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Adjunctive Neuromuscular Electrical Stimulation for Treatment-Refractory Dysphagia

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Objectives: Neuromuscular electrical stimulation (NMES) has been proposed as an adjunctive modality for the treatment of swallowing disorders. We present data from a prospective case series to define and measure effects of a systematic therapy for chronic pharyngeal dysphagia using adjunctive NMES.

Methods: Six adult patients with pharyngeal dysphagia received 15 sessions of a standardized protocol of swallowing exercises with adjunctive NMES. The patients completed clinical and instrumental baseline, posttreatment, and 6-month follow-up evaluations. Outcome measures included the proportion of patients who improved in clinical swallowing ability, functional oral intake, and change in body weight; patient perception of swallowing ability; and changes in kinematic aspects of swallowing.

Results: Significant change was demonstrated for clinical swallowing ability ($p < .042$), functional oral intake ($p < .02$), weight gain ($p < .026$), and patient perception of swallowing ability ($p < .043$). Hyoid and laryngeal elevation during swallowing demonstrated bolus-specific patterns of change. No patient experienced a treatment-related or swallowing-related complication. Patients (4 of 5) who were followed out to 6 months after treatment maintained functional gains.

Conclusions: A systematic therapy for chronic pharyngeal dysphagia using adjunctive NMES produced improvement in clinical swallowing ability and functional oral intake without significant weight loss or complications.

Key Words: case series, dysphagia, swallowing, treatment.

INTRODUCTION

Recently, transcutaneous neuromuscular electrical stimulation (NMES) was proposed as an adjunctive modality for the treatment of dysphagia. A search of published and presented studies identified 11 studies that incorporated NMES into the treatment for pharyngeal dysphagia.[1-11] Collectively, these studies included 206 patients treated with an NMES-based therapy. No patient complications were reported, and an average of 81% of treated patients were reported to demonstrate clinical improvement. Nine of these studies reported use of the VitalStim (model 5900, Chattanooga Group, Hixson, Tennessee) therapy protocol. However, in a recent survey,[12] we found that 90% of therapists who employed this clinical protocol used additional techniques in conjunction with the VitalStim therapy approach. One implication of this finding is that the VitalStim therapy approach is not systematic or well defined. As a result, clinicians may be applying adjunctive NMES to a variety of treatment approaches, some of which may not be well suited to take advantage of potential NMES benefits.

Although the majority of available data on the application of NMES for dysphagia therapy suggest positive outcomes in the absence of treatment-related complications, these studies uniformly suffer from poor design, limited methodology, and systematic investigator bias. Robey[13] has described a 5-phase model of clinical research structuring a series of clinical investigations leading to a comprehensive evaluation of any intervention. The initial phase in this model is to describe and measure any clinical effect of the intervention. In this regard, a phase I study should focus on a specified therapeutic outcome and delineate how this outcome is to be evaluated. Acceptable research designs to accomplish this goal include case reports, prospective case series, and retrospective studies. In a recent critique of available evidence of the VitalStim approach to dysphagia therapy, Logemann[14] supported a sequence of acceptable studies similar to that proposed by Robey.[13] Logemann[14] suggested that a series of systematic clinical investigations will lead to a clearer

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delineation of the role of NMES application in the treatment of dysphagia. Moreover, she suggests that the initial step should be a well-designed prospective case series to evaluate the potential for benefit, delineate any clinical effect, and identify any treatment-related complications.

In the present study, we report the results of a prospective case series using validated outcome measures representing different domains of treatment outcome. We specify a primary clinical outcome for the intervention, including how this outcome is to be assessed. We describe a specific and standard program of therapy that includes a consistent application of transcutaneous NMES. Finally, we report the measured clinical effect of this program of intervention across 6 patients with chronic pharyngeal dysphagia who obtained no benefit from prior therapies.

METHODS

Subjects. All patients who presented to an academic hospital outpatient dysphagia clinic over a 9-month period were screened for inclusion in the study. Selection of participants was based upon the following criteria: chronic (at least 6 months) impairment of swallowing, age 90 years or less at onset of treatment, a physician referral stating stable medical condition and ability to participate in an NMES-based treatment program, a Mini Mental State Examination (MMSE)[15] score of 23 or greater, a Functional Oral Intake Scale (FOIS)[16] score of 5 or less indicating significant limitations in functional oral intake of food and liquid, and videofluoroscopic evidence of pharyngeal dysphagia. Videofluoroscopic evidence of pharyngeal dysphagia was characterized by the presence of reduced hyolaryngeal elevation, reduced pharyngeal constriction, and/or reduced pharyngoesophageal segment opening. In addition, all study patients must have failed to respond to a previous trial of swallowing therapy. No patient could have received swallowing therapy within 3 months before participation in the present treatment study. Finally, all patients enrolled in the study were willing and able to attend daily treatment sessions for up to 3 weeks.

The local Institutional Review Board approved the study, and all enrolled patients signed an approved consent form.

Baseline Measures. Before the initiation of swallowing therapy, each patient completed a baseline evaluation to ensure inclusion criteria and to provide a pretherapy measure on outcome assessments. Baseline measures of outcome assessment included clinical and instrumental swallowing evaluation, documentation of weight, and patients' self-perception of swallowing ability. Clinical assessment of swallowing ability was completed with the Mann Assessment of Swallowing Ability (MASA).[17] Functional oral intake of food and liquid was documented with the FOIS.

An instrumental swallowing evaluation was completed via a videofluoroscopic swallowing evaluation. Instrumental data were used to confirm the presence of pharyngeal dysphagia, identify the starting food or liquid to be used in therapy, and document specific clinical indicators of airway compromise for each subject (eg, cough, throat clearing, eye watering). In addition, the fluoroscopic swallowing study was used in the evaluation of biokinematic changes following intervention. The standard materials attempted in these examinations included thin liquid, nectar thick liquid, and pudding (Varibar; E-Z-Em, Inc, Westbury, New York) in both 5-mL and 10-mL amounts. If it was deemed clinically appropriate (ie, did not place the patient at risk for airway compromise), the patients were offered a cup to drink self-selected volumes of liquids and a cracker coated with barium pudding to masticate and swallow. The sequence of materials swallowed was tailored to individual patients on the basis of clinical presentation. Attempts were made to present all materials to each patient. However, if, in the judgment of the examining speech-language pathologist and/or radiologist, the patient aspirated excessive amounts of any material, the study was terminated.

Nutritional status was estimated by body weight measured clothed but without shoes (Detecto Scale, Webb City, Missouri). To obtain a patient-focused outcome, each patient completed a perceptual evaluation of swallowing ability using a 10-mm visual analog scale and a line bisection task. For this study the continuum was "I can swallow anything I want (10)" versus "I cannot swallow at all (0)." The subjects were asked to indicate their response by marking a position on the line between the two.

Intervention. The treatment sessions were 1 hour per day, 5 days per week, for a maximum of 15 sessions. If the patient reached an adequate level of functional oral intake, which we defined as a level 6 on the FOIS (total oral diet including multiple consistencies without special preparation, but with specific avoidances or limitations), treatment could be terminated before completion of all 15 sessions. The treatment included daily transcutaneous NMES combined with functional swallowing activities. Electrical stimulation was delivered with a dual-channel electrotherapy system with pulsed current at a fixed pulse rate of 80 Hz and a pulse dura-

tion of 700 μs. (VitalStim model 5900, Chattanooga Group). A speech-language pathologist who was trained in the use of NMES provided the treatment.

Placement of Stimulating Electrodes. Before electrode placement, the skin on the anterior part of the neck of each patient was prepared with an alcohol pad to remove any substance that might interfere with electrode contact. Four stimulating electrodes, representing 2 channels, were placed on the anterior neck midline in a vertical arrangement. The thyroid notch was identified by palpation, and the first electrode was placed directly above the thyroid notch in the midline. The second electrode was placed immediately superior to the first. (Channel 1 was across the hyoid bone.) The third electrode was placed inferior to the thyroid cartilage (by palpation), and the fourth was placed directly inferior to the third. (Channel 2 was across the cricoid cartilage.)

Setting Amplitude for Neuromuscular Electrical Stimulation. Once the electrodes were positioned, electrical stimulation was introduced in an ascending-amplitude manner to determine the initial stimulation amplitude for treatment. This was determined by a combination of 1) a grabbing sensation reported by the patient, 2) palpation or visual identification of muscle contraction by the clinician, and/or 3) the presence of either voice change or an audible swallow. Any patient who demonstrated difficulty with verbal expression was offered a "stimulation ruler" system by which to indicate when the grabbing sensation had been attained. This stimulation ruler was modified from the commonly used pain ruler on which faces are used to depict different levels of intensity of stimulus. Once the target amplitude was identified, this level of stimulation was associated with a smiling face. During sessions, patients were asked to indicate at regular intervals whether the grabbing sensation fell below the optimal sensory response. If this occurred, the stimulation was increased until the patient and clinician again recognized the criteria for target amplitude.

Treatment Protocol. Each patient initially attended 2 pretreatment accommodation sessions to familiarize him or her with the equipment and procedures to be used and to establish the baseline amplitude for electrical stimulation. Accommodation sessions also served to minimize any anticipatory bias that might occur on commencing a new therapeutic activity. All subsequent treatment sessions were conducted for 1 hour and followed a standard protocol. All swallow attempts were completed under active electrical stimulation. In this protocol, a single swallowing strategy was taught to the patient to facilitate swallowing attempts, the criteria and steps for advancement were predetermined, and the program hierarchically incorporated issues of bolus volume, bolus consistency, eating rate, and amount of oral intake.

A simple swallowing technique was employed that required the patient to place the bolus in the mouth, close the mouth and breathe through the nose, and then swallow hard and fast with a single attempt. He or she was then instructed to keep the mouth closed and, if needed, inhale gently via the nose and clear the throat. This sequence was repeated until the bolus was swallowed or until the patient expectorated.

Materials. Each patient was required to progress along a preset food hierarchy that began with ice chips on the low end up to foods of the patient's preference on the high end. The starting level of material used in therapy was identified as the highest level on the hierarchy that did not cause aspiration or expectoration during the videofluoroscopic examination. From this starting point, the patients progressed through the food hierarchy according to performance and clinical impressions of safe swallowing (ie, no aspiration). Volume was increased initially, followed by type of material. If the initial material was deemed unsafe from the videofluoroscopic examination, the patient was allowed to initiate therapy using dry swallows. The final level of the food hierarchy allows the clinician to address any food avoidances, specific food difficulties, or nonfunctional behaviors such as persistent throat clearing.

Performance Monitoring. During each treatment session, the treating clinician recorded successful and unsuccessful swallow attempts. Successful swallow attempts were characterized by the absence of expectoration or clinical indicators of aspiration. Clinical indicators of aspiration (eg, coughing, throat clearing, change in respiratory rate) were noted for each patient during the fluoroscopic study, and this information was communicated in writing to the treating clinician so that he or she could monitor potential aspiration during therapy. Using these indicators, the treating clinician made on-line decisions to move forward or backward on the food hierarchy. If a patient demonstrated a successful swallow (no expectoration or clinical signs of aspiration) on 8 of 10 swallow attempts, the clinician advanced to the next-higher level on the food hierarchy. If a patient demonstrated clinical indications of aspiration on 3 of 5 swallow attempts, the clinician reverted to the next-lower level on the hierarchy.

Masking (Blinding). Intervention was administered by 2 experienced speech-language pathologists who were blind to the results of patients' baseline assessments and conducted therapy sessions as

per the predefined protocol. The only information available to the treating clinicians was a list of clinical indicators of aspiration, previously identified from instrumental baseline examinations, and the starting material and volume of food or liquid to be used during therapy.

The outcome assessor was blind to the results of the treatment sessions. This individual was not aware of the swallowing status of each patient at any evaluation, and had no information provided on progress during the treatment period.

Posttreatment Follow-Up. At the completion of therapy, all baseline evaluations were repeated to assess the immediate posttreatment outcome. Subsequently, the patients were asked to return for a repeat evaluation at 6 months after treatment.

Clinical Outcome of Treatment. The primary outcome measure for this study was the proportion of patients who improved in clinical swallowing ability, oral intake level, and body weight. Clinically meaningful improvement was defined a priori as an increase of 10 or more points on the MASA and an improvement of 2 or more points on the FOIS, without significant weight loss or dysphagia-related complications. These cut points were determined from previous data used in the development of these scales.[17]

The secondary outcome measures included a change in a patient's perception of swallowing ability over the treatment period, a change in hyoid and laryngeal elevation during swallowing, and the occurrence of any complications (eg, chest infection, dehydration).

To obtain biokinematic measures of hyoid and laryngeal elevation during swallowing, videofluoroscopic studies were digitized with a video capture board (Matrox RT.X100 Extreme; capture of approximately 30 frames per second with frame size 720 × 480 pixels) in a Core ACPI multiprocessor PC with Adobe Premier Pro digitizing software. All digitized videos were de-identified and assigned a computer-generated random number. For each material swallowed during the fluoroscopic examination, 2 video frames were selected from the digitized videos. One frame represented the resting, preswallow position of the hyoid bone and larynx. The second frame depicted the maximum upward and anterior displacement of each structure. Each picture frame was analyzed with a software measurement program (ImageJ 1.36).[18] A cursor was used to draw a line between the anterior-inferior corners of the second and fourth cervical vertebrae (C2 and C4). This line (within the image) was rotated as needed to a true vertical position. Subsequently, 3 points were marked on each digitized picture frame to measure hyoid and larynx displacement: 1) the anterior-inferior corner of C4, which served as an anchor point; 2) the anterior-inferior point of the hyoid bone; and 3) the anterior-superior subglottal corner of the tracheal air column.

The ImageJ program provides the calculated values of each point (x, y). The following formula was used to measure the extent of hyoid and larynx elevation: (y2 – y1) – (C4y2 – C4y1). In this formula, y1 and x1 represent the point coordinates in the resting position and y2 and x2 represent the coordinates at the point of maximum excursion of each structure. To convert these measures to millimeters, the vertical dimension of the third cervical vertebrae (C3) at the anterior border was measured and set to a constant distance of 15 mm with the image processing software. All resulting measures were considered relative to this calibration.

The mean and standard deviation of hyoid and laryngeal elevation during swallowing of each single bolus from the videofluoroscopic study were calculated for the baseline assessment and the posttherapy assessment.

Statistical Analysis. Patient demographics were reviewed with descriptive methods. Because of the small numbers and skewed distributions, any differences in performance noted on pretreatment and posttreatment evaluations were reviewed with nonparametric Wilcoxon signed rank tests for non-normally distributed samples. The χ^2 test was used for outcomes with discrete counts.

Estimations of clinical effects from treatment were calculated with the Hedges *g* statistic[19] for the predefined composite variable (no more than 10 points on the MASA and no more than 2 scale points on the FOIS) and for each of the primary clinical outcome measures (MASA, FOIS, weight change, patient's perception of swallowing ability). The Hedges *g* statistic was chosen to estimate clinical effect as it adjusts for sample size. Each point estimate is presented with the accompanying 95% confidence interval (CI).

Biokinematic data were compared only for those subjects who swallowed the same materials at both the pretherapy and posttherapy examinations. The subjects did not have to swallow all materials, but any given material had to have been swallowed before and after therapy by a subject to be included in the comparison. The reliability of kinematic measures was conducted with the intraclass correlation (ICC). Interjudge reliability was estimated via com-

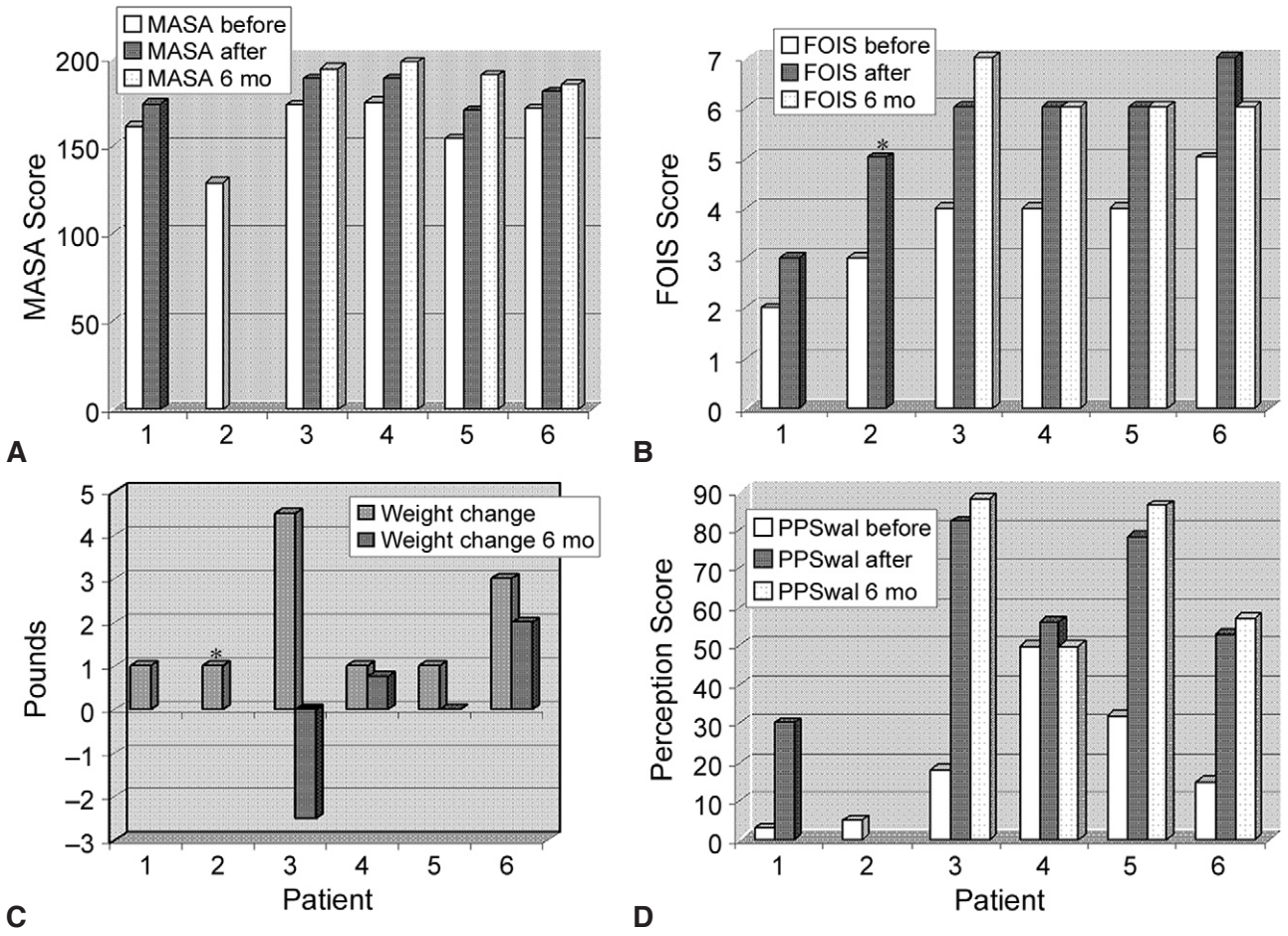

Fig 1. A) Change in Mann Assessment of Swallowing Ability (MASA) across treatment. **B)** Change in Functional Oral Intake Scale (FOIS) score after treatment. Asterisk — data point censored for time spent in study. **C)** Change in weight across treatment. Asterisk — data point censored for time spent in study. **D)** Change in patient perception of swallowing (PPSwal) across treatment.

parison of 2 independent judges' measurements of a randomly allocated sample of 20% of the swallows. Intrajudge reliability was estimated by having the same judge remeasure a randomly allocated sample of 20% of the swallows.

RESULTS

Baseline Characteristics. Six patients who presented with treatment-refractory pharyngeal dysphagia were enrolled in the study. The mean duration of dysphagia for the group was 5.1 years (range, 6 months to 15 years). Four subjects were male and 2 were female. The mean age of the group was 63.6 years (SD, 17.5; range, 33 to 85 years). Each of the patients was considered able to cognitively participate in intensive swallowing treatment; the mean MMSE score was 28.6 (SD, 2.8; range, 23 to 30). As a group, the patients demonstrated complicated medical histories. The primary diagnosis associated with dysphagia was stroke in 3 patients, head and neck cancer in 2 patients, and traumatic brain injury in 1 patient.

Follow-Up Evaluations. Immediate posttreatment data were complete for 5 of the 6 patients enrolled in the study. Over the course of the treatment, 1 patient suffered an unrelated adverse event and was withdrawn from the study. Data from this patient were censored for time spent in the study and were included in the results where possible. At 6 months after treatment, 4 of these 5 patients (80%) returned for follow-up reassessment. One subject was lost to follow-up because of advancing primary disease.

Number of Treatment Sessions and Electrical Stimulation Parameters. An average of 12 treatment sessions (range, 9 to 13) was provided to all subjects (not including the accommodation sessions). During each treatment session, the treating clinician recorded the electrical stimulation level. The mean amplitude of electrical stimulation was calculated for each session. The mean amplitude of the daily sessions for the group was 16.2 mA (range, 10 to 23 mA).

In-Treatment Performance. The average number of swallow attempts for all patients across all treat-

TABLE 1. SUMMARY OF CLINICAL EFFECTS FOR PREDEFINED COMPOSITE VARIABLE AND FOR EACH INDIVIDUAL CLINICAL OUTCOME MEASURE

Outcome Variable	*Hedges* g	*95% Confidence Interval*
Predefined composite	1.39	0.15 to 2.93
Mann Assessment of Swallowing Ability	1.34	0.59 to 1.45
Functional Oral Intake Scale	1.39	0.13 to 2.66
Weight	0.09	–1.04 to 1.22
Patient perception (visual analog scale)	1.86	0.51 to 3.21

ment sessions was 45.06 (SD, 27.5). On average, 79.3% (SD, 6.2%) of swallow attempts were considered successful. Over the course of treatment, patients advanced an average of 4.3 levels (SD, 0.52) on the 11-step food hierarchy.

OUTCOMES

Clinical Swallowing Ability (MASA). The MASA score for the group improved significantly between the pretreatment and posttreatment assessments ($p < .042$; Fig 1A). In total, 4 of 5 patients (80%) reached the defined primary end point of clinical improvement in swallowing ability. The mean MASA score increase for those 4 patients who reached this predefined end point was 21.25 (SD, 8.5). An additional patient also demonstrated an increase in the MASA score of more than 10 points; however, this patient did not advance in diet sufficiently to meet a priori criteria for clinically meaningful change.

Functional Oral Intake Scale. All 6 patients in the study significantly increased the range and amount of materials they consumed orally ($p < .02$). Five of the 6 patients (83%) raised their FOIS score by at least 2 scale points. The majority of patients progressed from a restricted single-consistency diet (level 4) to a full oral diet (level 6 or 7). In addition, 1 of the 2 patients not on an oral diet moved to oral feeding over the 3-week treatment period (Fig 1B).

Nutritional Outcomes: Weight Change. A significant weight increase was observed over the treatment period ($p < .026$). The average weight gain for the group was almost 2 lb, and 1 patient increased in weight by at least 4.5 lbs over the 3-week treatment period (Fig 1C).

Patient Perception of Swallowing. The 5 patients who completed the treatment protocol perceived significant improvement in their ability to swallow after treatment ($p < .043$). The mean increase in perception score for the group was 39.3, with a range of 6 to 64 points. No patient rated his or her posttherapy swallowing ability as lower than his or her baseline score (Fig 1D).

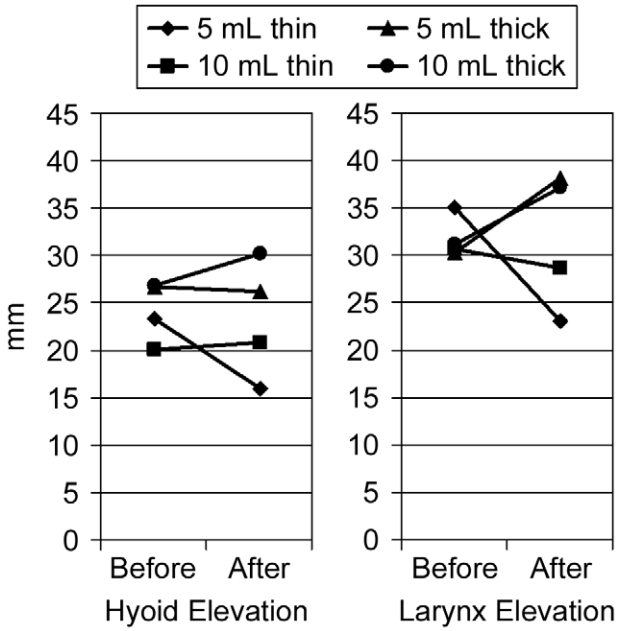

Fig 2. Biokinematic change in hyoid and laryngeal elevation across 4 liquid bolus conditions before and after therapy.

Estimation of Clinical Effect of Treatment. Moderate to large clinical effect sizes were noted for the predefined composite outcome ($g = 1.39$; 95% CI, 0.15 to 2.93) and for each individual outcome measure with the exception of weight change (Table 1). The effect size for the composite variable corresponds to a value of 92%, indicating that the average person in the NMES group would score higher than 92% of a control group that was initially equivalent.

Hyoid and Laryngeal Elevation. The only materials swallowed before and after therapy were thin and nectar thick liquids in both 5-mL and 10-mL amounts. Four patients swallowed 5 mL of thin liquid at both time points, and 3 patients swallowed the remaining 3 liquid materials at both time points.

Interjudge Reliability. Homogeneity of kinematic measurement between the 2 raters was demonstrated as an ICC of 0.89 (95% CI, 0.72 to 0.96)

Intrajudge Reliability. Consistency of kinematic measurement was demonstrated as an ICC of 0.94 (95% CI, 0.85 to 0.97).

Figure 2 presents the averages in hyoid and laryngeal elevation before and after therapy for each of these 4 materials. Both hyoid and laryngeal elevations varied across bolus volumes and materials between the baseline and posttherapy evaluations. Both structures demonstrated less elevation (8 and 11 mm, respectively) for swallowing of 5 mL of thin liquid in the posttherapy examination. Conversely,

TABLE 2. GROUP OUTCOMES OVER STUDY PERIOD

Outcome Measure	*Before Treatment*	*After Treatment*	*6-Month Follow-Up*
Mann Assessment of Swallowing Ability (mean ± SD)	160.5 ± 17.4	181.75 ± 8.5	191.7 ± 5.6
Functional Oral Intake Scale (median; range)	4; 2-5	6; 3-7	6; 6-7
Weight in pounds (mean ± SD)	144.0 ± 21.2	145.8 ± 27.6	138.0 ± 31.3
Patients' perception of swallowing (mean ± SD)	20.5 ± 17.8	59.8 ± 21.0	70.2 ± 19.6

both structures demonstrated increased elevation (4 and 7 mm, respectively) after therapy for swallowing of 10 mL of nectar thick liquid. Hyoid elevation changed little for the remaining materials (5 mL thick liquid and 10 mL thin liquid), but laryngeal elevation increased (average of 8 mm) during swallowing of 5 mL of nectar liquid after therapy. Thus, laryngeal elevation increased after therapy only for swallowing of thickened liquid materials.

Complications. No patient experienced any swallowing-related medical complication over the treatment period. One patient was withdrawn from the study after 9 treatment sessions for the occurrence of unrelated seizure activity. Swallowing treatment was terminated in this case because of side effects (increased drowsiness) from the seizure medication. The most commonly reported minor treatment complications were a burning sensation on increasing the stimulation amplitude (50%), skin irritation at the site of the electrodes (33%), a feeling of gastric fullness (33%), and neck soreness at the electrode site (17%). Coughing and expectoration were also common in session events, being noted in approximately 22% of treatment sessions.

Six-Month Follow-Up. Four of the 5 patients (80%) who completed the treatment protocol returned for review at 6 months after treatment. No statistically significant change was noted in clinical outcome measures from the posttreatment examination to follow-up (Table 2).

DISCUSSION

This study has shown that a specific program of swallowing therapy with adjunctive NMES delivered to a diverse group of patients with chronic, treatment-refractory pharyngeal dysphagia results in improved clinical and functional swallowing ability without significant weight loss or complications. Furthermore, we found that this protocol resulted in significant dietary change, including weight gain, and improved patients' perception of their swallowing ability. These clinical benefits were maintained in 4 patients out to 6 months after treatment.

Various factors may account for the clinical and functional gains realized by the patients in this study. The results of this prospective case series document positive clinical outcomes in multiple domains. However, these results do not address the relative contribution of the therapy procedures, NMES, or the combination of the two. Currently, we are evaluating outcomes of this therapy program with and without adjunctive NMES. The intention of this in-progress study is to systematically isolate and evaluate the contribution of NMES with the fixed parameters described in the present study.

Another, more pragmatic evaluation will be the comparison of the combined therapy and NMES program described in this study to more traditional dysphagia therapies. Kiger et al[7] reported no statistically significant differences between traditional dysphagia therapy techniques and the VitalStim approach to therapy. However, that report did not describe the contents of the VitalStim therapy. In view of the results of a recent large survey[12] revealing wide variation in the application of VitalStim therapy, interpretation of the negative results of that comparative study must be made cautiously. In addition, outcomes were assessed with nonvalidated and subjective scales. Thus, the study of Kiger et al[7] suffers from limitations similar to those that plague those studies claiming positive outcomes from VitalStim therapy. Delineation of the therapy process in all conditions and use of validated outcome measurement tools are important aspects of a thorough evaluation of the potential clinical benefit of adjunctive NMES in dysphagia therapy.

Physiologic change in the swallowing mechanism was assessed in reference to elevations of the hyoid bone and the larynx during swallowing. Reduction in these movements has been associated with pharyngeal deficits during swallowing. In the present study, elevation of these structures was evaluated during swallows of 4 liquid boluses. Both hyoid and laryngeal elevations for 5 mL of thin liquid were reduced after therapy. Conversely, laryngeal elevation was increased for thickened liquids only. Ludlow et al[20] demonstrated a lowering of the hyoid bone during rest in patients with pharyngeal dysphagia when transcutaneous electrical stimulation was applied. Laryngeal position did not reveal the same effect. Furthermore, swallowing performance in the subjects in the study of Ludlow et al[20] was inversely related to the degree of hyoid lowering. One speculative interpretation of that finding was that the low-

ered hyoid position facilitated a degree of resistance within the swallowing mechanism that might result in improved hyoid (and laryngeal) elevation after appropriate therapy. The results of laryngeal elevation in the present case series seem to support that speculation. Furthermore, the observation that increased elevation was noted only in the context of thickened liquids may support the concept of increased resistance in the swallowing mechanism. Thickened liquids, by virtue of their rheological properties, afford increased resistance to bolus movement and thus require more effort during swallowing.[21] To further understand the potential importance of this resistance hypothesis, these observations will need replication in a larger cohort of treated patients along with independent examination of the hypothesis.

Small-sample case series are not without limitations. Case series studies do not have internal controls and thus raise issues of patient selection and comparability with other populations. Although case series represent a relatively low level of evidence for treatment, we believe that given the level of published scientific literature on NMES for swallowing treatment, this design was an appropriate starting point to identify an effect of this specific dysphagia therapy with adjunctive NMES. The scientific sequence identified by both Robey[13] and Logemann[14] begins with a case series testing the clinical effect of a specific program of intervention. In the present report, we have defined both the behavioral treatment protocol and the NMES application protocol. The resulting clinical effect may be used to plan larger, comparative studies, and the amount of detail should permit replication by other clinical investigators.

The strengths of this study are the standardization of treatment procedures, the use of validated outcome measures, the minimization of observer bias in outcome evaluation by the blinding of the outcome assessor to the treatment progression, and the inclusion of multiple outcome measures, which reduces the potential for chance to influence the obtained outcomes. Moreover, the focus on a diverse sample of patients with chronic, treatment-refractory disorders who were not receiving additional treatments minimizes the influence of spontaneous recovery, minimizes cross-stimulation by co-occurring therapies, and offers some insights into the potential external generalization of obtained outcomes. Thus, although the results of this study do not speak to the efficacy of the described intervention, they do serve as a valid starting point for future larger and more controlled comparisons. Currently, we have small controlled studies in progress to compare this package of therapy to data-defined traditional therapy and to attempt to isolate the contribution of NMES beyond the effects of the behavioral program of intervention.

CONCLUSIONS

Results from series of carefully and systematically undertaken case studies have an important role in assembling evidence for more effective clinical practice. Case series reports provide the type of detailed preliminary findings essential to the design of higher-quality research studies. We present data from a prospective clinical series of cases treated with a specific program of dysphagia therapy with adjunctive NMES. The majority of patients treated with this approach demonstrated significant improvement in clinical and functional swallowing ability and in laryngeal elevation after intervention. Furthermore, 80% of these patients were able to maintain the clinical gains made out to 6 months after treatment. Although we acknowledge that this design is not a controlled study and does not constitute evidence of treatment efficacy, we believe the information from our case series adds significantly to the body of literature on electrical stimulation for swallowing rehabilitation and will help focus future research efforts in this area.

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