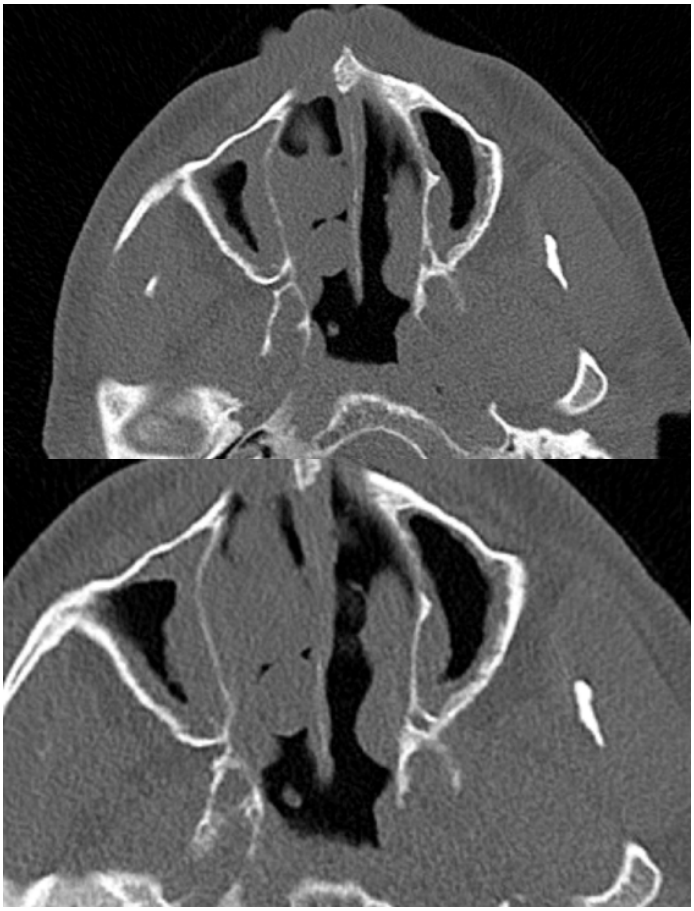

Figure 2. Paranasal sinus CT

DISCUSSION

CSS is a necrotizing vasculitis affecting small vessels. The prominent clinical features are asthma and eosinophilia, and there are many accompanying additional findings. There are 6 criteria determined by the American Society of Rheumatology to diagnose CSS. These are asthma, eosinophilia greater than 10% or 1500/mm³, paranasal sinusitis, pulmonary infiltration, histological vasculitis, and mononeuritis multiplex (2). The presence of 4 of these 6 criteria is sufficient to make a diagnosis. These diagnostic criteria were present in our patient, except for the tissue biopsy sample. The nasal polyp biopsy result was normal in the patient, but the presence of vasculitis could not be detected histologically since he did not accept renal biopsy. In addition to these criteria, the patient also had positivity for pANCA. The elevation of troponin suggested cardiomyopathy and myocardial infarction, which can be seen rarely in Churg Strauss disease, but myocardial infarction was ruled out in the patient. Although the patient had cholecystitis and had no known diagnosis of HT, high blood pressure values during his hospitalization, presence of erythrocyte (30 cells) in the complete urine examination at his admission and 3.73 g proteinuria in his 24-hour urine were also suggestive of glomerulonephritis. This is one of the additional diseases that can be seen in Churgs Strauss disease less commonly.

Corticosteroids are the first choice in the treatment of the disease (3), and in this patient, clinical findings and eosinophilia were improved with the use of steroids (4) (figure 3).

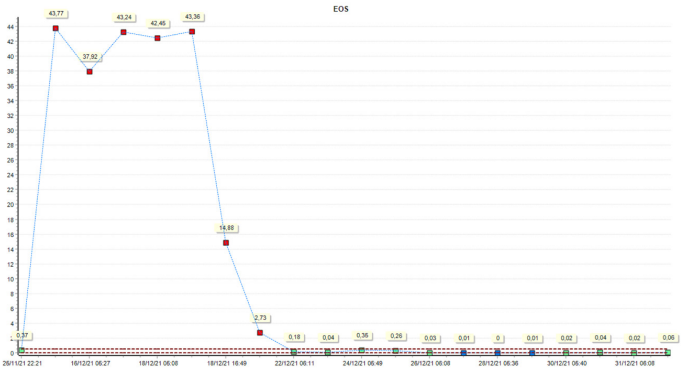

Figure 3. Eosinophil count

CONCLUSION

In relation to this disease, which belongs to the group of vasculitic diseases, it was found remarkable that eosinophilia showing a severe increase and worsening consciousness after leech therapy. Based on this case, it was also desired to draw attention to the fact that such side effects should not be ignored before leech therapy is recommended in vasculitic group diseases.

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Conflict of Interest: *The authors declare that they have no competing interest.*

Informed Consent: *Informed consent was taken from the patient.*

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