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Case Report

Dysphagia due to Lyme Disease: A Case Report

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Correspondence to
Eun Ji Lee
Department of Rehabilitation Medicine, Yeouido St. Mary's Hospital, The Catholic University of Korea, 10 63-ro, Yeongdeungpo-gu, Seoul 07345, Korea.
Email: eunjilv7@gmail.com

HIGHLIGHTS

- This rare case demonstrates dysphagia resulting from Lyme disease.
- The prevalence of Lyme disease is gradually increasing.
- Dysphagia should be considered a symptom of Lyme disease in affected patients.

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Dysphagia due to Lyme Disease: A Case Report

Pureum Kim,[1] **Seong Hoon Lim**,[1,2] **Jong In Lee**,[1] **Ho-geon Namgung**,[1] **Donghwan Kim**,[1] **Eun Ji Lee** [3]

[1]Department of Rehabilitation Medicine, Seoul St. Mary's Hospital, College of Medicine, The Catholic University of Korea, Seoul, Korea
[2]Institute for Basic Medical Science, Catholic Medical Center, The Catholic University of Korea, Seoul, Korea
[3]Department of Rehabilitation Medicine, Yeouido St. Mary's Hospital, The Catholic University of Korea, Seoul, Korea

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Eun Ji Lee
Department of Rehabilitation Medicine, Yeouido St. Mary's Hospital, The Catholic University of Korea, 10 63-ro, Yeongdeungpo-gu, Seoul 07345, Korea.
Email: eunjilv7@gmail.com

ORCID iDs
Pureum Kim
https://orcid.org/0009-0003-1820-6816
Seong Hoon Lim
https://orcid.org/0000-0002-5475-4153
Jong In Lee
https://orcid.org/0000-0002-8844-4807
Ho-geon Namgung
https://orcid.org/0009-0005-0211-4479
Donghwan Kim
https://orcid.org/0009-0005-0353-7414
Eun Ji Lee
https://orcid.org/0000-0002-3352-2417

ABSTRACT

Lyme disease is a multisystem infection that can affect the joints, heart, and nervous system when untreated. While it can present with cranial nerve palsy, dysphagia is rarely reported. This case highlights a rare instance of dysphagia in Lyme disease, typically known for neurological symptoms like facial nerve palsy. Despite the absence of erythema migrans or a documented tick bite, the patient's facial palsy, hearing loss, vocal cord paralysis, and dysphagia were attributed to Lyme disease. With the rising prevalence of Lyme disease, similar cases may increase, particularly in endemic regions of North America, Europe, and parts of Asia, emphasizing the need for early diagnosis and treatment in patients with unexplained dysphagia.

Keywords: Lyme Disease; Cranial Nerve Diseases; Dysphagia

INTRODUCTION

Lyme disease is an arthropod-borne infection caused by *Borrelia* species, predominantly *Borrelia burgdorferi* in the U.S. and *B. burgdorferi*, *Borrelia afzelii*, and *Borrelia garinii* in Asia and Europe [1,2]. Clinical manifestations range from localized skin reactions, such as erythema migrans, to systemic syndromes affecting joints, heart, and the central nervous system [3,4].

Neurological involvement, or neuroborreliosis, occurs in 15% of untreated cases, manifesting as meningitis, encephalitis, radiculitis, and cranial nerve deficits. Facial nerve palsy is the most frequent cranial nerve involvement, occurring in 80%–95% of these cases [5,6].

Dysphagia linked to Lyme disease is rare, with a few cases associated with lower motor neuron syndrome and brainstem encephalitis [7-9]. No cases have been reported where only cranial nerve palsy and dysphagia were present. We report a rare case of Lyme disease presenting with dysphagia and review the relevant literature.

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None.

Conflict of Interest
The authors have no potential conflicts of interest to disclose.

Author Contributions
Conceptualization: Lee EJ; Investigation: Nagmung HG; Project administration: Lim SH; Resources: Lee JI; Software: Kim D; Supervision: Lim SH; Validation: Lee EJ; Visualization: Kim D; Writing - original draft: Kim P; Writing - review & editing: Lee EJ.

CASE DESCRIPTION

A 63-year-old male presented with bilateral hearing impairment, left facial palsy, and pain. The patient had no significant past medical history, and tick exposure has not been confirmed. The patient was initially hospitalized due to chronic otitis media and a left ventilation tube was inserted along with antiviral treatment (acyclovir), but symptoms persisted. He was discharged but returned with worsening symptoms, including left vocal cord paralysis, requiring nasogastric tube placement for aspiration risk. Temporal bone computed tomography showed improvement in otitis media, but temporal bone magnetic resonance imaging (MRI) suggested pachymeningitis with thickened dura in the left middle fossa (**Fig. 1**).

With no improvement in symptoms 6 weeks after onset, additional laboratory tests were conducted for further evaluation and management, and the patient was readmitted to the hospital. Cerebrospinal fluid (CSF) analysis showed a pH of 7.0, white blood cell count of 5 cells/μL, red blood cell count of 560 cells/μL, glucose concentration of 80 mg/dL, and protein concentration of 32.5 mg/dL, findings that ruled out meningitis. Blood tests showed a white blood cell count of 9,400 cells/L, with segmented neutrophils at 84.05%, and a C-reactive protein level of 14.89 mg/dL, indicating inflammation. And blood tests for neurosarcoidosis, rheumatoid conditions, and infections like tuberculosis and Lyme disease were conducted. Broad-spectrum antibiotics and dexamethasone were initiated for pachymeningitis and chronic suppurative otitis media.

A videofluoroscopic swallowing study (VFSS) conducted six weeks after symptom onset showed aspiration on both puree and liquid textures, with a penetration-aspiration scale (PAS) score of 8, residual material in the pyriform sinus (**Fig. 2A**), decreased epiglottic movement, and upper esophageal sphincter dysfunction. Mylohyoid motor evoked potentials (MH-MEPs) were recorded using bipolar silver-silver chloride electrodes placed 2 cm lateral to the midline over each mylohyoid muscle, with a reference electrode on the mentum. Cortical stimulation was performed using a commercially available MagPro X100 (MagVenture, Lucernemarken, Denmark), employing the 70-mm figure-of-8 coil.

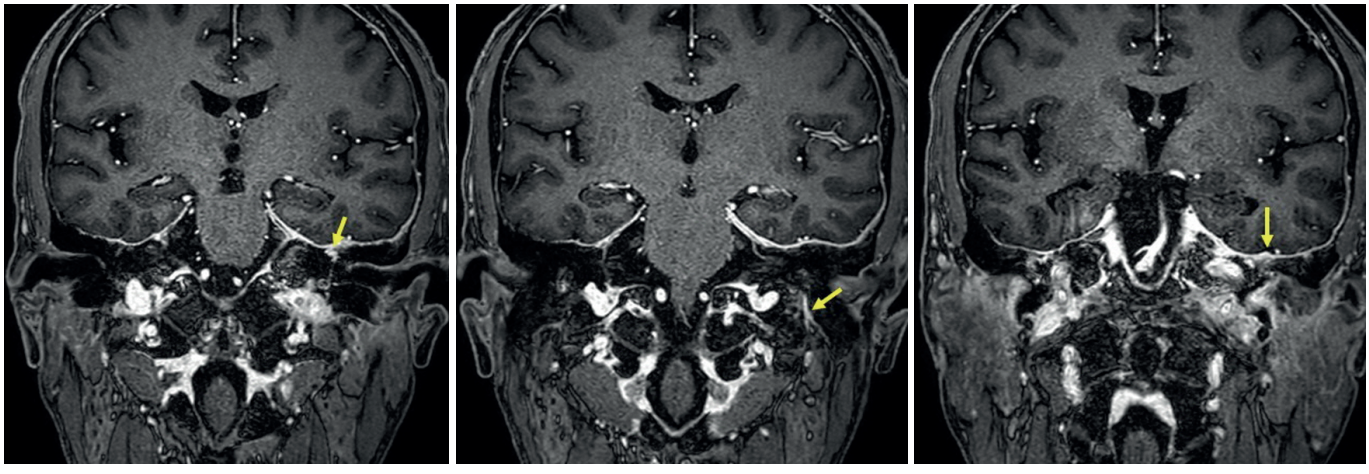

Fig. 1. Contrast-enhanced MRI showing left facial nerve and dural abnormalities.
Axial T1-weighted contrast-enhanced brain MRI was shown asymmetric enhancement left labyrinthine, geniculate ganglion and mastoid segment of left facial nerve and dural enhancement at left middle cranial fossa (arrows).
MRI, magnetic resonance imaging.

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Table 1. Motor evoked potential recordings from both mylohyoid muscles

Sites	Latency (millisecond)	Amplitude (millivolt)
Right mylohyoid	6.05	0.09
Left mylohyoid	No response	No response

The motor evoked potential study was performed by attaching electrodes to the mylohyoid muscle and stimulating the cortex, following the method described by Hamdy et al. [10]. No response was observed on the left side, while a slight delay in latency was observed on the right side. Normal reference values are latencies of 5 to 6 milliseconds and amplitudes of over 50 microvolts [11].

The optimal stimulation site ('hot spot') was located by positioning the coil tangentially over the mylohyoid motor cortex, approximately 2–4 cm anterior and 4–6 cm lateral to the vertex. Amplitude and latency of MH-MEPs were calculated from five sweeps at 120% of the resting motor threshold, with the dominant hemisphere identified as the side showing the highest amplitude. The Synergy electromyography/evoked potentials system (Medelec Co. Ltd., Bristol, UK) was used for MEP recordings, which were performed by a well-trained clinician. Motor evoked potential studies confirmed mylohyoid muscle dysfunction, more prominent on the left (**Table 1**) [10,11]. Repetitive transcranial magnetic stimulation (rTMS) was administered using a MagPro X100 (MagVenture), employing the 70-mm figure-of-8 coil. The patient received rTMS from a trained clinician, targeting the right mylohyoid muscle at 20 Hz for 5 seconds per train, with an intertrain interval of 55 seconds, 20 trains per session, totaling 1,000 pulses per day at 80% of the resting motor threshold, over a period of 10 days. However, a follow-up VFSS (about 8 weeks after symptom onset) showed persistent pharyngeal contraction weakness and aspiration (**Fig. 2B**).

Given the suspicion of immunoglobulin G (IgG) 4-mediated disease causing pachymeningitis, methotrexate was administered weekly, starting at 10 mg and increasing to 15 mg. Vocal cord paralysis showed no improvement, and injection laryngoplasty was performed.

Lyme disease serology later confirmed IgG 1:32 and IgM 1:16. The patient was treated with doxycycline 100 mg twice daily for 3 weeks. Two months after discharge, a VFSS showed reduced residue and improved aspiration with a PAS score of 3 in puree and liquid textures, allowing the patient to resume an oral diet (**Fig. 2C**).

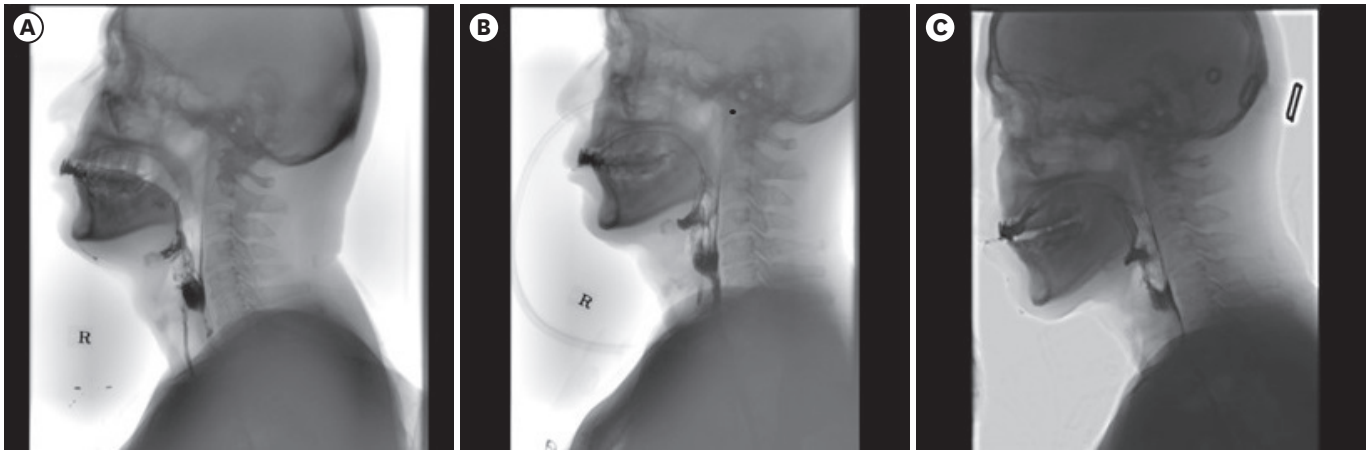

Fig. 2. .Sequential VFSS findings showing aspiration and recovery over time.
VFSS performed 6 weeks after symptom onset (A) and 8 weeks after symptom onset (post 10 days of repetitive transcranial magnetic stimulation) (B) showed aspiration on puree textures, with a PAS score of 8 and moderate to maximal residue at the vallecula and pyriform sinus. (C) Two months after discharge, a VFSS showed reduced residue and improved aspiration, with a PAS score of 3 in puree textures.
VFSS, videofluoroscopic swallowing study; PAS, penetration-aspiration scale.

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DISCUSSION

This case represents a rare occurrence of dysphagia associated with Lyme disease. Neurological manifestations of Lyme disease, termed neuroborreliosis, are the second most common clinical presentation after erythema migrans, but dysphagia has only been documented in rare cases [7-9]. While erythema migrans occurs in approximately 80% of Lyme disease cases, only 25% of patients recall a tick bite [12]. Therefore, the absence of a tick bite history or rash does not rule out the diagnosis.

The most common neurological manifestation of Lyme disease is cranial nerve involvement, with facial nerve palsy being the most frequent. However, other cranial nerves—including the oculomotor (III), trochlear (IV), trigeminal (V), hypoglossal (XII), olfactory (I), and vestibulocochlear (VIII) nerves—can also be affected. This broad range of involvement highlights the diverse neurological impact Lyme disease can have, extending beyond the typical presentation of facial nerve palsy to include a variety of cranial neuropathies, potentially leading to symptoms like visual disturbances, facial pain, impaired balance, and olfactory dysfunction [5].

In our patient, alongside facial palsy, there was hearing impairment, decreased gag reflex, left vocal cord palsy, and dysphagia, indicating involvement of the facial, vestibulocochlear, glossopharyngeal, and vagus nerves. Cranial nerve palsies can result from both viral and bacterial infections, but autoimmune mechanisms are also implicated in their development. Lyme disease, particularly in its neurological form (neuroborreliosis), is known to affect multiple cranial nerves, likely due to a combination of direct infection and immune-mediated inflammation [5,13,14]. This case reflects the complex neurological presentation of Lyme disease and highlights the importance of considering infectious and autoimmune processes in the differential diagnosis of cranial neuropathies. This aligns with the broader understanding that, beyond direct bacterial invasion, immune system dysregulation plays a critical role in the pathogenesis of neurological Lyme disease [15].

In our patient's case, dysphagia could be related to potential causes such as neurological symptoms of Lyme disease as well as localized muscle involvement like Lyme myositis. When comparing neuropathy and myopathy, it is significant that our patient exhibited sensory impairment, no autonomic symptoms, and minimal pain. Additionally, muscle enzymes, including serum creatine kinase and lactate dehydrogenase, were within the normal range, and there was no evidence of proximal muscle weakness. An electromyography could have been helpful for a more definitive assessment. However, due to the higher clinical probability of neuropathy, we did not consider further high-cost or invasive procedures, such as MRI or muscle biopsy.

Our patient received 10 sessions of rTMS. The effects of rTMS are generally known to promote the activation of swallowing-related muscles and neural plasticity, primarily in cases involving central nervous system lesions [16]. However, it has been suggested that rTMS may also facilitate functional recovery in peripheral nervous system lesions [17]. Given that it is a relatively safe and non-invasive treatment, we proceeded with patient agreement.

Given the increasing global incidence of Lyme disease and its neurological implications, Lyme disease should be considered in the differential diagnosis of cranial nerve palsies, particularly in regions where it is endemic. It has been reported that many cases of Lyme

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disease occur in the United States and Europe, and patients are also reported annually in countries such as Russia, China, and Japan [18-21]. The global incidence of Lyme disease is rising, notably in the United States, where cases increased from 35,198 in 2008 to 62,551 in 2022 [22]. In South Korea, although only 2 cases were reported in 2011, an estimated 45 cases occurred in 2023, potentially linked to climate change, such as global warming [23]. The upward trend in Lyme disease cases is expected to continue. This case emphasizes the importance of considering Lyme disease in patients with similar neurological symptoms, as early diagnosis and treatment are crucial for preventing long-term complications.

The patient presented without skin lesions suggestive of Lyme disease, and there was no confirmed history of tick exposure. Initial symptoms included facial palsy and hearing impairment, followed by progressive dysphagia. Prior to the diagnosis of Lyme disease, several interventions, including rTMS and laryngoplasty, were attempted for the severe and atypical dysphagia, without immediate effect. Despite these interventions, the patient's symptoms persisted, prompting further investigation into other potential etiologies. Lyme disease was subsequently confirmed through serologic testing, and treatment with doxycycline was initiated. The therapeutic effects of doxycycline in Lyme disease are typically observed over a period of several days to weeks [13]. In the present case, the patient demonstrated significant improvement in aspiration after two months of doxycycline therapy, as verified by VFSS, enabling the initiation of oral feeding. This suggests that the dramatic improvement in dysphagia may have been attributed to the treatment of Lyme disease, possibly in conjunction with delayed effects of prior interventions.

In conclusion, this case highlights the importance of considering Lyme disease in the differential diagnosis of unexplained dysphagia, even in the absence of typical symptoms, to ensure timely and appropriate treatment.

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