Clinical efficacy and safety of intrathecal methotrexate in the treatment of Balo's concentric sclerosis: A case report

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Introduction

Balo's concentric sclerosis (BCS) is a rare, progressive variant of multiple sclerosis characterized by concentric rings of demyelinated white matter and relatively preserved myelin alternating with each other. Intense immunosuppressive treatment may be indicated in patients with aggressive disease. In this study childhood onset BCS which benefited from intrathecal (IT) methotrexate (MTX) is presented.

Case report

A previously healthy 5 year-old boy was admitted with weakness of left arm/leg and aphasia. All laboratory tests were normal. Cerebrospinal fluid (CSF) oligoclonal bands were type 2 negative. Magnetic Resonance Imaging (MRI) showed multiple tumefactive and multi-layered demyelinating lesions (Figure1). Intravenous (IV) immunoglobulin, IV+oral steroids, plasma exchange were given sequentially. The left hemiplegia recovered partially, but right hemiplegia developed on the 7 days later. Immunosuppressive therapy with azathioprine and rituximab were given, but the disease progressed radiologically (Figure 2). ITMTX was started with 12,5 mg via lumbar puncture and repeated every two months. No side effects were observed in the patient. After 1 year of ITMTX treatment, his left hemiplegia had fully and right hemiplegia partially recovered, and he started to speak few words. Radiological remission was also observed (Figure 3). He has been treating with ITMTX for 18 months(9 doses) and there is no new lesion in the neuroimaging in the last 6 months.

Discussion

ITMTX was well tolerated, with no major safety or tolerability issues.
One of the noteworthy aspects of this treatment is the acceptance of patients to undergo repeated LP in order to receive ITMTX.

Stark et al. reported that significant adverse effects were not observed in their cohort despite long term treatment(3-10 years) because of the dosing frequency relative to the much higher dosing frequency used in patients with CNS malignancies.

The mechanism of action of MTX in MS is unknown. Mueller et al. suggested in an animal study that the mechanism of action of IT MTX in progressive MS may be a result of inhibition of glial scar formation (sclerosis).

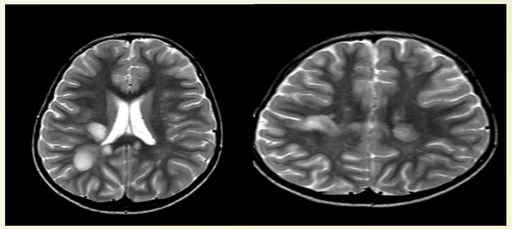

Figure-1. Magnetic Resonance Imaging (MRI) showed multiple tumefactive and multi-layered demyelinating lesions (Balo'sconcentric sclerosis) at the onset .

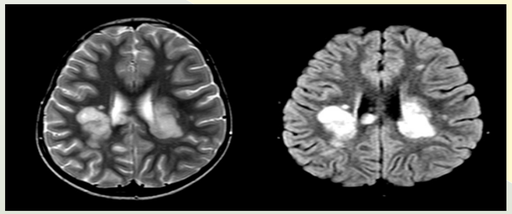

Figure-2. Increase in lesions size and new contrast-enhancing lesion (pre-ITMTX)

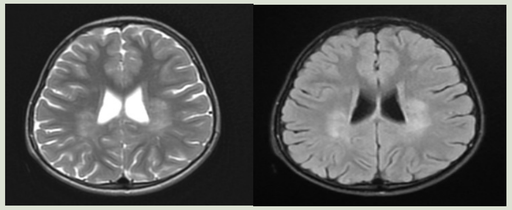

Figure-3. Posterior periventricular sequelae lesions (1 year after ITMTX)

Conclusion

The safety and the tolerability of long-term therapy with ITMTX in patients with treatment-resistant, progressive forms of MS were reported previously. There was evidence of disease stabilization in our patient and long-term ITMTX appears as a safe therapeutic option in advanced BCS even in the childhood.

References

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