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Bilateral Vocal Cord Paralysis Requiring Tracheostomy Due to Neuroborreliosis

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Neuroborreliosis can cause multiple cranial and peripheral neuropathies; however, involvement of both recurrent laryngeal nerves is rare. We report the case of a 90-year-old man who presented with dysphonia and right upper and lower extremity weakness. His course was complicated by bilateral vocal cord paralysis and respiratory failure requiring tracheostomy. The diagnosis of borreliosis was made by detection of IgM and IgG antibodies against *Borrelia burgdorferi* on enzyme immunoassay and Western blot. The patient received IV ceftriaxone for 2 weeks, followed by complete recovery of motor and vocal function over 2 months. Our case is the third report of bilateral vocal cord paralysis in the literature, and the first one, to our knowledge, presenting with respiratory failure requiring an artificial airway. Physicians should be aware of this unusual complication of neuroborreliosis. CHEST 2014; 146(5):e153-e155

ABBREVIATIONS: BVCP = bilateral vocal cord paralysis

Bilateral vocal cord paralysis (BVCP) of the adult is an uncommon diagnosis triggered by iatrogenic trauma (surgical and intubation-related), malignancy, demyelinating disorders, amyotrophic lateral sclerosis, and cryptogenic causes.[1] Neuroborreliosis as a cause of this condition is very rare, with seven cases of unilateral paralysis and, to our knowledge, only two cases of BVCP reported in the literature. We describe a case of neuroborreliosis complicated with right hemiparesis, BCVP, and respiratory failure.

Case Report

A 90-year-old man, who was visiting central Massachusetts, presented with new-onset right-sided weakness that was preceded by headache, voice hoarseness, and malaise of 4 weeks duration. The patient denied dyspnea, neck stiffness, or photophobia. He had a history of diabetes mellitus type 2, hypertension, and hypothyroidism. He had visited a physician during the first week of having these complaints and underwent multiple tests, including negative IgM and IgG enzyme immunoassay for *Borrelia burgdorferi*. Presenting physical examination demonstrated mild right upper and lower extremity weakness and disorientation to time. There was no labored breathing, use of accessory respiratory muscles, or stridor. Initial laboratory study results showed leukocytosis of 12.5 cells $\times 10^9$/L with neutrophilia and no band forms. CT with angiogram of the

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head and neck was normal. Lumbar puncture was attempted twice without success. Empirical treatment of meningitis with IV ceftriaxone, vancomycin, and ampicillin was initiated.

Five hours after presentation, the patient developed stridor and severe tachypnea. Chest radiography showed small left lower lobe atelectasis, and arterial blood gas measurements demonstrated respiratory acidosis (pH, 7.20; $Paco_2$, 71 mm Hg; Pao_2, 125 mm Hg; bicarbonate, 28.4 mmol/L; Fio_2, 0.40). The patient was intubated emergently. Questionable supraglottic edema was seen during intubation, and therapy with IV dexamethasone was initiated. A head MRI was normal. Thyroid-stimulating hormone and antinuclear antibodies levels were normal. Serologies for *Treponema pallidum*, hepatitis B and hepatitis C virus, and blood cultures were negative. Enzyme immunoassay showed elevated levels of IgM and IgG specific to *B burgdorferi*, representing recent seroconversion. The presence of both immunoglobin fractions was confirmed by Western blot. The diagnosis of neuroborreliosis was made, and treatment was narrowed to IV ceftriaxone.

On the second day, the patient passed a spontaneous breathing trial. Videolaryngoscopy did not reveal evidence of supraglottic edema, and he was extubated. Immediately, the patient had stridor and was reintubated. Two days later, the patient again passed a breathing trial and was extubated with otolaryngology at the bedside. Videolaryngoscopy postextubation showed the vocal cords in the paramedian position, with slight movement of the left cord and a very narrow glottic space. The slight movement seen on the left was considered functionally negligible. He was reintubated and subsequently underwent percutaneous tracheostomy. No subglottic lesions were seen on bronchoscopy. The patient transitioned to spontaneous breathing and was discharged to complete 2 weeks of antibiotics. Follow-up as an outpatient demonstrated complete resolution of the right hemiparesis and BVCP after 2 months, with subsequent tracheal decannulation and recovery of his voice.

Discussion

Borreliosis affects many organs, including the nervous system. *B burgdorferi* seeds the meninges early in the infection and can cause multiple neurologic manifestations weeks to months after the initial tick bite. It can affect the peripheral nervous system as well. Meningitis, encephalopathy, mononeuropathy, and polyneuropathy have all been described as clinical presentations.[2] When cranial neuropathies occur, the facial nerve is most commonly affected. Disease of the vagus nerve and its branches seems to be exceptionally uncommon. The diagnosis of neuroborreliosis is made by the potential exposure to infected *Ixodes* ticks, clinical symptoms, and positive two-tier serology for *B burgdorferi*, with or without presence of specific antibodies on cerebrospinal fluid.[3]

Documented vocal cord paralysis in neuroborreliosis is exceptional, with seven cases of unilateral paralysis published.[4-9] These patients had voice hoarseness and responded favorably to antibiotic therapy with complete recovery. BVCP is even rarer, with only two reports.[10,11] One case describes a woman presenting with hypoxemic respiratory failure treated with nasal oxygen and observation.[10] Complete recovery of vocal function occurred after 6 months. The other case describes a man presenting with central hypoventilation caused by neuroborreliosis who required a tracheostomy for administration of positive pressure ventilation; 3 months later, he developed bilateral sixth nerve and vocal cord palsies.[11] His neurologic deficits resolved over a year.

Ours is the third case of bilateral vocal cord paralysis caused by neuroborreliosis that has been published, to our knowledge. The patient presented with precipitous stridor leading to respiratory failure. Once the airway obstruction was resolved, the patient transitioned quickly to spontaneous breathing, confirming the absence of central hypoventilation or diaphragmatic paralysis. He might have suffered from unilateral cord paralysis initially, which evolved into a bilateral form after 4 weeks. To our knowledge, this is the first case severe enough to require an artificial airway for the treatment of neuroborreliosis-related BVCP. The patient had a complete recovery, indicating that antibiotics are an effective treatment, even when the paralysis is severe.

Interestingly, a case-series survey of 266 patients with Lyme disease showed that 4.9% of them experienced dysphonia.[12] However, impairment of vocal cord function was not confirmed. This could suggest that vocal cord paralysis caused by borreliosis is more common than previously believed and potentially unreported. Thirty-six percent of surveyed physicians from the American Broncho-Esophagological Association considered serology for *B burgdorferi* as a useful test in the evaluation of vocal cord paralysis.[13] Nonetheless, there are no large case series of vocal cord paralysis caused by borreliosis in the literature. In conclusion, neuroborreliosis may present with life-threatening

BVCP with respiratory failure, including prolonged need for an artificial airway.

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