Acute demyelination in childhood associated with TNF alpha-inhibitor therapy

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Abstract

Introduction: Tumor necrosis factor-alpha inhibitors (TNFi) have revolutionized the treatment of rheumatological autoimmune disorders. We report a rare, although serious concern with TNFi usage, demyelination of the central nervous system.

Material and methods: Case report.

Results: A 15-year old woman with history of polyarticular juvenile idiopathic arthritis stable on methotrexate and infliximab developed blurred vision and retro-orbital pain in her right eye. She had no preceding illness or fever and her arthritis was well controlled. Her examination confirmed right optic neuritis with severely impaired visual acuity (20/200), dyschromatopsia and right afferent papillary defect. Blood test results were within normal range, extensive infectious work-up and aquaporin-4 antibody were negative. A brain MRI performed one week after the onset of symptoms revealed asymmetric T2-hyperintensity and enlargement of the right prechiasmatic optic nerve, compatible with optic neuritis without abnormal enhancement, and the brain parenchyma appeared unremarkable. She declined to have a lumbar puncture, which had been proposed for routine studies, IgG index and oligoclonal bands. She was treated with pulse high-dose steroids and experienced rapid improvement. Infliximab was discontinued and methotrexate was increased to 25 mg weekly. After 3 years of follow-up her central acuity is 20/20 in the right eye. She has had no clinical or MRI evidence of additional demyelinating lesions.

Conclusions: TNFi medications can be associated with demyelination in the central and peripheral nervous system even in childhood. Close follow-up of these patients is mandatory to determine whether they have a monophasic or recurrent CNS disorder.