Expiratory Muscle Strength Training: Speech Production Outcomes in Patients With Multiple Sclerosis

T. Chiara, PhD, PT, D. Martin, PhD, PT, and C. Sapienza, PhD, CCC-SLP

Purpose. This study investigated the effect of expiratory muscle strength training (EMST) on voice production, dysarthria, and voice-related quality-of-life issues in persons with multiple sclerosis (PwMS). It was hypothesized that PwMS would have improved voice production and reduced voice-related quality-of-life issues following EMST. *Participants and Methods.* Seventeen participants with MS and 14 healthy (H) controls completed 8 weeks of EMST, followed by 4 weeks of no training. Analyzed outcomes as a function of EMST were maximal expiratory pressure (MEP), sustained vowel prolongation (SVP), words per minute (WPM) measured from connected speech, and quality-of-life indices related to the presence of the dysarthria and dysphonia. *Results.* PwMS had lower MEPs, shorter SVP, and less WPM than the controls prior to training. Following EMST, both groups had significant improvement in MEPs that stayed above baseline after training halted. EMST did not improve voice production or voice-related quality of life for PwMS. *Conclusion.* Respiratory muscle weakness is present in PwMS having mild- to moderate-level disability. EMST improved expiratory muscle strength but did not statistically change objective and subjective components of voice/speech production in PwMS.

Key Words: *Multiple sclerosis—Maximum expiratory pressure—Expiratory muscle strength training—Speech—Voice production—Voice-related quality of life.*

Multiple sclerosis (MS), a disease of neurogenic origin within the white and gray matter of the central nervous system, may affect the respiratory muscles, particularly the expiratory muscles and the laryngeal muscles.[1-5] Demyelination and axonal damage, present from the earliest stage of the disease, account for the progressive disability.[2,6,7] Dysfunctional neural control of the expiratory muscles results in generating less than adequate pressure for speech production, particularly for speech tasks that require sustained phonation or for connected speech that involves finer control of inspiratory and expiratory timing.[8-12]

Previous studies of respiratory muscle strength training in persons with multiple sclerosis (PwMS) have had mixed findings as to the change in respiratory muscles, including 1) no gains,[13] 2) relative gains that were not statistically significant,[14] and 3) relative gains that were statistically significant.[15-17] The type of training modality, which respiratory muscle group(s) received the training, and the level of disability of the participants varied in these studies. Prior to this current study, no investigation had examined respiratory muscle strength in PwMS early in the disease, the effect of respiratory muscle strength training on voice/speech production, or voice-related quality of life in PwMS.

Impaired communication in PwMS may occur because of difficulty in controlling the quality of the voice and in articulating words due to motor impairment in the muscles controlling speech as well as having insufficient subglottal pressure. Speech impairments in PwMS include motor control dysfunction such as dysarthria, particularly spastic, ataxia, or mixed dysarthria.[4,18-22] The earliest description of speech impairment associated with MS was scanning speech.[4] The symptoms of the speech production disorder can include abnormally long pauses between words or individual syllables of words. Words may be slurred or pronounced with a hypernasal sound quality.[21] In addition, impaired sound pressure generation in PwMS may occur due to the inability to generate sufficient expiratory muscle force. Weak phonation and disturbances of the respiratory cycle are 2 of 5 major characteristics of the speech impairment in PwMS, resulting from weakened expiratory and/or laryngeal muscles.[23]

The purpose of this study was to examine the effect of expiratory muscle strength training (EMST) on maximal expiratory pressure (MEP), a known index of expiratory muscle strength, and on certain features of voice/speech production in PwMS including prolongation of a vowel

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and production of words per minute (WPM) during the reading of a standardized passage. Evidence from strength-training paradigms used with the limbs indicates that individuals who start a program at a lower level of function have a larger capacity toward improvement.[24] Including the healthy (H) control group as a comparison group allowed us to ascertain if this finding was similar to the response of limb muscle.

It was hypothesized that PwMS would have a lower MEP than H controls prior to training. Second, both PwMS and H controls would significantly increase MEP following 8 weeks of training, but the PwMS would have a significantly greater training effect than the H controls. Third, following EMST, the PwMS would improve vowel prolongation and WPM during reading of a standardized passage. Increased expiratory pressure during reading would allow for a greater number of words to be produced per breath with fewer pauses by providing an active mechanism for generating pressure during speech. It has been reported that PwMS often have vocal fold weakness as evidenced by vocal fold bowing.[25,26] Loss of airflow at the glottis results in decreased laryngeal airway resistance thus making it more difficult to coordinate breath flow during speech. By increasing expiratory pressure during speech, a potential compensatory mechanism may exist allowing the PwMS to produce a greater number of words per minute. Fourth, the improvement in MEP for the PwMS would significantly correlate with improvements in voice-related quality-of-life measurements following the training. Finally, it was hypothesized that MEP would remain above baseline value following a period of no training for both the PwMS and H control groups.

METHODS

Participants

Seventeen participants with MS (3 males) and 14 H controls (2 males) participated in this study. Table 1 presents the demographic data for all participants.

Inclusion/Exclusion Criteria

Participants were accepted into the study if they were 20 to 59 years of age; had no known cardiac, pulmonary (eg, asthma, chronic obstructive pulmonary disease, pulmonary fibrosis), neurological (other than MS), or orthopedic conditions that adversely affected the respiratory muscle system or gas exchange system; were nonsmokers or had not smoked within the past 5 years (previous smokers had to produce a forced expiratory volume in the first second over a forced vital capacity of greater than 75%); had never received speech therapy for any speech or voice problem; and were native speakers of English. PwMS were required to have a definite diagnosis of MS as per Poser's criteria,[27,28] mild (1 to 3.5) to moderate (4 to 6.5) disability as assessed by the Expanded Disability Status Scale (EDSS),[29] no exacerbation in the 3 months prior to the initiation of the study, and sufficient facial muscle strength to achieve and maintain lip closure around a circular mouthpiece.

Table 1. Demographics of Persons With Multiple Sclerosis and Healthy Controls

	Persons With MS	Healthy Controls
Age (years)	48.9 ± 7.61	44.1 ± 7.64
Height (cm)	167.3 ± 9.0	165.7 ± 7.3
Weight (kg)	76.2 ± 20.7	78.1± 13.8
Characteristics of Persons With MS		
EDSS[a] score	3.62 ± 1.31 (1.5-6.5)	
Years from first symptom	11.76 ± 6.56 (1.25-24.25)	
Years from diagnosis	8.43 ± 6.17 (1.25-18)	

Mean ± SD, numbers in parentheses are ranges. MS = multiple sclerosis; EDSS = Expanded Disability Status Scale.
a. Expanded Disability Status Scale: 0 = *normal neurology* to 10 = *death due to MS*.[29]

Participant Attrition

One H control withdrew, and 5 PwMS withdrew or did not qualify for the study secondary to exacerbation or EDSS score greater than 6.5.

PROCEDURES

All participants were seen for 3 assessment sessions (pretraining, posttraining, and detraining) and once per week across the 8-week training phase of the protocol. To minimize the possibility of fatigue in the PwMS, each of the 3 main assessment sessions were completed in 2 visits whereas for the H controls the assessments were completed in 1 visit.

Maximal Expiratory Pressure

Maximal expiratory pressure, measured in cm H_20, was assessed using a handheld digital manometer (Micro Mouth Pressure Meter, MP01, Micro Direct Inc, Auburn, Me). Each maneuver was performed while standing. The participant, with the nose closed by a nose clip, inhaled fully to total lung capacity, placed the mouth around a disposable mouthpiece, then blew the air out as hard and fast as possible directly into the manometer. Participants

Table 2. Amyotrophic Lateral Sclerosis Severity Scale: Speech Scale

Speech	Ability	Points
Normal speech processes	Normal	10
Nominal abnormalities		9
Detectable speech disturbances	Perceived speech changes	8
Obvious speech abnormalities		7
Intelligible with repeating	Repeats message on occasion	6
Frequent repeating required		5
Speech combined with nonverbal communication	Speech plus nonverbal communication	4
Limits speech to one word response		3
Loss of useful speech	Vocalizes for emotional expression	2
Nonvocal		1
Tracheostomy		X

Other speech severity scales such as the Amyotrophic Lateral Sclerosis Functional Rating Scale have only one question addressing speech impairment in individuals with neurological problems.

were verbally coached strongly to produce the expiratory maneuver to the best of their ability. The task was performed a minimum of 3 to a maximum of 10 times. Previous investigators have recommended that as many as 10 times are needed to achieve proficiency in performing maximal maneuvers.[30] The top 3 measurements were averaged for the data analysis.

Acoustic Measures

Each participant was positioned in front of a digital to analog tape recorder (model: DTC-ZA5ES, Super Bit Mapping, Sony Corporation of America, New York, NY) connected in series to a microphone preamplifier (model 760x, dbx Professional Products, Sandy, UT) wearing a cardioid headworn microphone (model: SHURE, Shure Inc, Niles, Ill) placed 1 inch from the right corner of their mouth. Speech tasks were performed 3 times. Comfortable and loud sound pressure levels (SPL), the loud SPL (LSPL) task being twice as loud as the comfortable SPL (CSPL), include 1) sustained production of /a/ and 2) reading the standardized passage titled "My Grandfather."[4,31] Order of the tasks was randomized. Twice-as-loud level was a subjective choice of each participant, that is, what they felt was double their comfortable level of loudness during talking. All recordings were analyzed by digitizing the acoustic signal through a Santa Cruz sound card (Turtle Beach, Connected Audio, Yonkers, NY) and analyzed with Abode Audition (Syntillium Software Corporation, Phoenix, AZ).

Total sound duration, defined as the total time the vowel was audible, was measured from the sustained vowel production. Oscillographic display of the vowel production allowed for easy identification of vowel onset and offset. The mean value of the sustained vowel duration from the 3 trials of the CSPL and 3 trials of the LSPL was used for data analysis. The words per minute (WPM) value was obtained from the number of words in the passage divided by the time to read the passage multiplied by 60 sec/min, that is (200 words per passage/45 sec to read the passage) × (60 sec/min). The mean value of the WPM from 3 trials of the CSPL and 3 trials of the LSPL was used for data analysis.

Questionnaires

No MS disease–specific questionnaire exists as to the effect of dysarthria or influence of one's voice on the quality of life in PwMS. As dysarthria and the influence of one's voice on quality of life were outcomes of interest in this study, the Amyotrophic Lateral Sclerosis Severity Scale (ALSSS)[32-34] and the Voice-Related Quality of Life (VRQOL) measure were used.[35,36] The questionnaires used have high validity and reliability in other neurological disorders. However, these questionnaires have not been used previously with PwMS.

Dysarthria Measurement

Amyotrophic Lateral Sclerosis Severity Scale–Speech Subsection. The ALSSS was developed to provide an ordinal staging system and a means of rapid functional assessment for patients with ALS. The scale allows an examiner to evaluate symptoms numerically in speech impairment (Table 2).[32-34]

Quality of Life Measurement

Voice-Related Quality of Life Measure. The VRQOL measure was used to determine if voice impairment affected the participants' quality of life,[35,36] The VRQOL was used to measure the change in perception of one's voice on one's quality of life following EMST (Table 3). A total score (VT) and 2 subscale scores, physical (VP) and

Table 3. Description of Voice-Related Quality of Life Measure

Because of My Voice	How Much of a Problem Is This?
1. I have trouble speaking loudly or being heard in noisy situations	1 2 3 4 5
2. I run out of air and need to take frequent breaths when talking	1 2 3 4 5
3. I sometimes do not know what will come out when I begin speaking	1 2 3 4 5
4. I am sometimes anxious or frustrated (because of my voice)	1 2 3 4 5
5. I sometimes get depressed (because of my voice)	1 2 3 4 5
6. I have trouble using the telephone (because of my voice)	1 2 3 4 5
7. I have trouble doing my job or practicing my profession (because of my voice)	1 2 3 4 5
8. I avoid going out socially (because of my voice)	1 2 3 4 5
9. I have to repeat myself to be understood	1 2 3 4 5
10. I have become less outgoing (because of my voice)	1 2 3 4 5
The overall quality of my voice during the last 2 weeks has been (please circle)	
Poor Fair Good Very Good Excellent	

Written instructions: We are trying to learn more about how a voice problem can interfere with your day-to-day activities. On this paper, you will find a list of possible voice-related problems. Please answer all questions based on what your voice has been like over the past 2 weeks. There are no "right" or "wrong" answers. Considering both how severe the problem is when you get it and how frequently it happens, please rate each item below on how bad it is (that is, the amount of each problem that you have). Use the following scale for rating the amount of the problem; 1 = none, not a problem, 2 = a small amount, 3 = a moderate (medium) amount, 4 = a lot, 5 = problem is as "bad as it can be."[35,36]

social (VS), are presented as percents. The higher the percentage, that is, 100% compared to 75%, indicates less impact of voice on quality of life. Per the VRQOL guidelines, participants were to rate the influence of their voice based only on the 2 weeks prior to completing the questionnaire.[35,36] All variables of interest were examined over time and between the PwMS and the H controls.

Training and Weekly Assessments

Participants trained 5 days per week, once under the supervision of an investigator (TC) and 4 times at home with no supervision. Participants used a Positive Expiratory Pressure (PEP) threshold trainer (Threshold®PEP, Healthscan Products Inc, division of Healthdyne Technologies, Marietta, Ga). The investigators modified the Threshold®PEP trainer by exchanging the standard spring with springs that provided 4 and 8 times the normal resistance. The range of the modified trainers was 16 to 160 cm H_2O pressure. Participants were instructed to breathe in and out through the unit, maintaining their exhalation for at least 5 seconds, and to rest a minimum of 30 seconds to a minute between sets. Participants trained for 8 weeks, completing 4 sets of 6 repetitions once per day. One of the PwMS trained twice per day completing 2 sets of 6 repetitions per session after the third week of training secondary to increased fatigue. His change in MEP, vowel prolongation, and WPM were comparable to the other PwMS, so his data were included in the analyses. Weekly assessments were used to evaluate training (ie, correct use of the training tool and to remeasure the participants' MEP to adjust the training intensity). Training intensity was controlled at a set percentage of MEP, that is, 40% the first week, 60% the second week, and 80% the third through the eighth week. MEP was assessed identical to that during the 3 primary assessment sessions. Three to 7 measurements were taken, with the mean of the top 3 measurements within 10% of each other used for the adjusted intensity. Participants completed a training log, which requested information on number of sets and repetitions, rating of perceived exertion, date, and any anecdotal comments. Training compliance as measured by the participants' logs ranged from 90% to 100%.

Detraining

Participants returned their trainer to the principal investigator and were not exposed to the training protocol for 4 weeks. Participants were reassessed within 72 hours after completion of the detraining phase. The same assessment protocol was followed as outlined for the pre- and posttraining phases.

Statistical Analysis

Statistical analysis was completed using SuperANOVA and Statview (Abacus, Carmel, Calif). A 1-way analysis of variance (ANOVA) was used to examine demographic data, that is, age, height (in centimeters [cm]), and weight (in kilograms [kg]) between the 2 groups. A multiple regression analysis was used to examine the relationship between the demographic variables, MEP and delta MEP (ie, the change in MEP from pretraining to posttraining) in all participants as well as EDSS score, years from first symptom, and years from diagnosis in the PwMS. Gender,

Table 4. Maximal Expiratory Pressure

Group Effect		
PwMS	Healthy Controls	
80.55 ± 21.00	130.66 ± 41.58	
F = 23.90, *P* = .0001		
Assessment Effect: Means Comparison by Contrast		
Pretrain	Posttrain	Detrain
85.73 ± 36.45	114.73 ± 42.36	109.09 ± 37.60
F = 53.28, *P* = .0001		
Percent Change in MEP Between Assessments[a] Means Comparison by Contrast		
Pretrain – Posttrain	Posttrain – Detrain	Pretrain – Detrain
F = 95.08, *P* = .0001	*F* = 3.77, *P* = .057	*F* = 60.99, *P* = .0001
Percent Change in MEP Following Training		
PwMS = 40.4%	Healthy Controls = 29.2%	
Difference Between Assessments[b]		
Pre-Post vs Post-De	Pre-Post vs Pre-De	Post-De vs Pre-De
F = 59.22, *P* = .0001	*F* = 1.76, *P* > .05	*F* = 40.59, *P* = .0001

The interaction between group × assessments was not significant ($P > .05$). PwMS = persons with multiple sclerosis; MEP = maximal expiratory pressure; Pre = pretrain; Post = posttrain; De = detrain.
a. For example, [(posttraining-pretraining)/pretraining] × 100.
b. For example, (posttraining-pretraining) vs (posttraining-detraining).

a strong factor in distinguishing respiratory pressures between men and women, was not included in the regression analysis, as there were so few male (3 and 2) compared to female (14 and 12) PwMS and H, respectively. A 2-way repeated-measures ANOVA having a within-factor of assessment (pretraining, posttraining, and detraining) and 1 between-factor of group (PwMS and H) was used to examine MEP and the questionnaire data. A 3-way repeated-measures ANOVA having the within-factors of assessment and SPLs and the between-factor of group was used to examine the speech task acoustic data. Univariate analysis was performed on main effects if no interaction was found. Contrast by means comparisons was used to determine the significance between the levels in variables with greater than 2 levels. Significance level was set at a P value ≤ .05.

RESULTS

Demographics

There was no statistical difference in age, height, or weight between the PwMS and the H control groups. Regression analysis found that of the independent variables, group (ie, PwMS and H controls) had the greatest influence on MEP (R^2 = .395, adjusted R^2 = .368). Within the PwMS group, the EDSS score and years from first symptom were found to have the greatest influence on MEP (R^2 = .384, adjusted R^2 = .326). However, no independent variable had significant influence ($P > .05$) on the change in MEP.

Maximal Expiratory Pressure

Table 4 presents the mean ± standard deviation, the F value, and the P value for MEP for the main effects. Prior to and following EMST, the PwMS produced lower MEPs than the H controls. The difference in MEP from pretraining to posttraining for the PwMS was 26.04 ± 16.71 (40.4%) and for the H controls 32.14 ± 21.88 (29.2%). The difference in MEP within the PwMS separated as to level of disability found those with mild disability (37.73%) improved less than those with moderate disability (44.01%). The percent change in MEP ([posttrain – pretrain]/pretrain) × 100) was significantly different between the pre- to posttraining and pre- to detraining compared to posttraining to detraining.

Duration of Sustained Vowel Prolongation

Table 5 presents the mean ± standard deviation, the F value, and the P value for vowel prolongation. Interactions were found for SPL and group and for SPL and assessment. Sustained vowel prolongation was significantly shorter for both the PwMS and the H controls

Table 5. Sustained Vowel Prolongation

Interaction of Sound Pressure Level and Group			
PwMS – C	Healthy Controls – C	PwMS – L	Healthy Controls – L
12.22 ± 3.96	14.86 ± 4.99	14.76 ± 6.22	20.77 ± 8.52
$F = 5.07$, $P = .032$			

Unpaired *T* Test					
PwMS – Healthy Controls vs CSPL			PwMS – Healthy Controls vs LSPL		
Pretrain	Posttrain	Detrain	Pretrain	Posttrain	Detrain
$t = 1.232$ $P > .05$	$t = 2.037$ $P = .0509$	$t = 1.936$ $P > .05$	$t = 2.718$ $P = .0110$	$t = 2.375$ $P = .0244$	$t = 2.081$ $P = .0467$

Interaction of Sound Pressure Level and Assessment					
Pre – C	Post – C	De – C	Pre – L	Post – L	De – L
13.38 ± 4.86	13.60 ± 3.81	13.37 ± 5.28	16.22 ± 7.09	17.33 ± 6.96	19.14 ± 9.49
$F = 5.28$, $P = .0079$					

Univariate Test Between Sound Pressure Level and Assessment				
Pretrain C	Pretrain L	Post C	Post L	Detrain C
to pre L $F = 20.88$	to post C $F = 17.71$	to post L $F = 34.53$	to detrain C $F = 38.10$	to detrain L $F = 81.68$
to post L $F = 38.91$	to detrain C $F = 20.29$	to detrain L $F = 76.42$	to detrain L $F = 8.21$	
to post L $F = 82.86$	to detrain L $F = 20.55$	to detrain C NS, $P > .05$		
to post C NS, $P > .05$	to post L NS, $P > .05$			
to detrain C NS, $P > .05$				

The interactions between group × assessment × SPL and between group × assessment were not significant ($P > .05$). All univariate tests for the interaction between assessment × SPL were significant at $P = .0001$ except post L vs detrain L, $P < .0059$. Cells with strikethrough lines had no univariate comparisons. PwMS = persons with multiple sclerosis; C = comfortable; L = loud; SPL = sound pressure level.

during CSPL compared to the LSPL. Sustained vowel prolongation was significantly short at each assessment during the LSPL and at the posttraining assessment during the CSPL for the PwMS compared to the H controls. Whereas the CSPL returned toward baseline level at detraining, the LSPL increased further at the detraining assessment.

Words per Minute

Table 6 presents the mean ± standard deviation, the *F* value, and the *P* value for WPM. An interaction between group and SPL as well as a main effect for assessment were found. PwMS had no significant difference in the WPM at the CSPL and LSPL, whereas the H controls produced significantly more WPM at the CSPL compared to the LSPL. Significantly greater WPM was produced at the posttraining and detraining assessment sessions compared to the pretraining assessment, whereas a nonsignificant decrease was found from posttraining to detraining.

Questionnaires

Amyotrophic Lateral Sclerosis Severity Scale. Table 7 presents the contrast by means comparison of the difference in ALSSS scores in the PwMS as a combined group and in the mild and moderate subgroups, based on the EDSS scores.[29] Dysarthria, expected only in the PwMS if at all, was significantly greater compared to the H controls. Dysarthria of the mild and moderate groups had greater impact due to dysarthria on their lives than the H controls. Posttraining the PwMS reported significantly less impact of dysarthria. Although impact on

Table 6. Connected Speech: Reading "My Grandfather" Passage—Words per Minute

<table>
<tr><th colspan="6">Interaction Between SPL · Group</th></tr>
<tr><td>PwMS – CSPL</td><td colspan="2">Healthy Controls – CSPL</td><td colspan="2">PwMS – LSPL</td><td>Healthy Controls – LSPL</td></tr>
<tr><td>178.74 ± 29.22</td><td colspan="2">213.36 ± 22.15</td><td colspan="2">175.83 ± 28.57</td><td>199.19 ± 20.15</td></tr>
<tr><td colspan="6">F = 22.32, P = .0001</td></tr>
<tr><td colspan="3">PwMS CSPL vs LSPL</td><td colspan="3">Healthy Controls CSPL vs LSPL</td></tr>
<tr><td colspan="3">F = .33, P > .05</td><td colspan="3">F = 61.213, P = .0001</td></tr>
<tr><th colspan="6">Unpaired T Test Between Group and SPL Across Time</th></tr>
<tr><td colspan="3">PwMS – Healthy Controls vs CSPL</td><td colspan="3">PwMS – Healthy Controls vs LSPL</td></tr>
<tr><td>Pretrain</td><td>Posttrain</td><td>Detrain</td><td>Pretrain</td><td>Posttrain</td><td>Detrain</td></tr>
<tr><td>t = 4.387
P = .0001</td><td>t = 3.736
P = .0008</td><td>t = 3.193
P = .0037</td><td>t = 2.401
P = .0230</td><td>t = 2.789
P = .0092</td><td>t = 2.320
P = .0285</td></tr>
<tr><th colspan="6">Main Effect for WPM at Each Assessment</th></tr>
<tr><td colspan="2">Pretraining</td><td colspan="2">Posttraining</td><td colspan="2">Detraining</td></tr>
<tr><td colspan="2">185.12 ± 27.65</td><td colspan="2">193.18 ± 30.40</td><td colspan="2">197.04 ± 29.52</td></tr>
<tr><td colspan="6">F = 15.05, P = .0001</td></tr>
<tr><th colspan="6">Means Comparison for Time by Contrast</th></tr>
<tr><td colspan="2">Pre – Posttraining</td><td colspan="2">Post – Detraining</td><td colspan="2">Pre – Detraining</td></tr>
<tr><td colspan="2">F = 13.22, P = .0006</td><td colspan="2">F = 3.03, P > .05</td><td colspan="2">F = 28.91, P = .0001</td></tr>
</table>

The interactions between group vs assessment vs SPL, group vs assessment, and assessment vs SPL were not significant ($P > .05$). SPL = sound pressure level; PwMS = persons with multiple sclerosis; C = comfortable; L = loud; WPM = words per minute.

Table 7. Dysarthria in Individuals With Multiple Sclerosis

<table>
<tr><th colspan="6">PwMS as Group Compared to Healthy (H) Controls</th></tr>
<tr><td colspan="6">PwMS vs Healthy Controls: F = 13.32, P = .001</td></tr>
<tr><th colspan="6">Individuals With MS Separated by Level of Disability</th></tr>
<tr><td colspan="3">Mild (MI) PwMS</td><td colspan="3">Moderate (MO) PwMS</td></tr>
<tr><td colspan="3">F = 4.45, P = .04</td><td colspan="3">F = 9.15, P = .004</td></tr>
<tr><th colspan="6">Contrast by Means Comparison for Both Subgroups at Each Assessment</th></tr>
<tr><td>Pre MI</td><td>Post MI</td><td>De MI</td><td>Pre MO</td><td>Post MO</td><td>De MO</td></tr>
<tr><td>to post MI
F = 4.45
P = .0396</td><td>to de MI
NS
P > .05</td><td>to per MI
NS
P > .05</td><td>to post MO
F = 9.15
P = .0038</td><td>to de MO
NS
P > .05</td><td>to per MO
F = 12.46
P = .0009</td></tr>
<tr><td>to pre MO
F = 6.89
P = .0112</td><td>to post MO
NS
P > .05</td><td>to de MO
NS
P > .05</td><td>to pre H
F = 26.77
P = .0001</td><td>to post H
NS
P > .05</td><td>to de H
NS
P > .05</td></tr>
<tr><td>to pre H
F = 7.24
P = .0095</td><td>to post H
NS
P > .05</td><td>to de H
F = 5.03
P = .0290</td><td></td><td></td><td></td></tr>
</table>

A main effect for group was found with combined data for all PwMS. An interaction was found between group × time when the data for the PwMS were separated as to level of disability based on their EDSS score. PwMS = persons with multiple sclerosis; NS = not significant; MS = multiple sclerosis; Pre = pretrain; Post = posttrain; De = detrain.

one's life returned toward baseline ALSSS value for those with mild disability during the detraining period, the impact continued to be significantly less for those with moderate disability after the detraining period, with no significant difference found between them and the H controls.

Voice-Related Quality of Life Measure. Table 8 presents the data of VRQOL scores in the PwMS as a combined group and in the mild and moderate subgroups compared to the H controls. The VRQOL score (VT) as well as the subscale scores for social (VS) and physical (VP) were significantly lower in PwMS than the H controls at

Table 8. Voice-Related Quality of Life in PwMS and Healthy Controls

Total Score		*Social Subscore*		*Physical Subscore*	
PwMS	Healthy	PwMS	Healthy	PwMS	Healthy
85.42 ± 16.20	98.21 ± 3.44	90.01 ± 18.13	99.36 ± 2.79	81.81 ± 16.35	97.01 ± 5.89
F = 9.11, P = .005		F = 4.17, P = .051		F = 12.76, P = .001	
Voice-Related Quality of Life in PwMS Separated as to Level of Disability					
Total Score		*Social Subscore*		*Physical Subscore*	
Mild	Healthy	Mild	Healthy	Mild	Healthy
87.30 ± 14.06	98.21 ± 3.44	92.96 ± 16.35	99.36 ± 2.79	82.40 ± 15.17	97.01 ± 5.89
F = 5.02, P = .0335		F = 1.46, P > .05 (NS)		F = 8.13, P = .0082	
Moderate	Healthy	Moderate	Healthy	Moderate	Healthy
82.74 ± 18.89	98.21 ± 3.44	86.21 ± 19.96	99.36 ± 2.79	81.04 ± 18.10	97.01 ± 5.89
F = 8.13, P = .0082		F = 5.27, P = .030		F = 8.62, P = .0069	

No interaction was found between group and assessment when the multiple sclerosis (MS) data were combined or separated as to level of disability. A main effect for group was found with MS data combined and separated as to level of disability. PwMS = persons with multiple sclerosis; NS = not significant.

each assessment. The mild subgroup reported greater physical impact, whereas the moderate subgroup reported greater physical and social impact on their life due to their voice in comparison to H controls. EMST did not significantly improve the impact on quality-of-life of voice-related issues in PwMS as a group or separated as to their level of disability.

DISCUSSION

Several predictions were put forth concerning the effects of EMST in PwMS compared to H controls. First, it was hypothesized that the PwMS would have lower MEP than the H controls prior to training. At baseline, the PwMS MEP (64.47 ± 15.75) was 57.8% of the H controls (111.56 ± 38.11). These data agree with previous investigators reporting greater respiratory muscle weakness in PwMS than healthy controls, but slightly higher than that reported (maximal inspiratory pressure [MIP] 40% less and MEP 60% less) in patients with neuromuscular diseases, irrespective of the primary pathology, compared to healthy controls.[37-39] Additionally, these data agree with previous investigators reporting greater expiratory (PwMS MEP being 57.8% of H control) than inspiratory weakness (PwMS MIP being 69.47% of H control).[17,18,20] The level of respiratory muscle weakness of the PwMS was comparable to that of PwMS having a higher level of disability.[13-17] Presence of weakened respiratory muscles in PwMS having a mild level of disability (EDSS score, M ± SD: 3.6 ± 1.3) and moderate duration of disease (years from diagnosis, M ± SD: 8.43 ± 6.17) in comparison to previously studied patients with MS[13-17] suggests that strengthening of the respiratory muscles may be important to maintaining function as the disease progresses.

Second, it was hypothesized that training would result in significant improvement in MEP in the PwMS and the H control groups. Indeed, MEP did significantly increase for both groups following training. The improvement in PwMS was greater than previously reported in individuals having similar to greater disability[14,16,17] and similar to the gain in MIP in PwMS who received inspiratory muscle strength training.[15] Greater improvement in our participants may have occurred due to lower level of disability, higher training intensity compared to previous studies, or enhanced training specificity.[13-17] The gain in MEP in the H controls was comparable to previously reported data.[40] However, the percent change of our H controls was slightly greater than that reported in studies examining EMST in younger healthy adults.[41,42] Greater percent change in MEP in the H controls may have occurred because of their older age. Participants were possibly less active and may have had a tendency to breathe shallower, therefore having a greater potential for improvement. The "initial values" principle of exercise training states that those with the lowest initial values of a physiologic system have the greatest capacity for improvement in response to training.[24,43]

The third hypothesis, based on the concept that those who are less fit have a greater capacity to make improvement compared to more fit individuals,[24,43] was that the percent gains in the PwMS would be greater than that of the H control group. Following training, the percent gains in MEP were not statistically different (F = 1.30, P > .05) in the PwMS (43.6% ± 32.47%) compared to the H controls (32.13% ± 21.87%). Overall, due to skeletal muscle weakness and balance impairment, the PwMS were less active than the H control. Inactivity is known to cause deconditioning, which affects respiratory muscle as well as other skeletal muscle.[24,43] Presumably the PwMS should have had a potential for greater gain, as the physiological state of their muscles was lower than that of the H control group. Earlier investigators have also suggested that the gains in respiratory muscle strength in PwMS perhaps result from placing a challenge against their low level of physical activity.[13,14,16,17] The lack of statistical difference between the PwMS and the H controls may have been due to the small participant sample-size, as the duration, intensity, and frequency of training were comparable to or greater than in previous studies achieving similar gains in MEP.[14-17]

The fourth hypothesis was that improvement in expiratory muscle strength for the PwMS would translate to improvements in voice and speech production and reductions in the perception of impairment of dysarthria and dysphonia. Voice production was assessed by sustained vowel prolongation and speech production by reading the standardized passage "My Grandfather" at 2 SPLs. Before and after EMST, the PwMS performances during vowel prolongation were less than the H controls. EMST did not translate to an increase in vowel prolongation in the PwMS. Unexpectedly, the PwMS vowel production was greater following detraining. Lack of improvement in PwMS following EMST may be explained by uncoordinated laryngeal and expiratory musculature activity, although unstudied in this investigation. Greater vowel prolongation following detraining may have also been due to a practice effect that suggests that voice therapy alone may be of benefit to PwMS.

No other study has examined the effects of EMST on voice/speech tasks in patients with neuromuscular disease. Limited studies exist that describe speech therapy given to PwMS. The Lee Silverman Voice Treatment (LSVT) has been used in 1 study examining speech in 2 individuals with MS.[44] LSVT resulted in longer sustained vowel durations after 4 weeks of training and at a 6-month follow-up (ie, participant 1 pre- to posttraining increased by 13.2 seconds, and pre- to follow-up increased by 9.5 seconds; participant 2 pre- to posttraining increased by 6.7 seconds, and pre- to follow-up increased by 7.7 seconds).[44] Longer sustained vowel duration following LSVT than EMST may be due to the fact that LSVT focuses the training load on the larynx as opposed to the expiratory muscles, and it may be that PwMS improvement stemmed from enhanced glottal closure, a primary goal of LSVT. Although there have been other studies that have used EMST to impact speech, these studies have been with children with cerebral palsy and in adult vocal performers.[45,46]

The H control group lengthened their vowel prolongation following the training as well as in the detraining phase, and a practice effect is the likely reason. In agreement with previous studies, EMST increased prolongation of vocalization immediately following training in the H control group.[45-47] Further work needs to be done to determine if EMST is an effective treatment for PwMS having voice projection impairment.

In similar fashion to vowel prolongation, EMST resulted in an improvement in WPM only following detraining in the PwMS. This may have been a learning effect due to improved coordination or comfort in performing the desired skill. However, in the H control group, there was a reduction in WPM following training and detraining. A decline in WPM in the presence of increased expiratory pressure following training may have occurred if the participants were concentrating more on the loudness level than on the reading task. The airflow and air pressure that are available during speech influence word production. Few studies have examined word production rates in PwMS. Dysfunctional voice has been reported to occur in less than 25% to nearly 50% of the participants in studies involving PwMS.[3,4,22] Speech dysfunction in PwMS includes slow rate of speech, slower than normal oral diadochokinetic rates, expressive communication disorders, and production of fewer WPM than healthy populations.[3,4,22] In agreement with these previous investigators, PwMS compared to the H control group had produced fewer WPM.

Data on speech production, that is, WPM and duration of speech in PwMS, are very limited. Hartelius and associates, citing previous investigators, reported a mean ± standard deviation of 148 ± 36.5 and range of variation 58 to 235 WPM, whereas their participants produced 123 ± 24.6 with a range of 76 to 172 WPM.[19] Our participants with MS produced a greater number of WPM. Comparison is difficult between the previous studies and the current study, as different reading passages were used.

Currently, there are no data about the effect of EMST on reading rate in healthy populations. WPM reading rate has been reported as an M ± SD of 117 ± 24 and a range of variation between 121 and 221 WPM.[19] Previous investigators, comparing speech of PwMS to healthy individuals, report a WPM rate of 190 and 166 for male and female, respectively, for healthy individuals.[19] The WPM of the healthy participants in the current study was greater than those reported in prior studies. One reason for the difference may have been the reading task. However, to determine if this was the cause of the noted difference in WPM, further analysis of the 2 distinct passages would be necessary.

The final component of the fourth prediction was that the improvement in MEP for the PwMS would result in significant improvements in dysarthria and voice-related quality of life. Impact of the quality of one's voice assessed by the use of the ALSSS for dysarthria showed a difference between the PwMS and H control group. As a group, the PwMS had a weak trend ($F = 2.74$, $P = .073$) toward a significant change on the impact of dysarthria on quality of life following training. However, the moderate subgroup of the PwMS had a significant change in the impact of dysarthria on quality of life following training. Improvement may have resulted from the increased MEP, which would permit greater pressure support during speech. It can be argued that practice was not a factor as the participants were rating their daily speech.

The magnitude of the voice-related problems experienced by a patient and the importance that the patient places on those problems, that is, the impact that the voice disorder has on the patient's voice-related quality of life, are primary influences on decisions about the nature and intensity of treatment. There was no direct speech therapy in this study. However, it was hypothesized that increased MEP would ameliorate voice-related quality-of-life issues. As expected, the VRQOL measure was lower in PwMS compared to the H controls for the total score (VT), and the physical subscale (VP). This is in agreement with investigators who reported "lower VRQOL for laryngeal cancer patients than in normal patients."[48] EMST did not improve the VRQOL for the PwMS. No other study examining EMST, which included voice-related quality of life as an outcome measure, could be found. The inability to show improvement through the use of the VRQOL measure following the EMST may have been due to the lower disability of the participants, the limited influence that voice impairment had on the PwMS at the time of this study, an insensitivity of the VRQOL measure to assess voice impairment in PwMS, or that EMST does not impact voice-related quality-of-life issues. As the VRQOL measure has been used to assess treatment in patients with moderate to severe dysphonia finding improvement in voice-related quality of life,[49] future research in PwMS is warranted in patients presenting with greater dysfunction in their voice prior to EMST.

PwMS and H control participants' expiratory muscle strength remained above baseline at a significant level following 4 weeks of no training. Limited data exist as to the effect of detraining on respiratory muscle strength. Previous investigators reported that 18 weeks of detraining that followed 9 weeks of training resulted in the greatest loss of strength between the first and ninth week of detraining.[12] EMST in PwMS resulted in significant gains in MIP, which returned toward baseline level following 3 months of detraining.[14] Inspiratory muscle strength training with PwMS resulted in gains in MIP and MEP, which were maintained for 1 month following the commencement of training.[15] However, no true detraining period occurred, as the participants had continued to train at a lower level than during the training period; thus, this was more comparable to a low-level maintenance program than a detraining period.[15] The MEP of healthy participants who performed EMST for either 4 or 8 weeks remained above baseline after 4 and 8 weeks of detraining.[47] Further work is needed to determine the optimal training duration, the true rate of decline during a nontraining period, and the best possible maintenance training level.

Limitations of the Study

The total number of individuals with MS was limited (n = 17), resulting in a limited number of participants within each of the disability subgroups (mild = 10, moderate = 7). A greater number of individuals with MS may be needed to address whether EMST can improve MEP and components of speech production in PwMS. Lung and chest-wall compliance reduction may result from viscoelastic properties (fibrosis), spasticity of the respiratory muscles, or osteoarticular mobility. However, these factors were not examined in this study. Future studies examining the effect of respiratory muscle strength training should consider incorporating assessment of these factors.

CONCLUSION

It was shown that PwMS have expiratory muscle weakness in the presence of limited disability, as well as early in the disease process. Respiratory muscle weakness may impair speech in PwMS. EMST was able to improve expiratory muscle strength in PWMS and in H controls. However, no specific group changes occurred in vocalization, whereas vocalization at an LSPL showed gains after training and after a period of no training. Future research is needed to assess EMST in PwMS who have more speech impairment due to respiratory muscle weakness and to investigate other treatment techniques in comparison to EMST in PwMS. The results of this study may not be generalizable to all patients with MS, as the patients in this study had mild to moderate level of disability and were not selected based on presence or absence of speech impairment or degree of dysarthria.

REFERENCES

1. Filippi M, Rocca MA. MRI evidence for multiple sclerosis as a diffuse disease of the central nervous system. *J Neurol.* 2005; 252(Suppl 5):v16-v24.
2. Aboussouan LS. Respiratory disorders in neurologic diseases. *Cleve Clin J Med.* 2005;72(6):511-520.
3. Beukelman DR, Kraft GH, Freal J. Expressive communication disorders in persons with multiple sclerosis: a survey. *Arch Phys Med Rehabil.* 1985;66(10):675-677.
4. Darley FL, Brown JR, Goldstein NP. Dysarthria in multiple sclerosis. *J Speech Hear Res.* 1972;15(2):229-245.
5. Gosselink R, Kovacs L, Decramer M. Respiratory muscle involvement in multiple sclerosis. *Eur Respir J.* 1999;13(2):449-454.

6. Brück W. Inflammatory demyelination is not central to the pathogenesis of multiple sclerosis. *J Neurol*. 2005;252(Suppl 5): v10-v15.
7. Rovaris M, Gambini A, Gallo A, et al. Axonal injury in early multiple sclerosis is irreversible and independent of the short-term disease evolution. *Neurology*. 2005;65(10):1626-1630.
8. Colton R, Hirano M. *Understanding Voice Problems: A Physiological Perspective for Diagnosis and Treatment*. Baltimore, MD: Williams & Wilkins; 1990.
9. Hartman DE. Neurogenic dysphonia. *Ann Otol Rhinol Laryngol*. 1984;93(1 Pt 1):57-64.
10. Hixon T. *Respiratory Function in Speech and Song*. Boston, MA: College-Hill; 1987.
11. Proctor D. *Breathing, Speech, and Song*. New York, NY: Springer-Verlag Wien; 1980.
12. Romer LM, McConnell AK. Specificity and reversibility of inspiratory muscle training. *Med Sci Sports Exerc*. 2003;35(2): 237-244.
13. Wiens ME, Reimer MA, Guyn HL. Music therapy as a treatment method for improving respiratory muscle strength in patients with advanced multiple sclerosis: a pilot study. *Rehabil Nurs*. 1999;24(2):74-80.
14. Gosselink R, Kovacs L, Ketelaer P, Carton H, Decramer M. Respiratory muscle weakness and respiratory muscle training in severely disabled multiple sclerosis patients. *Arch Phys Med Rehabil*. 2000;81(6):747-751.
15. Klefbeck B, Hamrah Nedjad J. Effect of inspiratory muscle training in patients with multiple sclerosis. *Arch Phys Med Rehabil*. 2003;84(7):994-999.
16. Olgiati R, Girr A, Hugi L, Haegi V. Respiratory muscle training in multiple sclerosis: a pilot study. *Schweiz Arch Neurol Psychiatr*. 1989;140(1):46-50.
17. Smeltzer SC, Lavietes MH, Cook SD. Expiratory training in multiple sclerosis. *Arch Phys Med Rehabil*. 1996;77(9):909-912.
18. Hartelius L, Buder EH, Strand EA. Long-term phonatory instability in individuals with multiple sclerosis. *J Speech Lang Hear Res*. 1997;40(5):1056-1072.
19. Hartelius L, Nord L, Buder EH. Acoustic analysis of dysarthria associated with multiple sclerosis. *Clin Linguistics Phonetics*. 1995;9(2):95-120.
20. Hartelius L, Runmarker B, Andersen O. Prevalence and characteristics of dysarthria in a multiple-sclerosis incidence cohort: relation to neurological data. *Folia Phoniatr Logop*. 2000; 52(4):160-177.
21. Hartelius L, Runmarker B, Andersen O, Nord L. Temporal speech characteristics of individuals with multiple sclerosis and ataxic dysarthria: "scanning speech" revisited. *Folia Phoniatr Logop*. 2000;52(5):228-238.
22. Hartelius L, Svensson P. Speech and swallowing symptoms associated with Parkinson's disease and multiple sclerosis: a survey. *Folia Phoniatr Logop*. 1994;46(1):9-17.
23. Espir ML, Watkins SM, Smith HV. Paroxysmal dysarthria and other transient neurological disturbances in disseminated sclerosis. *J Neurol Neurosurg Psychiatry*. 1966;29(4):323-330.
24. Powers S, Howley. *Exercise Physiology: Theory and Applications to Fitness and Performance*. 6th ed. Boston, MA: McGraw Hill; 2005.
25. Lamotte D, Thoumie P. [Multiple sclerosis and botulinum toxin]. *Ann Readapt Med Phys*. 2003;46(6):299-302.
26. Rontal E, Rontal M, Wald J, Rontal D. Botulinum toxin injection in the treatment of vocal fold paralysis associated with multiple sclerosis: a case report. *J Voice*. 1999;13(2):274-279.
27. Poser CM, Brinar VV. Diagnostic criteria for multiple sclerosis. *Clin Neurol Neurosurg*. 2001;103(1):1-11.
28. Poser CM, Paty DW, Scheinberg L, et al. New diagnostic criteria for multiple sclerosis: guidelines for research protocols. *Ann Neurol*. 1983;13(3):227-231.
29. Kurtzke JF. Rating neurologic impairment in multiple sclerosis: an expanded disability status scale (EDSS). *Neurology*. 1983; 33(11):1444-1452.
30. Smeltzer SC, Lavietes MH. Reliability of maximal respiratory pressures in multiple sclerosis. *Chest*. 1999;115(6):1546-1552.
31. Darley FL, Aronson A, Brown JR. *Motor Speech Disorder*. Philadelphia, PA: W. B. Saunders; 1975.
32. Hillel AD, Miller RM, Yorkston K, McDonald E, Norris FH, Konikow N. Amyotrophic lateral sclerosis severity scale. *Neuroepidemiology*. 1989;8(3):142-150.
33. Yorkston KM, Mille RM, Strand EA. *Huntington's Management of Speech and Swallowing in Degenerative Diseases*. Tucson, AZ: Communication Skill Builder; 1995.
34. Yorkston KM, Strand E, Miller R, Hillel A, Smith K. Speech deterioration in amyotrophic lateral sclerosis: implications for the timing of intervention. *J Med Speech-Language Pathol*. 1993; 1(1):35-46.
35. Hogikyan ND, Sethuraman G. Validation of an instrument to measure voice-related quality of life (V-RQOL). *J Voice*. 1999; 13(4):557-569.
36. Hogikyan ND, Wodchis WP, Terrell JE, Bradford CR, Esclamado RM. Voice-related quality of life (V-RQOL) following type I thyroplasty for unilateral vocal fold paralysis. *J Voice*. 2000; 14(3):378-386.
37. Black LF, Hyatt RE. Maximal respiratory pressures: normal values and relationship to age and sex. *Am Rev Respir Dis*. 1969; 99(5):696-702.
38. Black LF, Hyatt RE. Maximal static respiratory pressures in generalized neuromuscular disease. *Am Rev Respir Dis*. 1971; 103(5):641-650.
39. Laghi F, Tobin MJ. Disorders of the respiratory muscles. *Am J Respir Crit Care Med*. 2003;168(1):10-48.
40. Gething AD, Williams M, Davies B. Inspiratory resistive loading improves cycling capacity: a placebo controlled trial. *Br J Sports Med*. 2004;38(6):730-736.
41. Sapienza CM, Davenport PW, Martin AD. Expiratory muscle training increases pressure support in high school band students. *J Voice*. 2002;16(4):495-501.
42. Suzuki S, Sato M, Okubo T. Expiratory muscle training and sensation of respiratory effort during exercise in normal subjects. *Thorax*. 1995;50(4):366-370.
43. Sharkey BJ. Intensity and duration of training and the development of cardiorespiratory endurance. *Med Sci Sports*. 1970;2(4):197-202.
44. Sapir S, Pawlas A, Ramig L, Seeley E, Fox C, Corboy J. Effect of intensive phonatory-respiratory treatment (LSVT) on voice in two individuals with multiple sclerosis. *J Med Speech-Language Pathol*. 2001;9(2):141-151.
45. Cerny F, Panzarella K, Stathopoulus E. Expiratory muscles conditioning in hypotonic children with low vocal intensity levels. *J Med Speech-Language Pathol*. 1997;5:141-152.
46. Hoffman-Ruddy B. *Expiratory Pressure Threshold Training in High-Risk Performers* [Dissertation]. Gainesville, FL: Communication and Speech Disorders, University of Florida; 2001.
47. Baker S. *Expiratory Muscle Strength Training and Detraining: Effects on Speech and Cough Production* [Dissertation]. Gainesville, FL: Communication and Speech Disorders, University of Florida; 2003.
48. *Chemo-Radiation Offers Better Post-Treatment Voice Quality for Laryngeal Cancer*. Retrieved from http://www.innovations-report.com/html/reports/medicine_health/report-32208.html.
49. University of Michigan Health System. *Botox Injections Provide Long-Term Relief and Better Quality of Life for Voice Disorder Patients. Lasting Effects Seen in Three-Year Study of People With Spasmodic Dysphonia*. Ann Arbor, MI: University of Michigan Health System. Retrieved from http://www.med.umich.edu/opm/newspage/2004/botox.htm.