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Case Report

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Toxocara canis-Associated Myelitis with Eosinophilic Pneumonia

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The existence of *Toxocara canis*-specific antibodies has recently been reported in patients with atopic myelitis. Here, we report the case of a 35-year-old male patient admitted with a chief complaint of right lower limb hypoesthesia lasting for a month. The patient was diagnosed with eosinophilic pneumonia 3 months ago, and a spine MRI revealed the presence of myelitis in the cervicothoracic cord. After confirming the presence of hyper-IgE-emia and *Toxocara canis* antibodies, the patient was treated with steroids and albendazole treatment, which improved his symptoms. To our knowledge, this is the first case of *Toxocara canis*-associated myelitis with eosinophilic pneumonia.

Key words: Myelitis, *Toxocara canis*, Eosionophilia, Pulmonary eosinophilia

INTRODUCTION

Atopic myelitis refers to myelitis characterized by hyper-immunoglobulin E (IgE)-emia and the presence of mite antigen-specific IgE [1]. Its clinical features include a young age of onset, a subacute or chronic nature, and sensory symptoms rather than weakness as the chief complaint [2]. Currently, *Toxocara canis* is thought to be the main cause of atopic myelitis [3, 4] and is known to lead to myelitis without systemic manifestations in most cases. Herein, we report a case of toxocariasis-associated atopic myelitis accompanied by eosinophilic pneumonia.

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CASE REPORT

A 35-year-old male patient was admitted with a chief complaint of hypoesthesia in the right leg lasting for 1 month. At first, the progress of the hypoesthesia was only monitored, as the symptoms were mild and there was no discomfort reported. One week later, the hypoesthesia progressed to a degree that the patient was unable to feel hot water. The patient did not complain of weakness, pain, paresthesia, dysuria, or erectile dysfunction. He had experienced pain in his left chest 3 months prior to his visit, and he had been admitted to the department of pulmonary medicine. At this point, the patient's eosinophil count was elevated to 13.3% (normal range is 1~5%) and a chest computed tomography (CT) scan revealed consolidation with ground-glass opacification in both lungs, which led to a diagnosis of eosinophilic pneumonia (Fig. 1). Chest pain was naturally improved after 2 weeks, and no other respiratory symptoms, such as cough, sputum, or dyspnea, were observed. Furthermore, the eosinophil count decreased to

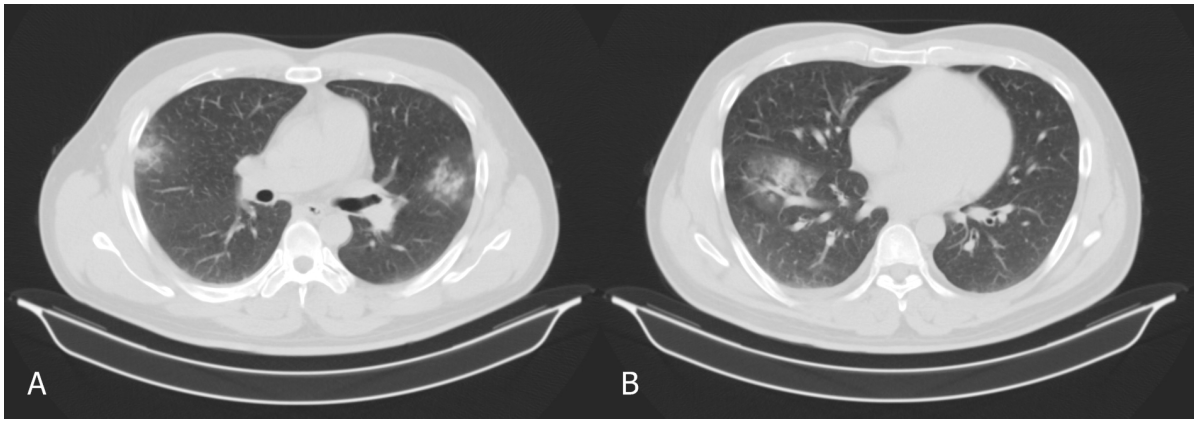

Fig. 1. Chest CT of the patient. Consolidation with ground-glass opacification was observed in both lungs.

7.7% and a chest radiograph revealed slight improvements in the patient's lungs. Thus, the patient's progress was monitored without any further treatment. No other disease history, such as asthma, atopic dermatitis, sinusitis, or arthritis was reported, and no symptoms such as photosensitivity, oral ulcers, or genital ulcers were observed. Moreover, the patient did not consume any raw meat other than raw fish, and reported no exposure to animals or pets. A neurological examination revealed that pain, touch, and vibration sensations were all decreased to approximately 25% of normal levels on the right side below the T8 level. Motor power was intact as revealed by a medical research council (MRC) grade of V, and the deep tendon reflex was found to be normal. The Babinski reflex and ankle clonus examinations revealed no pathologic reflexes.

Spinal magnetic resonance imaging (MRI) revealed a high T2 signal intensity lesion with focal enhancement at the C5-T3 level (Fig. 2). Brain MRI was normal. Furthermore, although a somatosensory evoked potential central conduction defect was observed at the thoracic level, motor evoked potential, brainstem auditory evoked potential, and visual evoked potential test results were all normal. The blood eosinophil count was 7.5%, which was elevated to a similar level as the patient's previous visit. The patient was negative for the presence of antinuclear, anti-Ro/La, anti-neutrophil cytoplasm (P-ANCA, C-ANCA), antiphospholipid, and aquaporin-4 antibodies. The level of angiotensin converting enzyme was 21 IU/L (normal range 20~70 IU/L), the level of vitamin B12 was 893.4 pg/ml (normal range 243~894 pg/ml), and the level of folate was 6.33 ng/ml (normal range 3.1~17.5 pg/ml). Test results for human immunodeficiency virus and human T-lymphotrophic virus were both negative. No white blood cells were detected in the cerebrospinal fluid (CSF), and the CSF protein level was 23.8 mg/dL (normal range 15~45 mg/dL). No oligoclonal bands were observed and the IgG index was 0.67. Serum IgE levels were elevated to 1,216 IU/ml (normal range 1.0~183.0 IU/ml), with antigen-specific IgE against *Dermatophagoides farina* and *Dermatophagoides pteronyssinus* highly elevated to above 100 IU/ml (highly positive). Serum *Toxocara canis* antibody levels by commercial enzyme-linked immunosorbent assay (Bordier Affinity Products SA, Crissier, Switzerland) were increased to 2.62 (normal range <0.847). We thus diagnosed the patient with *Toxocara canis*-associated myelitis and treated him with steroid pulse therapy and albendazole (400 mg b.i.d.) for 2 weeks. The patient was not treated with oral steroids. There were no specific side effects observed during the treatment period. Two months later, sensation in the right lower limb was increased to 50% of normal, which was a partial improvement from the time of admission. Blood eosinophil levels were also normalized to 3.5%. Currently, 6 months after treatment initiation, the patient's symptoms are maintained at a similar level.

DISCUSSION

Atopic myelitis was first described in 4 patients with acute myelitis accompanied by hyper-IgE-emia and atopic dermatitis [5]. Later in 1998, Kira et al. suggested the use of the term atopic myelitis to describe patients diagnosed with the condition, which commonly occurs in the cervical cord and often accompanies atopic dermatitis with normal CSF findings [1]. Yoon et al. compared 14 atopic myelitis patients with 15 non-atopic myelitis patients in 2009 and reported an earlier age of onset and a non-acute and slowly progressive disease course in atopic myelitis patients [2]. They also observed that motor symptoms were relatively mild, while sensory symptoms were predominant in atopic myelitis patients [2]. MRI findings revealed a high percentage of swelling, the predominance of large eccentric vs. central lesions in axial images, and focal lesions with gadolinium

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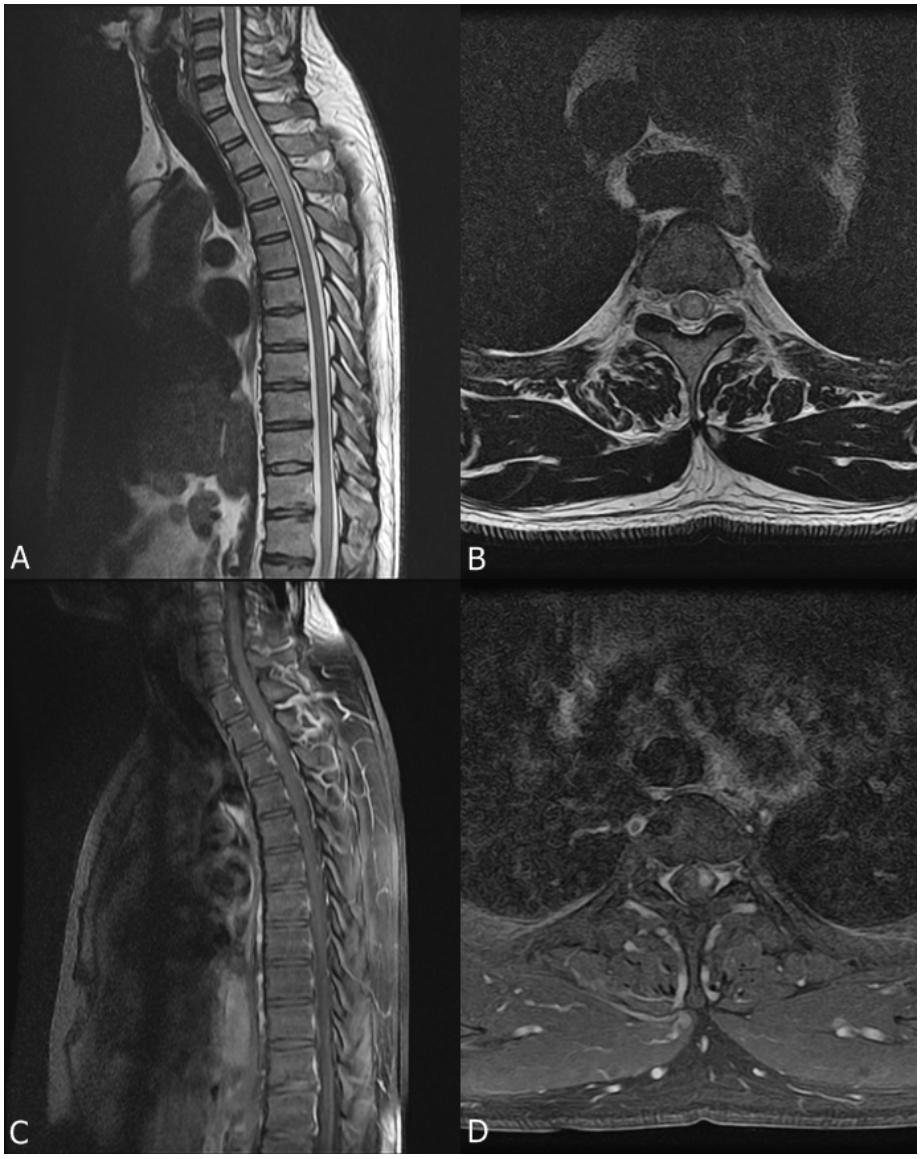

Fig. 2. Spine MRI of the patient. (A) Sagittal T2-weighted image (T2WI) showing a high signal intensity lesion in the cervicothoracic cord (C5-T3). (B) An axial T2WI showing diffuse swelling of the entire cord. (C) and (D) Sagittal T1-weighted enhancement image showing focal lateralized enhancement.

enhancement [2]. The specific IgE to *Dermatophagoides farina* and *Dermatophagoides pteronyssinus* was observed more often [1]. Isobe et al. provided the diagnostic criteria for atopic myelitis in 2012 [6]. The patient in this case study met all of the absolute criteria and one of the positive findings, as well as negative findings supporting the diagnosis of probable atopic myelitis. Moreover, although diffuse swelling and focal enhancement lesions were observed in the cervical cord, the patient complained only of sensory symptoms without weakness. Our other observations, such as a subacute onset and normal CSF findings, were consistent to previous reports of atopic myelitis.

Toxocara canis is a parasite commonly found in dogs. It can infect humans when soil contaminated with embryonated eggs, or raw liver or meat containing its larvae, is ingested [7]. Ingested larvae penetrate the host tissues using the systemic circulation and trigger eosinophilia and hyper-IgE-emia by inducing the immune-mediated hypersensitivity reaction [3]. Central nervous system involvement is rare. Jabbour et al. reported the detection of *Toxocara canis* antibodies in the blood and CSF of 17 patients [8]. They reported a subacute or chronic course of disease and the observation of nodular MRI enhancements, similar to atopic myelitis patients. Albendazole treatment, with or without steroids, improved the patients' symptoms and the MRI findings. *Toxocara*-specific IgG was observed in 31 patients of 33 Korean atopic myelitis patients (93.9%), suggesting that *Toxocara* may be the main cause of atopic myelitis [3]. In addition, there are case reports of *Toxocara canis* myelitis patients improving after the use of albendazole and steroids [4, 9]. Although there is a high

correlation between the presence of *Toxocara canis* and atopic myelitis, whether *Toxocara canis* is the definite cause of atopic myelitis is still not well understood [10]. Therefore, we could diagnosed the patient as *Toxocara canis*-associated myelitis and possitbility of atopic myelitis is still unclear. Interestingly, no systemic manifestations other than myelitis were observed in previous reports, and to our knowledge, the present case is the first one in which eosinophilic pneumonia was present. Eosinophilic pneumonia is an infiltrative lung disease with prominent eosinophilic infiltration and induced by eosinophil [10]. Systemic diseases, drug, toxic material, and infection such as Toxocariasis can cause eosinophilic pneumonia [11]. Toxocariasis is common in children, where respiratory symptoms such as diffuse interstitial pneumonia or severe asthma may occur, but the infection is rare in adults and the symptoms are mild or subclinical in many cases [12]. Although there is a possibility of false positive because of relative specificity (86%) and cross-reaction with *Trichinella spiralis* and trematode infections with the ELISA kit [13], clinical manifestation was consistent with previously reported *Toxocara canis*-associated myelitis cases.

Here, we present a case of myelitis accompanied by hyper-IgE-emia in a patient with a history of eosinophilic pneumonia. We confirmed the presence of *Toxocara canis* antibodies in the patient and treated him with steroids and albendazole, which improved his symptoms. Thus, if the above clinical features are observed in myelitis patients with an unknown cause, the patient must be diagnosed with atopic myelitis. Furthermore, myelitis caused by Toxocariasis should be suspected in such patients, especially those with a history of eosinophilic pneumonia.

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