Hourglass Constriction Neuropathy Affecting Suprascapular Nerve: Case report

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Introduction

Hourglass constriction (HGC) neuropathy is a neurologic condition caused by non-traumatic, non-compressive fascicular constrictions of one or more individual peripheral nerves. Differential diagnostic approaches for non-traumatic acute unilateral shoulder weakness are important for further treatments. We present a case of HGC affecting suprascapular nerve which were visualized by SHINKEI (nerve-SHeath signal increased with INKed rest-tissue RARE Imaging) magnetic resonance neurography (MRN) and describe the Result of nonsurgical treatments in this case.

Case

A 26-year-old female presented with a 5 month history of scapular pain and difficulty in elevating left arm over head. Past medical history revealed complete remission of non-Hodgkin's lymphoma 15 years ago and Kikuchi diseases 5 years ago. 10 days before the development of weakness, she experienced extremely severe pain in the shoulder girdle area with 3 days preceding flu-like symptoms. After the pain decreased over ten days, she noticed her weakness. Manual muscle tests revealed weakness of shoulder abductor and external rotator (Medical Research Council [MRC] Grade, 3/5), although elbow flexor and wrist extensor was normal. There were no abnormalities in sensory tests. On inspection, the suprapinatus and infraspinatus were mildly atrophied. Electromyography showed denervation of the supraspinatus and infraspinatus. Neck, chest, and abdominal computed tomography revealed normal findings. At 3 months after the onset, she visited other hospital. Cervical spine magnetic resonance imaging (MRI) revealed no disc herniation or foraminal stenosis (Fig. 1A), and shoulder MRI (Fig. 1B) demonstrated increased signal intensity of supraspinatus and infraspinatus suggesting denervation related edema. At that time, she was presumptively diagnosed as an idiopathic neuralgic amyotrophy (INA). At 5 months after the onset, she visited our clinic and performed highsolution MRN using SHINKEI protocol. MRN revealed three focal constrictions of the suprascapular nerve (Fig. 1C and 1D). Laboratory tests including cerebrospinal fluid analysis, anti-nuclear autoantibody, and anti-ganglioside antibodies were normal. Thus, she was diagnosed as a HGC neuropathy of suprascapular nerve rather than an INA. She was treated with oral steroid (prednisolone 10mg for 14 days) and ultrasonography guided perineural injection (triamcinolone acetonide 20mg) once, and then improved to near normal (motor strength of shoulder abduction and external rotation [MRC, 5-/5]) at the 10 months after the onset.

Discussion

Although HGC neuropathy is a rare entity and the exact etiology is still unclear, it should be considered as a potential cause of spontaneous nerve palsy, especially unilateral shoulder weakness. Because HGC neuropathy clinically mimic an INA, high resolution MRN should be performed to differentiate HGC neuropathy from INA.

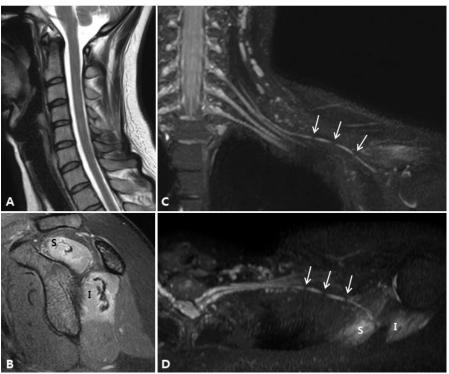


Fig. 1. Images of Case 1. (A) No evidences of cervical disc herniation or foraminal stenosis. (B) Sagittal 2D T-weighted fat suppression image demonstrates denervation-related edematous change of the supraspinatus (S) and infraspinatus (I) muscles. (C) Curved reformatted coronal 3D SHINKEI magnetic resonance neurography (MRN) reveals three focal constrictions (arrows) of the suprascapular nerve. (D) Curved reformatted axial 3D SHINKEI MRN reveals three focal constrictions (arrows) of the suprascapular nerve with denervation-related edematous change of the supraspinatus (S) and infraspinatus (I) muscles.