Movement Disorders
CLINICAL PRACTICE

Skill Training Resulted in Improved Swallowing in a Person with Multiple System Atrophy: An Endoscopy Study

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Dysphagia is a frequent symptom of multiple system atrophy (MSA). Current dysphagia management is compensatory, involving thickening liquids, postural maneuvers, or enteral feeding.[1] There are no studies of dysphagia rehabilitation in MSA.

We found that skill-training improved swallowing function in a 44-year-old man who developed progressive walking difficulty, urinary urgency, and orthostatic hypotension at age 41. No evidence of cognitive disturbance was found. Based on the clinical features of cerebellar ataxia, parkinsonism, and autonomic disturbance, a movement disorder specialist (SHK) diagnosed him with MSA cerebellar subtype. His scale for the assessment and rating for ataxia score was 15, unified Parkinson's disease scale part III score was 17, unified multiple system atrophy rating scale (UMSARS)-1 was 14, and UMSARS-2 was 20. Prominent cerebellar signs were wide-based gait with variable stance. Magnetic resonance imaging showed prominent pontocerebellar atrophy with hot cross bun sign. He developed dysphagia (choking on liquids) within one year of the onset of gait imbalance. He had no history of aspiration pneumonia.

Clinical swallowing evaluation revealed adequate bulbar muscle function. However, endoscopic evaluation revealed delayed swallowing, with the bolus passing over the epiglottis before swallowing onset (see Supporting Video 1). There was aspiration below the vocal folds and diffuse post-swallow pharyngeal residue, suggesting deficits in coordinating pharyngeal swallowing events and airway protection.

Biofeedback in strength and skill training (BiSSkiT) is a novel dysphagia treatment approach that has been studied previously in Parkinson's disease.[2] BiSSkiT targets motor control and swallowing precision by providing feedback regarding the timing and strength of muscle contractions.[2] This treatment differs from traditional swallowing rehabilitation, as it is task-specific, provides biofeedback, and calibrated increases in skill-requirement as proficiency improves.

The patient completed six one-hour BiSSkiT sessions over the course of six weeks. Surface electromyography provided biofeedback on a monitor regarding contractions of the suprahyoid muscles. The BiSSkiT software generated a target for the patient to "hit" during swallowing (see Supporting Video 1). The size of the target changed according to the patient's success, requiring the patient to monitor and modify swallowing muscle contractions. This has been hypothesized to activate parieto-premotor pathways, bypassing the basal ganglia and/or cerebellum and facilitating movement control.[2,3]

In addition to weekly therapy, the patient undertook daily home practice that included a smartphone video module with variably timed, audiovisual prompts to swallow saliva with a specific force (i.e., "swallow hard" or "swallow soft"), a skill targeted with BiSSkiT. As the goal was to facilitate increased cortical control over swallowing, we did not monitor for accuracy. The patient reported completing (on average) 60 cued swallows per week.

Swallowing-related outcomes were measured pre- and post-treatment. BiSSkiT performance improved post-treatment from 68% accuracy (target: 118 pixels) to 78% accuracy (target: 27 pixels), indicating improvements even as the task became more difficult. The patient had subjective improvement in swallowing symptoms such as decreased choking and coughing (per the SWAL-QOL). We also observed robust improvements in swallowing physiology (Table 1; see Supporting Video 1). Premature spillage and aspiration were eliminated, and post-swallow residue was reduced, allowing the patient to remain on an unrestricted diet.

This research is the first report of dysphagia rehabilitation in MSA-C. Although exploratory, results suggest potential benefit for others with MSA-C and warrants further investigation. By therapeutically targeting swallowing precision, the patient appears

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TABLE 1 Changes in functional swallowing parameters measured during flexible endoscopic evaluations of swallowing before and after six weeks of dysphagia rehabilitation

	Premature Spillage into the Pharynx	Vallecular Residue	Pyriform Sinus Residue	Epiglottic Residue	Laryngeal Vestibule Residue	Vocal Folds Residue	Subglottic Residue	PAS Score
Pre-treatment								
5cc thin liquid bolus	✓	✓	✓	✓	✓	✓	✓	8
Serial thin liquid bolus sips	✓	✓	✓	✓	✓	✓	✓	8
1 tsp mixed consistency (thin liquid/soft solid) bolus	✓	✓	✓	✓	✓	✓	-	8
Post-treatment								
5cc thin liquid bolus	-	✓	✓	✓	-	-	-	3
Serial thin liquid bolus sips	-	✓	✓	✓	✓	-	-	3
1 tsp mixed consistency (thin liquid/soft solid) bolus	-	-	-	-	-	-	-	1

Abbreviations: PAS, Penetration-Aspiration Scale.[5]

to have reorganized swallowing motor patterns to improve airway protection and swallowing efficiency, which translated into quality-of-life outcomes. The cerebellum governs the timing and coordination of swallowing-related muscle contractions[4] and enhancing the cortical control of swallowing might provide benefits in cases of cerebellar dysfunction. Further research is needed to understand the mechanisms for improvement in swallowing and refine treatment parameters.

Author Roles

1. Research Project: A. Conception, B. Organization, C. Execution; 2. Statistical Analysis: A. Design, B. Execution, C. Review and Critique; 3. Manuscript Preparation: A. Writing the First Draft, B. Review and Critique.

S.E.P.: 1A, 1B, 1C, 2A, 2B, 3A, 3B
J.S.S.: 1C, 3B
J.A.C.: 1C, 2A, 2B, 3B
S-H.K.: 3B
M.S.T.: 1A, 1B, 1C, 2A, 3A, 3B

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Supporting Information

Supporting information may be found in the online version of this article.

Supporting Video 1: Pre-treatment flexible endoscopic evaluation of swallowing demonstrating aspiration of liquids, example of BiSSkiT during therapy, and post-treatment evaluation of swallowing demonstrating improved function.