Case report

Toxocara canis myelitis involving the lumbosacral region: a case report

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Context: *Toxocara canis* is a parasite known to cause visceral larva migrans. The infection rarely affects the central nervous system but there have been several reports of myelitis caused by visceral larva migrans due to *Toxocara canis*. In previous reported cases, the lesions were located in the thoracic or cervical spinal cord. To the best of our knowledge, this is the first report of a lesion involving the lumbosacral region.

Findings: A 60-year-old man developed weakness and dysesthesia in the lower limbs. The symptoms resolved spontaneously, but recurred after five months. One month later, the patient developed pollakiuria and constipation. He was a dog owner and frequently ate raw chicken meat and beef liver. Sagittal T2-weighted image (T2WI) showed swelling and hyperintensity in the spinal cord from T10 to the lumbosacral region and focal nodular enhancement on the posterior segment of the lumbar spinal cord. Blood cell counts showed slight eosinophilia and elevated serum immunoglobulin E level. Cerebrospinal fluid examination showed slight pleocytosis with eosinophilia. Enzyme-linked immunosorbent assay showed high levels of anti-*Toxocara* antibodies in the serum and cerebrospinal fluid. In addition, confirmatory test by Western blot was positive. The patient was initially treated with intravenous methylprednisolone with slight improvement in muscle weakness. Albendazole was added with a second course of intravenous methylprednisolone. The muscle weakness in the lower limbs improved considerably, and swelling and hyperintensity on T2WI almost disappeared.

Conclusion: Our results suggest that *Toxocara canis* myelitis cannot be discounted even if the myelitis involves the lumbosacral region.

Keywords: *Toxocara canis*, Myelitis, Lumbosacral region, Visceral larva migrans, Albendazole

Introduction

Toxocara canis (*T. canis*) is a common parasite found in dogs that may infect humans through the consumption of contaminated rabbit, pork, beef, or chicken meat.^{1,2} Ingesting fruits and vegetables contaminated with dog feces containing *T. canis* eggs can also lead to infection in humans.² The parasite causes visceral larva migrans (VLM) in various organs such as the liver, lung, heart, skin, eye, and rarely in the central nervous system.¹ There have been several reports of myelitis caused by VLM due to *T. canis*,^{2–10} with lesions located in the thoracic or cervical spinal cord. To the best of our knowledge, there is no previous report of VLM due to

T. canis affecting the lumbosacral region. Here we describe a case of *T. canis* myelitis involving the lumbosacral spinal cord.

Case report

Seven months before admission, a 60-year-old man, who lived in Miyazaki prefecture in southern Kyusyu, Japan, developed weakness and dysesthesia in the lower limbs. The symptoms resolved after one week without any treatment. Five months later, the weakness and dysesthesia in the lower limbs recurred. The patient went to a hospital and T2-weighted image (T2WI) showed swelling and hyperintensity in the spinal cord from T10 to the lumbosacral region (Figure 1a, c). There was focal nodular enhancement in the posterior segment of the lumbar spinal cord after gadolinium

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Figure 1 Initial T2-weighted MRI taken one month prior to admission to our hospital. (a) Sagittal and (c) axial T2-weighted imaging shows swelling and hyperintensity of the spinal cord from T10 to the lumbosacral region (arrows). (b) Sagittal and (d) axial T1-weighted imaging shows focal nodular enhancement in the posterior segment of the lumbar spinal cord after gadolinium injection.

injection (Figure 1b, d). He was referred to the orthopedics unit as spinal tumor was suspected. However, his weakness and dysesthesia progressed rapidly, and one month later he developed pollakiuria and constipation. At that time, he was admitted to our hospital.

He had a history of hypertension and had suffered a left putamen hemorrhage at 54 years of age. He had no history of allergies. He had a pet dog for approximately 10 years and frequently ate raw chicken meat and beef liver. On neurological examination, in addition to the residual right hemiparesis caused by the left putamen hemorrhage, he had right-dominant weakness in the lower limbs and could not walk. He had dysesthesia below the L1 dermatome level, and numbness in the anogenital region, pollakiuria, and constipation. Deep tendon reflexes were slightly brisk in the right extremity, but Babinski signs were bilaterally negative and muscle tone was normal. Sagittal T2WI MRI showed that swelling and hyperintensity of the spinal cord had increased compared with the image taken one month earlier (Figure 2a). MRI of the brain was normal.

Laboratory investigations showed slight eosinophilia (white blood cell count 5290/mm³, eosinophils 9.6%,

basophils 0.9%, monocytes 6.2%, lymphocytes 38.8%, neutrophils 44.5%) and serum immunoglobulin E (IgE) level was mildly increased at 876 IU/ml (normal range; ≤ 232 IU/ml). Immunologic studies for autoimmune disorders including antinuclear antibodies, anti-Sjögren's syndrome A and B antibodies, proteinase 3-anti-neutrophil cytoplasmic antibodies, myeloperoxidase anti-neutrophil cytoplasmic antibodies, and anti-aquaporin 4 antibodies were negative. Levels of vitamin B12 and angiotensin-converting enzyme were within the normal range. Viral serology showed no evidence of cytomegalovirus, Epstein-Barr virus, herpes simplex virus, or varicella-zoster virus. Antibodies to human T-cell lymphotropic virus type 1 (HTLV-1) by gelatin particle agglutination (PA) test were present in the serum and cerebrospinal fluid (CSF). However, on retesting using indirect immunofluorescence (IF) test, it was positive only in the serum. The monoclonal integration band of HTLV-1 and CSF cytology was not observed, and the neopterin concentration in the CSF was within the normal range. Examination of the CSF showed 8 cells/ μ l with 21.5% eosinophils, glucose 57 mg/dl (normal range; 50–75 mg/dl), and protein 31.5 mg/dl (normal range; 10–40 mg/dl). The immunoglobulin G (IgG) index was 1.12 (normal range; ≤ 0.73) and oligoclonal bands (OCB) were present, although myelin basic protein was negative. CSF cultures yielded no bacterial or fungal growth.

Figure 2 Sagittal T2-weighted MRI before and after treatment at our hospital. (a) Swelling and hyperintensity had increased compared with the images taken one month earlier (Fig. 1a; arrows). (b) After the anthelmintic therapy and the second course of intravenous methylprednisolone, the swelling and hyperintensity had almost disappeared (arrows).

Figure 3 Results of enzyme-linked immunosorbent assay (ELISA). Levels of anti-*Toxocara* antibodies in the serum and CSF were considerably higher than the negative control and the levels of anti-*Ascaris* antibodies.

On the basis of eosinophilia, the history of contact with a dog, and the history of eating raw chicken meat and beef liver, parasitic infection was considered. Enzyme-linked immunosorbent assay (ELISA), which uses larval excretory-secretory antigen-coated microtiter plates,¹¹ detected significantly greater levels of anti-*Toxocara* antibodies in the serum and CSF than those in the negative control. Low levels of anti-*Ascaris* antibodies were detected in the serum as well as in the CSF (Figure 3). Confirmation test for *Toxocara* in the serum by Western blot (WB) was positive in both high and low molecular weight zones (Fig. 4). Based on these findings the patient was diagnosed with myelitis caused by *T. canis* infection.

The initial treatment was the same as for myelitis, with intravenous methylprednisolone (1000 mg/day for 3 days). The patient's muscle weakness improved slightly but he could still not walk. Dysesthesia, pollakiuria, and constipation did not improve. After the diagnosis of *T. canis* myelitis, and 8 days after the initial treatment, the patient received albendazole (600 mg/day for 4 weeks) with a second course of intravenous methylprednisolone (1000 mg/day for 3 days). The dose (10–15 mg/kg of body weight) and the duration of albendazole treatment was followed by the regimen suggested by Yoshikawa *et al.*¹² There was marked improvement in the muscle weakness in the lower limbs and the patient

Figure 4 Positive result yielded by *Toxocara* Western blot IgG kit (LDBIO Diagnostics, Lyon, France) in the serum.

was able to walk using a cane. The dysesthesia remained but numbness in the anogenital region, pollakiuria, and constipation improved. On follow-up T2WI two weeks later, swelling and hyperintensity of the spinal cord had almost disappeared (Figure 2b) and re-examination of the CSF showed that pleocytosis and eosinophilia had resolved (4 cells/ μ l with 5.5% eosinophils, glucose 66 mg/dl, protein 17.6 mg/dl, and IgG index 0.55). Three weeks later, the patient was discharged. After discharge, his muscle weakness and sensory disturbances persisted, but the MRI performed two and eight months after discharge, the swelling and hyperintensity of the spinal cord were no longer observed.

Discussion

Diagnosis of myelitis caused by VLM due to toxocariasis has been generally based on the following four findings:¹³ (1) a high titer of anti-*Toxocara* antibody in the CSF; (2) a high titer of anti-*Toxocara* antibody in the serum; (3) eosinophilia in the serum or CSF; and (4) clinical and radiological improvement after anthelmintic therapy. Recently, Jabbour *et al.* suggested that the presence of eosinophilia is not considered to be essential but supportive for the diagnosis,² whereas specific MRI features might be essential for the diagnosis. The patient presented here fulfilled all four criteria and the specific MRI features, with elevated anti-*T. canis* antibody and eosinophilia in the serum and CSF, and clinical and radiological improvement after anthelmintic therapy. Moreover, the patient resided in Miyazaki Prefecture where food-borne parasitic diseases remain endemic.¹¹ A seroprevalence study revealed that 3.6% of healthy

population in Miyazaki Prefecture and 1.6% of healthy population in Japan are seropositive.¹⁴ Because the patient owned a dog and frequently ate raw chicken meat and beef liver, he had a high risk for toxocariasis.

Atopic myelitis (AM) and HTLV-1-associated myelopathy/tropical spastic paraparesis (HAM/TSP) were considered in the differential diagnosis. Eosinophilia and elevated IgE levels support a diagnosis of AM, but the patient had no history of atopic disease. Furthermore, the patient had OCB in the CSF, which is not a feature of AM.¹⁵ However, in some previous case reports of *T. canis* myelitis, OCB were present.^{3–5}

The tests for HTLV-1 in the serum and CSF were positive by the PA test. Therefore, HAM/TSP was another possible diagnosis. However, the IF test for confirmation were positive only in the serum.¹⁶ Clinically, the disease progression was too acute and there was no spasticity or pathological reflexes in the lower limbs. In addition, the neopterin concentration in the CSF (often used as a marker of disease activity in HAM/TSP¹⁷), eosinophilia, and improvement after anthelmintic therapy also countered a diagnosis of HAM/TSP.

For the parasite, although the relation between HTLV-1 antibody and *Strongyloides stercoralis* have been reported,¹⁸ relation between *T. canis* infection has not been revealed. There were four cases of *T. canis* infection in the past 10 years in our department, but the HTLV-1 antibody was positive only in this case. There appears to be no obvious relation between HTLV-1 antibody and *T. canis* infection, but larger samples and further investigations are needed.

In our patient, anti-*Toxocara* and anti-*Ascaris* antibodies were detected in the serum and CSF by ELISA, and we have performed a confirmatory test by WB only for *Toxocara*. He frequently ate raw chicken meat and beef liver and it is possible that he ate raw beef liver infected with *T. canis* and *Ascaris suum* on separate occasions and developed a co-infection. However, there are no case reports in the literature of co-infection with these two parasites. It is also possible that a single roundworm infection could cause a second variant reactivity on ELISA as a cross-reaction.¹⁹ ELISA indicated significantly higher levels of anti-*Toxocara* antibodies than those of *Ascaris*. These findings support the conclusion that the myelitis was associated with VLM due to *T. canis* rather than with a co-infection of these two parasites. Nevertheless, to confirm these findings, western blotting for *Ascaris* should be performed.

The MRI findings of *T. Canis* myelitis have been previously reported in the literature.^{2–10} Single long cord lesions with focal nodular enhancement in the posterior

or posterolateral segment of the spinal cord, and relatively short segmental involvement, as in the present case, are thought to be characteristic findings.^{2,9–10} In all previous reports, the lesions were located in the thoracic or cervical spinal cord, most frequently in the thoracic spinal cord. There is no previous report of a lesion in the lumbosacral region. The mechanism of the myelitis caused by VML is not well understood but is suspected to be a direct invasion of the larvae of *Toxocara spp.* or mechanical damage caused by an immune-mediated inflammatory response.¹ There is no specific description of confinement of lesions to the cervical and thoracic spinal cord. Information on more cases is required to better understand the mechanism of this disease.

Conclusion

We reported a case of *T. Canis* myelitis involving the lumbosacral region. Our results suggest that *T. Canis* myelitis cannot be discounted even if the myelitis is located in the lumbosacral region. In patients with eosinophilia in the serum or CSF, characteristic MRI findings, and relevant history, myelitis caused by VLM due to *T. canis* should be included in the differential diagnosis, and *Toxocara* serology of the serum and CSF should be performed.

Conflict of interest

There are no conflicts of interest to declare.

References

- Glickman LT, Schantz PM. Epidemiology and pathogenesis of zoonotic toxocariasis. *Epidemiol Rev* 1981;3:230–50.
- Jabbour RA, Kanj SS, Sawaya RA, Awar GN, Hourani MH, Atweh SF. *Toxocara canis* myelitis: clinical features, magnetic resonance imaging (MRI) findings, and treatment outcome in 17 patients. *Medicine (Baltimore)* 2011;90(5):337–43.
- Duprez TP, Bigaignon G, Delgrange E, Desfontaines P, Hermans M, Vervoort T, et al. MRI of cervical cord lesions and their resolution in *Toxocara canis* myelopathy. *Neuroradiology* 1996;38(8):792–5.
- Strupp M, Pfister HW, Eichenlaub S, Arbusow V. Meningomyelitis in a case of toxocariasis with markedly isolated CSF eosinophilia and an MRI-documented thoracic cord lesion. *J Neurol* 1999;246(8):741–4.
- Goffette S, Jeanjean AP, Duprez TP, Bigaignon G, Sindic CJ. Eosinophilic pleocytosis and myelitis related to *Toxocara canis* infection. *Eur J Neurol* 2000;7:703–6.
- Kumar J, Kimm J. MR in *Toxocara canis* myelopathy. *AJNR Am J Neuroradiol* 1994;15(10):1918–20.
- Eberhardt O, Bialek R, Nägele T, Dichgans J. Eosinophilic meningomyelitis in toxocariasis: case report and review of the literature. *Clin Neurol Neurosurg* 2005;107(5):432–8.
- Umehara F, Ookatsu H, Hayashi D, Uchida A, Douchi Y, Kawabata H, et al. MRI studies of spinal visceral larva migrans syndrome. *J Neurol Sci* 2006;249(1):7–12.
- Lee IH, Kim ST, Oh DK, Kim HJ, Kim KH, Jeon P, et al. MRI findings of spinal visceral larva migrans of *Toxocara canis*. *Eur J Radiol* 2010;75(2):236–40.
- Kaido M. Ascarid myelopathy. *Neurol Med* 2012;77:243–50. [Article in Japanese]

- 11 Maruyama H, Noda S, Nawa Y. Emerging problems of parasitic diseases in southern Kyushu, Japan. *Jpn J Parasitol* **1996**;45(3): 192–200.
- 12 Yoshikawa M, Koyama N, Hontsu S, Yamamoto Y, Mikasa K, Kimura H. Lessons from eight cases of adult pulmonary toxocar-
iasis. *Respirology* **2011**;16(6):1014–15.
- 13 Finsterer J, Auer H. Neurotoxocarosis. *Rev Inst Med Trop Sao Paulo*. **2007**;49(5):279–87.
- 14 Kondo K, Akao N, Ohyama T, Okazawa T. Sero-epidemiological investigation of toxocarasis in Asian area. In: Yamaguchi T, Araki T, Chen ER, (ed.) *The Fifth Asian-Pacific Congress for Parasitic Zoonoses: The Organizing Committee of Asian-Pacific Congress for Parasitic Zoonoses Publishing*; **1998**. p. 65–70.
- 15 Isobe N, Kanamori Y, Yonekawa T, Matsushita T, Shigeto H, Kawamura N, Kira J. First diagnostic criteria for atopic myelitis with special reference to discrimination from myelitis-onset mul-
tiple sclerosis. *J Neurol Sci* **2012**;316(1–2):30–5.
- 16 Hinuma Y, Nagata K, Hanaoka M, Nakai M, Matsumoto T, Kinoshita KI, *et al.* Adult T-cell leukemia: antigen in an ATL cell line and detection of antibodies to the antigen in human sera. *Proc Natl Acad Sci U S A* **1981**;78(10):6476–80.
- 17 Yamano Y, Sato T. Pathophysiology, treatment and biomarkers for HTLV-1-associated myelopathy/tropical spastic paraparesis (HAM/TSP). *Nippon Rinsho* **2013**;71(5):870–75. [Article in Japanese]
- 18 Nakada K, Kohakura M, Komoda H, Hinuma Y. High incidence of HTLV antibody in carriers of *Strongyloides stercoralis*. *Lancet* **1984**;1(8377):633.
- 19 Watthanakulpanich D. Diagnostic trends of human toxocarasis. *J Trop Med Parasitol* **2010**;33:44–52.