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Management of Dysphagia in Neurodegenerative Disease

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Abstract

Purpose The purpose of this review was to provide an overview of approaches to dysphagia management for patients with neurodegenerative disease.

Recent Findings Recent literature focused on dysphagia management for persons with neurodegenerative disease suggests implementation of a proactive, multidisciplinary approach to dysphagia evaluation and treatment tailored based on the specific neurodegenerative disease diagnosis. More research to determine the efficacy of interventions (compensatory and rehabilitative) has been conducted for certain disease groups (e.g., Parkinson's disease) than others (e.g., Alzheimer's disease). Generally, there is a need for more research focused on rehabilitative approaches to dysphagia treatment in neurodegenerative disease populations.

Summary This manuscript provides an overview of the research focused on dysphagia intervention approaches in Parkinson disease, amyotrophic lateral sclerosis, multiple sclerosis, Alzheimer's disease and related dementias, Huntington's disease, and myasthenia gravis. Future research studies should determine best practice for dysphagia management in understudied neurodegenerative disease populations in order to guide clinical decision-making.

Keywords Neurodegenerative · Dysphagia · Swallow · Deglutition · Management · Treatment

Introduction

Neurodegenerative diseases result from progressive degeneration or loss of neurons over time due a variety of causes. In 2017, it was estimated that neurodegenerative diseases affected 4.7 to 6.0 million individuals in the United States [1]. Aging is the primary risk factor for many neurodegenerative diseases, and as people develop longer life expectancies, it can be expected that the prevalence of neurodegenerative diseases will increase as well. Improved diagnostic paradigms that identify neurodegenerative diseases in the preclinical stage may also result in higher prevalence [2]. Development of dysphagia, or swallowing dysfunction, is a common symptom in individuals with neurodegenerative diseases. The goal for management of dysphagia in individuals with neurodegenerative disease is to prolong swallow function longer into disease progression in order to improve quality of life. Dysphagia etiologies, as well as hallmark dysphagia characteristics, can vary depending upon the patient's underlying neuropathology. Therefore, dysphagia management across neurodegenerative disease populations also varies and should be tailored to each patient. This article will provide an overview of oropharyngeal dysphagia evaluation and management across the most common neurodegenerative diseases including Parkinson disease (PD), amyotrophic lateral sclerosis (ALS), multiple sclerosis (MS), Alzheimer's disease and related dementias (ADRD), Huntington's disease (HD), and myasthenia gravis (MG).

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Evaluation of Dysphagia in Neurodegenerative Disease

By definition, neurodegenerative diseases are progressive in nature; consequently, it is imperative to understand the patient and care partner goals of care, expectations for disease

progression, and known implications the disease process on swallowing function prior to initiation of dysphagia management [3, 4]. This is of significance as dysphagia onset, profile, and severity are not homogenous amongst neurodegenerative disease diagnoses. Given this crucial prerequisite knowledge, initial screening for dysphagia may be conducted by nursing staff who are also often responsible for implementing safe swallowing strategies and recommendations from the speech language pathologist (SLP). Once a swallowing workup is initiated, the first step in identifying dysphagia is for the SLP to complete a thorough clinical swallow evaluation (CSE). The CSE may be conducted at bedside or a variety of other environments depending on medical setting and will provide the (SLP) clinician with data that will impact diagnosis and treatment planning, including medical history, cognitive status, cranial nerve function, and respiratory function [5]. If appropriate, the clinician will then proceed to present liquid and food boluses to the patient and will assess for safety and efficiency of the swallow during this process using validated clinical tools [6, 7]. Based on these findings, the clinician will then determine the necessity for an instrumental assessment; the videofluoroscopic swallow study (VFSS) and fiberoptic endoscopic evaluation of swallowing (FEES) are the most commonly used diagnostic tools that allow for visualization of the swallow and development of a plan of care [8, 9]. While the CSE is beneficial for the above reasons, it is typically insufficient to determine a plan of care for an individual with neurodegenerative disease given the high occurrence of silent (asensate) aspiration and the frequent underreporting of dysphagia previously observed in those with PD, ADRD, and ALS [10–13]. Whenever possible, an instrumental evaluation should be completed at the time of diagnosis (to obtain a baseline for swallowing function) and then at regular intervals across disease progression.

Supplemental instrumentation may provide additional information regarding swallowing physiology or swallowing related functions such as lingual strength or cough/respiratory function. High-resolution pharyngeal manometry (HRPM), or intranasal insertion of a narrow catheter lined with pressure sensors, may be used to further characterize the amplitude, duration, and coordination of muscular contraction during swallowing [14–16]. Measures of tongue strength and endurance are of value given evidence for a decline in physiological reserve within the muscles of the tongue and the crucial role of lingual musculature in swallowing [17, 18]. Finally, respiratory measures such as peak airflow, peak cough airflow, maximum expiratory pressure, and maximum inspiratory pressure may be obtained given the frequency of sensorimotor cough dysfunction amongst neurodegenerative diagnoses and the significant role respiratory function plays in protecting the airway [19–22].

It is also important for the evaluating clinician to incorporate the use of a patient-reported outcome measures (PROMs) to provide insight into the patient's (or care partner's) perception of swallowing function and the impact of dysphagia on quality of life. Selection of a specific PROM will depend on the patient's neurodegenerative diagnosis, as some PROMS are diagnosis-specific, as well as the goals for PROM use [23]. While the patient's perspective of their own swallow function is important to assess, questionnaires and patient interviews may not be a reliable option to screen or evaluate for dysphagia given underreporting of dysphagia [24••]. In addition to the impact of swallowing on patient quality of life, there is a growing body of evidence revealing the significant burden of dysphagia on the care partners of patients with neurodegenerative disease as well as other patient populations [25]. As such, evaluation of caregiver burden is now an additional use of PROMs that can be incorporated into dysphagia assessment [26].

In summary, the CSE, instrumental swallowing assessments, supplemental instrumentation, and PROMS will provide the clinician the opportunity to comprehensively assess swallowing-related functions, swallowing physiology, and the impact of swallowing on quality of life in persons with neurodegenerative disease. These findings should then be integrated with other factors, such as overall health status, cognitive functioning, disease prognosis, and patient/family desires, to determine risk of dysphagia-associated morbidities and to guide dysphagia management.

Overview of Dysphagia Management Approaches in Neurodegenerative Disease

Intervention options for patients with neurodegenerative disease will vary depending on diagnosis and these other aforementioned factors but can be expected to involve some or all of the following: diet modification, surgical or medical interventions, behavioral interventions, and/or alternative means of nutrition. These various approaches will be described in more detail specific to each primary neurodegenerative disease condition.

Proactive Approach to Dysphagia Care

Currently, the most common approach to dysphagia management is reactive in nature, meaning interventions are implemented only after dysphagia is identified. Initial referrals for SLP involvement in a patient's case often occur following a hospitalization during which a patient is suspected of or diagnosed with dysphagia. A shift in the paradigm of care of persons with neurodegenerative disease to proactive dysphagia management entails SLP clinician involvement at the time of disease diagnosis has been previously proposed [27]. This proactive approach allows for detection of dysphagia early, which allows for

implementation of intervention approaches that build functional physiologic reserve across systems contributing to healthy swallowing function and takes advantage of the potential to improve swallowing function and maintain it longer into disease progression. The patient is also able to actively engage in their disease process which can relieve some of the care partner burden by including the patient in treatment decisions and advanced care planning specific to feeding and swallowing (see Table 1).

Oral Care

Another important factor to consider across neurodegenerative populations is oral health. Poor oral health is a predictor of pneumonia development and salivary production, an important factor in maintaining optimal oral health, declines in those with neurodegenerative diseases [28]. Oral care is important to incorporate into a patient's plan of care to prevent poor oral health and reduce risk of pneumonia development. Good oral care includes (but is not limited to) regular dental visits, regular teeth and tongue brushing with fluoride toothpaste, regular flossing, removal of dentures when sleeping, use of mouthwash if needed (e.g., xerostomia), and removal of excess mucous secretions. It is important to note for those patients unable to tolerate an oral diet that it is critical to continue with good oral care (keeping the oral cavity clean and moist) even if nothing is being taken by mouth as secretions may still be aspirated. Any clinician involved in the patient's care can play an important role in ensuring appropriate referrals to dental providers take place to support this.

Multidisciplinary Team

It is also important to note that comprehensive multidisciplinary teams are imperative when making decisions regarding the patient's plan of care [29, 30]. Dysphagia management in patients with neurodegenerative disease is multifactorial and requires expertise from different fields for specialized treatment. It has been shown that a multidisciplinary approach to treatment not only improves objective outcomes (i.e., reduces adverse events) but also increases patient satisfaction [29, 30]. Common multidisciplinary teams include (and are not limited to) SLPs, occupational therapists, physical therapists, physicians, physician assistants, nurse practitioners, nurses, neurologists, nurses, social workers, and dieticians. Otolaryngologists play an important role along with speech language pathologists specific to voice and swallowing changes in patients with neurodegenerative disease. Referrals for evaluation of voice and swallowing changes may initially be to otolaryngology but multidisciplinary clinics that include the SLPs ensure comprehensive approaches to diagnosis and treatment planning. Otolaryngologists will perform surgical interventions when indicated and will collaborate with the SLP before and after the procedures to ensure optimal outcomes.

Parkinson's Disease

Parkinson's disease is characterized by misfolding and aggregation of alpha-synuclein proteins, resulting in the accumulation of Lewy bodies within the nervous system, thus causing neuronal loss in the substantia nigra, and resultant dopamine deficiency within the striatum [31, 32]. This neuropathology eventually manifests in clinical symptoms including motoric disturbances such as tremor, rigidity, bradykinesia, postural change, salivary pathology including xerostomia, hyposalivation, and drooling, as well as depression, malnutrition, dehydration, and cognitive decline [33–38]. In a recent meta-analysis, oropharyngeal swallowing dysfunction was found to impact 82% of PD patients [24••]. The neuropathology of PD has been shown to result in various disturbances to the swallowing mechanism, including delayed initiation and completion of laryngeal vestibule closure, and reduced pharyngeal constriction [39, 40]. These impairments contribute to increased likelihood of pharyngeal residue (inefficiency) and aspiration during swallowing [41, 42]. Furthermore, in addition to the direct impairments in swallow physiology observed, respiratory safety is further affected via impairments to airway

Table 1 Differences between a reactive versus proactive approach to dysphagia management

Reactive approach	Proactive approach
• Treatment initiated after dysphagia diagnosis	• Early, subclinical swallowing changes are identified
• Focuses on compensatory/adaptive strategies that do not result in lasting change to swallowing	• Builds functional physiologic reserve for swallowing and targets improvement in swallowing physiology
• Potential misalignment with patient's goals of care which can lead to increased care partner burden	• Engages patient in decision-making specific to dysphagia to increase self-efficacy
• Increased risk of developing pneumonia, dehydration, and malnutrition	• May prevent onset of negative health outcomes and maximize oral intake for longer

clearance mechanisms such as urge to cough, cough pressures, cough peak airflow extent, and consistency [21, 43, 44]. Consequently, aspiration pneumonia is a leading cause of mortality among PD patients [45–47].

Management of PD-associated dysphagia has been primarily behaviorally focused as pharmacological and surgical interventions have historically shown little benefit and occasionally negative effects on the swallowing mechanism [48, 49]. Among the pharmacological interventions, Levodopa and Carbidopa remain the current primary medications to increase dopamine levels within the striatum. The benefits of these treatments to generalized dyskinesias seen in PD are established in the literature; however, there is currently little evidence to support the use of these medications to improve swallow function [48, 50–52]. Meanwhile, deep brain stimulation (DBS) remains the primary surgical intervention to manage PD and is often selected when individuals do not (or no longer) respond to pharmacological approaches [53]. DBS consists of surgical placement of electrodes primarily in the subthalamic nucleus or the globus pallidus to provide stimulation to brain regions associated with dopamine release [53]. Although more recent approaches to DBS involving other regions have elicited marginal improvements in duration of vocal fold closure, support for DBS in improving swallow function continues to be significantly limited [54]. In addition to DBS, laryngoplasty, which has typically been used to treat glottal insufficiency in PD, has revealed promising results for reducing dysphagia symptoms based on improvements in PROM scores [55]. However, more work on this topic is needed to understand the true impact of this approach on swallow function.

Alternatively, behavioral interventions for dysphagia management remain the primary approach to managing the effects of PD on swallow function. Compensatory and adaptive interventions, while well-established in the field, have few studies performed exclusively in PD populations. Among the most significant are the studies by Robbins, Logemann, and colleagues examining the effects of chin down posture, as opposed to thickened liquids on aspiration of liquid boluses in patients with PD with or without dementia [56, 57]. This study revealed that moderately thick liquids best minimized aspiration when compared to mildly thick liquids and chin down posture with thin liquids. However, in a subset of these patients followed for three months, incidence of pneumonia was higher in those taking moderately thick liquids compared to those on mildly thick liquids.

Among the behavioral interventions researched, the majority of recent studies focus on restorative, strength-based interventions. Of these approaches, respiratory muscle strength training (RMST) has been the most widely studied [58, 59]. This intervention draws from the rationale that individuals with PD have reduced expiratory airflow and pressure during coughing and are consequently less equipped to protect the airway from invasion of penetrated or aspirated material [60]. Among the RMSTs, expiratory muscle strength training (EMST) has been studied more than its counterpart, inspiratory muscle strength training (IMST). Efficacy studies regarding the use of EMST have revealed support for improved expiratory pressure, volumes, cough physiology, and penetration–aspiration outcomes. In addition to the effects on airway protection, there is evidence that EMST may have positive implications for swallowing physiology, as use of the EMST150 device has been found to result in engagement of swallow-related structures including the submental muscles, pharynx, and the velum. More recently, IMST has also been studied with the rationale that increased volume of inspired air may contribute to more effective airway clearance. Studies have revealed improvements in the endurance of inspiratory musculature, maximum inspiratory pressure, maximum phonation time, peak subglottic pressure, and trivial improvements to voluntary peak cough flow [61, 62].

Other strength-based rehabilitative interventions have been studied in PD. Among these, Lee Silverman Voice Treatment (LSVT), although primarily designed as an intervention for vocal intensity and intelligibility, has been examined as an intervention for dysphagia in two studies [63–66]. This preliminary work supports that LSVT may elicit improvements in several aspects of swallow physiology; however, further research is needed at a larger scale to support the efficacy of this intervention. Neuromuscular electrical stimulation (NMES) [67, 68] has also been applied to PD-associated dysphagia in a few studies, with inconclusive results regarding the contribution of this modality as a supplement to traditional exercise-based therapy [69, 70]. Finally, lingual strengthening has been examined in a single study as an adjunct to RMST, revealing improvements in lingual strength and maintenance of swallow function [71].

In addition to these strength-based interventions, use of interventions that prioritize coordination during swallowing is emerging. A recent clinical trial examining the effects of sensorimotor training for airway protection (smTAP) as compared to EMST revealed that only smTAP was successful in upregulating reflexive airway protection [72, 73]. Additionally, air stacking has been examined in combination with EMST; revealing that the combination of these interventions provided the most benefit to reflexive and voluntary peak cough-flow [74]. Finally, a recent study examined the effects of video-assisted swallow therapy (VAST), or bio-feedback provided via flexible endoscopy [75]. This intervention did not out-perform traditional dysphagia therapy in regards to swallow safety; however, it was successful in promoting improvements to pharyngeal clearance and Swal-QoL scores.

Additionally, considering the progressive nature of PD, several studies have examined the costs and benefits of

alternative means of nutrition (e.g., feeding tube placement). The most recent longitudinal research has placed median survival time post placement of a percutaneous endoscopic gastronomy (PEG) at 344 days [76]. While a PEG may allow for continued administration of medications and a consistent source of nutrition, there are adverse events associated with PEG placement in this population including PEG site infection, buried bumpers, and aspiration pneumonia [76, 77]. Given this, careful, multidisciplinary consideration of the costs and benefits of alternative means of nutrition in patients with PD continues to be recommended.

Amyotrophic Lateral Sclerosis

Amyotrophic lateral sclerosis (ALS) is a motor neuron disease characterized by loss of upper and lower motor neurons resulting in progressive muscle weakness and wasting. Symptoms at onset may initially present as limb muscle weakness (i.e., spinal onset); however, bulbar onset is estimated in 25–30% of cases with symptoms presenting as dysarthria, dysphagia, or dysphonia [78]. Survival rate in persons with ALS is poor, with life expectancy approximately 2–4 years following onset and < 10% of persons with ALS with survival beyond 10 years. Dysphagia is estimated to occur in 85% of persons with ALS leading to malnutrition, dehydration, respiratory failure, and reduced quality of life [79]. Goals for dysphagia management should include maintaining the ability to eat for as long as possible while minimizing aspiration risk.

Swallowing in ALS is characterized by physiologic impairments that threaten safety and efficiency, highlighting the necessity of comprehensive examination (including direct visualization of swallowing function) and routine monitoring [80]. Provisional assessment guidelines have been established by the Northeast ALS group and emphasize multidisciplinary care with incorporation of baseline swallowing, speech, and communication evaluations. This includes PROMs, dietary intake, bulbar function measures (e.g., lingual strength), pulmonary function, and screening for aspiration risk and airway defense capacity [81].

Current dysphagia management approaches for persons with ALS include dietary modifications, non-oral tube feeding, and compensatory strategies. Malnutrition is a negative prognostic indicator for survival in ALS; therefore, timely placement of PEG tubes is necessary for ensuring adequate nutrition and hydration intake, facilitating stabilization of weight, and maintaining medication access [82, 83]. Compensatory strategies to improve swallowing safety or efficiency include the throat clear, chin tuck, head turn or head tilt postures, although emphasis has been placed on individualizing the combination or necessity of these strategies based on each individual's swallowing impairment profile determined by instrumental assessment [84].

Evidence regarding exercise-based interventions targeting swallowing pathophysiology is limited to date. Treatment approaches have historically avoided exercise under the premise that it may induce overuse injuries, thereby resulting in worse function. However, a recent review on the effects of therapeutic physical exercise suggests that it may slow the progression of muscle deterioration in persons with ALS, which may lead to improved performance in activities of daily living [85]. Studies of EMST as a proactive intervention to build functional reserve for persons with ALS (particularly those in earlier stages of the disease process), although preliminary, have illustrated that it is feasible and well-tolerated in this population [86–88]. Findings illustrate maintained or improved inspiratory and expiratory pressure generation and peak cough flow production with mixed effects on swallowing safety and efficiency. Since the goal of exercise in this population is maintenance rather than recovery of function, these findings suggest that therapeutic exercise may be used to slow dysphagia progression; however, further investigation is needed.

Persons with ALS will experience continued decline in respiratory and swallow function throughout the course of the disease, which may lead to consideration of surgical interventions especially in advanced stages [89••]. Factors regarding decision-making for procedures to support respiration (e.g., tracheostomy) or to prevent aspiration (e.g., laryngotracheal separation, narrow field or central part laryngectomy, or total laryngectomy) are complex and necessitate discussion addressing goals of care as early as possible. Tracheostomy may facilitate increased respiratory support; however, a cuffed tracheostomy tube does not in itself prevent aspiration of secretions and pharyngeal swallowing pressures may be reduced. Aspiration prevention surgeries have mixed reports with respect to post-intervention patient and care partner quality of life and burden as well as consequences for speech and communication. Further research studies on operative interventions to manage dysphagia in this population are needed to more thoroughly characterize outcomes, benefits, and long-term management solutions.

Alzheimer's Disease and Related Dementias (ADRD)

Alzheimer's disease is the most common cause of dementia, which is defined as progressive loss of cognitive functioning, most notably related to memory and abstract thinking, that impacts the ability to perform activities of daily living [90]. Other types of dementia include vascular,

lewy body, frontotemporal, and mixed (combination of Alzheimer's disease and vascular dementia). Alzheimer's disease is characterized by beta amyloid deposition and neurofibrillary tangles that lead to loss of synapses and neurons, whereas vascular dementia results from cerebral tissue ischemia causing gliosis and demyelination [91, 92]. Life expectancy for persons with ADRD varies from several years to 10 + years depending upon the age of diagnosis and the dementia subtype [91, 93].

Dysphagia has been estimated to occur in up to 84% of those with ADRD and is caused by deficiencies in sensory and/or motor processes that may be compounded by malnutrition and sarcopenia [94]. These impairments can manifest in oral or pharyngeal phase deficits and impact both the safety and efficiency of the swallow, with changes in swallowing physiology varying by dementia type and severity. Individuals with dementia also frequently experience difficulty with self-feeding, placing them at higher risk of illness due to feeding dependency [95, 96]. There is emerging evidence to suggest that changes in swallowing, beyond those expected with aging, begin early and worsen with disease progression.

Despite the negative health sequelae resulting from dysphagia, there are currently no effective treatments in patients with AD/ADRD that have lasting impact on swallow physiology or are sustainable. Currently, the most common approach to managing swallowing disorders in dementia is through reactive approaches, which can be either compensatory or adaptive [27]. Compensatory strategies may involve postural changes (e.g., chin tuck). Adaptive modifications limit the bolus volume or method of administration (e.g., single sips or drinking from a cup instead of a straw), enhance or alter the taste of foods and liquids, or involve changing the consistencies of foods and liquids. Additionally, controlling the mealtime environment by eliminating distractions, encouraging self-feeding, and using spaced-retrieval training may all result in increased oral intake [97].

Although enteral feeding support in the form of feeding tube placement may be discussed as an option for those with dementia, this practice is largely discouraged due to research revealing serious adverse effects of feeding tube placement in advanced ADRD [98]. Although there are many factors that may influence an individual's response to feeding tube placement, including age, sex, and dementia stage at time of placement, there is currently no evidence to support feeding tube placement as a means for sustaining or prolonging life in those with advanced dementia [99].

Similar to other neurodegenerative conditions, persons with ADRD may benefit from proactive approaches to dysphagia management. Although more research on proactive therapy is needed in this population, it is believed that strength- and skill-based rehabilitative treatments have the potential to positively impact the trajectory of swallowing decline in ADRD. Specifically, in the early stages of ADRD, motor learning is still intact and participation in exercise programs is achievable [100–102]. There is also a role for a proactive building of functional physiologic reserve prior to dysphagia onset in this ADRD in order to prolong the ability to independently eat and drink [27].

Multiple Sclerosis

Multiple sclerosis (MS) is a chronic autoimmune disease of the central nervous system, predominantly characterized by demyelination. As repeated MS relapses occur, damage becomes diffuse and causes brain volume loss, which leads to physical function impairments [103]. Medications are primarily used to treat MS and reduce the occurrence of relapses; however, dysphagia may be an adverse effect of these medications [104]. Oral stage impairments in swallowing include reduced tongue strength, difficulty forming a cohesive bolus, and reduced sensitivity. Pharyngeal stage impairments include increased pharyngeal delay time, reduced tongue base retraction, reduced pharyngeal contraction, decreased laryngeal elevation, impaired cough reflex, and reduced or delayed relaxation of the upper esophageal sphincter (UES) [105].

Current dysphagia treatment in MS includes compensatory and rehabilitative approaches; however, the future study of proactive methods of dysphagia management for this population will be vital to enhanced care. Enteral nutrition, or nutritional tube support, is a common practice for MS patients who have severe dysphagia. More studies are needed to assess the overall effects, with consideration for age, sex, disease duration, and type of MS. Specific to rehabilitative approaches, one randomized controlled trial of 20 patients with MS compared a combination of sensorimotor exercises and swallowing maneuvers (terms "Traditional Dysphagia Therapy") to "usual care" (postural adjustments and dietary modifications) over six weeks and reported greater change in swallowing outcomes (Mann Assessment of Swallowing Ability (MASA) scores, safety, and efficiency) in those who received rehabilitative approaches [106]. EMST has also been studied in MS with evidence of increased maximum expiratory pressures (MEPs), mixed results specific to swallowing safety, and improved Swal-QoL scores [107].

Neuromuscular electrical stimulation over three weeks was found to reduce post-swallow residue and occurrence of aspiration in 25 patients with MS [108, 109]. Two pilot studies have shown that transcranial direct current stimulation (tDCS) may improve safety, maximum expiratory pressures, dysphagia outcome and severity scale (DOSS) scores, and DYMUS scores [110, 111]. Botulinum toxin injections to the

Table 2 Dysphagia interventions in neurodegenerative populations

Specific intervention	Intervention type	Outcomes across neurodegenerative populations
Thickened liquids	Adaptive	Parkinson's disease and ADRD • Moderately thick (i.e., honey thick) liquids—most effective in reducing aspiration • No reduced risk of pneumonia despite reduced aspiration [52] • Other adverse events (e.g., UTI, dehydration, and fever) [52] Huntington's disease • Moderately thick and mildly thick are effective in reducing aspiration [108] • Pureed as well as minced and moist diets useful for reducing risk of choking Myasthenia gravis • Pureed diet useful for improving swallow function during myasthenic crisis [114•]
Eating strategies and environmental modifications	Adaptive	ADRD • Minimizing distractions results in improved caloric intake • Slower pace and longer timeframe for eating is helpful for oral intake • Encouraging self-feeding is critical Huntington's disease • Monitoring feeding rate and bolus size for increased tolerance of oral intake [107, 108]
Sensory enhancement techniques	Adaptive	ADRD • Changes in taste, temperature, and volume may be useful • Thermal–tactile techniques stimulate afferent sensory receptors, but may be short-lived
Chin down posture	Compensatory	Parkinson's disease and ADRD • Reduced risk of aspiration/penetration compared to thickened liquids Huntington's disease • Reduced risk of aspiration/penetration [104]
Spaced retrieval training	Compensatory or restorative	ADRD • Increased oral intake [87]
Expiratory muscle strength training (EMST)	Restorative	Parkinson's disease • Improved respiratory measures [56] • Improved volitional cough measures [57] • Improved penetration–aspiration outcomes [117•, 118, 119] • Potential effect on swallowing musculature [120, 121] Multiple sclerosis • Improved maximum expiratory pressures (MEPS) and SWAL-QOL scores • Mixed results for airway invasion Amyotrophic lateral sclerosis • Increased expiratory pressures [77, 78] • Improved peak cough flow
Inspiratory muscle strength training (IMST)	Restorative	Parkinson disease • Improved inspiratory pressure [56, 57] • Improved inspiratory muscle endurance [56, 57] • Improved max inspiratory pressure [56, 57] • Trivial improvements to voluntary peak cough flow [56, 57] Amyotrophic lateral sclerosis • Increased inspiratory pressure and peak cough flow when combined with expiratory muscle strength training [79]

Table 2 (continued)

Specific intervention	Intervention type	Outcomes across neurodegenerative populations
Lingual strengthening	Restorative	Parkinson disease • Improvements to lingual strength and maintenance of swallow function *when used in combination with RMST* [62]
Neuromuscular electrical stimulation (NMES)	Restorative	Multiple sclerosis • Reduced post-swallow residue, aspiration occurrences, and patient-reported swallow exertion [96]
Sensorimotor training for airway Protection (smTAP)	Restorative	Parkinson disease • Upregulation of reflexive airway protection (cough airflow, expiratory volume, and urge to cough) [63]
Lee Silverman Voice Treatment (LSVT LOUD ®)	Restorative	Parkinson disease • Improvements in lingual propulsion, tongue base retraction, airway clearance, oral transit time, oral efficiency, UES opening extent and duration, peak cough, and expiratory flow based on *two pilot studies*
Visually assisted swallow therapy (VAST)	Restorative	Parkinson disease • Improved swallow efficiency compared to traditional therapy [66] • Equivalent effects on swallow safety as traditional therapy [66]
Air stacking	Restorative	Parkinson disease • Improvements to reflexive and voluntary peak cough-flow *when used in combination with EMST* [65]
Transcranial direct current stimulation (tDCS)	Restorative	Multiple sclerosis • Improved swallowing safety and maximum expiratory pressures (MEPs) [100] • Improved dysphagia outcome and severity scale (DOSS) and DYMUS scores [101]
Botulinum toxin (Botox)	Medical	Multiple sclerosis • Improved safety during swallowing [102]

cricopharyngeal muscle have also been shown to improve swallowing safety in MS. While these studies are promising for use of rehabilitative and medical dysphagia management approaches in MS, additional research with larger cohorts is needed [112].

Huntington's Disease

Huntington's disease (HD) is an inherited neurodegenerative disease that occurs from a mutation on a gene (cytosine–adenine–guanine trinucleotide repeats in the Huntington gene) and affects more than 30,000 people in the United States [113]. Life expectancy for HD is within 10 to 30 years following symptom onset, with pneumonia being the most common cause of death. Primary characteristics of those with HD include chorea, cognitive decline, and psychiatric disturbances [114•].

Dysphagia characteristics in this population include impaired coordination in both oral and pharyngeal phases of swallowing, reduced bolus control, involuntary oral movements leading to increased risk of choking, reduced and delayed swallow trigger, pharyngeal residue, and penetration/aspiration [115]. Due to the choreic nature of HD, weight loss is a common characteristic and often requires collaboration between dietitians and other clinicians as patients may benefit from high-calorie diets [116].

Depending on severity of disease and cognitive ability of the patient, current approaches to dysphagia treatment are largely adaptive and compensatory. Adaptive treatments in this population focus on modification of liquids and/or solids for increased tolerance of oral intake as well as a slower feeding rate [117•, 118]. Compensatory approaches include postural strategies (e.g., chin tuck) and maneuvers (Mendelsohn and Masako have been shown to be feasible) to increase safety and/or efficiency during swallowing [114•, 119–121]. The efficacy of rehabilitative interventions in this population is largely unknown.

Myasthenia Gravis

Myasthenia gravis (MG) affects approximately 36,000 to 60,000 people in the United States and is caused by disruption of neurotransmitter, acetylcholine, that leads to muscle weakness and fatigue. With treatment, individuals with myasthenia gravis have a normal life expectancy. There

are two main types of MG: ocular (affects muscles around the eyes and eyelids) and generalized MG (more common, spreads to other muscles) [122].

Dysphagia characteristics for those with MG typically include prolonged bolus mastication, reduced bolus formation, prolonged transport, oral residue, delayed swallow trigger, pharyngeal residue, and penetration/aspiration [123]. Myasthenic crises can lead to possible respiratory failure and endotracheal intubation that could further impact swallowing abilities. Current literature regarding effective treatments for dysphagia in patients with MG is sparse; however, it appears that dietary modification to increase tolerance of oral intake is most frequently used [124]. While physical training and exercise may be safe in persons with MG and can result in improved muscle strength and daily function, this has yet to be examined specific to treatment for dysphagia [125, 126].

For a summary of dysphagia interventions in neurodegenerative disease populations, please see Table 2.

Other Rare Neurodegenerative Diseases

Although there are other rare neurodegenerative diseases that lead to dysphagia (e.g., oculopharyngeal muscular dystrophy, Prion diseases, Friedreich ataxia, and progressive supranuclear palsy), there is limited research focused on optimal approaches to dysphagia management for persons with these conditions. Therefore, general principles for proactive, multidisciplinary management of dysphagia should be applied to these populations until future research elucidates the most efficacious interventions.

Conclusions

Evidence-based approaches to dysphagia evaluation and management are critical for individuals with neurodegenerative disease. While adaptive or compensatory approaches to dysphagia management are necessary to optimize the safety and efficiency of swallowing while maintaining oral intake, additional work can be done to establish efficacy of rehabilitative approaches that can optimize swallowing function for longer into disease progression. The field will benefit from additional research to determine best practice for dysphagia management based on well-designed efficacy and effectiveness studies across neurodegenerative conditions.

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Declarations

Conflict of Interest The authors declare that they have no conflict of interest.

Human and Animal Rights and Informed Consent All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee (Institutional Review Board of the University of Wisconsin-Madison, reference number: 2011–0322) and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

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Papers of particular interest, published recently, have been highlighted as:
- • Of importance
- •• Of major importance

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