CASE REPORT

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A Case of Multiple Sclerosis Presenting with Thoracic Lhermitte's Sign

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ABSTRACT Lhermitte's sign (LS) is a transient symptom consisting of an electric shock-like sensation radiating ascending along the spinal cord during forward flexion of the neck. It may be associated with sensory complaints such as hypo or hyperesthesia. Although it is more common in cervical lesions, it can also be seen less frequently in thoracic lesions. LS is classified as one of the paroxysmal pain syndromes of multiple sclerosis (MS). It can also be seen in cervical spondylosis, disc herniation, trauma, spinal cord malignancies, vitamin B12 deficiency, cisplatin toxicity and radiation myelotoxicity. In cases of thoracic myelitis, LS should be considered and confirmed by neuroradiological and neurophysiological examinations. In this article, we aimed to present a case of MS with Lhermitte sign due to a lesion in the thoracic spinal cord, as a rare but possible example of spinal cord pathology in the thoracic region.

Keywords: Spinal kord; myelitis; Lhermitte; multiple sclerosis

Lhermitte's sign (LS) was first described in 1924 as a transient symptom consisting of an electric shock-like sensation radiating along the spinal cord during forward flexion of the neck. It can be associated with sensory complaints such as hypo- or hyperesthesia.[1] Although it is more common in cervical lesions, it can also be seen less frequently in thoracic lesions. LS is classified as one of the paroxysmal pain syndromes of multiple sclerosis (MS).[2] We aimed to present a case of MS with LS due to a lesion in the thoracic spinal cord.

CASE REPORT

A 22-year-old female patient was admitted to the neurology outpatient clinic with numbness and tingling sensation in both feet for two weeks. The patient stated that when she bent forward with her body, there was an electric feeling that started in her waist and spread downwards. Trauma, urinary and stool incontinence was not described. There was no features in the patient's medical and family history. In her neurological examination, hypoesthesia was detected in the bilateral distal lower extremities and plantar response could not be obtained on the right. Routine blood tests were within normal limits. After thoracolumbar magnetic resonance imaging (MRI) showed a lesion at the T4 level that was hyperintense on T2-weighted images, isointense on T1-weighted images, and showed contrast enhancement (Figure 1).

On cranial MRI, pericallosal, periventricular, subcortical, and cortical white matter hyperitense fusiform lesions in fluid-attenuated inversion recovery sequence (Figure 2). There is no lesion and contrast agent involvement was observed on cervical MRI. Lumbar puncture was performed to the patient with the prediagnosis of MS. The cerebrospinal fluid (CSF) opening pressure was 140 mmH20 and its appearance was clear. No cells were seen in the Thoma

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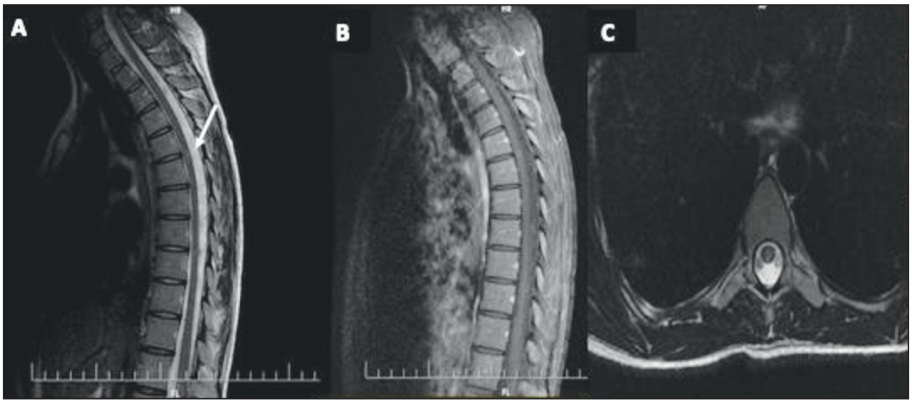

FIGURE 1: (A) Hyperintense lesion at T4 vertebral level in thoracic sagittal T2, axial (C), sagittal T2 **(A)** and faint contrast-enhanced T1**(B)** and axial sequence **(C).**

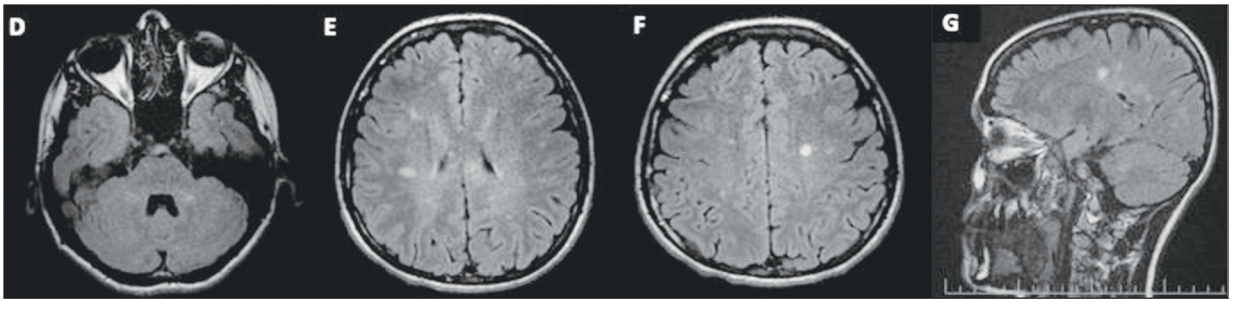

FIGURE 2: Hyperintense lesions at the pontocerebellar junction on the left **(D)**, periventricular **(E)** on the right, and cortical **(F)** on the left in the axial T2 FLAIR, perpendicular pericallosal lesions in the saggital T2 FLAIR **(G).**
FLAIR: Fluid-attenuated inversion recovery.

slide. In CSF biochemistry, protein was found to be 24 mg/dL, glucose 56 mg/dL (simultaneous blood glucose 78 mg/dL). There was no feauture in vasculitis panel, viral etiolgy, brucella agglutination, venereal disease research laboratory, angiotensin converting enzyme. The oligoclonal band examined in CSF was positive and immunoglobulin G index was 0.66. The patient received 1,000 mg intravenous methylprednisone for 10 days. In the visual evoked potential study, the P100 wave latency was 126 ms in the right eye, and the P100 wave latency was 125 in the left eye. There was no electrophysiological finding suggesting abnormality in medial and tibial somatosensory evoked potentials. Interferon Beta-1a treatment was started with the diagnosis of MS in the patient whose complaints almost completely resolved on the 10th day of the treatment. Informed consent was obtained from the patient for the case report.

DISCUSSION

The Lhermitte sign was first described as a subjective electrical sensation radiating down the spine and into the extremities of patients with cervical demyelinating lesions. Although it is often associated with MS previously; it can also be seen in cervical spondylosis, disc herniation, trauma, spinal cord malignancies, vitamin B12 deficiency, cisplatin toxicity and radiation myelotoxicity.[3] Even though this symptom is typically associated with cervical pathology, it has not been reported in the literature in patients with thoracic lesions, except for rare cases of tumor or deformity.[4] It occurs especially due to hyperexcitability as a result of irritation and inflammation of the posterior and lateral parts of the spinal cord.[5] Lesions are seen on cervical MRI in 92% of MS patients with LS. On the other hand, LS may not be detected in 53% of patients with MRI findings.[6] Although LS

has been described in detail in MS patients, it is not pathognomonic for the disease. However, there are studies showing that it can be an early symptom of MS in the absence of other signs and symptoms.[7] While it is seen in approximately one third of MS patients, its frequency is uncertain. In a study of 694 MS patients, it was indicated that approximately 16% of the patient cohort experienced LS, and this was strongly associated with neuroradiological and neurophysiological abnormalities.[8]

The electric sensation that occurs during trunk flexion originates from the thoracic spinal cord. Classical LS and LS that emerged with trunk flexion have different features. Senses evoked by neck flexion; it begins in the neck and spreads vertically down the spine and sometimes to the toes and arms. Sensations triggered by trunk flexion are circumferentially radiated around the thoracic or abdominal trunk, usually in T6-T8 dermatomes. This symptom may indicate that MS plaques are localized in the thoracic cord even when thoracic MRI is negative.[9]

The absence of abnormal sensations during cervical flexion in our patient and the appearance of the symptom with trunk flexion suggested that there may be any spinal pathology in the thoracolumbar region. Given the frequency of occurrence of this particular symptom, we present this case as a rare but possible example of spinal cord pathology in the thoracic region.

Historically, thoracic etiologies of LS are much less common than cervical ones. Therefore, it has the potential to be overlooked during clinical evaluation. Since LS is a well-recognized but less-explored symptom in MS patients, its presence in other spinal and radicular pathologies should especially be questioned during neurological examination.[10] Considering the findings confirmed by neuroradiological and neurophysiological examinations, it should not be forgotten that LS may occur in thoracic myelitis cases.

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Conflict of Interest

No conflicts of interest between the authors and / or family members of the scientific and medical committee members or members of the potential conflicts of interest, counseling, expertise, working conditions, share holding and similar situations in any firm.

Authorship Contributions

Idea/Concept: *Melis Doğanay Öcalan, Gülin Morkavuk;* ***Design:*** *Melis Doğanay Öcalan, Gülin Morkavuk;* ***Control/Supervision:*** *Gülin Morkavuk, Alev Leventoğlu;* ***Data Collection and/or Processing:*** *Melis Doğanay Öcalan, Gülin Morkavuk;* ***Analysis and/or Interpretation:*** *Melis Doğanay Öcalan, Gülin Morkavuk, Alev Leventoğlu;* ***Literature Review:*** *Melis Doğanay Öcalan;* ***Writing the Article:*** *Melis Doğanay Öcalan;* ***Critical Review:*** *Gülin Morkavuk, Alev Leventoğlu;* ***References and Fundings:*** *Melis Doğanay Öcalan;* ***Materials:*** *Melis Doğanay Öcalan, Gülin Morkavuk, Alev Leventoğlu.*

REFERENCES

1. Lhermitte J, Bollack J, Nicolas M. Les douleurs à type de décharge électrique consécutives à la flexion céphalique dans la sclérose en plaques: Un cas de forme sensitive de la sclérose multiple. Rev Neurol. 1924;39:56-62. [Crossref] [PubMed]
2. Oh J, Vidal-Jordana A, Montalban X. Multiple sclerosis: clinical aspects. Curr Opin Neurol. 2018;31(6):752-9. [Crossref] [PubMed]
3. Kempster PA, Rollinson RD. The Lhermitte phenomenon: variant forms and their significance. J Clin Neurosci. 2008;15(4):379-81. [Crossref] [PubMed]
4. Hills A, Al-Hakim M. Lhermitte sign as a presenting symptom of thoracic spinal pathology: a case study. Case Rep Neurol Med. 2015;2015:707362. [Crossref] [PubMed] [PMC]
5. Lhermitte J. Multiple sclerosis: Sensation of electrical discharge as early symptom. Arch Neurol 1929;22(1):5-8. [Crossref]
6. Gutrecht JA, Zamani AA, Slagado ED. Anatomic-radiologic basis of Lhermitte's sign in multiple sclerosis. Arch Neurol. 1993;50(8):849-51. [Crossref] [PubMed]
7. Kanchandani R, Howe JG. Lhermitte's sign in multiple sclerosis: a clinical survey and review of the literature. J Neurol Neurosurg Psychiatry. 1982;45(4):308-12. [Crossref] [PubMed] [PMC]
8. Beckmann Y, Özakbaş S, Bülbül NG, Kösehasanoğulları G, Seçil Y, Bulut O, et al. Reassessment of Lhermitte's sign in multiple sclerosis. Acta Neurol Belg. 2015;115(4):605-8. [Crossref] [PubMed]
9. Reder AT. Thoracic flexion provokes circumferential dysesthesia: A symptom of thoracic cord lesions in MS. Mult Scler. 2021;27(7):1008-13. [Crossref] [PubMed]
10. Khare S, Seth D. Lhermitte's sign: the current status. Ann Indian Acad Neurol. 2015;18(2):154-6. [Crossref] [PubMed] [PMC]