Skill-Based Dysphagia Training as an Intervention for Individuals with Huntington's Disease

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INTRODUCTION

Impaired swallowing (dysphagia) is highly prevalent in Huntington's disease (HD), associated with the leading cause of death aspiration pneumonia.[1,2] There is growing evidence to support rehabilitation for other motor and cognitive symptoms of HD [3-5]; however, sparse research has evaluated swallowing rehabilitation in this population.[6] This study evaluated an innovative skill-based swallowing training protocol in individuals with HD.

METHODS

+ Inclusion criteria: Adults with a clinical diagnosis of HD, identified symptoms of dysphagia as screened by the EAT-10 questionnaire (score of ≥ 3), and adequate cognition to participate in therapy.

+ A within-subject A-B-A design was utilised to include two-week blocks of no treatment pre-therapy as baseline and post-therapy for retention:

Day 0 Assessment 1	→ Lead in period	Day 14 Assessment 2	→ Treatment period	Day 28 Assessment 3	→ Retention period	Day 42 Assessment 4

+ Twelve participants completed 10 sessions of daily skill-based therapy over 2 weeks using Biofeedback in Strength and Skill Training software and surface electromyography hardware.

+ Swallowing outcomes were assessed using: the Timed Water Swallowing Test (TWST), Test of Masticating and Swallowing Solids (TOMASS), pharyngeal manometry, videofluoroscopy, ultrasound and the Swallowing Quality of Life Questionnaire (SWAL-QoL).

(Image used with consent)

RESULTS

+ **Feasibility of this therapy:** All participants (n = 12) completed the full protocol and improved in task performance. There were no adverse effects.

+ **Swallowing outcome measures:** Videofluoroscopic assessment of swallowing biomechanics demonstrated significant improvements in liquid bolus transit times, and upper oesophageal sphincter distension decreased post-therapy (Table 1). There were no significant treatment effects observed in the ultrasound, TWST or TOMASS data; however subjectively, five patients reported clinical improvements, including elimination of overt signs of aspiration post-therapy. Manometric measures of timing and amplitude moved closer to normative data, but no significant treatment effect was found.

+ **Swallowing related quality of life:** There were significant improvements in 3 out of 4 parameters of the SWAL-QoL questionnaire ($p < 0.05$). These improvements were maintained two weeks post-therapy.

+ **Skill retention:** There was no measurable deterioration in swallowing following daily therapy; however, with few treatment effects observed, we were unable to evaluate whether treatment effects on swallowing biomechanics are retained post-therapy cessation.

Table 1. Summary of videofluoroscopic outcomes pre- and post- therapy

Outcome measure	Bolus	Est. Change	p – value
Oral transit time (s)	Liquid	0.05	0.15
	Puree	-0.06	0.87
Pharyngeal transit time (s)	Liquid	0.05	0.26
	Puree	-0.11	0.44
Total transit time (s)	Liquid	0.09	**0.05***
	Puree	-0.18	0.51
Aryepiglottic closure (s)	Liquid	0.02	0.09
	Puree	-0.02	0.11
Upper oesophageal sphincter opening duration (s)	Liquid	0.04	0.32
	Puree	-0.09	0.09
Upper oesophageal sphincter distension (mm)	Liquid	0.18	0.13
	Puree	-0.58	**0.02***
Pharyngeal constriction ratio (mm²)	Liquid	0.02	0.36
	Puree	0.003	0.82
Hyoid excursion (mm)	Dry	0.33	0.83
	Liquid	2.25	0.28
	Puree	-1.01	0.36

CONCLUSIONS

+ This exploratory research demonstrated that this skill-based training is a feasible intervention for individuals with HD, with observed changes to task performance and evidence of improved patient perceptions of their swallowing impairment.

+ Although there was little evidence to suggest that this training was effective in altering swallowing biomechanics in these patients, the number of positive swallowing outcomes that did not reach statistical significance may suggest a lack of statistical power.

+ Further research is required to evaluate the effectiveness of skill-based approaches to change swallowing biomechanics using longer treatment protocols, larger samples of individuals with more severe swallowing dysfunction to fully explore the potential benefits of swallowing rehabilitation in this patient population.

[1]Schindler et al., (2020) *Scientific Reports*, 10, 1-8; [2]Heemskerk & Roos, (2012) *PLoS Currents*, 4; [3]Quinn et al., (2020) *Neurology*, 94, 942-956; [4]Fritz et al., (2017) *Journal of Huntington's Disease*, 6, 217-235; [5]Andrews et al., (2015) *Neurodegenerative Disease Management*, 5, 155-164 ; [6]Burnip et al., (2018) *Journal of Huntington's Disease*, 9, 1-12.