·Letter to the Editor·

Incidental frontoparietal parasagittal meningioma in a patient with acute bilateral optic neuritis

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Received: 2014-01-21 Accepted: 2014-04-22

DOI:10.3980/j.issn.2222-3959.2014.06.28

Lim SH, Moon SH, Kim MM. Incidental frontoparietal parasagittal meningioma in a patient with acute bilateral optic neuritis. *Int J Ophthalmol* 2014;7(6):1064-1066

Dear Sir,

Bilateral optic neuritis is not a rare condition in children following a viral syndrome [1]. In contrast, simultaneous bilateral optic neuritis without known systemic disease has been considered a rare condition in adults [2]. In addition, incidental extra-axial meningioma in a patient with bilateral optic neuritis has not been documented. The authors present a first case report on incidental parasagittal meningioma with bilateral optic neuritis.

A 40-year-old male presented with acute simultaneous and worsening visual loss and ocular pain in both eyes that started five days prior to the clinic visit. He had globe tenderness and worsening of ocular pain on ocular movement. He also had Uhthoff's symptom, a transient visual obscuration associated with elevation in body temperature. The patient reported the appearance of gray cloud or veil front of his eyes, with particular involvement of the central field. There were no co-morbid medical conditions, and past medical history was not significant.

On presentation, his best corrected visual acuity (BCVA) was counting finger in both eyes; the patient recognized only the first plate of Ishihara pseudoisochromatic plates. The pupillary reactions showed mid-dilated pupil with diminished pupillary light reflex in both eyes. Fundus examination revealed mild disc edema with surrounding peripapillary edema in both eyes. However, there were no signs of disc hemorrhage, sheathing of retinal vessels, or periphlebitis, suggesting anterior ischemic optic neuropathy, sarcoidosis, or Behcet's disease. Optical coherence tomography (OCT) also demonstrated peripapillary retinal nerve fiber layer swelling. Findings on fluorescein angiography revealed fluorescein leakage bounded by edematous disc in the late phase (Figure 1A). Visual field tests showed generalized field loss (mean deviation, -32.47 dB in the right eye, -33.16 dB in the left eye) in both eyes (Figure 1C). He showed no neurological deficit and there were no signs of meningeal irritation or symptoms of transverse myelitis.

Thus, we made a provisional diagnosis of atypical bilateral optic neuritis secondary to demyelination. In order to exclude multiple sclerosis, neuromyelitis optica (NMO), and other etiologies, we advised the patient to undergo chest X-ray and magnetic resonance imaging (MRI, with gadolinium enhancement and fat suppression techniques). Laboratory measurements included a liver function test, electrolyte levels, angiotensin converting enzyme, antinuclear antibody (ANA), fluorescent treponemal antibody-absorption (FTA-ABS), venereal disease research laboratory test (VDRL for syphilis), and anti-NMO IgG antibody assays. However, the cerebrospinal fluid was not evaluated because the patient refused to undergo lumbar puncture. Serologic tests and anti-NMO IgG were normal and negative.

Axial T1-weighted MRI showed incidental right frontoparietal parasagittal meningioma with homogeneous enhancement (Figure 2A). Coronal T1-weighted MRI showed meningioma filling the angle between the falx and convexity dura, characteristic of parasagittal meningioma with patent superior sagittal sinus (Figure 2B). Axial and coronal T1 fat saturation MRI showed increased intensity of both optic nerve sheaths. (Figure 2C, 2D). In addition, sagittal T1 fat saturation MRI showed concomitant parasagittal meningioma with optic nerve sheath enhancement (Figure 2E). T2 weighted image did not show distension of perioptic subarachnoid space. And MRI images did not show demyelinating plaques, suggesting multiple sclerosis in white matter or compressing lesions, such as a suprasellar mass or optic nerve sheath meningioma.

The patient received intravenous injection of methylprednisolone of 1000 mg per day, in divided doses for 3d, followed by oral prednisolone at 60 mg per day for 11d, followed by a gradual dose reduction according to the Optic Neuritis Treatment Trial regimen [3]. Seven days after administration of systemic IV steroid, his BCVA recovered

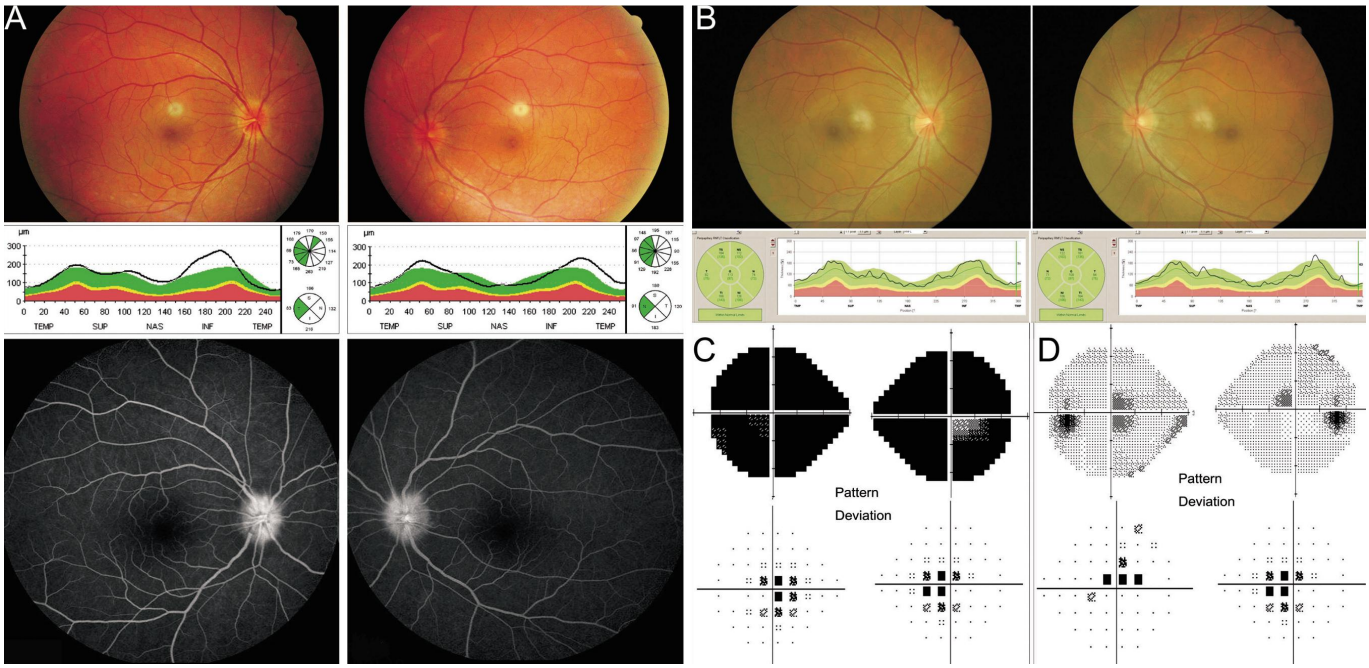

Figure 1 Ophthalmic findings and visual fields of the patient on presentation (A, C) and two weeks after treatment (B, D).

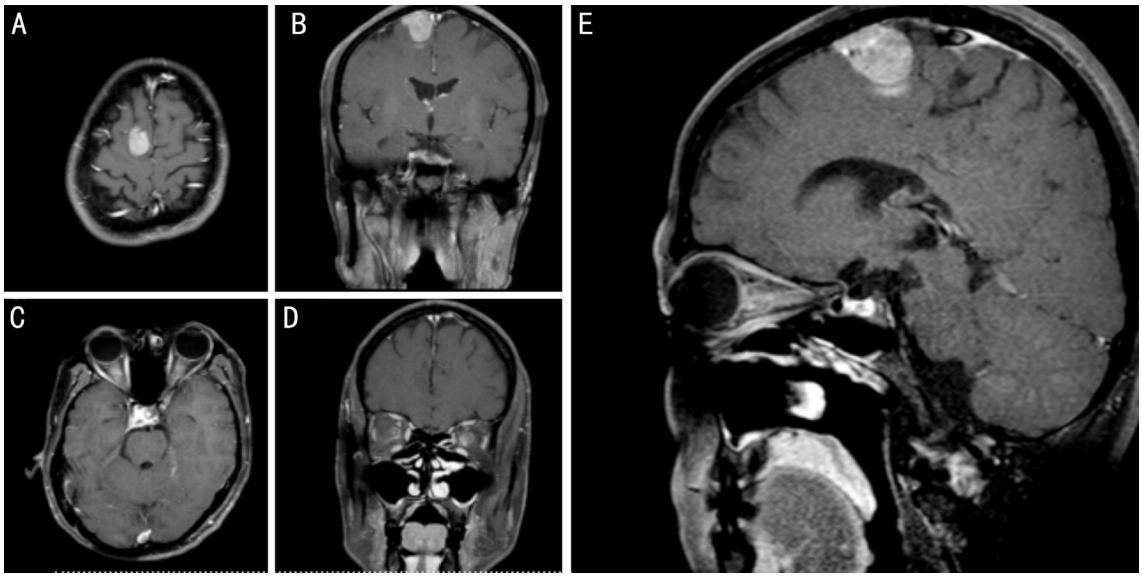

Figure 2 Magnetic resonance (MR) images of the patient.

to 20/40 in both eyes, and fundus examination showed decreased peripapillary disc swelling. Two weeks after administration of systemic IV steroid, his BCVA was 20/25 in the right eye and 20/20 in the left eye, and his visual field showed marked improvement, compared to examination before treatment (Figure 1D). Fundus examination showed minimal peripapillary swelling. However, the amount of swelling was markedly reduced, as shown by OCT (Figure 1B). Gamma knife treatment of the extra-axial meningioma was planned. However, he wanted a second opinion at another hospital and the treatment at our hospital was deferred.

We present a atypical bilateral optic neuritis with incidental meningioma. To the best of our knowledge, this is a first case report on incidental parasagittal meningioma with bilateral optic neuritis. When evaluating patients with acute bilateral optic neuropathy, physicians should consider other etiologies, including immunodeficiency, viral infection, NMO, multiple sclerosis, paraneoplastic autoimmune optic neuritis, and vaccination-induced optic neuritis [4-7]. In this patient, there was no evidence for transverse myelitis, immunodeficiency, or neoplasm according to history or clinical findings on presentation. In addition, a serologic test for anti-NMO IgG antibody (aqua-porin 4 antibody) using a cell-based assay was negative. Therefore, the authors made a diagnosis of idiopathic acute demyelinating bilateral optic neuritis with incidental extra-axial meningioma.

Meningiomas are mostly very slow growing tumor and the majority are asymptomatic through life [8]. Whether this parasagittal meningioma is a part of this clinical phenotype or not remains uncertain. Then, why bilateral optic neuritis occurs in this patient and what is possible mechanisms of

these changes? The authors hypothesized the mechanisms including meningioma-associated inflammation, peritumoral edema or incidentaloma. However, the exact pathogenesis remains unknown. The first possible mechanism is inflammation associated with meningioma. The recent published article suggested that meningioma-associated mediators may trigger an inflammatory reaction combined with vasculopathy leading to oculomotor neuropathy mimicking ophthalmoplegic migrane [9]. In similar, Barrett *et al* [10] also reported that recurrent third nerve palsy as the presenting feature of neurofibromatosis 2. And histopathologic findings showed increased cyclooxygenease-2 expression in meningioma [11]. These findings may suggest the some association between inflammatory reaction and meningioma. The second possible mechanism is peritumoral edema effect. Some authors suggested increased expression of aqua-porin4 (AQP4) is associated with peritumoral edema in meningiomas [12,13]. And they hypothesized AQP4 over-expression could lead to abnormal water transport and edema formation in meningiomas and increased AQP4 level is also closely related to the expression of VEGF [12,13]. We thought that these changes might affect the anti-NMO IgG antibody assay in our patient and these changes might disturb the axoplasmic flow in optic nerve.

The differential diagnosis between papilledema and optic neuritis is also important. This patient did not complain headache or nausea suggesting intracranial hypertension. Visual field defects generally show the enlarged blind spot and pupillary light reflex are not affected in papilledema. In contrast, the visual field results of the patient were generalized depression and pupillary reaction showed diminished light reflex in both eyes. Cerebrospinal fluid (CSF) pressure and MRV examination are also important to diagnose the papilledema, however, these were not examined in this patient. The recent published article about OCT image suggested the retinal pigment epithelium (RPE)/Bruch's membrane (BM) is commonly deflected inward in papilledema, because the RPE/BM angulation is presumed to be caused by elevated pressure in the subarachnoid space[14]. On the contrary, the RPE/BM showed negative angulation in this patient. Thus the authors could exclude papilledema with increased intracranial pressure.

The previous study [2] reported that acute bilateral optic neuritis without myelopathy typically shows improvement with corticosteroid therapy without immunomodulatory therapy and had good prognosis. These findings were compatible with our results. In addition, the action of corticosteroids in brain tumors includes reduction in vascular permeability, inhibition of tumor formation, and decreased CSF production. These mechanisms might be some helpful to recover visual symptoms in this patient.

Only two patients enrolled in the optic neuritis treatment trial (ONTT) study were found to have compressive lesions; therefore, it could be argued that, although MRI is always performed, it is seldom necessary [15]. In this case report, extra-axial meningioma with bilateral acute optic neuritis was found incidentally. This case report highlights the importance of neuroradiologic imaging and careful neurologic examination when evaluating atypical bilateral optic neuritis in adults.

ACKNOWLEDGEMENTS

Conflicts of Interest: Lim SH, None; **Moon SH,** None; **Kim MM,** None.

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