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Rare nervous system involvement in an anti-myelin oligodendrocyte-positive case: spinal leptomeningeal involvement

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Abstract

Anti-myelin oligodendrocyte (MOG)-positive cases have a widespread clinical presentation. MOG autoantibodies are associated with acute disseminated encephalomyelitis, multiple sclerosis, clinically isolated syndrome, neuromyelitis optica, isolated optic neuritis, and transverse myelitis. Our patient had bilateral optic neuritis with acute tetraparesis. Cranial magnetic resonance imaging (MRI) showed leptomeningeal contrast involvement and contrast enhancement in all cranial nerve nuclei. Spinal MRI revealed leptomeningeal contrast enhancement, and a contrast-enhancing lesion was found in the cervical spinal cord.
This case is worth presenting because spinal leptomeningeal involvement is rare in anti-MOG-positive patients.

Keywords: Anti-MOG, Tetraparesis, Spinal, Leptomeningeal enhancement

Introduction

Anti-myelin oligodendrocyte (MOG) is found on the outer surface of the myelin sheath and on oligodendrocytes in the central nervous system [1-3]. Anti-MOG-positive cases have a widespread clinical presentation. MOG autoantibodies are associated with acute disseminated encephalomyelitis (ADEM), multiple sclerosis (MS), clinically isolated syndrome, neuromyelitis optica (NMO), isolated optic neuritis (ON), and transverse myelitis (TM) [2, 4-7]. Here, we report the case of a patient with spinal leptomeningeal contrast enhancement, which has rarely been reported.

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Informed Consent
The authors stated that the written consent was obtained from the patient presented with images in the study.

Conflict of Interest
No conflict of interest was declared by the authors.

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Case presentation

Written informed consent was obtained from the patient to publish this study. An 18-year-old female patient was admitted with complaints of progressive blurred vision, arm weakness, and gait disturbance. An eye examination revealed a bilateral relative afferent pupillary defect, and she could see only bilateral hand motion. Bilateral papilledema was also observed during the fundus examination (Figure 1). Motor strength was bilaterally 4/5 in upper extremities and 2/5 bilaterally in lower extremities, deep tendon reflexes were globally hypoactive, and the plantar response was neutral. Urinary retention occurred the next day. Cranial magnetic resonance imaging (MRI) showed leptomeningeal contrast involvement and contrast enhancement in all cranial nerve nuclei (Figure 2). Spinal MRI revealed leptomeningeal contrast enhancement (Figure 3), and a contrast-enhancing lesion was found in the cervical spinal cord (Figure 4). A lumbar puncture was performed. Ten monocytes were detected in cerebrospinal fluid (CSF), CSF protein was 112 mg/dL, and CSF cytology was negative. Angiotensin-converting enzyme, tuberculosis-Polymerase Chain Reaction (PCR), salmonella, brucella, syphilis tests, CSF cytological examination, and aquaporin-4 antibody test results were negative. The IgG index was 0.79. The CSF oligoclonal band was type 2-positive.

Figure 1: Bilateral papilledema in the patient's fundus upon examination

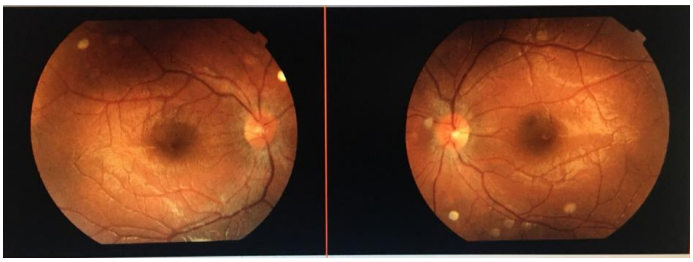


Figure 2: Brain MRI images showing linear contrast enhancement in axial (a) and sagittal (b) postcontrast series, which is inconsistent with leptomeningeal involvement.

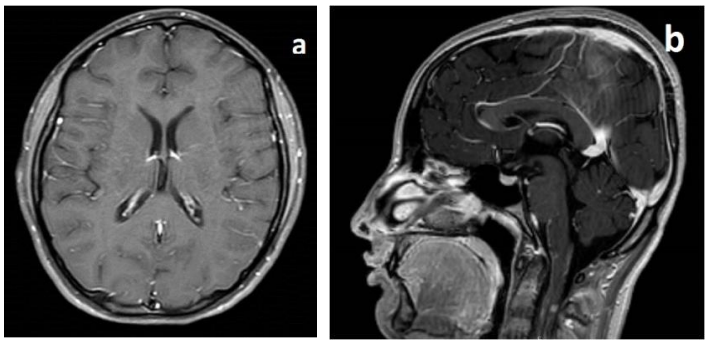


Figure 3: Linear contrast enhancement was consistent with leptomeningeal involvement in the patient's spinal lumbar (a) and cervical (b) postcontrast T1A images

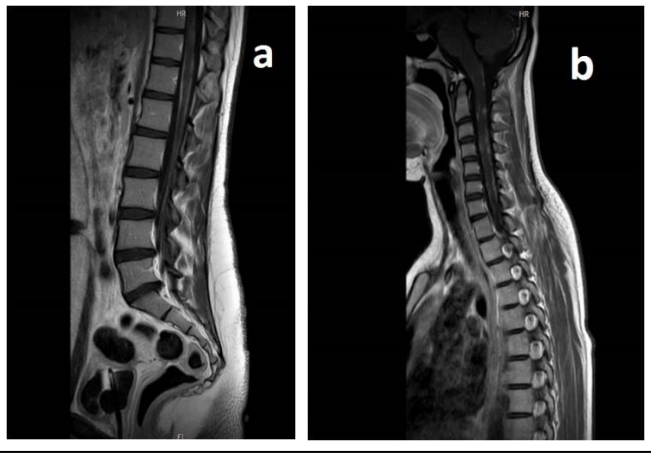
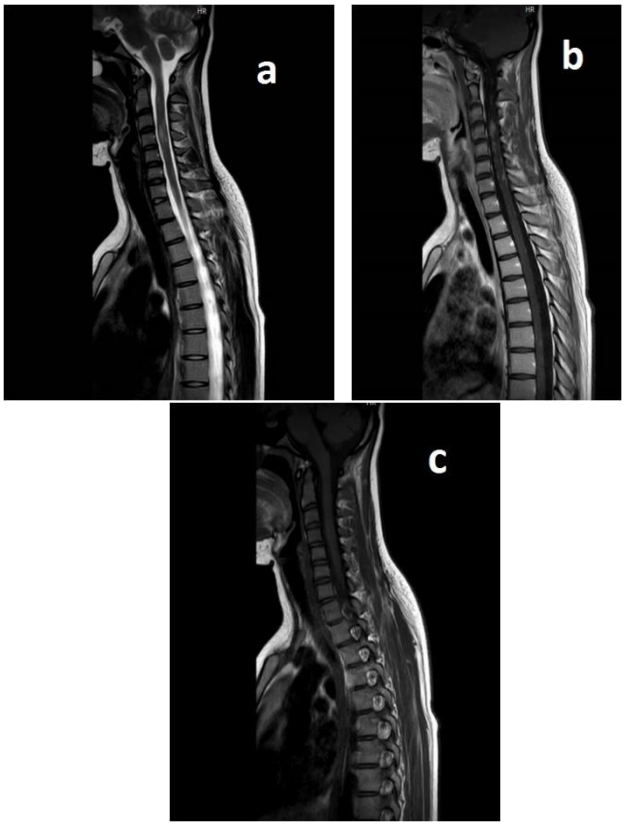


Figure 4: Cervical sagittal MRI sequences of T2A (a), post-contrast (b) and pre-contrast (c). T2A images showed a mild expansile lesion with contrast enhancement in the patient's cervical spinal cord.



The patient was started on 1000 mg/day of intravenous methylprednisolone, and after 10 days, treatment was continued with 1 mg/kg/day of oral methylprednisolone. Approximately 10 days after treatment initiation, the patient's visual acuity had improved to 0.5 in the right and left eyes, and the muscle strength was bilaterally 5/5 in the proximal upper extremities, 4/5 in the distal extremities, and 4/5 in the lower extremities. She was discharged on 1 mg/kg/day of oral methylprednisolone. During the outpatient clinic follow-up, the anti-MOG result was positive, and azathioprine was started. Our patient responded well to steroids and azathioprine, and her clinical symptoms completely resolved. We followed the patient for 2 years, and no new episodes developed.

Discussion

Information on antibody-mediated diseases in the central nervous system has increased in the last decade. One of these antibodies is the MOG antibody. Anti-MOG has been identified in diseases with different clinical spectra, including seronegative neuromyelitis optica spectrum disease (NMOSH), ON, TM, ADEM, and encephalopathic diseases [8-11]. Our patient had bilateral ON and tetraparesis and responded well to steroid and azathioprine treatment, and her clinical symptoms subsequently resolved completely.

The interesting feature of this patient is the presence of spinal leptomeningeal involvement. Leptomeningeal involvement has recently been recognized as an important feature in MS and AQP4-positive NMOSH [12]. However, few anti-MOG-positive cases have been reported with leptomeningeal involvement as case reports in the literature. Reported rare cases with leptomeningeal involvement are associated with cranial leptomeningeal involvement [13-15].

There are only a few patients with spinal leptomeningeal involvement, such as in our case [16, 17]. The spinal cord was involved in two different ways on the MRI from patients with a myelitis episode; one is T2 signal hyperintensity exceeding three vertebral segments, and the other is involvement not exceeding two vertebral segments. Most lesions involved white matter, with more than 50% of the cord in the axial section, and this was accompanied by cord edema. The lesions involved the entire spinal cord, but conus involvement was specific. A patched, cloud-like enhancement with unclear borders was observed [18].

Inflammation in CNS demyelinating diseases is often associated with blood–brain barrier disruption. Leptomeningeal enhancement, which indicates an abnormally permeable leptomeningeal blood barrier, was accompanied by intraparenchymal blood–brain barrier breakdown during an acute myelitis episode, as visualized by contrast enhancement on T1-weighted imaging. This finding suggests that meningeal inflammation may have occurred as a bystander reaction following MOG–IgG-related parenchymal inflammation associated with subpial demyelination [18].

Conclusion

Our case shows that there might also be spinal leptomeningeal involvement in anti-MOG-positive myelitis patients. As the literature on these subjects increases, the clinical and radiological findings in anti-MOG patients will be more clearly defined.

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