CLINICAL IMPLICATIONS OF BASIC RESEARCH

The Effects of Lingual Intervention in a Patient With Inclusion Body Myositis and Sjögren's Syndrome: A Longitudinal Case Study

Georgia A. Malandraki, PhD, CCC-SLP, Andrew Kaufman, BS, Jacqueline Hind, MS, Stephanie Ennis, MS, Ronald Gangnon, PhD, Andrew Waclawik, MD, JoAnne Robbins, PhD

ABSTRACT. Malandraki GA, Kaufman A, Hind J, Ennis S, Gangnon R, Waclawik A, Robbins J. The effects of lingual intervention in a patient with inclusion body myositis and Sjögren's syndrome: a longitudinal case study. Arch Phys Med Rehabil 2012;93:1469-75.

Objective: To report the 5-year course of a patient's swallowing disorder in the context of progressive neuromuscular disease and the effectiveness of a lingual strengthening treatment program.

Design: This is a case report that describes a lingual treatment protocol that was repeated 3 times over a 5-year period with and without maintenance periods.

Setting: The study was completed in 2 settings—an outpatient swallowing clinic at an acute care hospital and the patient's home.

Participant: The subject was a 77-year-old woman who was diagnosed with inclusion body myositis and Sjögren's syndrome.

Intervention: The patient participated in an intensive 8-week lingual strengthening protocol 3 times (at years 1, 4, and 5) and a subsequent maintenance program twice (at years 4 and 5).

Main Outcome Measures: Three outcome measures were collected during the study: (1) lingual manometric pressures at the anterior and posterior tongue, measured by using a lingual manometric device, (2) airway invasion measured by using an 8-point Penetration-Aspiration Scale, and (3) clearance of the bolus measured by using a 3-point residue scale.

Results: Isometric lingual strengthening was effective in maintaining posterior tongue lingual pressure and Penetration-Aspiration Scale scores during the treatment periods. Residue scale scores did not significantly change during treatment.

Conclusions: We conclude that, in this patient, lingual strengthening slowed the progression of disease-related lingual strength loss and extended functional swallowing performance. Thus, this type of intervention may hold promise as an effective swallowing treatment option for patients with neurodegenerative inflammatory diseases such as inclusion body myositis and Sjögren's syndrome.

Key Words: Inclusion body myositis; Longitudinal studies; Neuromuscular diseases; Rehabilitation; Swallowing.

INCLUSION BODY MYOSITIS (IBM) is the most common idiopathic inflammatory myopathy seen in individuals older than 50 years.[1,2] It accounts for approximately 30% of all inflammatory myopathies,[2,3] and it manifests as slowly progressive proximal and distal weakness. Dysphagia (ie, difficulty swallowing) is reported to occur in 21% to 65% of patients with IBM due to oropharyngeal and esophageal muscle involvement and can cause weight loss, nutritional deprivation, and even aspiration.[1,4-7] Typical dysphagia symptoms seen in patients with IBM include the feeling of pharyngeal obstruction (globus sensation) usually caused by cricopharyngeal dysfunction, airway penetration and aspiration, nasal regurgitation, reduced tongue control, and reduced laryngeal elevation during swallowing.[5,8]

Sjögren's syndrome may be observed in patients with IBM.[9] It constitutes an autoimmune rheumatic disorder affecting moisture-producing glands in the body such as the lacrimal and salivary glands.[10] Typical symptoms include xerostomia, xerophthalmia, fatigue,[10] as well as swelling of the salivary glands and tooth loss.[11] Oropharyngeal and esophageal dysphagia frequently have been reported in patients with Sjögren's syndrome.[12-14] In this syndrome, most commonly patients will experience difficulty chewing and manipulating food in the oral cavity because of xerostomia, as well as esophageal motility difficulties.[12-14]

Although the effect of strengthening and aerobic exercise in idiopathic inflammatory myopathies has been controversial[15,16] because of the concern regarding increased muscle inflammation, more recent studies report relatively positive outcomes for patients with IBM.[17-21] To date, these investigations had focused on the limb muscles.

The case reported herein focuses on a patient diagnosed with IBM and associated Sjögren's syndrome who was evaluated

List of Abbreviations

IBM	inclusion body myositis
IOPI	Iowa Oral Performance Instrument
P/A	penetration-aspiration
1RM	1 repetition maximum

From the Program of Speech and Language Pathology, Department of Biobehavioral Sciences, Teachers College, Columbia University, New York, NY (Malandraki); Geriatric Research Education and Clinical Center, William S. Middleton Memorial Veterans Hospital, Madison, WI (Malandraki, Hind, Robbins); Departments of Medicine (Malandraki, Hind, Robbins), Biostatistics and Medical Informatics and Population Health Sciences (Gangnon), and Neurology (Waclawik), University of Wisconsin, Madison, WI; F. Edward Hebert School of Medicine, Uniformed Services University, Bethesda, MD (Kaufman); and University of Washington, Seattle, WA (Ennis).

Supported in part by the Department of Veterans Affairs, Veterans Health Administration, Office of Research and Development, Rehabilitation Research and Development Service.

The contents do not represent the views of the Department of Veterans Affairs or the US government.

A commercial party having a direct financial interest in the results of the research supporting this article has conferred or will confer a financial benefit upon the author or one or more of the authors. Robbins discloses a relationship with Swallow Solutions, LLC, which is focused on clinical research in dysphagia.

Correspondence to Georgia A. Malandraki, PhD, CCC-SLP, Dept of Biobehavioral Sciences, Teachers College, Columbia University, 1052A Thorndike Hall, 525 West 120th St, New York, NY 10027, e-mail: *malandraki@tc.columbia.edu.* Reprints are not available from the author.

In-press corrected proof published online on Apr 4, 2012, at www.archives-pmr.org.

0003-9993/12/9308-00080$36.00/0
doi:10.1016/j.apmr.2012.02.010

and received treatment in the outpatient swallow clinic of an acute care hospital over an 8-year period. A formal 8-week lingual therapy protocol was implemented 3 times during the course of 5 years. This program has been effectively applied to healthy elderly adults and stroke patients with dysphagia, resulting in improved swallowing and quality-of-life outcomes.[22,23] The aim of the present case study was to determine the effectiveness of this protocol and a subsequent maintenance program in the presence of IBM and Sjögren's syndrome. Because of the relentlessly progressive nature of IBM, leading to myodegenerative changes, muscle and functional gains typically seen in healthy elders[24] and in patients with acute neural lesions (such as stroke) were not expected in this patient. However, the clinician/researcher authors hypothesized that lingual therapy, with a goal of strengthening the related musculature, would enable the patient to maintain lingual strength and functional swallowing performance during the treatment and maintenance periods.

METHODS

Patient Swallowing History

The patient was a 77-year-old woman who in 2001 began noticing progressive leg weakness and difficulty walking. Soon after the development of these initial symptoms, the patient underwent extensive evaluations in the neuromuscular clinic of the same hospital (February 2002), including a muscle biopsy (fig 1), and she was diagnosed to have IBM. The patient was also under rheumatological care for Sjögren's syndrome. She initially refused a therapeutic trial of immunotherapy, but since 2006, she has been on a small dose of methotrexate (7.5mg weekly) and prednisone (5mg daily).

A few months after her initial presentation, she began to experience swallowing difficulties, and in December 2002 she was referred to the Outpatient Swallow Service of the University of Wisconsin Hospital and Clinics for a comprehensive swallowing evaluation, including a clinical noninstrumental assessment, lingual manometric measurements, and a videofluoroscopic swallow study. Initially, the patient presented with mild symptoms, including mildly reduced upper esophageal sphincter opening and base of tongue retraction and minimal pharyngeal residue. No aspiration or penetration of the laryngeal vestibule was observed at that time. Dietary and compensatory modifications were suggested and were successful for several years. Three years later, the patient returned with more severe swallowing complaints, including delayed initiation of swallowing, and at that time, the first 8-week cycle of the standardized lingual strengthening intervention was initiated. General progression of the patient's swallowing and motor symptoms over the years, as well as times when cycles of the lingual strengthening protocol were recommended, is shown in table 1. Different interventions in addition to the lingual strengthening regimen that were attempted with the patient through the course of treatment can be viewed in the same table. In later years, the patient also started suffering from memory changes and cognitive difficulties.

In general, the patient had a steady decline in her swallowing function and eating habits, which appeared to respond to several treatment methods (some more than others) for months or years at a given time. The present study presents the descriptive results of a standardized lingual strengthening treatment protocol that was implemented at 3 time points, and herein is referred to as *the intervention*. All procedures followed were in accordance with the ethical standards of the Helsinki Declaration of 1975, as revised in 1983.

Design

The intervention was initiated and completed at 3 time points during the 5-year course. Detraining (no strengthening) was applied twice after the second course of treatment with a period of maintenance between detraining periods. Maintenance was applied twice after the second and third courses of treatment. The reason why maintenance was applied only during the second and third cycles of treatment, and detraining only after the second cycle, was because the subject was a clinical patient who was followed for many years. Initially, there was no intent to include this subject into a research protocol. The authors after reviewing this patient's progress decided to present these noteworthy data. Figure 2 displays the time course of all 3 intervention cycles.

Outcome Variables and Instrumentation

During the study, 3 key outcome variables were recorded. The first was lingual manometric pressures at the anterior and posterior tongue that were measured by using the Iowa Oral Performance Instrument[a] (IOPI), a handheld device with an

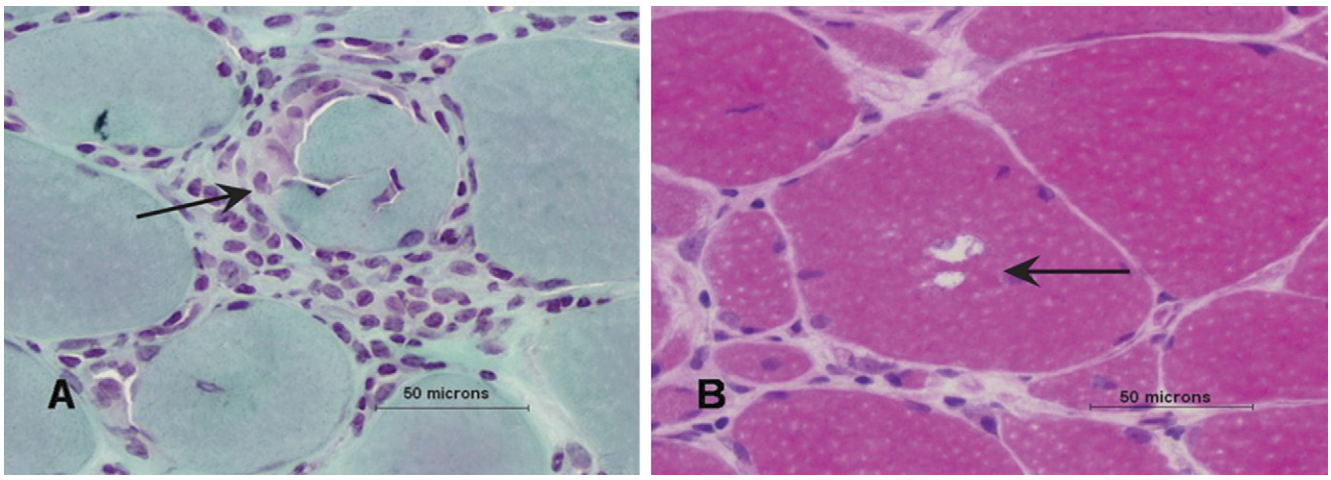

Fig 1. Muscle biopsy. (A) Inflammatory changes in endomysium. Focal invasion of a nonnecrotic muscle fiber by inflammatory cells (arrow). Modified Gomori Trichrome stain. (B) Muscle fiber containing rimmed vacuoles (arrow). Hematoxylin and eosin stain.

Table 1: General Progression of Patient's Swallowing, Eating, and Motor Status*

Date	Swallowing-Related Complaints	VFSS Findings	Lingual Manometric Pressures (range or mean)	Suggested Treatment at the Time	General Motor Ability	Eating Habits
December 2002	Sticking sensation, coughing	Reduced UES opening, reduced BOT retraction, pharyngeal residue	No pressures collected	Dietary modification, pacing during meals, treatment of xerostomia	Golfer, unassisted gait	Avoids beef, pork, and potatoes
March 2005	More severe sticking sensation, more frequent coughing	Reduced UES opening, reduced BOT retraction, pharyngeal residue, delayed pharyngeal swallow	Anterior: 37kPa Posterior: 34kPa	Lingual strengthening protocol (first cycle), CP injection	More difficulty walking	No specific food description
June 2007 July 2007	Same symptoms Symptoms exacerbated	No VFSS completed	No pressures collected Anterior: 25kPa Posterior: 22kPa	EGD, Shaker exercise	Use of a cane	1 can of nutritional supplement daily to supplement Only soft/smooth food such as yogurt/jello due to UES dilation
June 2009	Same symptoms — more severe, plus wet voice	Reduced UES opening, reduced BOT retraction, pharyngeal residue, aspiration and penetration with thin liquids, more severely delayed pharyngeal swallow	Anterior: 23–28kPa Posterior: 17–23kPa	Lingual strengthening protocol (second cycle), postural adjustments	Use of a walker	Uses L head turn most of time, takes more time to eat dry and solid foods
April 2010	Symptoms even more exacerbated	Reduced UES opening, reduced BOT retraction, pharyngeal residue, aspiration and penetration with thin liquids, more severely delayed pharyngeal swallow	Anterior: 21–25kPa Posterior: 19–21kPa	Lingual strengthening protocol (third cycle), postural adjustments	Use of a walker	No specific food description

Abbreviations: BOT, base of tongue; CP, cricopharyngeus muscle; EGD, esophagogastroduodenoscopy; L, left; UES, upper esophageal sphincter; VFSS, videofluoroscopic swallow study.
*Pressures are reported as were documented in patient's medical records.

air-filled bulb sized to simulate a 5-cm^3 bolus pressed between the tongue and the hard palate. The pressure measurements were obtained biweekly either at our clinic or at the patient's home. Normative data indicate that scores of 40kPa or above are considered within normal limits for older people.[25,26] The IOPI measurements were obtained by 3 raters (during the 3 different exercise periods) who were all trained and experienced in IOPI administration.

The other 2 outcome variables were swallowing bolus flow data, specifically the direction of bolus flow (airway invasion: aspiration or penetration) and the clearance of the bolus, measured by scores on the Penetration-Aspiration (P/A) Scale[27,28] and a residue scale,[29] respectively. These data were obtained from the videofluoroscopic swallow studies performed at baseline, weeks 4 and 8 of the intervention, and before and after detraining and maintenance periods. During the videofluoroscopic swallow evaluations, at least 4 bolus types were administered: 5mL thin liquid from a spoon, thin liquid patient-administered from a cup (Varibar Thin),[b] 5mL semisolid from a spoon (Varibar Pudding),[b] and a cookie (Wanda's Barium Cookies).[c] All swallows were recorded with the KayPentax Digital Swallowing Workstation,[d] for later analysis. The 8-point P/A Scale (1 indicating no penetration/aspiration and 2–8 indicating increasing degrees of penetration/aspiration)[27,28] was used to evaluate airway invasion during swallowing. The 3-point residue scoring system (0=no residue, 1=coating, and 2=pooling)[29] was utilized to quantify bolus clearance from the oropharynx. Scoring of swallows was performed by 2 experienced speech and language pathologists (J.H. and S.E.), and 10% of all swallows were reanalyzed by another experienced speech and language pathologist (G.A.M.). In addition, 10% of the swallows were reanalyzed by the same judges (J.H. and S.E.) to assess intrajudge reliability.

Intervention

Eight-week lingual intervention implemented at 3 time points. The intervention, completed 3 times during the 5-year period, consisted of an 8-week isometric progressive lingual strengthening program comprising compressing an air-filled bulb between the tongue and the hard palate by using the IOPI.

Baseline maximum pressures (1 repetition maximum [1RM])[30] were obtained for each tongue location (anterior and posterior) by obtaining 2 sets of data (3 IOPI trials) that differ by less than 5% to account for variability. For the swallowing bolus flow data, videofluoroscopic swallow studies were performed at all 3 baseline assessments, as well as at weeks 4 and 8 of the exercise regimens and before and after detraining and maintenance periods. During the 3 courses of treatment, the patient had agreed to not be enrolled in any other type of swallow therapy.

During week 1 of the intervention, the target value of each lingual repetition was 60% of the 1RM. For the remaining 7 weeks of the program, the target value was increased to 80% of the 1RM. At the end of weeks 2, 4, and 6, the baseline was remeasured and the 80% target value was recalculated. At those visits, there was an additional 30-minute intervention session during which the placement of the IOPI, any technical problems, and the first training session for that day was performed. The care provider/roommate also was present during this session to ensure that she knew exactly how to help the patient perform the exercises at home, if necessary.

The subject exercised the anterior and posterior parts of the tongue for 3 sets of 10 repetitions 3 times per day 3 days a week as recommended for the strength training of striated musculature by the American College of Sport Medicine.[31]

Detraining and maintenance programs. At the end of the second course of treatment in 2008, detraining was completed for 8 weeks to determine how long the patient would continue

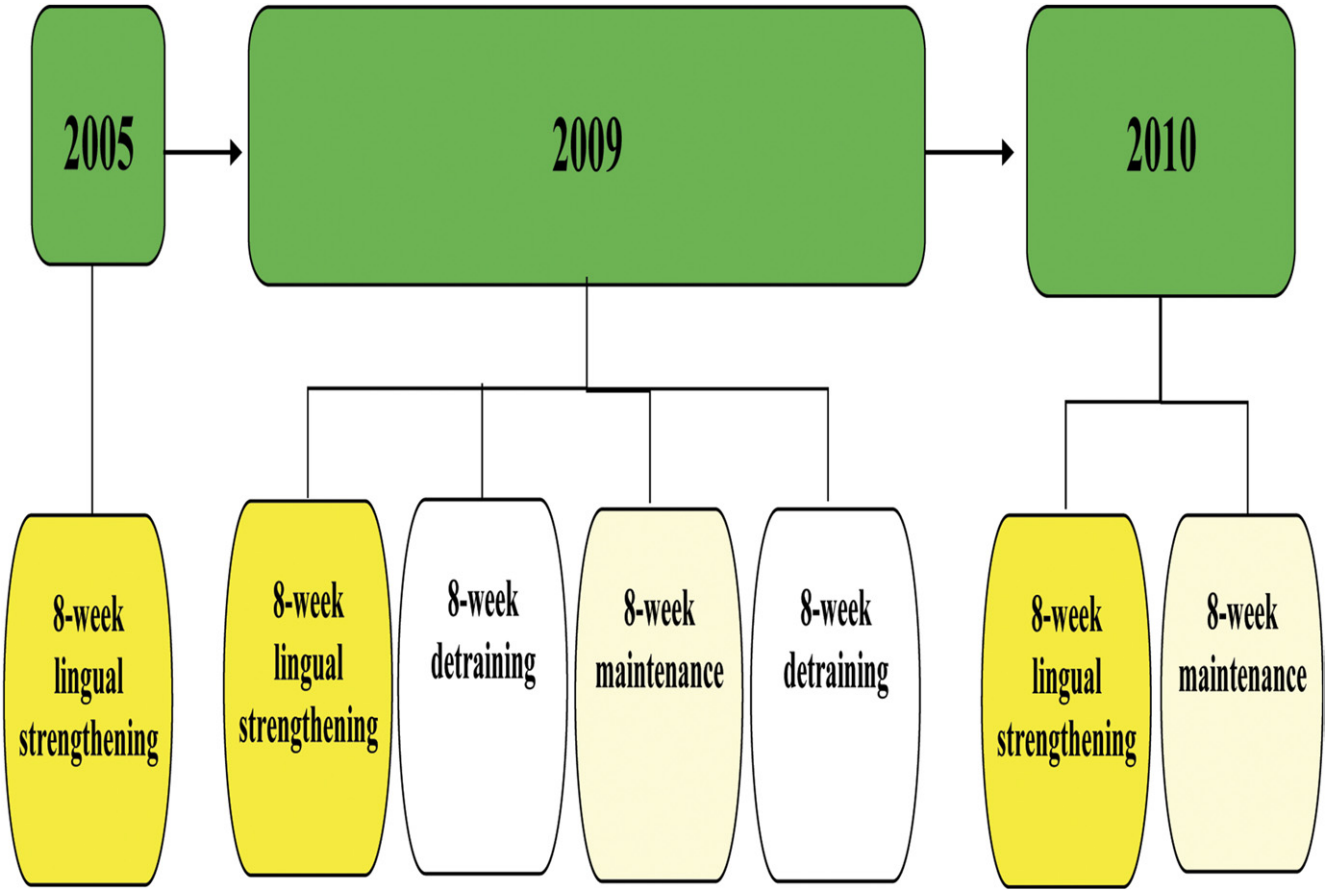

Fig 2. Timeline of all 3 interventions.

to benefit from the exercise regimen effects and if/when she would reach/decline baseline levels again. Detraining also was applied for 8 weeks postmaintenance after the second course of treatment. The maintenance program was based on previous research showing positive outcomes of maintaining skeletal muscle strength and size (knee extensors) in older men exercising at 80% of 1RM 1 day per week for 6 months, following 12 weeks of progressive resistance training.[32] Similarly, our maintenance consisted of training at a rate of 3 sets of 10 repetitions once per day for 1 day a week. The intervention was performed at 80% of maximum strength. This maintenance program was applied twice, after the second and third courses of treatment. The maximum strength was collected once at the beginning of the maintenance program and was considered the maintenance baseline value. It was then reassessed biweekly throughout the maintenance program, as well as 8 and 16 weeks following the maintenance program. Videofluoroscopic swallowing studies also were completed at baseline and at week 8 of the maintenance program.

Intervention fidelity—compliance. A practical measure of fidelity included a daily recording journal that the patient had to complete at home. In this journal, the patient recorded with a checkmark every time she was completing her exercises. Review of the journal at the end of the study showed that she had completed 90% of all her training sessions.

Reliability. Ten percent of all blinded scoring data (P/A Scale scores and residue scale scores) were reanalyzed by the same rater and a different rater (G.A.M. and J.H.) and the percentage of agreement was calculated. Interrater and intrarater reliability for the P/A scale and the residue scale scores were less than 90% within 1 scale score across all swallowing evaluations. The interrater reliability for the IOPI measurements was not calculated for the present study, because this was part of regular clinical patient care; however, previous studies have demonstrated very good interrater reliability on IOPI measurements.[33]

Statistical Analysis

Anterior and posterior isometric lingual pressure measurements for all 3 intervention periods and P/A Scale scores for the latter 2 periods were modeled by using a generalized additive regression model with smoothing parameters chosen by generalized cross-validation.[34-36] The reason P/A Scale scores for the latter 2 periods were modeled was because these 2 intervention cycles comprised both an intervention period and a maintenance period. Results are presented as estimated mean isometric pressure with pointwise 95% confidence intervals. All analyses were performed by using R.[37]

RESULTS

Isometric Lingual Pressures

Figures 3A and B show maximum anterior and posterior tongue strength, respectively, over the course of subject observation and treatment periods. The patient demonstrated signif-

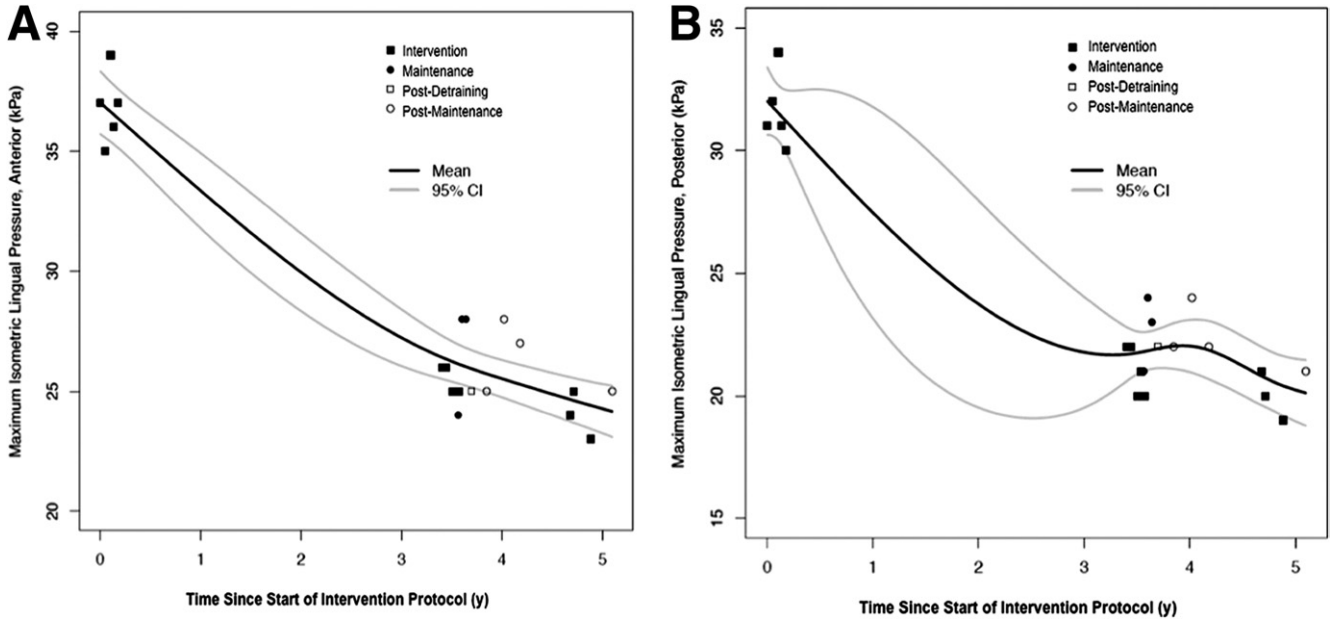

Fig 3. Actual and estimated mean isometric lingual pressure values with pointwise 95% CIs. (A) Anterior tongue. (B) Posterior tongue. Abbreviation: CI, confidence interval.

icant changes in lingual strength over time ($P < .001$ both anteriorly and posteriorly). Anterior isometric pressures declined over the entire period, but the rate of decline was lower during the last 2 years (when the subject received 2 cycles of treatment and maintenance) compared with the projected decline observed over the 3 years between the first and second cycles of treatment when the subject did not return to the clinic. Posterior isometric pressures actually stabilized during the second cycle of intervention with declines during the breaks between the cycles of intervention.

Swallowing Physiology Results

Figure 4 shows mean P/A Scale scores over the course of the latter 2 treatment periods when similar protocols were used (intervention and maintenance periods). The patient demonstrated significant variations in P/A Scale scores over time ($P = .03$). The mean P/A Scale score was stable and relatively low during the second course of treatment. There was a marked worsening in mean P/A Scale scores in the year between the second and third exercise cycles. After the third treatment period (16wk of exercise and maintenance), mean P/A Scale scores returned to the level of the prior year. Residue scale scores did not significantly change across time.

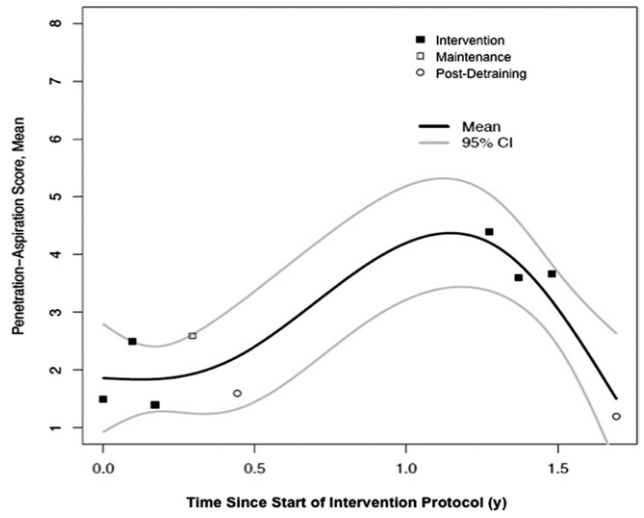

Fig 4. Actual and estimated mean P/A Scale scores with pointwise 95% CIs. Abbreviation: CI, confidence interval.

DISCUSSION

The present study shows the descriptive effectiveness of an intensive 8-week progressive isometric lingual intervention regimen in the treatment of oropharyngeal dysphagia of a patient with IBM and associated Sjögren's syndrome. To our knowledge, this is the first study supporting the role of lingual strengthening in delaying the disease progression and maintaining functional swallowing performance in a patient with these medical conditions causing multiple health challenges. Although this case report presents the data from only 1 subject, the results warrant serious consideration. The lingual strengthening intervention performed in this study previously has been shown to significantly increase strength and improve swallowing outcomes in both healthy adults and patients with stroke.[22,23] However, in the case of neuromuscular disease, it traditionally has been assumed unreasonable to expect significant gains in strength.

The effect of strengthening and aerobic exercise in idiopathic inflammatory myopathies has been considered controversial[15,16] because of apprehension over increased muscle inflammation. However, in recent years, more studies report relatively positive outcomes for patients with IBM. Researchers state that well-controlled programs of both strengthening and aerobic nature are safe and can lead to strength maintenance or even strength and muscle gains and increased quality of life in some patients with IBM without causing an increase in serum creatinine kinase concentrations or other histological markers of disease.[17-21]

Here, the subject was treated and followed over a significant period of time. Data are presented from early in the disease

process as well as the current, more advanced state. Results reveal that the patient had diminished decline progression and preservation of swallowing performance during treatment periods, as well as during maintenance. Figures 3A and B show a substantial drop in lingual pressure generation during the 3 years between the first cycle and the second cycle of the strength-building protocol when the patient refrained from treatment. Although this decline appears constant when graphed, without data during this period, the authors recognize that the slope of decline may well be less uniform. For instance, the subject could have maintained the strength from 2005 for a period up until immediately before the start of the second cycle. A search of the medical history suggests, however, that the decline in overall body strength was steady with no dramatic changes noted during that period. Taking that into consideration, it appears that the subject did, indeed, maintain her lingual strength relatively well during treatment and maintenance periods. Results show that the patient was able to maintain her mean P/A Scale scores within 1 point, except for the last treatment cycle, during which she showed improvement.

Study Limitations

Despite the promising results of the present case report, there are some limitations that need to be considered. First, this is a single case report, which significantly limits the generalizability of the results. A study with larger sample size would be necessary before these findings could be generalized to patients with IBM. Another limitation includes the missing data points during periods that the subject did not return to the clinic. This is related to the fact that this subject was a clinical care patient and initially there was no intent to include her research protocol. The authors decided to present this noteworthy data after reviewing this patient's progress. Furthermore, because of the nature of this research study, we were unable to collect interrater reliability data on the IOPI measurements. Finally, the dual diagnosis complicates the interpretation of findings. For instance, whole airway invasion decreased during strengthening periods; however, residue remained the same. The latter may be attributed to the Sjögren's syndrome, with dryness of the epithelium permitting adherence of material to the oropharynx despite the change in strength.

CONCLUSIONS

The present case report supports the role of progressive resistance isometric lingual strengthening regimens in the maintenance of lingual pressure generation and swallowing performance for patients with neuromuscular diseases such as IBM, even in the presence of associated Sjögren's syndrome. Further investigation is warranted to determine whether this treatment can be generalized to other patients with the same underlying pathology, as well as the dose-response–specific information including optimal frequency and intensity of strengthening interventions for patients with neuromuscular diseases.

Acknowledgments: We thank Abby Duane, BS, for helping with manuscript preparation and Susi Nehls, BS, for her editing contributions.

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Suppliers

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