Abrupt Onset of Dysphonia Caused by a Huge Aortic Arch Aneurysm and Feasible Rehabilitation: A Case Report

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거대 대동맥궁 동맥류로 인한 급성 발성 이상 및 적용 가능한 재활: 증례보고

조준모 · 강시현 · 김돈규 · 서경묵 · 범재원[1]

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Abstract

Ortner's syndrome or cardiovocal syndrome refers to vocal cord paralysis caused by compression of the left recurrent laryngeal nerve by cardiovascular disorders. We present the case of an 88-year-old woman who complained of dysphonia after surgery under spinal anesthesia for an open reduction and internal fixation, following an intertrochanteric fracture of the femur. Laryngoscopy revealed left vocal cord immobility in the paramedian position, and a computed tomography scan revealed a 6.5-cm fusiform aneurysm in the aortic arch. Needle electromyography demonstrated abnormal spontaneous activities with reduced recruitment of the left thyroarytenoid muscle. After a month of active inpatient rehabilitation that consisted of balance and gait training with a rolling walker, she could walk using a walker under supervision. Thus, we first suggest a feasible rehabilitation strategy in geriatric patients with Ortner's syndrome confirmed by laryngeal electromyography.

Key Words

Aortic aneurysm, Dysphonia, Recurrent laryngeal nerve

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Introduction

Ortner's syndrome or cardiovocal syndrome refers to vocal cord paralysis caused by compression of the left recurrent laryngeal nerve by cardiovascular disorders such as thoracic aortic aneurysms. In a previously published report, a 77-year-old male suffered an aortic aneurysmal rupture one month after the onset of dysphonia, which required emergency surgery with aortic arch replacement, whereas another 85-year-old woman underwent elective aortic arch replacement.[1]

In several reports, the authors described cases of Ortner's syndrome presenting with vocal symptoms mainly in geriatric patients.[2-6] Meanwhile, we describe a geriatric case of Ortner's syndrome confirmed by laryngeal electromyography, and subsequently suggest a feasible rehabilitation strategy.

Case Report

An 88-year-old woman was hospitalized for rehabilitation after surgery for an open reduction and internal fixation following an intertrochanteric fracture of the right femur at another hospital. She had a history of hypertension, tuberculosis, pneumothorax, and osteoporosis. Common symptoms of aortic aneurysms, such as severe chest or back pain, cardiopulmonary symptoms, or dysphagia, were absent.

After surgery under spinal anesthesia, the patient complained of dysphonia, which did not improve for about 4 weeks until she was transferred to our hospital. Dysphonia may suggest paralysis of the vocal cords whose movements are innervated by the recurrent laryngeal nerve. Laryngoscopy revealed left vocal cord immobility in the paramedian position and there was no mass lesion (Fig. 1). A contrast-enhanced computed tomography (CT) scan of the neck and chest showed a 6.5-cm fusiform aneurysm with mural thrombi in the aortic arch, which was presumed to compress the left recurrent laryngeal nerve (Fig. 2A). Laryngeal electromyography revealed abundant positive sharp waves and fibrillation

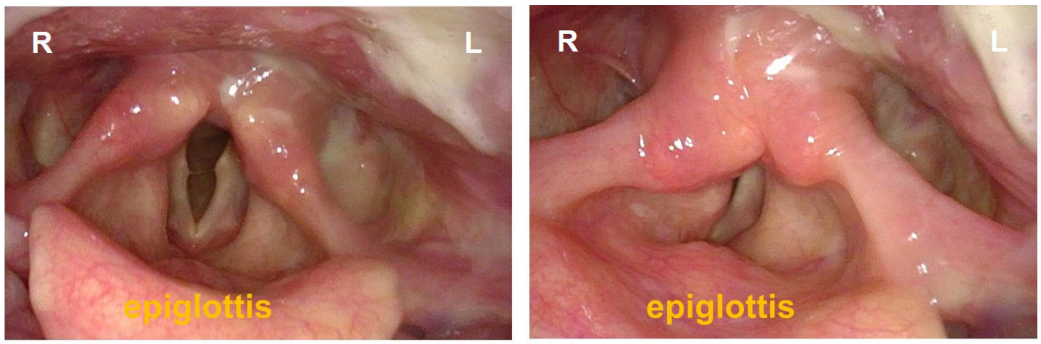

Fig. 1. Laryngoscopy revealed left vocal cord immobility in the paramedian position, and there was no mass lesion. (R: right side, L: left side)

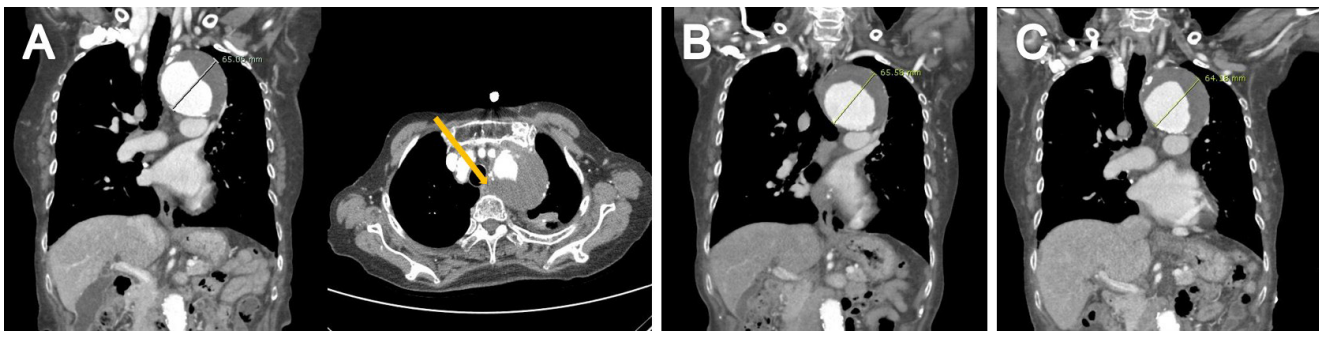

Fig. 2. A contrast-enhanced computed tomography (CT) scan. (A) Coronal and axial CT scan showed a 6.5cm saccular aneurysm with mural thrombi in the aortic arch which could compress the left recurrent laryngeal nerve (arrow) around the tracheoesophageal groove. (B) A follow-up CT a month later demonstrated no significant change in aneurysm size. (C) A follow-up CT 2.5 months after the first CT scan and 3 weeks after the second femur surgery revealed that the aneurysmal size was decreased little smaller.

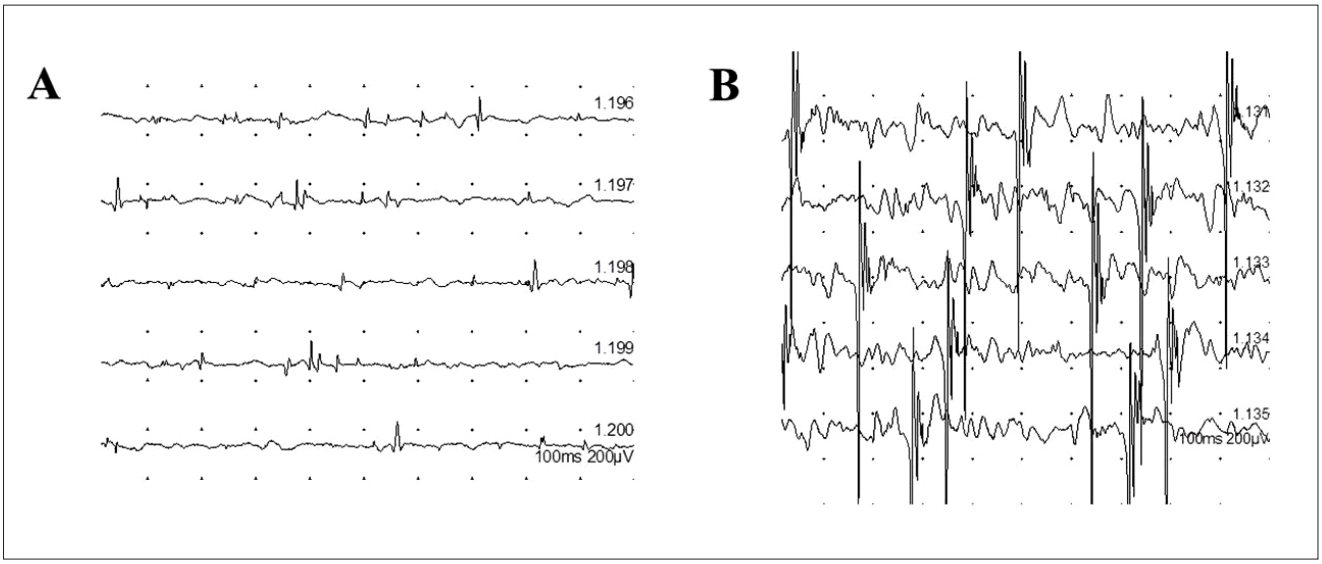

Fig. 3. Laryngeal electromyography revealed positive sharp waves and fibrillation potentials (A) with reduced recruitment (B) at the left thyroarytenoid muscle.

potentials with reduced recruitment of the left thyroarytenoid muscle (Fig. 3), which was consistent with partial axonal loss of the left recurrent laryngeal nerve. There was no abnormality on electromyography in the left cricothyroid muscle.

After consulting with a cardiologist and a cardiac surgeon, we considered performing a surgery for the high risk of rupture. Although the patient and caregivers were fully aware of the possible risks, they refused surgery due to the patient's old age. We decided to observe the patient's symptoms and implement a rehabilitation protocol. The heart rate was monitored to prevent tachycardia and reduce the risk of rupture during rehabilitation. The systolic blood pressure was set below 130 mmHg in the ward although an appropriate rate pressure product was not established from previous studies. Medications (losartan 50 mg, hydrochlorothiazide 12.5 mg, and isosorbide mononitrate 40 mg per day) were administered in consultation with a cardiologist. After a month of active inpatient rehabilitation that consisted of balance and gait training with a rolling walker, she could walk using a walker under supervision. A follow-up CT was performed, which showed no significant change in aneurysm size (Fig. 2B). The patient's dysphonia improved slightly. We warned the patient that the aortic artery aneurysm can suddenly rupture, which would require emergency heart surgery.

The patient sustained a fracture in the left intertrochanteric region following a fall at another hospital, and underwent an open reduction and internal fixation with an intramedullary nail. Another CT scan was performed three weeks after the second surgery, corresponding to two and a half months after the first CT scan. Although the aneurysm was slightly smaller (Fig. 2C) than the one detected earlier, dysphonia was slightly worse. However, the dysphonia became better as her general weakness was improved in the hospital course. Thus, we thought that dysphonia might be due to general weakness after surgery and not caused by aggravation of left recurrent laryngeal neuropathy. She underwent rehabilitation which consisted of balance and gait training. About one year after the first surgery, dysphonia was much improved.

Discussion

Aortic arch aneurysm is a rare cause of recurrent laryngeal nerve palsy, and only 5% of patients with thoracic aortic

aneurysm present dysphonia.[4] The prognosis of Ortner's syndrome, especially without surgical treatment, has not yet been investigated. However, this medical condition is life-threatening and many cases have been reported with aneurysmal rupture after the onset of dysphonia.[1-3] Thus, if a patient complains of dysphonia with symptoms such as severe chest or back pain, cardiopulmonary symptoms, or dysphagia that may accompany an aortic aneurysm, clinicians should be aware of the cardiovascular causes of dysphonia to avoid delays in diagnosis.

In our case, vocal cord palsy caused by endotracheal intubation could be ruled out because spinal anesthesia was performed during surgery. Laryngoscopy, CT scan or magnetic resonance imaging, and laryngeal electromyography are needed to diagnose Ortner's syndrome.[7] If there is no need for immediate surgical intervention and dysphonia is not aggravating, rehabilitation such as gait and balance training can be safely conducted. A previous article reported a poor recruitment rate and good compliance to the exercise intervention for older adults with small abdominal aortic aneurysm.[8] To the best of our knowledge, our case report is the first to report active inpatient rehabilitation in a patient with a large aortic arch aneurysm.

This study has several limitations. First, the severity of the dysphonia was not evaluated by a quantitative assessment tool but by the subjective symptom of the patient. A previous report suggested the use of auditory-perceptual evaluation such as the Grade, Roughness, Breathiness, Asthenia, Strain (GRBAS) scale, and measurement of the maximum phonation time.[9] Second, a follow-up test such as a CT scan or electromyography at the time of dysphonia improvement could not be conducted because the patient was lost to follow-up. Partial axonal loss of the recurrent laryngeal nerve would be recovered if the size of the aneurysm decreased. Quantitative voice measurement with long-term follow-up is also needed to evaluate the prognosis.

The definitive treatment for Ortner's syndrome is surgical removal of the aortic aneurysm. However, it is often not possible to perform surgical treatment in geriatric patients due to their underlying medical condition. Meanwhile, there are difficulties in applying intensive rehabilitation programs because a high systolic blood pressure in geriatric patients can increase the risk of aortic aneurysmal rupture. If dysphonia is aggravated, an urgent CT scan will be essential to evaluate the change in the size of the aortic aneurysm. Therefore, rehabilitation guideline, including continuous monitoring of the rate pressure product, is necessary to recommend and supervise exercise intensity. Feasible and effective rehabilitation will pave the way to improve the physical function of patients with aortic arch aneurysms.

REFERENCES

1. Ohki M. Thoracic saccular aortic aneurysm presenting with recurrent laryngeal nerve palsy prior to aneurysm rupture: A prodrome of thoracic aneurysm rupture? Case Rep Otolaryngol 2012;2012:367873
2. Lydakis C, Thalassinos E, Apostolakis S, Athousakis E, Michou E, Kontopoulou E. Hoarseness as imminent symptom of aortic aneurysm rupture (Ortner's syndrome). Int Angiol 2006;25:231-233
3. Nishimura Y, Okamura Y, Uchita S, Honda K. Abrupt rupture of an aortic arch aneurysm into the pulmonary artery. Eur J Cardiothorac Surg 2009;36:212-213
4. Teixido MT, Leonetti JP. Recurrent laryngeal nerve paralysis associated with thoracic aortic aneurysm. Otolaryngol Head Neck Surg 1990;102:140-144
5. Elzamzamy UA, Joharjy IA. Thoracic aortic aneurysm presenting only as vocal cord paralysis. Neurosciences (Riyadh) 2007;12:245-248
6. Zangirolami AC, Oliveira FV, Tepedino MS. Ortner's syndrome: Secondary laryngeal paralysis caused by a great thoracic aorta aneurysm. Int Arch Otorhinolaryngol 2015;19:180-182
7. Akbulut S, Inan R, Demir MG, Cakan D. Laryngeal electromyography is helpful for cardiovocal syndrome. Acta Medica (Hradec Kralove) 2016;59:29-32
8. Gunasekera RC, Moss J, Crank H, Mitchell PA, Nawaz S, Tew GA. Patient recruitment and experiences in a

randomised trial of supervised exercise training for individuals with abdominal aortic aneurysm. J Vasc Nurs 2014;32:4-9

9. Omori K. Diagnosis of voice disorders. JMAJ 2011:54:248-253