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Case report

Bell-shaped sensory impairments of all modalities in a neurosarcoidosis patient

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Abstract

We describe a 45-year-old man with neurosarcoidosis complaining of bell-shaped tightening and pain with sensory disturbance of superficial and deep sensations. The patient showed subacute progressive sensory impairment in bilateral C7-Th12 dermatomes. Triceps and patellar tendon reflexes were decreased. Chest X-ray revealed bilateral hilar lymphadenopathy without pleural effusion. There was abnormal accumulation of gallium in the bilateral hilar lymph nodes, parotid glands, and lacrimal glands on scintigraphy. Examination of bronchoalveolar lavage fluid showed an elevated CD4/CD8 ratio. Transbronchial lung biopsy showed non-caseating granulomas with many epitheloid cells and occasional Langhans giant cells without any necrotic lesion. The tuberculin reaction was negative, and elevation of serum lysozyme and IgG level were seen. These findings fulfilled the clinical criteria for sarcoidosis. Spine MRI demonstrated no abnormality. Studies of short-latency somatosensory evoked potentials showed delayed N13 latency and absent N19 and N28 potentials bilaterally. A nerve conduction study revealed no abnormality. The patient's muscle strength was normal through the entire clinical course. Therefore, we consider that his sensory impairment was caused by peripheral neuropathy, especially in the dorsal root region. Neurosarcoidosis is important for differentiating bell-shaped sensory impairments of all modalities.

Keywords: Neurosarcoidosis; Sensory impairment; Dorsal root

1. Introduction

Bell-shaped (forme suspension) sensory impairment affects the trunk and upper limbs that correspond with successive dermatomes. Cervical syringomyelia or intramedullary tumors cause bell-shaped sensory impairment, which usually shows dissociated sensory disturbances. While sarcoidosis could affect the sensory nerve system, there have been only a few reports about bell-shaped sensory impairment during neurosarcoidosis [1–3]. Moreover, there have been no reports of bell-shaped sensory impairment of all modalities during sarcoidosis. In this report, we present a 45-year-old man with neurosarcoidosis who had bell-shaped impairment of superficial and deep sensations. We emphasize that the presence of bell-shaped sensory impairment of all modalities helps diagnosis of neurosarcoidosis.

2. Case report

A 45-year-old man had noted paresthesia in the umbilical region. He manifested subacute progressive paresthesia. Two weeks later, he was admitted to our hospital. On physical examination, his body temperature was 36.6 °C, blood pressure was 128/86 mmHg, and pulse was regular (78/min). There were no cardiac murmurs, rhonchus or crackles. He had no skin lesions or superficial lymphadenopathy. Neurological examination revealed sensory impairment of all modalities in the bilateral C8-Th10 dermatomes, although worse in the left side, that made him feel paresthesia. There was no abnormality in the cranial nerves, muscle tone, power, or deep tendon reflexes. Pathological reflexes were not shown. He had no def-

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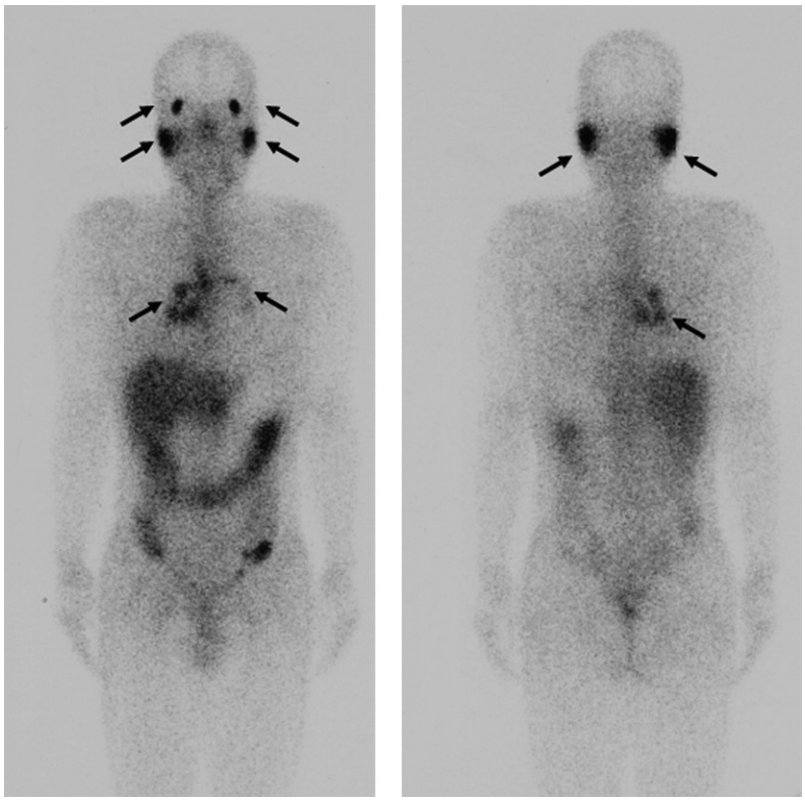

Fig. 1. ^{67}Ga scintigraphy showing bilateral abnormal accumulation in lacrimal glands, parotid glands and hilar lymph nodes (arrows).

inite cerebellar ataxia. Neither Spurling's sign nor Lasègue's sign was noted. He did not complain of any bowel or bladder dysfunction. Laboratory studies failed to detect evidence of diabetes mellitus, collagen disease, or viral infection. The white blood cell (WBC) count was 4500/μl with 9.9% eosinocytes. Serum IgG, lysozyme, and soluble interleukin (IL)-2 receptor levels were elevated (1150 mg/dl, 10.4 μg/ml, and 887 U/ml, respectively). Serum IgE and angiotensin-converting enzyme (ACE) levels were normal. The CD4/CD8 ratio (4.43) in the blood was elevated. Cerebrospinal fluid (CSF) examination revealed pleocytosis (38/μl for lymphocytes), elevation of total protein (106 mg/dl), and normal glucose (46 mg/dl). Oligoclonal studies were negative. The reaction of a tuberculin test was negative. Ophthalmic examination detected uveitis without symptoms. Chest radiography revealed bilateral hilar lymphadenopathy that was worse in the right side than the left side, with a normal lung field and without the presence of pleural effusion. During a chest CT scan, paratracheal and bilateral hilar lymphadenopathy were observed. Gallium scintigraphy demonstrated abnormal accumulation in bilateral lacrimal glands, parotid glands, and hilar and mediastinal lymph nodes (Fig. 1). Examination of bronchoalveolar lavage fluid showed an elevated CD4/CD8 ratio (4.78). Since these findings suggested sarcoidosis, a transbronchial lung biopsy was performed. Histological findings of the lung tissue showed non-caseating epithelioid granulomas without a necrotic region that was compatible with sarcoidosis (Fig. 2). Gd-enhanced brain and spinal MRI showed no abnormalities. Motor and sensory nerve conduction studies including F-wave revealed normal function. On examination of short-latency somatosensory evoked potentials (SSEP), the bilateral N13 latency was delayed (Rt.: 14.19 ms, Lt.: 14.19 ms; normal values: 11.88–13.46 ms) and both bilateral N19 and N28 were not evoked.

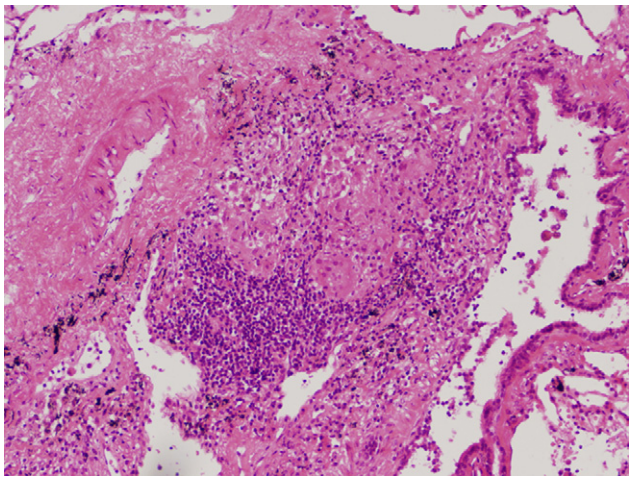

Fig. 2. Biopsy of lung tissue showing non-caseating epithelioid cell granulomas and occasional Langhans giant cells without any necrotic lesion (hematoxylin-eosin stain, original magnification × 100).