Treatment for upper esophageal sphincter dysfunction in a patient with poststroke dysphagia

A case report

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Abstract
Rationale: Botulinum toxin injection is a widely used procedure for the treatment of the dysfunction of the upper esophageal sphincter (UES). Although the injection can be guided by ultrasound, electromyography, or computed tomography, such techniques cannot determine the exact extent of narrowed UES and ensure that the narrowed extent is fully covered by the treatment. This report describes a dual guiding technique with ultrasound and the balloon catheter in a patient with poststroke dysphagia to improve these weaknesses.

Patient concerns: The patient was admitted to a rehabilitation hospital 2 weeks postcerebral infarction.

Diagnoses: Clinical presentation of the patient included severe hemiplegia and dysphagia. The fiberoptic endoscopic evaluation of swallowing (FEES) revealed penetration/aspiration when swallowing 1 ml water and 1 ml yogurt and pooling in the postcricoid region.

Interventions: Balloon catheter dilatation procedures and Botulinum toxin injection were performed. We used a dual guiding technique with ultrasound and the balloon catheter to determine the whole segment of UES dysfunction by locating the lowest level of the impaired UES opening and to reduce difficulty in differentiating UES from adjacent tissues during Botulinum toxin injection.

Outcomes: No persistent progress was observed on the symptoms and volume of the balloon during dilatation. The patient showed quick responses after Botulinum toxin injection. The postinjection balloon catheter dilatation showed an increased maximum volume (preinjection, 5.5 ml vs. postinjection, 14 ml), and the patient was able to eat yogurt, congee, or semi-solid food 100–150 ml 4 weeks after the injection.

Lessons: The dual guiding method holds several advantages, suggesting that it may be considered as a promising choice in dealing with UES dysfunction.

Abbreviations: CT = computed tomography, EMG = electromyography, FEES = fiberoptic endoscopic evaluation of swallowing, UES = upper esophageal sphincter, VFS = videofluoroscopy.

Keywords: balloon catheter dilatation, botulinum toxin injection, dysphagia, ultrasound

1. Introduction
The dysfunction of the upper esophageal sphincter (UES) can significantly hinder the recovery of patients with dysphagia. Botulinum toxin injection is a widely used procedure for the treatment of UES dysfunction.

The UES is composed of cricopharyngeus, thyropharyngeus (part of inferior pharyngeal constrictor), and cervical esophagus, forming a high-pressure zone that extends 3–4 cm. Between the pharynx and the esophagus, UES generates a high-pressure zone with a length 3.0–4.5 cm measured by manometry. Nevertheless, a study showed that UES needs a surgical incision of 6 cm to achieve complete ablation of the UES pressure, indicating the involvement of a longer segment. Thus, an effective injection for UES dysfunction requires the determination of the exact extent of narrowed UES and ensuring that the narrowed extent is fully covered by the treatment. Although Botulinum toxin injection can be guided by using ultrasound, electromyography (EMG), or computed tomography (CT), such techniques cannot achieve these goals. We report here a 51-year-old patient with dysphagia after cerebral infarction, who received Botulinum toxin injection by dual guiding with ultrasound and the balloon catheter to successfully relieve UES dysfunction. To the best of our knowledge, this is the first report on performing Botulinum toxin injection by such a dual guiding procedure to precisely locate the extent of the narrowed UES. Written informed consent for publication of this case study (patient information and images) has been obtained from the legally authorized representative of this patient.

2. Case presentation
A 51-year-old male driver with dysphagia after cerebral infarction was admitted to our hospital. He had a cerebral
infarction (left radiological crown area) 2 years ago, and the sequela was a speech impairment. The patient could not eat liquid and solid food and was fed through a nasogastric tube. He had left hemiplegia (Brunnstrom stage 1 in upper and lower limbs) and was too weak to sit in an upright position and did not receive a barium swallow test (videofluoroscopy, VFS). The fiberoptic endoscopic evaluation of swallowing (FEES) revealed penetration/aspiration when swallowing 1 ml water and 1 ml yogurt and pooling in the postcricoid region (the portion of the hypopharynx that is located posteriorly to the larynx). Then, a balloon catheter dilatation procedure with ultrasound was used to monitor and confirm the dysfunction of the upper esophagus, with the lowest level at vertebrae C6 (Fig. 1A and Supplemental Video 1, http://links.lww.com/MD/C898). We used methods introduced in a study[8] to identify the anatomical landmark of C6 vertebrae with ultrasonography.

The dilatation was performed with a urethral catheter. The patient needs nebulization of budesonide if performing dilatation through the nose. Equipment needed are urethral catheterization pack containing a urethral catheter (14–16 Fr); 10 ml or 20 ml syringe; 10 ml or 20 ml normal saline; anesthetic spray, e.g., 1% lidocaine. The common procedures of the balloon dilatation with a Foley catheter are as follows:

1) Patient lying on back, or sitting;
2) Insert the catheter into the esophagus through the nose or mouth;
3) Inflate the balloon of the catheter by using 2.5–15 ml of normal saline with a 10 or 20 ml syringe and keep the plunger against the reflux of normal saline from the balloon;
4) Gently pull up the catheter until the balloon reaches the narrowed segment of the upper esophageal sphincter (UES) where the balloon is blocked;
5) Hold against the resistance 20–30 s;
6) Ask the patient to swallow his own saliva (dry swallow) one or more times until the inflated balloon passes through UES;
7) Pump liquid from the balloon immediately after passing UES;
8) Increase the volume of the balloon 0.5–1 ml during the next dilatation;
9) One session contains 5–10 dilatations;
10) Perform no more than one session per day.

The position of the balloon was monitored through ultrasound. The catheter was gently pulled up until the balloon reached the functionally narrowed segment of the UES and then was held against the resistance for 20–30 s. Afterwards, the patient was asked to swallow his own saliva one or more times until the inflated balloon passed through the UES. The volume of the balloon was increased by 0.5–1 ml during the next dilatation. One session contained 5–10 dilatations.

We performed the balloon catheter dilatation for seven sessions within 4 weeks. No persistent progress was observed on the symptoms and volume of the balloon during dilatation (Table 1). Under ultrasound guiding, Botulinum toxin A (Lanzhou Biological Products Institute, China) was injected into the UES (total of 60 units, two sites) after a written informed consent was acquired. The protocol was approved by the ethics

Table 1

| Changes in balloon volumes in the patient. |
|-------------------------------------------|
| Before injection | After injection |
| Number of days | Balloon volume | Number of days | Balloon volume |
|-----------------|----------------|----------------|----------------|
| 42              | 3.5 ml         | 20             | 5.5 ml         |
| 38              | 4.0 ml         | 25             | 7.0 ml         |
| 31              | 5.0 ml         | 33             | 10.0 ml        |
| 23              | 2.4 ml         | 61             | 10.0 ml        |
| 21              | 4.0 ml         | 63             | 10.0 ml        |
| 16              | 4.5 ml         | 81             | 14.0 ml        |
| 12              | 3.9 ml         |                |                |

Figure 1. Balloon catheter in the esophagus revealed by ultrasound. (A) Ultrasound showed that the dilated balloon of a Foley catheter was blocked by the UES. Arrowhead: The catheter inside the UES. Arrow: The inflated balloon. Sharp triangle: C5 or C6 vertebral body. SCM: Sternocleidomastoid. See Supplemental Digital Content 1 for a dynamic demonstration; the video showed that the inflated balloon (with 4 ml of normal saline) could not pass through the UES when the catheter and balloon were pulled up. (B) Ultrasound showed a Foley catheter (with the undilated balloon) in the esophagus. The shape of the catheter changed during ultrasound monitoring, and here the catheter exhibited a round shape. (C) The catheter showed the anterior wall and posterior wall, whereas the lateral walls of the catheter could not be seen. Arrow: the catheter. Arrowhead: the needle (diameter 0.51 mm, length 60 mm), see Supplemental Digital Content 2 for a clear display of the needle.
committee of the hospital (No. 20180104), in accordance with the Declaration of the World Medical Association. Prior to injection, we used the dilated balloon to locate the lowest level of UES dysfunction. During injection, the esophagus was easily identified through the ultrasound when the catheter (with the undilated balloon) was indwelled in the esophagus (Fig. 1B and C, and Supplemental Video 2, http://links.lww.com/MD/C899).

The patient required eating solid and liquid food 11 h after the injection. Five days after injection, a follow-up FEES revealed residues in the vallecular and pyriform sinuses and penetration but no aspiration after swallowing 3 ml water, and residues but no penetration and aspiration after swallowing 3 ml yogurt. The postinjection balloon catheter dilatation showed an increased maximum volume (preinjection, 5.5 ml vs. postinjection, 14 ml, Table 1), and the patient was able to eat yogurt, congee, or semi-solid food 100–150 ml 4 weeks after the injection. His swallowing function improved further, and no complications were observed after 6 months of follow-up.

3. Discussion
UES dysfunction treatment through balloon dilatation using an indwelling bladder catheter was introduced in a previous study.9 Such a dilatation procedure exerts radial and axial (sheering) forces to the UES with the distended balloon and a pull-up maneuver, similar to the endoscopic balloon catheter dilatation introduced in previous reports."10"

The accurate injection of Botulinum toxin into the narrowed segment of the UES is crucial for this treatment."11" In the present case, the balloon catheter guided by ultrasound showed two advantages. First, it could locate the exact segment of the narrowing as the target of injection. We used ultrasound to monitor the balloon dilatation with a Foley catheter, and thus provided a dynamic view of the movement of an inflated balloon within the UES lumen. The exact location of the UES narrowing was then clearly observed. Second, the UES could be clearly identified from adjacent tissues when a catheter was inside the esophagus during Botulinum toxin injection. The thickness of UES is only about 2.4–4.57 mm,"4,12" and the dual guiding procedure can ease the precise injection into the thin UES wall but more closely approximates a kidney shape,"13" this irregular shape also increases the difficulty of accurate injection into UES walls if no direct visualization methods are available.

It is also possible to inject iodine contrast medium into the balloon and monitor its movement by using X-ray fluoroscopy or CT. Compared with fluoroscopy and CT, the present dual guiding procedure has no ionizing radiation and can visualize blood vessels as well as nerve branches during injection.

The abnormal muscle tonic activity of UES can be identified with EMG to determine targets of Botulinum toxin injection."14–16" During EMG-guided injection, however, it is risky to insert EMG needle multiple times to look for the caudal part of the narrowed UES.

Compared with the endoscopic balloon, a Foley catheter is readily available. Taken together, the method introduced here holds several advantages over the aforementioned approaches. Thus, it can be considered as a promising choice in dealing with UES dysfunction.

The symptoms of the patient were relieved quickly, indicating that the precise localization of injection sites led to a rapid effect of Botulinum toxin on UES dysfunction. The improvement lasts 6 months without any side effects, suggesting the safety of the dual guiding procedure.

The present dual guiding procedure can be used to confirm the presence of UES dysfunction, when a patient cannot receive VFS due to various reasons and FEES reveals a possibility of UES dysfunction. During VFS, UES opening can be seen at the level of approximately 5 mm below the surface of the true vocal folds under normal conditions."17,18" UES dysfunction is identified by the sign “Cricopharyngeal bar” and “jet effect” as the obstructive appearance."18" and related inefficiency of bolus transit through UES."18" UES dysfunction can also be inferred with FEES. For example, residual materials in the postcricoid region after the “white out” phase of FEES."19" indicate possibility of impaired UES opening."20" and a resistance against the advanced endoscope through the UES is another sign of UES dysfunction."18"

For this patient, the blocked balloon at the level of C6 vertebral body was clearly shown with dynamic ultrasonography, demonstrating the caudal end of UES dysfunction. UES dysfunction may involve in the thyropharyngeus and cervical esophagus besides cricopharyngeus,"13" and a barium swallow test cannot determine the caudal part of the impaired UES opening.

In summary, to the best of our knowledge, this case report is the first described Botulinum toxin injection into the UES under dual guiding with ultrasound and a balloon catheter. The whole length of the narrowed UES can be determined by the guiding procedure, and the catheter inside the esophagus (with the undilated balloon) facilitates the identification of the UES during injection. The present dual guiding procedure can visualize blood vessels as well as nerve branches during injection without ionizing radiation. These advantages suggest that it may be considered as a promising choice in dealing with UES dysfunction.

Acknowledgments
This work was supported by the Beijing Natural Science Foundation (Grant No. 17L20019) and the National Key Technology Support Program of China (Grant Nos. 2015BAI06B02 and 2018YFC2001401).

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