Case Report

Ewha Med J 2012;35(2):135-139
pISSN 2234-3180 / eISSN 2234-2591

EMJ
THE EWHA MEDICAL JOURNAL

Long Term Improvement of Dysphagia in Lateral Medullary Infarction: A Case Report

Jung Joong Yoon, Soo Jeong Han, Sung-Kyun Hwang[1], Hye-Kyung Jung[2]

Departments of Rehabilitation Medicine, [1]Neurosurgery, and [2]Internal Medicine, Ewha Womans University School of Medicine, Seoul, Korea

This report concerns a male patient suffered from refractory dysphagia after subarachnoid hemorrhage. A 49-year-old man admitted with severe headache followed by mental change. Imaging studies revealed that subarachnoid hemorrhage was located in basal cistern, and demonstrated ruptured vertebral dissecting aneurysm. After operation, the patient recovered well except severe dysphagia. Initial VFSS showed aspiration in fluid trial, penetration in semisolid bolus, and large amount of pharyngeal residue with poor relaxation of upper esophageal sphincter. For about 5 months, his symptom and several follow-up VFSS findings did not show marked improvement by various treatments. On magnetic resonance imaging for further evaluation of his brain lesion, an old infarction in right lateral side of medulla was found. He kept dysphagia rehabilitation more than one year, and his symptom improved to the level of oral feeding at last. **(Ewha Med J 2012;35(2):135-139)**

Key Words: Medullary infarction; Swallowing difficulty; Video fluoroscopic swallowing study

Introduction

Dysphagia is one of the perplexing symptoms of many neurologic diseases. The salient purposes of early diagnosis and rehabilitation of dysphagia is to avoid aspiration pneumonia, dehydration, malnutrition, and sepsis, which are potentially fatal complications [1]. The prognosis of dysphagia depends on the causative disease, and can vary from no improvement to the recovery of normal feeding [2].

During the normal swallowing process, the brainstem regulates signals from the cerebrum, to the oral cavity and pharyngolaryngeal region to allow the bolus to transit the swallowing pathway safely. The swallowing centers of cerebrum are in the sensorimotor cortex, the prefrontal cortex, and the insular region, and the right hemisphere is known to participate more than the left [3,4].

We report a case of refractory dysphagia in a patient who first admitted by subarachnoid hemorrhage (SAH). Dysphagia in this case was not nearly improved until about 5 months after onset despite traditional rehabilitation, functional electrical stimulation, and twice botulinum toxin injections, but the patient finally could eat per oral by continuous rehabilitative dysphagia therapy.

Case

A 49-year-old man with no specific medical history visited our emergency room with an aggravated headache of one week's duration and followed by loss of

Received: June 13, 2012, Accepted: July 5, 2012

Corresponding author: Soo Jeong Han, Department of Rehabilitation Medicine, Ewha Womans University School of Medicine, 1071 Anyangcheon-ro, Yangcheon-gu, Seoul 158-710, Korea
Tel: 82-2-2650-5035, Fax: 82-2-2650-5145
E-mail: ocrystal@ewha.ac.kr

consciousness. On neurologic exam, he showed normal pupil light reflex, intact motor and sensory functions, and normoreflexive deep tendon reflexes. Babinski's sign and ankle clonus were also negative on both sides. A computed tomography (CT) revealed that SAH was located in basal cistern (Fig. 1A) and also showed ventricular dilatation. Cerebral angiography on the day of admission demonstrated a dissecting aneurysm of the right vertebral artery, fusiform dilatation of aneurysm started from distal to the origin of the posterior inferior cerebellar artery (PICA). The aneurysm was treated by endovascular trapping of vertebral artery distal to PICA. After operation, brain CT revealed the posthemorrhagic hydrocephalus, which was resolved with permanent ventriculoperitoneal shunt procedure. Postoperatively, the Levin tube was inserted for tube feeding because of his hoarseness and dysphagia, and we needed to evaluate the characteristics of dysphagia.

During the first video fluoroscopic swallowing study (VFSS) performed at 40 days after SAH, only a small amount of fluid could pass the upper esophageal sphincter (UES) during the pharyngeal phase because the UES was not relaxed. Semi-solid and solid bolus could not pass the UES and remained as large amount retention at the vallecular and pyriform sinuses. Fluid aspiration and semi-solid penetration were also observed (Fig. 2A). He took 20 days of electrical stimulation therapy (EST) at the tongue base to stimulate mylohyoid, digastrics, geniohyoid and thyrohyoid muscles for 30 minutes a day, 5 days a week after the first VFSS. According to follow-up VFSS after EST, no improvement was observed (Fig. 2B). The patient then received traditional dysphagia rehabilitative therapy such as oromotor facilitation, pharyngeal swallowing exercise, education of compensatory strategies, and functional electrical stimulation (FES) with Vital-stim (Chattanooga Group, Austin, TX, USA) for 2 weeks. At 2nd follow-up VFSS with inducing UES relaxation by foley catheter-ballooning, we found no difference as compared with the previous study, except for the disappearance of semi-solid penetration. He maintained treatment by FES and dysphagia rehabilitation, and injecting 80 units of botulinum toxin at the posterior side (40 units) and each lateral side (20 units) of UES by endoscopy. After that, a fourth VFSS was performed on the 115th day of onset, but results were not improved. A fifth VFSS on the 129th day conducted after injecting the same amount of botulinum showed a minimal amount of fluid passed UES but that aspiration was accompanied. And a semi-solid bolus could not pass the UES which resulted in a large amount of retention (Fig. 2C). On the 141th day, percutaneous endoscopic gastrostomy was performed, and traditional dysphagia rehabilitation and EST three to four times a week were maintained. During a neurologic examination of the patient performed at this time, only dysarthria, a decline of gag and cough

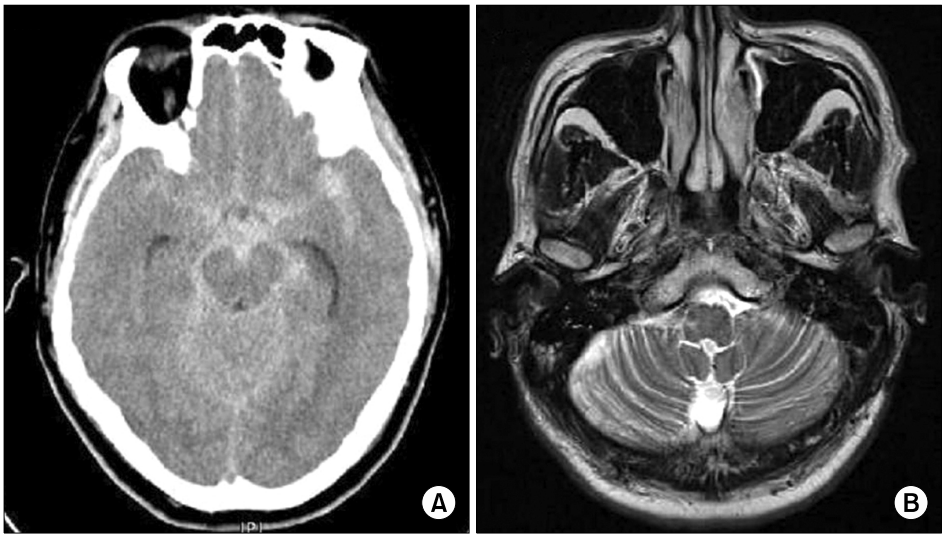

Fig. 1. Brain images of the patient. (A) Initial computed tomography image shows high density in basal cistern. (B) Axial T2-weighted magnetic resonance imaging shows high signal intensity in right lateral side of medulla.

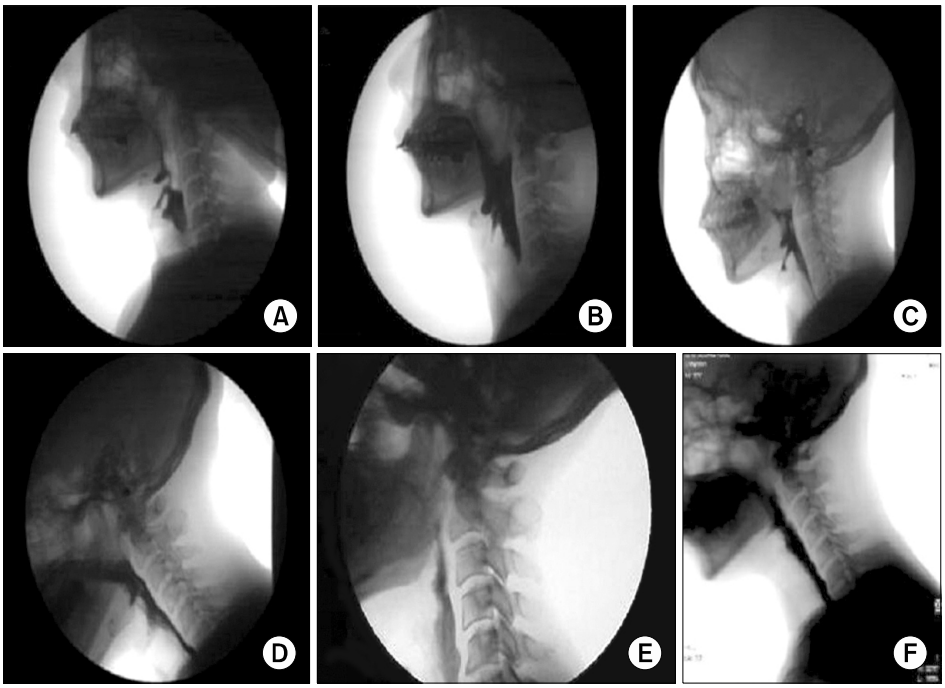

Fig. 2. Serial video fluoroscopic swallowing study images of the patient. (A) Large amount retention with penetration and cricopharyngeal hypertonicity is observed. (B) Bolus still cannot pass the upper esophageal sphincter (UES). (C) Only a few amount of fluid pass the UES and penetration is still shown. (D) Small amount of semi-solid is passing the UES. (E) The amount of UES-passing semi-solid is increased. (F) UES opening is still intact and there is no aspiration or penetration.

reflexes, minimal left nasolabial fold flattening and right uvular deviation were observed; deep tendon reflex was normoreflexive and the upper motor neuron sign was absent. On the left side, 10 to 30 percent sensory impairment to light touch and pain were observed, but no impairment was observed by manual motor testing, or pain and vibration sensory exams. On the 145th day of onset, brain magnetic resonance imaging (MRI) was performed to further evaluate his swallowing difficulty, and eventually we found an old infarction in the right lateral side of medulla (Fig. 1B). The patient continuously received traditional rehabilitative dysphagia therapy, and EST at digastrics muscle and thyrohyoid muscle. We first verified the improvement of the symptom at 8 months after onset. Some amount of semi-solid as well as fluid could pass UES without any aspiration or penetration (Fig. 2D). He kept rehabilitation therapy for dysphagia, and showed progressive improvement of the symptom. During the VFSS performed at 14 months after onset, some solid could pass UES and amount of vallecula and pyriform sinus retention decreased compared with few months ago (Fig. 2E). Final VFSS finding at 20 months after onset showed increased amount of UES-passing bolus without any aspiration or penetration (Fig. 2F).

Discussion

Dysphagia can be caused by various mechanisms, and may cause aspiration pneumonia and dehydration. It occurs frequently with stroke patients, especially who has lesion in the brainstem. Treatments of dysphagia involve sensory stimulation of the perioral area, oral and pharyngeal muscle strengthening, compensation by position correction, biofeedback therapy, electrical stimulation and operative treatment [5]. Of these, FES is often used and its effects in terms of reducing retentions and improving aspiration have been demonstrated on several studies [6]. In our case, the patient did not show any improvement after functional electrical stimulation for a month, which agrees with the findings of Shaw et al. [7] who found that FES had no effect on patients with severe dysphagia and poor cricopharyngeal relaxation.

Our patient was not able to especially relax the UES and this was not improved by several methods. The UES is controlled by the cricopharyngeal muscle and dysphagia occurs when this cricopharyngeal muscle be-

comes hypertonic. Cricopharyngeal hypertonicity is known to be caused by cerebrovascular disease, amyotrophic lateral sclerosis, pharyngeal diverticulum, but particularly after a stroke affecting the brainstem causing dysphagia (at a frequency of 50%) [8]. Treatments for cricopharyngeal hypertonicity are balloon dilatation, cricopharyngeal myotomy and pharyngeal plexus neurectomy but success rates are not satisfactory. Recently, botulinum toxin therapy has often been adopted. For example, Ahsan et al. [9] reported obtaining a good effect after injecting botulinum toxin into the cricopharyngeal muscle of a dysphagia patient who was unresponsive to traditional rehabilitative therapy. However, this patient did not gain an afferent effect after two 80 unit botulinum toxin injections separated by interval of 18 days.

In our patient, only SAH in both sylvian fissure, the basal cistern and the foramen magnum on brain CT was observed. Furthermore, he did not show any definite improvement for several months by continuous traditional dysphagia rehabilitative therapy, FES and botulinum toxin injection. Accordingly, we decided to perform brain MRI to search for another lesion on the 145th day, and at this time we found an old infarction at the right lateral side of the medulla. Vertebro-basilar artery aneurysm can present SAH and vascular infarction at the brainstem, but it is uncommon to occur simultaneously as in this case [10,11]. Dysphagia of this patient was caused mainly by poor relaxation of the cricopharyngeus muscle, which is innervated by the recurrent laryngeal nerve and pharyngeal plexus. We are inclined to believe that poor UES relaxation in this case was mediated by a medullary infarction because the recurrent laryngeal nerve originate from the vagus nerve and brain MRI showed that the medullary lesion included the region for motoneuron of vagus nerve, and impaired opening of UES is a common finding in patients with medullary infarction [12]. In addition, right medullary infarction could explain the dysarthria, impaired gag and cough reflexes, hypoesthesia of the left upper and lower limbs, minimal left nasolabial fold flattening and the right uvular deviation. According to the report of Prosiegel et al. [13], 30% of Wallenberg syndrome patients depend on tube feeding after functional swallowing therapy over 3 year period. Although there are many reports about effectiveness of rehabilitation, the evidence to support the medical effectiveness of common dysphagia treatment is limited [14]. Aydogdu et al. [15] reported in a study of lateral medullary infarction that the severity and long-term persistence of dysphagia are improved eventually by the role of remaining intact ipsilateral premotor neurons and the contralateral center. So we performed long-term dysphagia therapy to our patient, and he came to oral feeding at the time after one year from initial rehabilitation. The opening of the UES depends on the appropriate relaxation of the cricopharyngeal muscle and movement of the larynx during pharyngeal swallowing [12]. The total period of dysphagia rehabilitation of our patient include electrical stimulation therapy to enhance laryngeal movement, and it may be contributed to improvement of swallowing by laryngeal muscle strengthening, swallowing reflex promotion and reducing distance between hyoid bone and thyroid cartilage [16].

In the described case, the patient presented both SAH and medullary infarction. The only symptom of the patient was long lasting severe dysphagia caused by lateral medullary infarction. The effectiveness of swallowing treatment for dysphagia of lateral medullary infarction was unclear, but the patient's symptom was improved by long term dysphagia rehabilitative therapy. The diagnostic and therapeutic approach of this case may be helpful for appropriate management in the case of similar dysphagia patient.

References

1. Perlman AL. Dysphagia in stroke patients. *Semin Neurol* 1996;16:341-348.
2. Daniels SK, Brailey K, Priestly DH, Herrington LR, Weisberg LA, Foundas AL. Aspiration in patients with acute stroke. *Arch Phys Med Rehabil* 1998;79:14-19.
3. Hamdy S, Mikulis DJ, Crawley A, Xue S, Lau H, Henry S, et al. Cortical activation during human volitional swallowing: an event-related fMRI study. *Am J Physiol* 1999;277(1 Pt 1):G219-G225.
4. Zald DH, Pardo JV. The functional neuroanatomy of voluntary swallowing. *Ann Neurol* 1999;46:281-286.

5. Langmore SE, Miller RM. Behavioral treatment for adults with oropharyngeal dysphagia. *Arch Phys Med Rehabil* 1994;75:1154-1160.
6. Park YJ, Yang HS, Cha DY. Neuromuscular electrical stimulation therapy for patients with chronic dysphagia caused by stroke. *J Korean Acad Rehabil Med* 2007;31: 636-641.
7. Shaw GY, Sechtem PR, Searl J, Keller K, Rawi TA, Dowdy E. Transcutaneous neuromuscular electrical stimulation (VitalStim) curative therapy for severe dysphagia: myth or reality? *Ann Otol Rhinol Laryngol* 2007;116:36-44.
8. Lee CK, Kim JA. Pattern of post-stroke swallowing disorder according to the brain lesion. *J Korean Acad Rehabil Med* 2001;25:193-201.
9. Ahsan SF, Meleca RJ, Dworkin JP. Botulinum toxin injection of the cricopharyngeus muscle for the treatment of dysphagia. *Otolaryngol Head Neck Surg* 2000; 122:691-695.
10. Han TR, Kim JH, Kim HS, Paik NJ. Functional outcomes of subarachnoid hemorrhage and other stroke. *J Korean Acad Rehabil Med* 1992;16:139-146.
11. Murakami K, Takahashi N, Matsumura N, Umezawa K, Midorikawa H, Nishijima M. Vertebrobasilar artery dissection presenting with simultaneous subarachnoid hemorrhage and brain stem infarction: case report. *Surg Neurol* 2003;59:18-22.
12. Bian RX, Choi IS, Kim JH, Han JY, Lee SG. Impaired opening of the upper esophageal sphincter in patients with medullary infarctions. *Dysphagia* 2009;24:238-245.
13. Prosiegel M, Holing R, Heintze M, Wagner-Sonntag E, Wiseman K. Swallowing therapy: a prospective study on patients with neurogenic dysphagia due to unilateral paresis of the vagal nerve, Avellis' syndrome, Wallenberg's syndrome, posterior fossa tumours and cerebellar hemorrhage. *Acta Neurochir Suppl* 2005;93: 35-37.
14. Foley N, Teasell R, Salter K, Kruger E, Martino R. Dysphagia treatment post stroke: a systematic review of randomised controlled trials. *Age Ageing* 2008;37:258-264.
15. Aydogdu I, Ertekin C, Tarlaci S, Turman B, Kiylioglu N, Secil Y. Dysphagia in lateral medullary infarction (Wallenberg's syndrome): an acute disconnection syndrome in premotor neurons related to swallowing activity? *Stroke* 2001;32:2081-2087.
16. Park YJ, Yang HS, Cha DY. Neuromuscular electrical stimulation therapy for patients with chronic dysphagia caused by stroke. *J Korean Acad Rehabil Med* 2007;31: 636-641.