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Diabetic vagal mononeuropathy manifesting with isolated dysphagia

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Abstract

Vagal mononeuropathy is very rare. Diabetes mellitus is one of the causes of this rare disease condition. Here we report a 44-year-old woman who presented with an idiopathic vagal mononeuropathy and was finally diagnosed with diabetic vagal mononeuropathy. She presented with isolated dysphagia without hoarseness or other symptoms related with vagal dysfunction. Except for diabetes mellitus, no abnormalities were found by routine and specific checkups including brain imaging, gastroscopy, electromyography, and laryngoscopy. Finally, 12 days later, she abruptly developed hoarseness without other cranial nerve dysfunction.We suggest that her neurological symptoms originated from diabetes affecting the vagus nerve in isolation. Clinicians should pay attention to this association, especially when they encounter a patient with diabetes mellitus with sudden idiopathic dysphagia even without problems of vocalization.

INTRODUCTION

Vagal mononeuropathy is very rare and usually combined with other cranial neuropathies such as glossopharyngeal or accessory neuropathy.[1] Thus, purely isolated vagal mononeuropathy has been rarely reported. In addition, the majority of vagal mononeuropathy co-presents with changesof the voice (i.e. hoarseness).[2] Although most vagal mononeuropathy is idiopathic, diabetes mellitus (DM) and viral infection are considered possible etiologies.[2,3]

Recently, we saw a patient with an isolated vagal mononeuropathy, initially presenting only with dysphagia. The patient had no voice problem during the initial 11 days of disease course. Therefore, we suspected other etiologies of the swallowing problem such as from neuromuscular or esophageal origins. After the patient additionally developed hoarseness and was diagnosed with overt DM, we diagnosed diabetic vagal mononeuropathy.

CASE REPORT

A forty-four-year-old female came to our clinic because of dysphagia. One day prior, she abruptly noticed difficulty in swallowing both solids and liquids. At the time, she could not swallow any kinds of food at all. Neither voice change nor other autonomic symptoms existed at this time. She denied any previous medical problems, the family history was negative. However, she noted symptoms of fatigue, polydipsia, and polyuria for several months.

On neurological examination, cranial nerve function was normal. She showed normal bilateral gag reflexes with symmetric movements of soft plate, uvula and tongue. Other components of neurological examination, such as motor and sensory examination, cerebellar functions, and reflex, were also normal. In addition, there were no features suggestive of involvement of the parasympathetic autonomic nervous system, such as pupillary dysfunction, problems in bowel or sphincter, and cardiac arrhythmia.

Except fasting serum glucose level (191 mg/dL), she showed normal laboratory values, including complete blood count, liver function test, kidney function test, urine analysis, chest X-ray, and electrocardiogram. Serum level of acetylcholine receptor antibody was within normal limits. Serum anti-ganglioside antibody tests were negative. Both laryngeal electromyography (EMG) and repetitive nerve stimulation showed no abnormal findings. Magnetic resonance imaging of the brain, computerized tomography of the neck and cerebrospinal fluid analysis were also normal. Gastroscopy revealed no structural lesions of the

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esophagus and stomach. Video esophagographic swallowing study showed that some remnants of food stayed near the epiglottis with aspiration into the airway. Laryngoscopy revealed no abnormalities in valleculae and airways and identified normal movement of bilateral vocal cords and the pharyngeal wall. Nasogastric tube was inserted to prevent further aspiration of food.

To exclude DM and glucose intolerance state, we checked hemoglobin A1C (HgbA1c) and performed oral glucose tolerance test. Her HgbA1c level was 7.0 (%) and the result of oral glucose tolerance test was compatible with overt DM. Plasma glucose level was 242 mg/dL at 2 hours after a 75g glucose load.

On the 12th day of symptom onset, she abruptly developed hoarseness of speech. We thought that her dysphagia and hoarseness were originated from isolated vagal mononeuropathy. Although she complained of slight dizziness especially during standing, autonomic function testing showed no definite evidence of dysautonomia related with vagal lesion. Specifically, heart rate variability was normal obtained in resting state, deep breathing, standing, and Valsalva maneuver.

We diagnosed diabetic vagal mononeuropathy and prescribed oral hypoglycemic agents with blood glucose monitoring, and oral steroid agent (prednisolone 30mg per day). With control of DM, about 3 weeks after the initial symptom onset, voice and dysphagia started to improve. Symptoms completely disappeared 2 months after onset.

DISCUSSION

Our patient initially presented with isolated dysphagia for both liquid and solid foods. Therefore, we at first thought that the dysphagia was either from structural pathology of the esophagus or a neuromuscular disease. We excluded dysphagia of esophageal origin, because of negative structural test results of the esophagus. The differential diagnosis amongst neuromuscular origins is varied and difficult. Tests for myasthenia gravis, variants of Gullain-Barré syndrome as well as for central nervous system lesions were negative. The subsequent development of hoarseness, enabled us to suspect that dysphagia originated from a vagal mononeuropathy. Newly diagnosed DM was subsequently considered as the most likely cause.

Vagal neuropathy is usually reported as a component of multiple cranial neuropathies, especially co-involvement with anatomically adjacent glossopharyngeal or accessory nerves.[1] Theoretically, a patient with isolated vagal mononeuropathy can manifest with palatal and pharyngeal palsy, dysphagia, dysautonomia, vocal cord paralysis, and various combinations of these features.[1] Vagal mononeuropathy has been rarely described.[2,3] Of these reports, the majority of cases presented with hoarseness due to lesions of the recurrent or superior laryngeal nerve.[3,4] Thus, either hoarseness or change of the voice is considered as an essential feature of the vagal mononeuropathy.

Interestingly, our patient suggests that vagal mononeuropathy can present with isolated dysphagia and mimic various neuromuscular or esophageal diseases especially in the early stage. We could identify only two case series of isolated vagal mononeuropathy.[2,5] Berry *et al*.[2] reported a patients with vagal mononeuropathy presenting with voice problems and had various degrees of vocal cord EMG abnormalities. Our patient developed hoarseness at a later stage of disease and showed good outcome. The same study[2] suggested only two kinds of specific etiologies, DM (2 / 25) and viral origin related with upper respiratory infection (3/25). Other cases (20/25) were regarded as idiopathic. Amin *et al*.[5] proposed a separate entity, "post viral vagal neuropathy" with 5 idiopathic vagal mononeuropathy occurring after viral infection. All of their cases had phonation problems and only one had concomitant swallowing problems.

There is no evidence regarding the effect of steroids on diabetic vagal mononeuropathy or other types of diabetic neuropathies.[6] Therefore, the prescription of steroids to our patient can only be viewed as empirical for this rare clinical entity.

In conclusion, our case suggests that vagal mononeuropathy can present with isolated dysphagia. Clinicians should consider DM as one of the etiologies of vagal mononeuropathy when there is no other etiological explanation.

DISCLOSURE

Conflicts of interest: None

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