

Swallowing apraxia in a patient with recurrent ischemic strokes

A case report

Yeo Joon Yun, MD, Yong Jae Na, MD, Seung Hoon Han, MD, PhD*

Abstract

Rationale: Swallowing apraxia is defined as dysfunction in oral phase caused by the deficit in the coordination of tongue, lip, and chin movements, without motor weakness, sensory loss, and cognitive decline and has not been reported yet.

Patient concerns: A 69-year-old male with personal medical history of ischemic stroke about 10 years ago newly developed right striatocapular infarction. He had a problem in the oral phase of swallowing after recurrent ischemic strokes.

Diagnoses: He was diagnosed as swallowing apraxia via bed side examination and videofluoroscopic swallowing study.

Intervention: Videofluoroscopic swallowing study was done in this case.

Outcomes: Symptoms and findings of VFSS were not improved after 2 months treatment.

Lessons: This case implies that a clinician should be alert to swallowing apraxia as a possible cause when a patient with recurrent strokes complains of oral phase dysfunction of swallowing and considers proper diagnostic option such as videofluoroscopic swallowing study.

Abbreviations: BSE = bed side swallowing evaluation, MRI = magnetic resonance imaging, PEG = percutaneous endoscopic gastrostomy, VFSS = videofluoroscopic swallowing study.

Keywords: oral phase dysfunction, swallowing apraxia, videofluoroscopic swallowing study

1. Introduction

Dysphagia, or deglutition problem, is commonly manifested symptom in stroke survivors with unilateral or bilateral hemispheric and brain stem infarctions. Dysphagia has been identified as a risk factor for aspiration pneumonia, malnutrition, and dehydration. In case of brain stem infarction, laryngeal penetration and aspiration were observed because of functional deficit in pharyngeal phase.^[1] On the other hand, motor cortex

infarction often causes functional deficit in oral phase and it make difficulty in initiation of swallowing.^[1]

Swallowing apraxia is one of many causes that lead to dysphagia and the other one which similar to it is buccofacial apraxia. Swallowing apraxia is characterized by dysfunction in oral phase due to deficit in the coordination of tongue, lip, and chin movements without motor weakness, sensory loss, and cognitive decline.^[2] On the other hand, buccofacial apraxia is a movement disorder that involves orofacial structure in the absence of paresis. Swallowing apraxia has been observed before bolus transfer during the oral stage of swallowing and it has been characterized as a delay in initiation of bolus transfer regardless of lingual movement.^[1]

It is known that swallowing apraxia has been associated with strokes occurred in periventricular white matter and left anterior cortex.^[3,4] Many characteristics of swallowing apraxia are similar to other forms of apraxia such as limb apraxia, apraxia of speech, and buccofacial apraxia.^[1] However, unlike other forms of apraxia, swallowing apraxia has been reported to be unrelated to posterior parietal lobule.^[3] Authors experienced an interesting case with a problem in the oral phase of swallowing after recurrent ischemic strokes and report with a literature review.

2. Method

This is a case report and ethics committee or institutional review board approval was not obtained. We obtained informed consent about publishing a case report before patient discharge. However, it was not possible to gain additional informed consent for case details because the paper was written after discharge.

Editor: N/A.

Written informed consent was obtained for publication of this case report from the patient.

The Ethics Approval/Institutional Review Board (IRB) is not needed because this is not a case of new intervention is performed.

The authors have no funding and conflicts of interest to disclose.

Department of Rehabilitation Medicine, Hanyang University College of Medicine, Seoul, Republic of Korea.

* Correspondence: Seung Hoon Han, Department of Rehabilitation Medicine, Hanyang University Guri Hospital, 249-1 Gyomun, Guri, Kyunggi, 471-701, Republic of Korea (e-mail: pmrdr@hotmail.com).

Copyright © 2019 the Author(s). Published by Wolters Kluwer Health, Inc. This is an open access article distributed under the terms of the Creative Commons Attribution-Non Commercial-No Derivatives License 4.0 (CCBY-NC-ND), where it is permissible to download and share the work provided it is properly cited. The work cannot be changed in any way or used commercially without permission from the journal.

How to cite this article: Yun YJ, Na YJ, Han SH. Swallowing apraxia in a patient with recurrent ischemic strokes. *Medicine* 2019;98:39(e17056).

Received: 14 April 2019 / Received in final form: 9 July 2019 / Accepted: 12 August 2019

<http://dx.doi.org/10.1097/MD.00000000000017056>

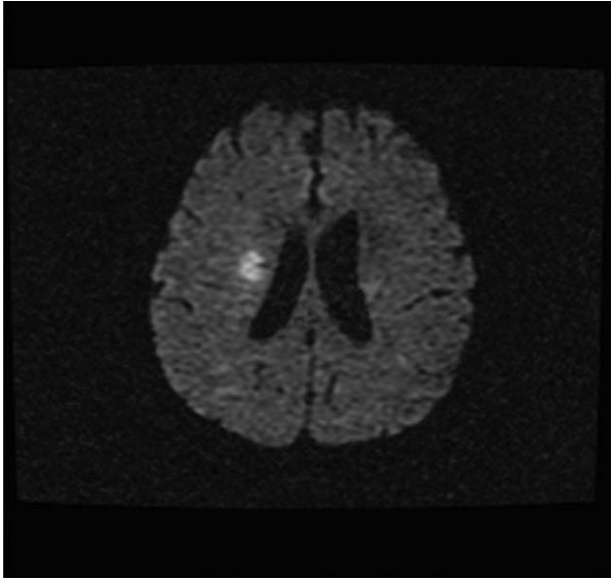


Figure 1. Brain magnetic resonance image taken at the second attack of recurrent strokes. Right striatocapular infarction is observed.

3. Case description

A 69-year-old male with personal medical history of ischemic stroke about ten years ago newly developed right striatocapular infarction (Fig. 1). At that time, he presented left-sided hemiparesis and mild dysarthria. However, functional deficit in activities of daily living and swallowing problem were not developed.

At 2 weeks after this second attack, he abruptly manifested right-sided hemiparesis, aggravated speech disturbance, and severe cough with dysphagia. Brain magnetic resonance imaging (MRI) taken immediately showed acute infarction at left internal capsule and fronto-parietal cortex and stenosis of left proximal internal carotid artery (Fig. 2). Therefore, he underwent stent placement at left carotid artery.

On physical examination performed after 3 days from the intervention, his mental status was alert and his cognition was good. He had no definite motor weakness or sensory deficit on face or all extremities. He could walk independently. However, he had motor-type aphasia and swallowing problem. He could mutter to himself although the authors could not understand it. After the third attack, he was fed by a nasogastric tube to prevent complications such as aspiration pneumonia and malnutrition.



Figure 2. Brain magnetic resonance images taken after the patient abruptly manifested aggravated speech disturbance and severe cough with dysphagia. Acute infarction at left internal capsule (A) and fronto-parietal cortex (B) and stenosis of left proximal internal carotid artery (C).

When his mouth was empty, voluntary movement of his tongue and lips appeared relatively good. However, when he tried to swallowing a food, his tongue and lip did not move well. When the examiner brought a spoon of yogurt close to his mouth, he opened his mouth and kept it in his mouth. However, he did not start to move his tongue and lips nor mix it, although his mouth was filled with yogurt. The drooling was also observed simultaneously.

The patient was able to move his lips and tongue on command when his mouth was empty. However, he failed to move them when food was in his mouth. At his initial examination of videofluoroscopic swallowing study (VFSS), oral phase dysfunction was observed. Tongue motions for searching and mixing a food had not been observed even after the barium was taken into his mouth. He could not squeeze it from the oral cavity to the pharynx. Even premature leakage was not observed. Therefore, next steps of examination evaluating the pharyngeal and esophageal phase of swallowing could not be performed.

Although he had been treated with oral motor facilitation and electrical stimulation with VitalStim for 2 months, his symptoms and findings of VFSS were not improved at all. The authors recommended percutaneous endoscopic gastrostomy (PEG) for him. However, he refused it and was discharged from the hospital.

4. Discussion

Authors report a patient who could not initiate a swallowing after recurrent ischemic strokes. The patient was able to move his lips and tongue on command when his mouth was empty. However, he failed to move them when food was in his mouth. The patient was unable to perform functional movements such as locking the food in the mouth by his lips or mixing it with saliva by his tongue or passing the food from the oral cavity to the pharynx by squeezing it. As a result, the patient failed to proceed any further swallowing process after the oral phase.

The lingual discoordination could also be considered as a possible reason in terms of oral phase delay due to inappropriate tongue movements in oral transfer.^[5] However, in this case, the patient showed grossly abnormal delays in oral phase. Therefore, it is more reasonable to diagnose the patient with swallowing apraxia rather than lingual discoordination, which shows brief periods of incoordination during oral phase of 2 to 3 seconds.^[2,5] Moreover, the movements of lips and tongue were close to normal without food in his mouth and any cognitive impairment and sensory loss were not observed. Limb and buccofacial apraxia which is different types of disorders of praxis system were not observed. Apraxia of speech could not be evaluated due to motor aphasia.

VFSS also known as modified barium swallow examination, captures sequential videoradiographic images of barium contrast-impregnated food and liquid as they are transported during oral cavity, pharyngeal cavity and esophagus in real time. As VFSS permits the visualization of bolus flow in relation to structural movement throughout the upper aerodigestive tract in real-time,^[7] it is considered to be the major instrument of choice to diagnose swallowing problem. Clinicians are able to observe the effects of various bolus volumes, bolus texture and compensatory strategies on swallowing physiology.^[7]

Contrast to VFSS, BSE also known as simple screening test, performed to inpatient at the bedside. First, examiner should check the oral cavity in order to define the normalcy of structure

and any obvious physical impairments. Then, swallowing can be evaluated by several methods such as dry swallowing, repetitive saliva swallowing test, water swallow test and swallowing provocation test. Many of these test are relatively easy to perform and are extremely useful in obtaining a rough picture of the swallowing condition.^[8]

Authors considered that the comparison between bedside swallowing evaluation (BSE) and VFSS results is playing a key role in differentiating swallowing apraxia from problems of oral phase due to other diseases. Because other disease such as buccofacial apraxia, speech apraxia can be tested using different stimuli and modalities whereas swallowing apraxia only be tested with swallowing.^[1] In this case, problems in the oral phase of swallowing were not expected because his tongue, lips, and chin movements were close to normal in BSE. However, the chance of being diagnosed with swallowing apraxia increased with severe delay in oral phase of swallowing and no lingual motion in VFSS. On the other hand, other reasons should be considered instead of swallowing apraxia if deficits in the coordination of tongue, lips, and chin movements were found in BSE and problems of oral phase were also detected in VFSS.

Some investigators believe that swallowing apraxia is not a true apraxia as deglutition is a semivegetative function and is not a 'learned skilled movement'.^[1] Also, Geschwind^[6] define apraxia as disorder in learned movement.^[1] Whether to classify swallowing apraxia as true apraxia is still a controversial issue. In addition, the study of the subtype of swallowing apraxia has not been reported yet in our knowledge. In our case, a patient did not show any tongue movement, but according to some reports, there are some cases where patients with normal tongue movement show symptoms of swallowing apraxia. Therefore, it is recommended to have further study on the subtype of swallowing apraxia.

Saito et al^[9] also described about swallowing hesitation. They classified swallowing hesitation into 3 types. First one is corticobulbar tract and swallowing hesitation. Second type is left primary motor cortex and "Rippling" swallowing hesitation. Last one is left frontal lesions responsible for "stasis" swallowing hesitation which is similar to our case. In their cases, patients also showed other types of apraxia such as ideomotor apraxia or buccofacial apraxia. Saito et al^[9] concluded "stasis" swallowing hesitation is due to broad lesion in the left hemisphere especially middle frontal gyrus. Similarly, in our case, patient showed broad left hemispheric lesion like Internal capsule and fronto-parietal cortex infarction plus ICA stenosis. In conclusion, both "stasis" swallowing hesitation and swallowing apraxia are due to broad lesion of left hemisphere but hesitation and apraxia are distinguished by a difference of main lesion.

Theoretically, if a patient has problems with oral phase, a compensatory swallowing technique like neck extension can be used for the patient to pass food smoothly from oral cavity to pharynx. However, if the intact function of pharyngeal phase is not ensured as this case, this technique should not be applied to the patient because the risk of asphyxia cannot be excluded. It is recommended to provide adequate nutrition to the patient via a feeding tube such as PEG tube if dysphagia is expected to be prolonged.

Because he had unique problem in the oral phase of swallowing process with no motor, sensory, and cognitive impairment, the authors conclude that the characteristics of his dysphagia was compatible with swallowing apraxia.

5. Conclusions

In this case, patient could not initiate a swallowing after recurrent ischemic stroke. He could move tongue or lips correctly when there is no food, but, in case of food inside in mouth, he failed to move tongue or lips. Authors could diagnose this symptom with swallowing apraxia via bed side examination and videofluoroscopic swallowing study. This case implies that a clinician should be alert to swallowing apraxia as a possible cause when a patient with recurrent strokes complains of oral phase dysfunction of swallowing and considers proper diagnostic option such as videofluoroscopic swallowing study.

Author contributions

Data curation: Yong Jae Na.

Writing – original draft: Yeo Joon Yun.

Writing – review & editing: Seung Hoon Han.

References

- [1] Daniels SK. Swallowing apraxia: a disorder of the Praxis system? *Dysphagia* 2000;15:159–66.
- [2] Robbins J, Levine RL, Maser A, et al. Swallowing after unilateral stroke of the cerebral cortex. *Arch Phys Med Rehabil* 1993;74:1295–300.
- [3] Robbins J, Levine RL. Swallowing after unilateral stroke of the cerebral cortex: preliminary experience. *Dysphagia* 1988;3:11–7.
- [4] Malandraki GA, Sutton BP, Perlman AL, et al. Neural activation of swallowing and swallowing-related tasks in healthy young adults: An attempt to separate the components of deglutition. *Hum brain mapp* 2009;30:3209–26.
- [5] Daniels SK, Brailey K, Foundas AL. Lingual discoordination and dysphagia following acute stroke: analyses of lesion localization. *Dysphagia* 1999;14:85–92.
- [6] Geschwind N. The apraxias; neural mechanisms of disorders of learned movement. *Am Sci* 1975;63:188–95.
- [7] Bonnie MH, Bronwyn J. The videofluorographic swallowing study. *PMC* 2008;19:769–85.
- [8] Satoshi H, Yasushi S. Screening tests in evaluating swallowing function. *JMAJ* 2011;54:31–4.
- [9] Tsukasa S, Keisuke H, Hajime N, et al. Clinical characteristics and lesions responsible for swallowing hesitation after acute cerebral infarction. *Dysphagia* 2016;31:567–73.