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Pharyngeal dystonia misdiagnosed as cricopharyngeal dysphagia treated with pharmacotherapy

Ho Eun Park[1*], Je-Sang Lee[1], Jin A Yoon[1], Ra-Yu Yun[1], Yong Beom Shin[1†]

Pusan National University Hospital, Department of Rehabilitation Medicine[1]

Introduction

The occurrence of dysphagia after stroke is very common. Although pharyngeal functions recover during stroke recovery, cricopharyngeal dysphagia (CPD) tends to remain. However the pathogenesis of focal pharyngeal dystonia caused by stroke is unclear and it hardly ever report. We report a rare case of dysphagia caused by focal pharyngeal dystonia after stroke that was successfully treated through pharmacotherapy. We also discuss a mechanism of a successful pharmacologic approach to central dysphagia.

Case report

A 43-year-old female was admitted to our rehabilitation clinic complaining excessive drooling and dysphagia. A year ago, the patient underwent coil embolization for SAH due to left vertebral artery aneurym rupture, and she was later confirmed to have lesions of the pons, medulla, and cerebellum due to left PICA infarction on brain magnetic resonance image. Gag reflex was diminished and left vocal cord palsy was confirmed, but the pronunciation and articulation were clear. At 3 months of onset, percutaneous endoscopic gastrostomy (PEG) was performed and there was no improvement after aggressive dysphagia therapy such as neuromuscular electrical stimulation. Activity of daily living was independent, but drooling, voice quality, and dysphagia resulted in significant quality of life deterioration. At 1 year and 2 months of onset, the videofluoroscopic swallowing study (VFSS) showed no progression from the pharyngeal to the esophageal phase. An otolaryngologist diagnosed cricopharyngeus muscle hypertonicity and underwent botulinum toxin injection under general anesthesia, but symptoms did not improve. After 3 months after botulinum toxin injection, on laryngoscopy, focal pharyngeal dystonia was confirmed rather than CPD. For management of dystonia, Trihexyphenidyl 2mg, clonazepam 0.5mg, and gabapentin 100mg three times a day were started to be considered as dysphagia caused by focal dystonia in pharyngeal muscle after brain lesion. Drooling showed a significant improvement in the first week of dosing, and at the 3rd weeks, the bolus transit to esophagus (Figure 1). The patient felt uncomfortable with PEG and removed the tube after orogastric tube training. Failure to take the medication for 3 days due to the loss of drug caused the drooling and food retention, and the washout period was not available due to fear of discontinuation of medication. There is no further improvement, but progress is being made without adverse effects (Figure 2).

Discussion

In patients with medulla or pons lesions, swallowing disorders have been reported to be particularly ineffective for botulinum toxin injection, suggesting that CP muscle itself is not the only problem. We suggest that pharyngeal dystonia should be considered in

addition to CPD in the case of dysphagia caused by dysinnervation of central nervous system. In addition to VFSS, esophageal manometry and electromyography should be performed to determine the treatment of central dysphagia.

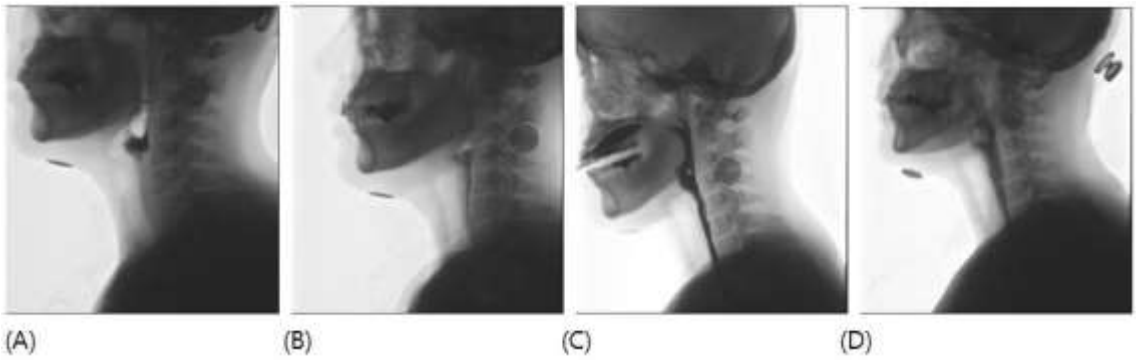

Fig 1. Videofluoroscopic swallowing study according to the time of administration of medication; (A) Before; (B) After 1 week; (C) After 3 weeks; (D) After 7 weeks

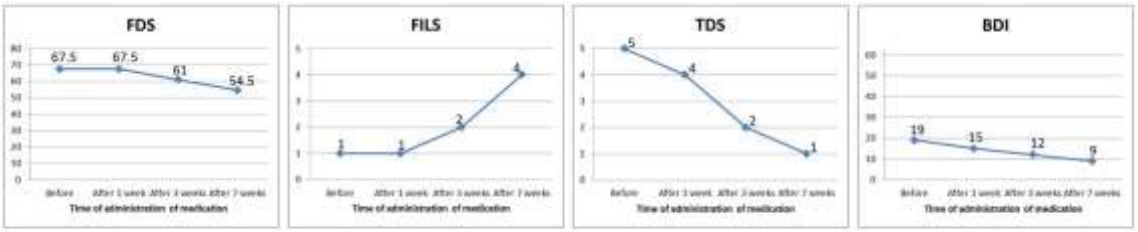

Fig 2. Changes in indices according to administration of medication; (A) Functional dysphagia scale (FDS); (B) Food intake level scale (FILS); (C) Teacher drooling scale (TDS); (D) Beck depression inventory (BDI)