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A Novel Approach to Severe Chronic Neurogenic Dysphagia Using Pharyngeal Sensory Electrical Stimulation

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Abstract: The treatment options for severe chronic neurogenic dysphagia are limited. A patient, after resection of medulla oblongata hemangioblastoma, who failed to respond to 7 mos of traditional dysphagia rehabilitation therapy, was treated with prolonged pharyngeal sensory electrical stimulation for 39 sessions over 57 days. For the first time, this case report showed improvement in hypopharyngeal peak pressure (9.1 *vs.* 90.8 mm Hg) using high-resolution manometry. Reductions in the penetration and aspiration scale, secretion, and residue of the vallecular and pyriform sinus were verified by videofluoroscopic swallowing study and flexible endoscopic evaluation of swallowing. The Functional Oral Intake Scale score increased from 1 to 6. No adverse event was observed. This case report presented a potential therapeutic protocol for severe chronic neurogenic dysphagia, which might be instructive for clinical practice.

Key Words: Pharyngeal Electrical Stimulation, Dysphagia, Hemangioblastoma, Hypopharyngeal Peak Pressure, Hypopharyngeal Contractility

(*Am J Phys Med Rehabil* 2023;102:e32–e35)

Hypopharyngeal weakness, insufficient laryngo-hyoid complex elevation, and upper esophageal sphincter (UES) dysfunction are great challenges in the rehabilitation of severe chronic neurogenic dysphagia. Severe chronic neurogenic dysphagia refers to those patients with severe dysphagia secondary to central nervous system disease for more than 3 mos. Severe chronic neurogenic dysphagia is associated with increased pneumonia, malnutrition, mortality, and cost.[1–3] Regarding hypopharyngeal weakness, no proven evidence for effective treatments exists yet.[4,5]

Pharyngeal electrical stimulation (PES) is a novel neurostimulation tool for dysphagia.[6–9] However, no studies investigated the effects of PES on pressures along the pharynx and upper esophagus, and no standardized recognized PES protocol for treating severe chronic neurogenic dysphagia exists. In addition, the localization of the PES electrodes is cumbersome. PES requires the catheter surface electrode to be accurately placed on the hypopharyngeal surface. The intubation approaches vary in different studies and include rough calculations according to the participant's height, inserting a length of 15 to 16 cm through the nose, intubating under gastrointestinal x-ray or laryngoscopy, or placement 3 cm above the UES after high-resolution manometry.[6–10]

Pharyngeal sensory electrical stimulation (PSES) is a modified neurostimulation tool. Compared with PES, PSES electrode placement is facilitated using electromyography (EMG). Regarding the stimulation parameters, the PES uses square wave, pulse width 0.2 msec, frequency 5 Hz, and 10 mins per day, whereas PSES uses mixed triangular and square wave, pulse width 10 msecs, frequency 5 Hz, and 10 mins per day. It was proposed that wide pulse duration might enhance the evoked sensory volley to the central nervous system[11–13] and reduce the fatigability of contractions.[14] Meanwhile, the triangular waveform and wide pulse duration could synergistically stimulate the denervated muscle.[15,16] The standard treatment period for PES is three consecutive days, but evidence is lacking for PSES. Here, PSES was applied to a severe chronic neurogenic dysphagia patient with hypopharyngeal weakness to investigate its safety and therapeutic effects.

This case conforms to all CARE guidelines and reports the required information accordingly (see CARE checklist, Supplemental Digital Content 1, http://links.lww.com/PHM/B853).

CASE PRESENTATION

This case involves a 31-yr-old male patient with a history of recurrent headaches and dizziness 4 yrs before admission on November 14, 2020. The magnetic resonance imaging showed hemangioblastoma in the dorsolateral medulla oblongata (Supplementary Figs. 1A and B, Supplemental Digital Content 3, http://links.lww.com/PHM/B855). The patient underwent hemangioblastoma removal on November 15, 2020. After surgery, repeated magnetic resonance imaging showed no intracranial hemorrhage, hydrocephalus, and abnormal tissue (Supplementary Fig. 1C, Supplemental Digital

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This research was funded by the National Natural Science Foundation of China (grant numbers 81972159 and 81672259), the Natural Science Foundation of Guangdong Province of China (grant numbers 2019A1515010388 and 2020A1515010881), and the Clinical Research Special Fund Project of the Third Affiliated Hospital of Sun Yat-sen University (Voyage Plan) (YHJH201909).

Ethics approval: The research proposal was approved by the ethics committee of The Third Affiliated Hospital of Sun Yat-sen University (no. [2021]02-259-01).

Informed consent: The patient signed a written informed consent for this study.

Financial disclosure statements have been obtained, and no conflicts of interest have been reported by the authors or by any individuals in control of the content of this article.

Supplemental digital content is available for this article. Direct URL citations appear in the printed text and are provided in the HTML and PDF versions of this article on the journal's Web site (www.ajpmr.com).

ISSN: 0894-9115
DOI: 10.1097/PHM.0000000000002116

Content 3, http://links.lww.com/PHM/B855). But the patient had persistent symptoms of dysphagia due to lingual sensorimotor deficits, cricopharyngeal dysfunction, and poor swallowing coordination. Besides that, the patient suffered from orthostatic hypotension, dystaxia, and hypodynamia. Even though the patient underwent routine swallowing rehabilitation, including transcutaneous neuromuscular electrical stimulation (20 min/day, 5 days/wk), cold thermal stimulation (20 min/day, 5 days/wk), tongue exercises (20 min/day, 5 days/wk), and catheter balloon dilation (8 times/day, 5 days/wk) for 7 mos, he still could not eat orally. Therefore, PSES was delivered as a supplemental treatment, whereas neuromuscular electrical stimulation and cold thermal stimulation were discontinued due to cost and unsatisfactory treatment results.

Before starting the PSES, high-resolution manometry (ManoScan 360, Ltd, Los Angeles, CA)[5,17] showed the hypopharyngeal peak pressure decreased (9.1 mm Hg) and the residual pressure of the UES relaxation increased (210.1 mm Hg) (Fig. 1, top row, white arrow). Flexible endoscopic evaluation of swallowing (ATMOS; MedizinTechnik GmbH & Co KG, Lenzkirch, Germany) showed right vocal cord paralysis, aspiration of saliva (Murray secretion severity rating scale = 4),[18] severe residue (Yale Pharyngeal Residue Severity Rating Scale),[19] aspiration (penetration and aspiration scale = 7[20]; Fig. 1, top row, red arrow), and an inability to eat orally (Functional Oral Intake Scale = 1). Videofluoroscopic swallowing study[21] (PLD8100C, gastrointestinal X-ray machine; Perlove, Zhuhai, China) showed that contrast agent could not enter the esophagus and aspiration, even when a catheter balloon placed in the pharynx aids swallowing (penetration and aspiration scale = 7; Modified Barium Swallowing Measurement Tool for Swallow Impairment[22] Pharyngoesophageal Segment Opening = 3; Fig. 1, top row, green arrow), and significantly reduced laryngohyoid complex elevation (Modified Barium Swallowing Measurement Tool for Swallow Impairment, Hyoid Motion = 2^{22}; Laryngeal Elevation = 2^{22}). (For more details, refer to Table 1 and Fig. 1, top row; see also Supplementary Materials, Supplemental Digital Content 2, http://links.lww.com/PHM/B854).

After receiving written informed consent from the patient, PSES was tried on May 21, 2021. The PSES device (ZIMMER, Neu-Ulm, Germany) contained a tube with two pairs of electrodes (the stimulus electrodes, placed at 8.8 cm apart from the reference electrodes, are also used to record the EMG signals), a control panel for regulating parameters, and a portable EMG device. The stimulus was a mixed waveform with a frequency of 5 Hz and a pulse width of 10 msecs (Supplementary Fig. 2A, Supplemental Digital Content 4, http://links.lww.com/PHM/B856). The first time placing the electrodes, a laryngoscope guided them into the pharyngeal cavity through the nose. The stimulus electrode ring was

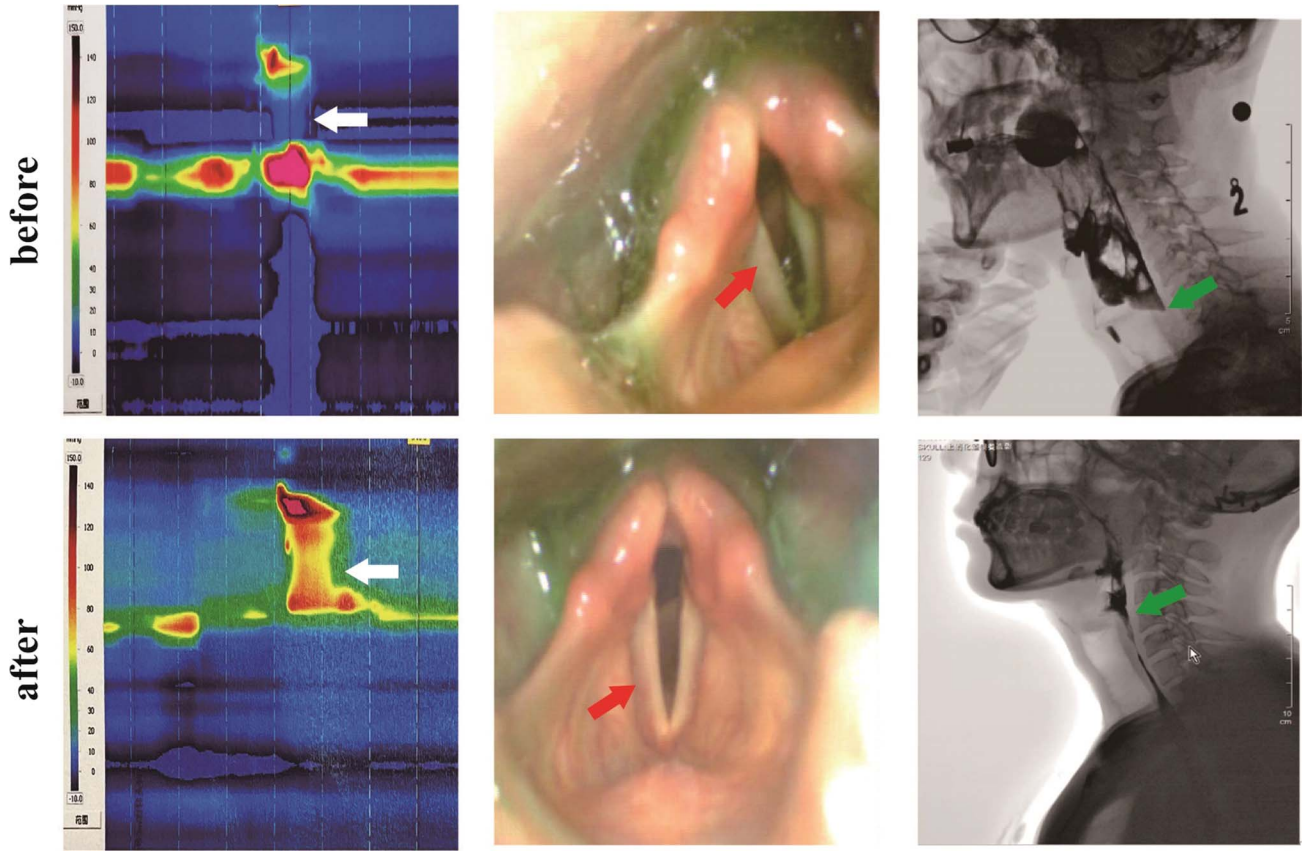

FIGURE 1. Auxiliary images before and after PSES. High-resolution manometry showed an increase in the hypopharyngeal peak pressure (white arrow) from 9.1 mm Hg to 90.8 mm Hg after PSES. Flexible endoscopic evaluation of swallowing showed improvement in aspiration (red arrow) before and after PSES. Videofluoroscopic swallowing study showed improvement in the pharyngoesophageal segment opening (green arrow) before and after PSES.

TABLE 1. Clinical evaluation before and after the PSES

Clinical Evaluation	Before	After
Functional oral intake scale	1	6
Penetration aspiration scale	7	1
Modified Barium Swallowing Measurement Tool for Swallow Impairment (pharyngoesophageal segment opening/hyoid motion/laryngeal elevation)	3/2/2	1/1/0
Murray secretion severity rating scale	4	2
Yale Pharyngeal Residue Severity Rating Scale (vallecular)	3	2
Yale Pharyngeal Residue Severity Rating Scale (left/right pyriform)	4/5	3/3
Hypopharyngeal peak pressure, mm Hg	9.1	90.8
Hypopharyngeal contraction duration, milliseconds	350	1106
Velopharyngeal peak pressure, mm Hg	118.9	157.7
Velopharyngeal contraction duration, milliseconds	816	1000
UES residue pressure, mm Hg	210.1	90.4
UES relaxation duration, milliseconds	Absent	70

placed within 1 cm of the piriform sinus, and the reference ring was placed in the upper esophagus (Supplementary Fig. 2B, Supplemental Digital Content 4, http://links.lww.com/PHM/B856). At this time, the pharyngeal EMG stably dropped below 20 μV, indicating good contact between the stimulation electrode and the pharyngeal mucosa (Supplementary Fig. 2C, Supplemental Digital Content 4, http://links.lww.com/PHM/B856). The depth of the stimulation electrode was 15 cm from the nostril to the piriform sinus. For subsequent electrode placement, the tube was inserted from the same nostril guided by the EMG without the laryngoscope. When the EMG signal decreased for the first time and stabilized below 20 μV, the location of the tube was fixed. When the patient could swallow, the EMG value increased above 20 μV during swallowing (Supplementary Fig. 2D, Supplemental Digital Content 4, http://links.lww.com/PHM/B856).

After the catheter position was fixed, the current intensity (CI) was detected. The CI was gradually increased from 0.5 mA. The perception threshold (PT) was the lowest CI at which the patient could feel the stimulation. The maximum tolerance threshold (MTT) was the CI at which the patient felt pain and discomfort and did not wish to increase further. The stimulus CI was calculated as PT + (0.75 × [MTT − PT]). The stimulation duration was 10 min/day. PSES was conducted every day, except for weekends, holidays, or when the patient felt uncomfortable. The endpoint of the PSES was set at 200 mL/time oral intake in consideration of nutrient requirements and potential swallowing function improvement. A qualified physiatrist performed PSES in the laryngoscopy examining room and disinfected it after each use. When the patient could orally take porridge 200 mL/time on July 16, 2021, PSES was discontinued. After the end of PSES, routine rehabilitations, including tongue exercises (20 min/day, 5 days/wk), catheter balloon dilation (8 times/day, 5 days/wk), and therapeutic feeding (thickened liquids with chin tuck, 30 min/day, 5 days/wk), were continued until hospital discharge to further ameliorate dysphagia caused by poor swallowing compliance and coordination, lingual weakness, and UES dysfunction. Ten minutes per day of PSES was applied in 39 sessions over 57 days. The stimulation intensities varied across sessions (Supplementary Fig. 2E, Supplemental Digital Content 4, http://links.lww.com/PHM/B856). The stimulus CI decreased from 20 to 4.4 ± 1.2 mA. No adverse reactions occurred.

The swallowing function was reevaluated after the last PSES. High-resolution manometry, videofluoroscopic swallowing study, and flexible endoscopic evaluation of swallowing were performed by the same licensed speech language pathologist and qualified physiatrist before and after treatment. The results indicated improved hypopharyngeal peak pressure (90.8 mm Hg) (Fig. 1, bottom row, white arrow), hypopharyngeal contraction duration, velopharyngeal peak pressure, velopharyngeal contraction duration, residual pressure of UES relaxation (90.4 mm Hg), and duration of relaxation, vocal cord mobility, secretion (Murray secretion severity rating scale = 2), residue, aspiration (penetration and aspiration scale = 1; Fig. 1, bottom row, red arrow), UES dysfunction (Modified Barium Swallowing Measurement Tool for Swallow Impairment, Pharyngoesophageal Segment Opening = 1; Fig. 1, bottom row, green arrow), and laryngohyoid complex elevation (Modified Barium Swallowing Measurement Tool for Swallow Impairment, Hyoid Motion = 1, Laryngeal Elevation = 0). (For more details refer to Table 1 and Fig. 1, bottom row).

Despite the cessation of PSES, the swallowing function continued to improve gradually with routine rehabilitation (Supplementary Fig. 3, Supplemental Digital Content 5, http://links.lww.com/PHM/B857). The patient could take most types of food (Functional Oral Intake Scale = 6) orally 120 days after discharge without adverse events. The timeframe of the case from the onset of illness until follow-up is presented in Supplementary Figure 4 (Supplemental Digital Content 6, http://links.lww.com/PHM/B858).

DISCUSSION

This case report showed the potential of PSES in the treatment of dysphagia for the first time. Hypopharyngeal peak pressure between tongue base and UES under high-resolution manometry was the best predictor of residue and aspiration.[2,3] Increased hypopharyngeal peak pressure contributed mostly to the improvements in residual, penetration, and aspiration scale and overall swallowing function. Surprisingly, UES dysfunction was also improved, which

might be associated with increased hypopharyngeal peak pressure that provides sufficient bolus driving force, and improved laryngo-hyoid complex elevation that provides sufficient up-forward movement.[2,23] Thus, PSES might be a novel treatment for patients with UES dysfunction and hypopharyngeal weakness.[4,24]

Moreover, despite the cessation of PSES, the swallowing function continued to improve gradually. The possible reason is that after PSES, the hypopharyngeal contractility was improved, allowing for oral intake. Through continuous oral intake therapy, the compliance and coordination of swallowing were reconstructed, and the neuroplasticity of the swallowing cortex was triggered. Thus, the swallowing function continued to improve and maintained.

Pharyngeal sensory electrical stimulation electrodes can be accurately located within 5 mins with a laryngoscope and EMG device. Compared with the stimulation intensity of PES, which often exceeds 10 mA in patients,[6–8] the stimulation intensity of PSES is relatively lower at 4.4 ± 1.2 mA, which may be due to the modified hybrid waveform and 10-msec pulse width of PSES. The gradually declined stimulation intensity might indicate restored pharyngeal sensation, which is associated with residue and aspiration, consistent with previous findings.[25] The underlying mechanism of PSES might be similar to that of PES, which can modulate the organization and behavior of the swallowing network.[8–10] Future studies are needed to investigate the mechanism of PSES.

The limitation is that this is a before-after case without randomized control. Nevertheless, the patient did not improve after 7 mos of routine therapy and began to recover gradually after PSES. Thus, the therapeutic effect might be mainly caused by the prolonged PSES, and self-recovery may contribute relatively minor. The PSES protocol in severe chronic hypopharyngeal weakness is likely longer than the standard protocol of PES. Further studies with large sample randomized controlled trials should be adopted to clarify the effect of PSES on severe chronic neurogenic dysphagia with poor hypopharyngeal contractility.

CONCLUSION

This case report explored the possible role of a novel therapeutic protocol for severe chronic neurogenic dysphagia with hypopharyngeal weakness, suggesting that PSES might be a potential therapeutic option for severe chronic neurogenic dysphagia with hypopharyngeal weakness. Further investigations with a large sample size and randomized control trials are needed to validate the effectiveness of PSES on severe dysphagic patients.

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