ACRM
AMERICAN CONGRESS OF
REHABILITATION MEDICINE

Archives of Physical Medicine and Rehabilitation

journal homepage: www.archives-pmr.org

Archives of Physical Medicine and Rehabilitation 2021;102:895-904

ORIGINAL RESEARCH

Classification of Stroke Patients With Dysphagia Into Subgroups Based on Patterns of Submental Muscle Strength and Skill Impairment

Karen B. Ng, PhD,[a,b] Richard D. Jones, PhD,[a,b,c,d,e] Esther Guiu Hernandez, MS,[a,b] Phoebe Macrae, PhD,[a,b] Maggie-Lee Huckabee, PhD[a,b]

From the [a]University of Canterbury Rose Centre for Stroke Recovery and Research, Christchurch; [b]School of Psychology, Speech and Hearing, University of Canterbury, Christchurch; [c]Department of Electrical and Computer Engineering, University of Canterbury, Christchurch; [d]New Zealand Brain Research Institute, Christchurch; and [e]Medical Physics and Bioengineering, Christchurch Hospital, Christchurch, New Zealand. Current affiliation for Ng, Laura Fergusson Brain Injury Trust, Christchurch, New Zealand.

Abstract

Objectives: To identify and characterize subgroups of stroke patients with clinical signs of dysphagia, based on swallowing-related strength and skill impairments of the submental muscle group.

Design: Prospective observational study.

Setting: Inpatient rehabilitation centers and community dwellings.

Participants: Individuals ($N=114$), including stroke patients with dysphagia ($n=55$) and 2 control groups including myopathic patients with dysphagia ($n=19$) and healthy volunteers ($n=40$) were included in this study.

Interventions: Not applicable.

Main Outcome Measures: Novel clinical assessment of strength (force generation) and skill (spatial and temporal precision of muscle activation) of the submental muscle group during swallowing and nonswallowing behaviors, using surface electromyography and dynamometry.

Results: Hierarchical cluster analysis revealed 4 clusters, which could be broadly characterized as cluster 1: intact strength and skill, cluster 2: poor strength and poor nonswallowing skill, cluster 3: poor strength, and cluster 4: poor strength and poor swallowing skill. Membership in cluster was significantly associated with medical diagnosis ($P<.001$). The majority of healthy and myopathic participants were assigned to clusters 1 and 3, respectively, whereas stroke patients were found in all 4 clusters. Skill outcome measures were more predictive of cluster assignment than strength measures.

Conclusions: Although healthy and myopathic participants demonstrated predominantly homogeneous swallowing patterns of submental muscle function within their etiology, several subgroups were identified within stroke, possibly reflecting different subtypes of swallowing function. Future research should focus on the nature and rehabilitation needs of these subtypes. Assessment of skill in swallowing may be an important but overlooked aspect of rehabilitation.

Archives of Physical Medicine and Rehabilitation 2021;102:895-904

Swallowing impairment, or dysphagia, is a common and serious consequence of stroke, leading to increased risk of aspiration pneumonia, dehydration, and malnutrition.[1] Stroke patients with dysphagia present with a diverse range of biomechanical impairments,[2] commonly categorized based on the anatomic location of presenting symptoms (eg, oral, pharyngeal, or esophageal

---

Presented to the European Society of Swallowing Disorders, Dublin, Ireland, September 25-29, 2018.

Supported by the Commonwealth Scholarship and Fellowship Fund, a University of Canterbury Doctoral Scholarship, and a Post Graduate Publishing Bursary from the Canterbury Medical Research Foundation.

Disclosures: The commercial software used in this study (Biofeedback in Strength and Skill Training) was developed at the University of Canterbury Rose Centre for Stroke Recovery and Research and is owned by the University of Canterbury.

0003-9993/21/$36 - see front matter © 2020 by the American Congress of Rehabilitation Medicine
https://doi.org/10.1016/j.apmr.2020.11.014

dysphagia). Although swallowing biomechanics in dysphagia are fairly well-defined, little is known about the pathophysiological mechanisms underlying these impairments.

Evidence from limb and motor speech research indicates that cortical and peripheral lesions result in distinct patterns of pathophysiological features.[3-6] In the corticospinal system, damage at the level of the muscle primarily causes weakness, and both weakness and decreased skill are crucial factors contributing to disability after cortical damage.[7-9] Stroke patients have impaired ankle motor coordination regardless of weakness, suggesting that deficits in strength and skill should be evaluated separately.[8] In the corticobulbar system, stroke can be associated with multiple types of motor speech disorders (eg, spastic, ataxic, or flaccid dysarthrias),[5] that are produced by lesions across the neuraxis.[3] Given the heterogeneity of biomechanical impairments after stroke, it is reasonable to surmise that there may be several subtypes of dysphagia, similar to that in other corticobulbar behaviors.[10,11]

Few studies have investigated the relative contributions of strength and skill to swallowing. Given the precise coordination and timing needed for safe swallowing, and the substantial role of the cortex in modulating the swallowing response,[12] impairment in centrally-mediated skill has been proposed as a factor that can contribute to poststroke dysphagia.[13-18] Skill is defined as the ability to precisely modulate timing and amplitude of muscle activity[8,19] and has previously been measured using surface electromyographic (sEMG) protocols.[18,20-23] The spatial and temporal aspects of muscle contraction are displayed on a screen, providing the patient with augmented biofeedback of their performance. Participants control the timing and force of their movement during swallowing-related behaviors to place their response cursor in an on-screen target. In a case study involving a brainstem stroke patient with dysphagia, performance accuracy was significantly reduced compared with healthy controls.[18] However, limited generalization can be made from the results of a single patient. The study did not measure strength levels, leading to the possibility that weakness confounded the results.

Because impaired hyoid movement may be associated with greater risk of airway invasion and pharyngeal residual,[24] submental muscle weakness may contribute toward dysphagia.[17,25] Submental muscle contraction can be estimated using videofluorographic swallowing studies (VFSS). However, the underlying cause of swallowing impairment can only be inferred, not directly measured, from visualization of biomechanical movement.[26] For example, decreased hyoid displacement seen on VFSS could be caused by weakness, but it could also reflect poor coordination or other neuromuscular deficits of motor control.[10] Because many of the submental muscles that elevate the hyoid are also involved in opening the jaw, a jaw-opening force test using muscle dynamometry could provide a more direct measure of isometric strength.[27]

The ability to classify strength and skill impairment patterns in dysphagia would represent an important step toward improving diagnostic specificity and developing treatments tailored to each patient's needs. In this study, we designed a novel clinical assessment to assess strength and skill in the submental muscle group using sEMG and dynamometry. The aim of this research was to identify and characterize subgroups of stroke patients with dysphagia. To assist in characterization of subtypes, 2 control groups (healthy controls and myopathic patients) were chosen based on their underlying swallowing physiology. It was expected that healthy participants would have relatively intact strength and skill and that patients with peripheral lesions (myopathy) would have submental function primarily characterized by weakness.[28-30] Weakness in the submental muscles has been found to be the main mechanism of dysphagia in patients with inflammatory myopathy.[30] We further hypothesized that due to the heterogeneous nature of stroke, stroke patients with dysphagia could be classified into several subgroups, some that overlap with control groups and some that are stroke-specific clusters.

List of abbreviations:

sEMG	**surface electromyography**
TOMASS	**Test of Masticating and Swallowing Solids**
TWST	**Timed Water Swallowing Test**
VFSS	**videofluoroscopic swallowing study**

Methods

Participants

Three groups of participants were recruited between November 2015 and September 2017: (1) stroke patients; (2) patients with inflammatory myopathy (inclusion body myositis, myotonic dystrophy, or oculopharyngeal muscular dystrophy); and (3) healthy controls without a history of dysphagia or any neurologic, structural, or muscular disorders that might affect swallowing. Participants were either self-selected from advertisements or were referred from nursing homes and rehabilitation facilities. Stroke and myopathic patients were included if they had dysphagic presentation on a clinical swallowing evaluation conducted by the primary researcher. (K.N.) Dysphagic presentation was defined as meeting 2 of the following 3 criteria: (1) score of 3 or higher on the 10-item Eating Assessment Tool (EAT-10)[31]; (2) deficits in the structural integrity, symmetry, sensation, and movement on cranial nerve examination; and (3) consistent overt signs during oral intake trials such as coughing, throat clearing, vocal and respiratory changes immediately after swallowing, and multiple swallows. As the focus was not on defining the specific biomechanics of swallowing, VFSS were not completed. Additional inclusion criteria included age of 50 years or older (in order to reflect the predominant age range for stroke)[32] and ability to follow simple verbal directions (assessed informally by primary researcher). Participants were excluded if they had a history of temporomandibular joint disorders. The study was approved by the appropriate regional Human Ethics Committee.

There are no established guidelines for calculating sample size for cluster analyses.[33] However, based on consultation with a statistician, it was recommended that a sample of approximately 20 participants should be recruited for each cluster. Because healthy controls and myopathic patients were expected to be assigned into their own clusters, and stroke patients were expected to be assigned to several clusters, recruitment of 20 healthy, 20 myopathic, and 60 stroke participants was targeted. Recruitment of healthy participants was later increased to 40 to ensure a normative sample with adequate age and sex representation.

Instrumentation

sEMG and biofeedback software

A triode surface electrode patch[a] with 2-cm interelectrode distance was attached to the prepared skin surface underneath the chin to measure electrical activity of the submental muscle group. The 2 recording electrodes were placed at midline, with the ground electrode oriented laterally. sEMG signals were recorded and the root-mean-square envelope of sEMG calculated by a portable device.[b] A custom-developed software, Biofeedback in Strength and Skill Training (BiSSkiT),[c] plotted a real-time waveform on a computer screen, with time in seconds on the x-axis and amplitude in µV on the y-axis.

Dynamometry

A compact dynamometer[d] was secured to the chin and head using a custom-made strap (fig 1). A molded chin cup made of dental putty was adhered to the dynamometer sensor plate. The band encircling the head and vertical straps were adjustable and were securely positioned on each participant so that the dynamometer was held tightly under the chin to minimize jaw opening. A hand-held monitor displayed the maximum force generated at each trial.

Experimental procedure

After receipt of written informed consent, all participants completed 4 assessment tasks, as well as 2 tests of oral intake. Because the dynamometer and sEMG electrodes could not be secured under the chin at the same time, half of the participants completed the jaw-opening strength task before the 3 counter-balanced sEMG tasks, and the other half completed the sEMG tasks before using the dynamometer. All swallows were performed with saliva, as the swallowing skill task involved submaximal levels of muscle activation that could interfere with safe bolus ingestion.

Jaw-opening strength

The dynamometer was calibrated to zero, and then secured under the participant's chin. Verbal instructions were: "Gradually increase your jaw-opening force over 1 second until reaching maximum force. Hold for 2 seconds, and then relax." Participants completed 5 trials, with a break of approximately 1 minute between trials.

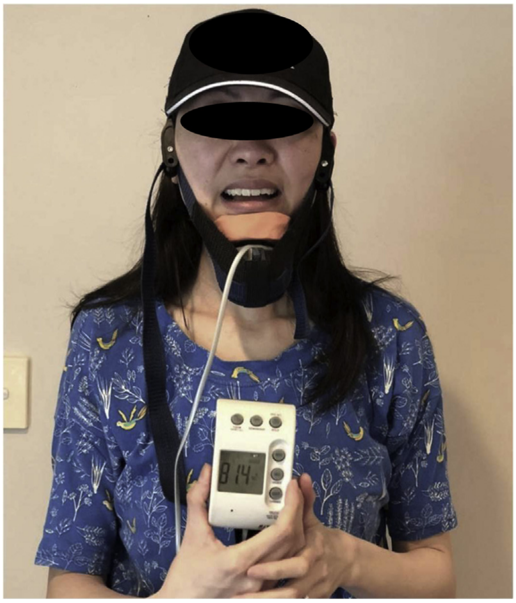


Fig 1 A compact dynamometer secured under the chin and to the head using custom-made, adjustable head straps was used for measuring jaw-opening strength. Maximum force generated at each trial was displayed on the monitor.

Swallowing strength

Participants were instructed to perform 5 effortful swallows, given the verbal directions "Swallow hard with all the muscles in your mouth and throat," followed by 5 regular effort swallows, at a rate of approximately 1 every 30 seconds.

Swallowing skill

Using the Biofeedback in Strength and Skill Training software, the y-axis on the screen was calibrated so that the maximum value equaled the average sEMG amplitude of 5 effortful swallows. A square target appeared in the center of the screen (fig 2); the height of the target box was calculated as 30% of the y-axis height, with a 1:1 height-to-width ratio. The participant was instructed to "swallow so that the peak of your waveform falls in the center of the square."

Jaw-opening skill

The y-axis was recalibrated to the mean peak sEMG amplitude from 5 trials of maximum jaw-opening. As with the swallowing precision task, a target appeared on screen. Participants were instructed to "open your jaw so that the peak of the waveform falls in the center of the square." In both skill tasks, participants completed 10 trials each with approximately 30 seconds between trials.

Tests of oral intake

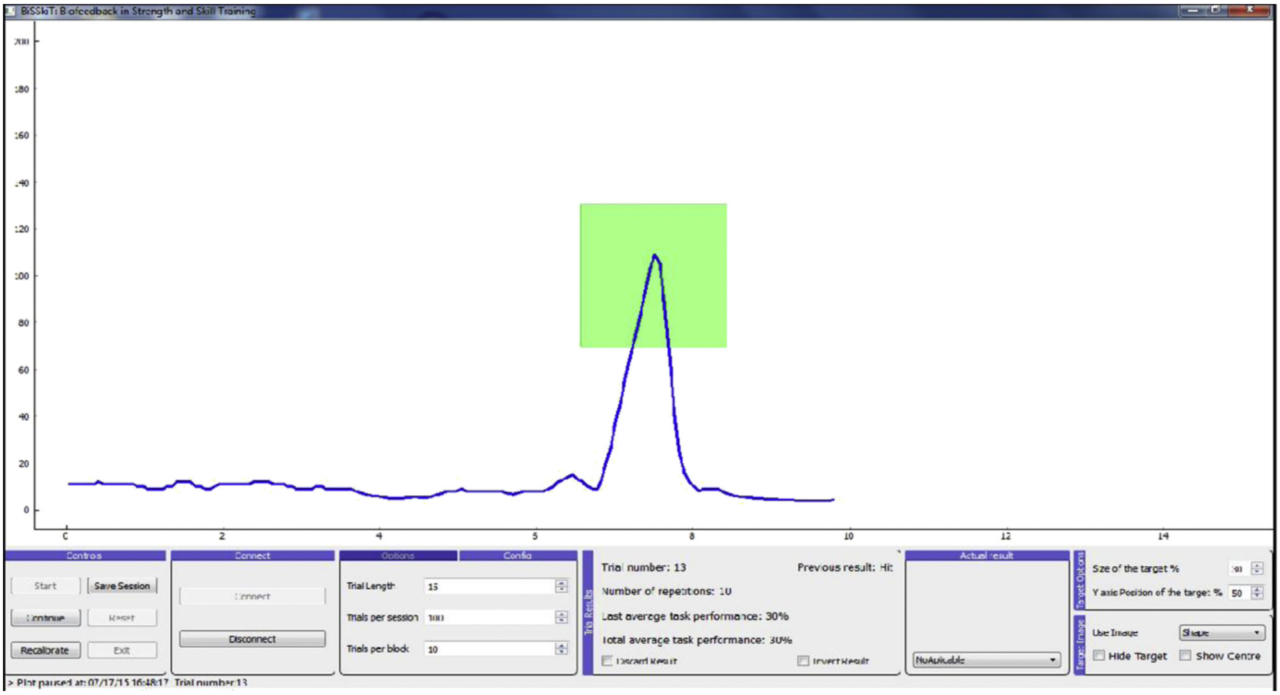
Participants took part in the timed water swallowing test[34] (TWST) and Test of Mastication and Swallowing Solids (TOMASS).[35] Results from these tests were not included in the cluster analysis.

Outcome measures

Raw data were analyzed to produce 8 strength and skill outcome measures for each participant (table 1).

Statistical analyses

Hierarchical cluster analysis using Ward's method and Euclidean distance was completed on the 8 strength and skill outcome variables to partition participants into subtypes, by maximizing similarities within groups and differences between groups.[36] Twenty-six cluster validation indices, including the silhouette statistic[37] and gap statistic,[38] were used to evaluate intracluster compactness and intercluster separation, with the optimal number of clusters decided by the method of majority rule.[39] Comparison of the association between cluster and diagnostic group membership was analyzed using Fisher's exact task. Cluster profiles were described by examining each cluster's mean scores on outcome measures. External validity of cluster solution was evaluated by comparing performance on TWST and TOMASS between clusters using multivariate analysis of variance, followed by univariate analyses of variance on continuous variables and generalized linear mixed effect models for binomial distributions with cluster as fixed effect and participant as random effect for binary variables. The classification and regression tree technique was used to identify which variables best predicted cluster assignment and the threshold cutoff values used for decision making. Analyses were completed in RStudio, version 1.1.442.[e]



Trial Length: 15

Trials per session: 100

Trials per block: 10

Trial number: 13

Previous result: Hit

Number of repetitions: 10

Last average task performance: 30%

Total average task performance: 30%

☐ Discard Result

☐ Invert Result

NoApplicable

Size of the target %: 30

Y axis Position of the target: %: 50

Use Image: Shape

☐ Hide Target

☐ Show Centre

> Plot paused at: 07/17/15 16:48:17 Trial number:13

Fig 2 Screenshot of Biofeedback in Strength and Skill Training software during skill assessment.

Results

A total of 122 participants met the inclusion criteria and were enrolled in the study. Eight stroke patients were unable to complete the study because of fatigue (n=2), severity of dysphagia (n=4), and severity of cognitive impairments (n=2). A total of 114 participants were included in the final analyses.

Demographics

Thirty-six participants had supratentorial stroke, 9 had infratentorial stroke, and 10 had unknown lesion location (table 2). The myopathic group comprised 12 participants with myotonic dystrophy, 6 with inclusion body myositis, and 1 with oculopharyngeal muscular dystrophy. The age of the stroke patients was greater than both healthy controls ($P<.001$) and myopathic patients ($P<.001$), but there was no significant difference in age between the healthy and myopathic groups. The stroke group had a shorter dysphagia duration and lower (less severe) self-reported mean score on the EAT-10 than myopathic participants. TWST and TOMASS scores were comparable between the stroke and myopathic patient groups ($P>.05$ for all TWST and TOMASS scores, except for number of swallows; $P<.01$).

Table 1 Strength and skill outcome measures

Task	Instrumentation	Outcome Measures	Definition	Unit
Jaw-opening strength	Dynamometer	JF	Mean JF	Newtons
Swallowing strength	sEMG	Normalized ES amplitude	Mean peak amplitude (μV) of ES, divided by mean peak amplitude (μV) of regular effort swallows	Ratio
Swallowing skill	sEMG with biofeedback	SHR	Frequency of hits divided by total number of trials and multiplied by 100	%
		STE	Mean time interval (s) between the center of the target and response peak, divided by total screen width (30s) and multiplied by 100	%
		SAE	Mean difference in amplitude (μV) between center of the target and response peak, divided by screen height (μV) and multiplied by 100	%
Jaw-opening skill	sEMG with biofeedback	JHR	Frequency of hits divided by number of trials and multiplied by 100	%
		JTE	Mean time interval (s) between the center of the target and response peak, divided by total screen width (30s) and multiplied by 100	%
		JAE	Mean difference in amplitude (μV) between center of the target and response peak, divided by screen height (μV) and multiplied by 100	%

NOTE. A "hit" was defined as the waveform peak falling inside the target. A smaller temporal or amplitude error represented increased accuracy.
Abbreviations: ES, effortful swallowing; JAE, jaw-opening amplitude error; JF, jaw-opening force; JHR, jaw-opening hit rate; JTE, jaw-opening temporal error; SAE, swallowing amplitude error; SHR, swallowing hit rate; STE, swallowing temporal error.

Table 2 Participant demographics and swallowing characteristics

Characteristic	Stroke (n=55)	Myopathy (n=19)	Healthy (n=40)	*P* Value
Age, y				<.001[*]
Mean ± SD	78.4±9.28	64.6±8.75	69.0±9.91	
Range	55-94	52-80	51-88	
Women, n (%)	18 (33)	11 (58)	20 (50)	.09[*]
Stroke lesion location, n (%)				
Supratentorial, right	19 (35)			
Supratentorial, left	17 (31)			
Infratentorial	9 (16)			
Unknown	10 (18)			
Myopathy type, n (%)				
Myotonic dystrophy		12 (63)		
Inclusion body myositis		6 (32)		
Oculopharyngeal muscular dystrophy		1 (5)		
Dysphagia duration, n (%)[†]				<.001[‡]
<1 mo	18 (33)	0 (0)		
1-3 mo	7 (13)	0 (0)		
4-12 mo	4 (7)	0 (0)		
>12 mo	26 (47)	19 (100)		
Oral intake status, n (%)				.03[‡]
Nonoral	4 (7)	1 (5)		
Modified oral	23 (42)	2 (11)		
Full oral	28 (51)	15 (79)		
EAT-10 score, mean ± SD	12.24±8.55	18.32±8.86		.01[‡]
TWST score, mean ± SD				
Volume/swallow	13.82±7.56	18.95±9.17	23.89±8.93	<.001[*]
Time/swallow	3.83±2.68	3.52±3.00	1.55±.47	<.001[*]
Volume/time	5.54±5.05	7.79±5.37	17.17±7.97	<.001[*]
TOMASS score, mean ± SD				
Bites	3.68±1.94	4.19±1.64	2.90±1.08	.01[*]
Masticatory cycles	101.19±45.86	93.75±35.55	52.00±16.51	<.001[*]
Swallows	2.77±1.73	4.69±3.36	2.28±1.01	<.001[*]
Time	97.47±44.34	101.05±33.16	45.49±15.93	<.001[*]

[*] Main effect of group.
[†] Patient report of the duration of dysphagia symptoms.
[‡] Difference between stroke and myopathic groups

Hierarchical cluster analysis

Results of the cluster analysis are displayed graphically (fig 3). A 3-cluster solution was recommended by 5 cluster validation indices, a 4-cluster solution was recommended by 9 indices, a 5-cluster solution was recommended by 2 indices, and a 6-cluster solution recommended by 3 indices. A 4-cluster solution was chosen because this was the most frequently recommended cluster arrangement, based on multiple validation indices. Examination of the performance profiles also suggested that a 4-cluster solution was optimal compared with 3 clusters, as it divided up a cluster of stroke patients into 2 separate clusters (clusters 2 and 4) with different patterns of submental skill performance.

Each cluster had different proportions of stroke, myopathic, and healthy participants (table 3) and also demonstrated different patterns of performance (fig 4). Membership in a diagnostic group and cluster was significantly associated ($P<.001$). Clusters were characterized as follows: cluster 1 consisted mainly of healthy participants, with relatively good performance on all strength and skill tasks; cluster 2 comprised predominantly stroke patients and was characterized by deficits in all strength and skill measures, except for relatively intact swallowing amplitude error; cluster 3 comprised stroke, myopathy, and healthy participants and was characterized by below-average scores in strength tasks but relatively intact precision; and cluster 4 consisted entirely of stroke patients and had deficits in all strength and precision measures, but relatively intact jaw-opening hit rate and jaw-opening amplitude error.

External validity of cluster solution

External validity was measured by comparing clusters on outcomes (TWST and TOMASS) that were not used in the cluster analysis. Due to severity of dysphagia, 7% (4 of 55) of stroke and 5% (1 o f19) of myopathic patients did not complete the TWST, 24% (13 of 55) of stroke and 11% (2 of 19) of myopathic patients did not complete the TOMASS, and 24% (13 of 55) of stroke and 5% (1 of 19) of myopathic patients did not complete either test. There was missing TWST data for 1 healthy participant, and another healthy participant declined to consume the cracker for the TOMASS due to gluten allergy. Of those who completed the oral intake tests, performance was significantly different between clusters for both TWST (Pillai's trace=.30, $F_{1,90}=12.64$; $P<.001$) and TOMASS (Pillai's trace=.51, $F_{3,81}=4.05$; $P<.001$).

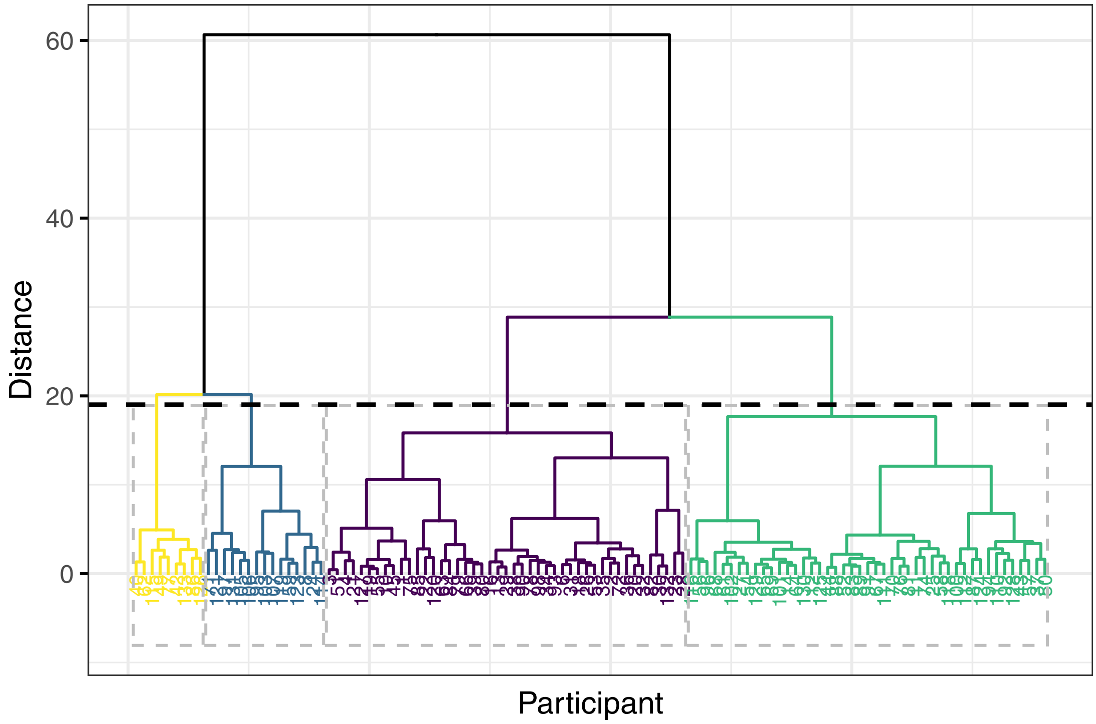


Fig 3 Hierarchical cluster analysis of all participants, visually displayed as a dendrogram. Starting at the bottom, each participant is initially considered to be an individual cluster. Clusters are progressively combined based on their similarity, and this step is repeated until all participants are members of 1 cluster. The height of the vertical lines reflect the dissimilarity at which clusters of participants were merged. Clusters merged together with a short vertical line are similar to each other, whereas clusters connected with a tall vertical line have greater differences. Cutting of the dendrogram branches at the dashed line resulted in 4 clusters, as calculated using cluster validation indices. The optimal number of 4 clusters was calculated using 26 different methods to determine the most frequently proposed number of clusters.

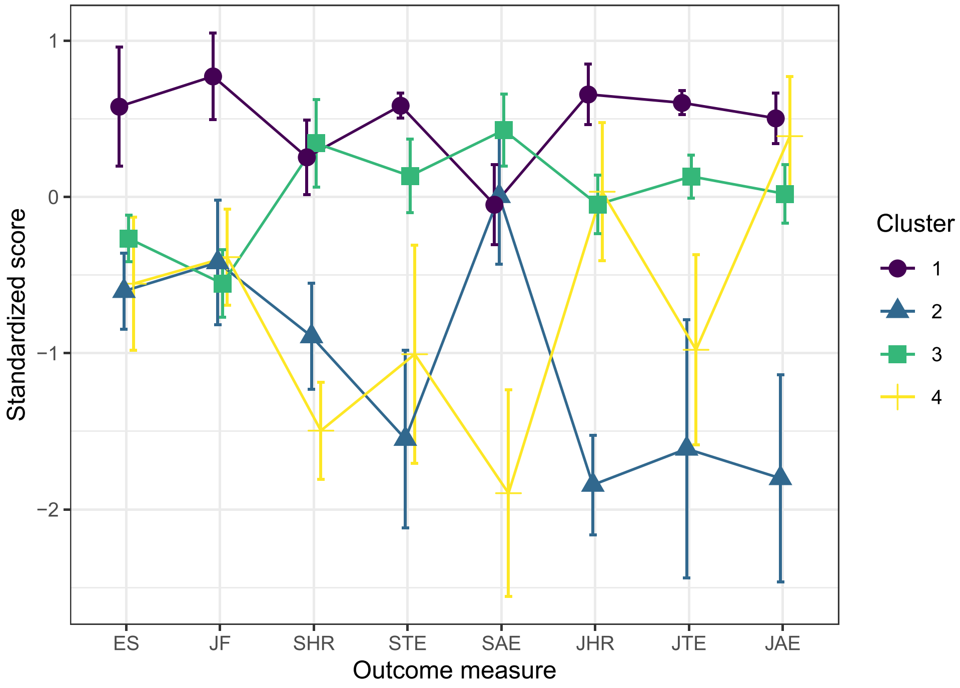


Fig 4 Standardized mean scores and 95% confidence intervals for strength and skill variables. Scores for SAE, STE, JAE, and JTE have been reversed for ease of interpretation, so that a score below zero reflects greater impairment. Abbreviations: ES, effortful swallowing; JAE, jaw-opening amplitude error; JF, jaw-opening force; JHR, jaw-opening hit rate; JTE, jaw-opening temporal error; SAE, swallowing amplitude error; SHR, swallowing hit rate; STE, swallowing temporal error.

Table 3 Diagnostic membership and assessment performance of the 4 clusters

	Cluster			
Characteristic	1 (n=45)	2 (n=15)	3 (n=45)	4 (n=9)
Diagnostic group, n (%)				
Healthy	34 (85)	0 (0)	6 (15)	0 (0)
Stroke	9 (16)	14 (25)	23 (42)	9 (16)
Myopathy	2 (11)	1 (5)	16 (84)	0 (0)
Strength variables, mean ± SD				
ES (ratio)	3.0±1.7	1.4±0.6	1.9±0.7	1.5±0.7
JF (Newtons)	97.1±25.6	64.0±20.0	60.3±20.0	65.0±11.1
Skill variables, mean ± SD				
SHR (%)	49.4±19.9	20.7±15.3	51.6±23.4	5.6±10.1
STE (%)	2.4±1.5	14.2±5.7	4.9±4.33	11.2±5.0
SAE (%)	17.1±6.0	16.7±5.5	13.7±5.4	30.1±6.0
JHR (%)	71.9±14.6	15.3±13.0	55.9±14.0	57.8±13.0
JTE (%)	1.4±0.5	6.1±3.2	2.4±1.0	4.8±1.7
JAE (%)	11.9±3.3	25.9±7.3	14.9±3.8	12.6±3.0

Abbreviations: ES, effortful swallowing; JAE, jaw-opening amplitude error; JF, jaw-opening force; JHR, jaw-opening hit rate; JTE, jaw-opening temporal error; SAE, swallowing amplitude error; SHR, swallowing hit rate; STE, swallowing temporal error.

On the TWST, cluster 1 ingested a greater volume per swallow than cluster 2 ($P<.05$) and had a shorter swallowing time and larger swallowing capacity than clusters 2, 3, and 4 ($P<.01$). On the TOMASS, cluster 1 required fewer bites than cluster 2 ($P<.05$) and fewer masticatory cycles and less time to finish the cracker than clusters 2, 3, and 4 ($P<.01$).

Classification and regression tree

The classification tree in figure 5 demonstrates that the most predictive variables (and cutoff values) for clustering participants were jaw-opening temporal error of 1.7% or 0.5 seconds, jaw-opening hit rate of 35% accuracy, and swallowing hit rate of 15% accuracy.

Discussion

Cluster analysis methods were used to explore underlying, previously undefined patterns in submental strength and skill among participants with and without dysphagia, irrespective of diagnosis. Results from the study identified the presence of 4 subgroups in stroke patients, with skill impairment being a key factor in determining cluster assignment. Previous research has used cluster analysis to identify subtypes of limb movement impairment after stroke.[40-42] Our findings reinforce that the heterogeneous nature of stroke can also be seen in swallowing function[43,44] and are consistent with neurophysiological studies identifying several types of underlying mechanisms in neurogenic dysphagia.[11]

Previous research suggests that the underlying cause of dysphagia is different depending on lesion location (central vs peripheral).[11,17] The 3 diagnostic groups were expected to have different patterns of submental muscle functioning due to differences in underlying swallowing physiology. Results show that this expectation was met. Membership in diagnostic group and cluster was significantly associated. The majority (85%) of healthy participants were assigned to the "intact" cluster, whereas the majority (84%) of the patients with damage at the level of the muscle (myopathy) were assigned to the "weak" cluster. Patients with central damage (stroke) demonstrated heterogeneous deficits in submental skill and strength, and were spread out across all 4 clusters. This provides preliminary internal validation of the cluster solution and confirms that the outcome variables chosen in this study were sensitive to pathophysiological differences expected between participants.

The classification tree demonstrated that skill measures of hit rate and spatiotemporal error were more accurately able to predict cluster assignment than strength. This is likely due to greater differences in skill scores between clusters than strength scores, resulting in skill tasks being better able to discriminate between clusters. In addition, none of the healthy participants were assigned to clusters 2 and 4 (clusters with relatively poor skill). This suggests that the clinical assessment had specificity for measures of skill, in that participants with intact precision were not incorrectly assigned. The clusters differed significantly on measures that were derived from clinical tests of functional ingestion (TWST and TOMASS) and were not used in the cluster analysis, providing initial support for the external validity of the cluster solution.

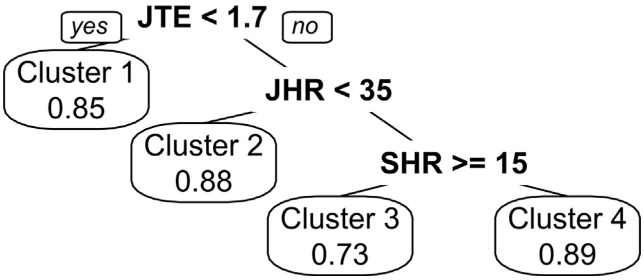


Fig 5 Classification tree showing variables and cutoff scores that best predict cluster assignment. Participants who matched the splitting rule at the top of each split were assigned to the left branch. Numbers under the clusters indicate probability that participants matching the splitting rule are correctly classed in that cluster. Abbreviations: JHR, jaw-opening hit rate; JTE, jaw-opening temporal error; SHR, swallowing hit rate.

This study highlights the differential contribution of strength and skill impairments to dysphagia. The skill assessment target was calibrated to each participant's maximum muscle activation during swallowing and jaw-opening, so that precision of muscle contraction could be measured without potential confound of weakness. The results reinforce the notion that dysphagia may be associated with both weakness and deficits in precise coordination.[10,17,18,22,45] The majority of stroke patients in this sample had submental muscle weakness, but demonstrated differing levels of skill impairment, providing support to the idea that strength and skill impairments are independent and separate contributors to functioning after stroke.[6,8,19,46]

Our findings have potential implications on dysphagia rehabilitation. Approximately one-fifth of the stroke patients were assigned to the "intact" cluster. Another two-fifths were assigned to the "weak" cluster. The final two-fifths of the stroke patients were assigned to clusters 2 and 4, with deficits in both strength and skill. Interestingly, there was no evidence of a cluster characterized by decreased skill only. One explanation for this absence could be that, even though both strength and skill contribute to swallowing, the ability to move precisely precedes the ability to generate maximum force. This may mean that for certain patients with both strength and skill impairments, skill training (ie, improving accuracy in timing and execution of swallowing behavior)[18,22,23,47,48] may need to be initiated before working on strengthening exercises. Prescribing only strength training when a patient has skill deficits may cause unintended adverse consequences.[49,50] This preliminary evidence of skill impairment in dysphagic individuals suggests that accurate and specific diagnosis of swallowing pathophysiology is fundamental to the effective management of dysphagia.

Study limitations

There were several limitations in this exploratory study. For the healthy controls, normal swallowing function was confirmed only via self-report. For the dysphagic patients, we used a clinical swallowing evaluation and not an instrumental assessment to determine presence of dysphagia as part of the inclusion criteria, and some patients may have been excluded from the sample. However, compared with VFSS, clinical swallowing evaluations underdiagnose dysphagia,[51] and therefore the patients in this study likely had dysphagia and met the inclusion criteria.

Submental sEMG amplitude was used as a proxy measure of swallowing strength, although the relationship between strength and sEMG amplitude remains unclear.[52] The participant subtypes in this study were based on the functioning of only 1 muscle group, the submental muscles. Whether these subtypes are reflected in other muscles used for swallowing is unknown and requires further investigation.

In addition, 13% of stroke patients were unable to participate in the clinical assessment, resulting in a stroke sample that may be biased toward those with mild-moderate impairments. Further research is warranted before applying these results to a more severely dysphagic population. There was also a lack of detailed information about stroke lesion location. Lesion location is associated with incidence and severity of swallowing impairment.[53] However, the relationship between lesion location and strength and skill patterns is unknown. This should be explored in future studies.

Finally, there were demographic differences between the 3 diagnostic groups. Stroke patients were older, had more recent onset of dysphagia, and were more likely to be on a modified diet. Despite this, TOMASS and TWST scores demonstrated that functional swallowing ability was better in the healthy group and was not significantly different between the dysphagia groups. Differences in dysphagia duration are to be expected given the development of dysphagia in an acute lesion such as stroke,[54] compared to the chronic, progressive nature of myopathy.[55,56]

Conclusions

Four clusters of stroke patients were identified based on clinical measures of submental muscle force and skill during swallowing-related behaviors. The results suggest that both strength and skill impairments might be present, and may occur independently in stroke patients with dysphagia. Measures of skill appeared to be more predictive of cluster assignment than strength measures, suggesting that assessment of muscle-activation precision in swallowing may be an important but overlooked aspect of dysphagia rehabilitation.

Suppliers

a. Triode surface patch; Thought Technology Ltd.
b. NeuroTrac Simplex; Verity Ltd.
c. Biofeedback in Strength and Skill Training; University of Canterbury.
d. Commander PowerTrack; JTech.
e. RStudio, version 1.1.442; RStudio, Inc.

Keywords

Deglutition; Deglutition disorders; Rehabilitation; Stroke

Corresponding author

Karen Ng, PhD, The University of Canterbury Rose Centre for Stroke Recovery and Research, Leinster Chambers, Level One, 249 Papanui Road, Merivale, Christchurch 8140 New Zealand. *E-mail address:* karen.b.ng@gmail.com.

References

1. Martino R, Foley N, Bhogal S, Diamant N, Speechley M, Teasell R. Dysphagia after stroke: incidence, diagnosis, and pulmonary complications. Stroke 2005;36:2756-63.
2. Daniels SK, Huckabee M-L, Gozdzikowska K. Dysphagia following stroke. 3rd ed. San Diego: Plural Publishing; 2019.
3. Duffy JR. Motor speech disorders: substrates, differential diagnosis, and management. 2nd ed. St. Louis: Elsevier; 2005.
4. Darley FL, Aronson AE, Brown JR. Clusters of deviant speech dimensions in the dysarthrias. J Speech Hear Res 1969;12:462-96.
5. Darley FL, Aronson AE, Brown JR. Differential diagnostic patterns of dysarthria. J Speech Hear Res 1969;12:246-69.
6. Kitago T, Krakauer JW. Losing control: brain vs spinal cord. Neurology 2010;74:1250-1.

7. Wirth B, Van Hedel HJA, Curt A. Ankle dexterity remains intact in patients with incomplete spinal cord injury in contrast to stroke patients. Exp Brain Res 2008;191:353-61.
8. Van Hedel HJA, Wirth B, Curt A. Ankle motor skill is intact in spinal cord injury, unlike stroke: Implications for rehabilitation. Neurology 2010;74:1271-6.
9. Tomita Y, Usuda S. Temporal motor coordination in the ankle joint following upper motor neuron lesions. J Phys Ther Sci 2013;25:539-44.
10. Huckabee M-L, Lamvik-Gozdzikowska K. Reconsidering rehabilitation for neurogenic dysphagia: strengthening skill in swallowing. Curr Phys Med Rehabil Rep 2018;6:186-91.
11. Ertekin C. Physiological and pathological aspects of oropharyngeal swallowing. Mov Disord 2002;17:S86-9.
12. Miller AJ. The neurobiology of swallowing and dysphagia. Dev Disabil Res Rev 2008;14:77-86.
13. Huckabee M-L, Macrae P. Rethinking rehab: skill-based training for swallowing impairment. SIG 13 Perspect Swallowing Swallowing Disord 2014;23:46-53.
14. Huckabee M-L, Kelly B. [Models of rehabilitation in dysphagia management: a case for swallowing apraxia and spastic dysphagia] [German]. In: Stanschus S, editor. [Rehabilitation of dysphagia] [German]. Idstein, Germany: Schulz-Kirchner Verlag GmbH; 2006. p 137-71.
15. Huckabee M-L, Lamvik K, Jones R. Pharyngeal mis-sequencing in dysphagia: characteristics, rehabilitative response, and etiological speculation. J Neurol Sci 2014;343:153-8.
16. Daniels SK. Swallowing apraxia: a disorder of the praxis system? Dysphagia 2000;15:159-66.
17. Paik N-J, Kim SJ, Lee HJ, Jeon JY, Lim J-Y, Han TR. Movement of the hyoid bone and the epiglottis during swallowing in patients with dysphagia from different etiologies. J Electromyogr Kinesiol 2008;18:329-35.
18. Stepp C.E, Britton D, Chang C, Merati A.L, Matsuoka Y. Feasibility of game-based electromyographic biofeedback for dysphagia rehabilitation. In: Proceedings of the 5th International IEEE/EMBS Conference on Neural Engineering; April 27-May 1, 2011; Cancun (Mexico), p. 233-236.
19. Canning CG, Ada L, O'Dwyer NJ. Abnormal muscle activation characteristics associated with loss of dexterity after stroke. J Neurol Sci 2000;176:45-56.
20. Malloy JR, Valentin JC, Hands GL, et al. Visuomotor control of neck surface electromyography in Parkinson's disease. NeuroRehabilitation 2014;35:795-803.
21. Hands GL, Stepp CE. Effect of age on human-computer interface control via neck electromyography. Interact Comput 2016;28:47-54.
22. Athukorala RP, Jones RD, Sella O, Huckabee M-L. Skill training for swallowing rehabilitation in patients with Parkinson's disease. Arch Phys Med Rehabil 2014;95:1374-82.
23. Perry SE, Sevitz JS, Curtis JA, Kuo SH, Troche MS. Skill training resulted in improved swallowing in a person with multiple system atrophy: an endoscopy study. Mov Disord Clin Pract 2018;5:451-2.
24. Steele CM, Bailey GL, Chau T, et al. The relationship between hyoid and laryngeal displacement and swallowing impairment. Clin Otolaryngol 2011;36:30-6.
25. Perlman A, Grayhack JP, Booth BM. The relationship of vallecular residue to oral involvement, reduced hyoid elevation, and epiglottic function. J Speech Hear Res 1992;35:734.
26. Clark HM. Clinical decision making and oral motor treatments. ASHA Lead 2005;10:8-35.
27. Tohara H, Wada S, Sanpei R, et al. Development of a jaw-opening sthenometer to assess swallowing functions. Jpn J Gerodontol 2011;26:78-84.
28. Leonard R, Kendall K, Johnson R, McKenzie S. Swallowing in myotonic muscular dystrophy: a videofluoroscopic study. Arch Phys Med Rehabil 2001;82:979-85.
29. Palmer P, Neel AT, Morrison L. Swallowing characteristics in patients with oculopharyngeal muscular dystrophy. J Speech Lang Hear Res 2010;53:1567-79.
30. Langdon PC, Mulcahy K, Shepherd KL, Low VH, Mastaglia FL. Pharyngeal dysphagia in inflammatory muscle diseases resulting from impaired suprahyoid musculature. Dysphagia 2012;27:408-17.
31. Belafsky PC, Mouadeb DA, Rees CJ, et al. Validity and reliability of the Eating Assessment Tool (EAT-10). Ann Otol Rhinol Laryngol 2008;117:919-24.
32. Kissela BM, J.C. K, Alwell K, et al. Age at stroke. Neurol 2012;79:1781-7.
33. Dolnicar S. A review of unquestioned standards in using cluster analysis for data-driven market segmentation, Conference Proceedings of the Australian and New Zealand Marketing Academy Conference. Geelong, Australia: Deakin University; 2002.
34. Hughes TA, Wiles CM. Clinical measurement of swallowing in health and in neurogenic dysphagia. QJM 1996;89:109-16.
35. Huckabee M-L, McIntosh T, Fuller L, et al. The Test of Masticating and Swallowing Solids (TOMASS): reliability, validity and international normative data. Int J Lang Commun Disord 2018;53:144-56.
36. Tan P-N, Steinbach M, Kumar V. Cluster analysis: basic concepts and algorithms, Introduction to data mining. New York: Pearson Addison Wesley; 2005. p 487-568.
37. Rousseeuw PJ. Silhouettes: A graphical aid to the interpretation and validation of cluster analysis. J Comput Appl Math 1987;20:53-65.
38. Tibshirani R, Walther G, Hastie T. Estimating the number of clusters in a data set via the gap statistic. J R Stat Soc Ser B 2001;63:411-23.
39. Charrad M, Ghazzali N, Boiteau V, Niknafs A. NbClust: an R package for determining the relevant number of clusters in a data set. J Stat Softw 2014;61:1-36.
40. Woytowicz EJ, Rietschel JC, Goodman RN, et al. determining levels of upper extremity movement impairment by applying cluster analysis to upper extremity Fugl-Meyer assessment in chronic stroke. Arch Phys Med Rehabil 2017;98:456-62.
41. Kinsella S, Moran K. Gait pattern categorization of stroke participants with equinus deformity of the foot. Gait Posture 2008;27:144-51.
42. Mulroy SJ, Gronley JK, Weiss W, Newsam CJ, Perry JP. Use of cluster analysis for gait pattern classification of patients in the early and late recovery phases following stroke. Gait Posture 2003;18:114-25.
43. Daniels SK, Schroeder MF, McClain M, Corey DM, Rosenbek JC, Foundas AL. Dysphagia in stroke: development of a standard method to examine swallowing recovery. J Rehabil Res Dev 2006;43:347-56.
44. Daniels SK, Schroeder MF, DeGeorge PC, Corey DM, Foundas AL, Rosenbek JC. Defining and measuring dysphagia following stroke. Am J Speech Lang Pathol 2009;18:74-81.
45. Brodsky MB, McFarland DH, Dozier TS, et al. Respiratory-swallow phase patterns and their relationship to swallowing impairment in patients treated for oropharyngeal cancer. Head Neck 2010;32:481-9.
46. Ada L, O'Dwyer N, Green J, Yeo W, Neilson P. The nature of the loss of strength and dexterity in the upper limb following stroke. Hum Mov Sci 1996;15:671-87.
47. Martin-Harris B, Focht, Garand KL, McFarland D. Optimizing respiratory-swallowing coordination in patients with oropharyngeal head and neck cancer. Perspect ASHA Spec Interes Groups 2017;2:103.
48. Yeates E. Improvements in tongue strength and pressure-generation precision following a tongue-pressure training protocol in older individuals with dysphagia: three case reports. Clin Interv Aging 2008;3:735-47.
49. Garcia J, Hakel M, Lazarus C. Unexpected consequence of effortful swallowing: case study report. J Med Speech Lang Pathol 2004;12:59-66.
50. Clark HM. Neuromuscular treatments for speech and swallowing: a tutorial. Am J Speech Lang Pathol 2003;12:400-15.
51. Mann G, Hankey GJ. Initial clinical and demographic predictors of swallowing impairment following acute stroke. Dysphagia 2001;16:208-15.

52. Stepp CE. Surface electromyography for speech and swallowing systems: measurement, analysis, and interpretation. J Speech Lang Hear Res 2012;55:1232-47.
53. Suntrup S, Kemmling A, Warnecke T, et al. The impact of lesion location on dysphagia incidence, pattern and complications in acute stroke. Part 1: dysphagia incidence, severity and aspiration. Eur J Neurol 2015;22:832-8.
54. Mann G, Hankey GJ, Cameron D. Swallowing function after stroke. Stroke 1999;30:744-8.
55. Oh TH, Brumfield KA, Hoskin TL, Stolp KA, Murray JA, Bassford JR. Dysphagia in inflammatory myopathy: clinical characteristics, treatment strategies, and outcome in 62 patients. Mayo Clin Proc 2007;82:441-7.
56. Oh TH, Brumfield K a, Hoskin TL, Kasperbauer JL, Basford JR. Dysphagia in inclusion body myositis: clinical features, management, and clinical outcome. Am J Phys Med Rehabil 2008;87: 883-9.