Effect of Sensory Discrimination Training on Structure and Function in Patients With Focal Hand Dystonia: A Case Series

Nancy N. Byl, PhD, PT, FAPTA, Srikantan Nagajaran, PhD, Alison L. McKenzie, PhD, PT

ABSTRACT. Byl NN, Nagajaran S, McKenzie AL. Effect of sensory discrimination training on structure and function in patients with focal hand dystonia: a case series. Arch Phys Med Rehabil 2003;84:1505-14.

Objective: To measure the effects of sensorimotor training based on the principles of neuroplasticity for patients with focal hand dystonia.

Design: Case series of 3 subjects with focal hand dystonia of the left hand, compared with age-matched normative controls.

Setting: Outpatient clinic.

Participants: Three consecutive clinic patients—musicians with focal hand dystonia—who described a history of repetitive practice and performance (2 women; ages, 23y and 35y; 1 man; age, 24y).

Intervention: Subjects were asked to stop performing the tasks that caused the abnormal movements, to participate in a wellness program (aerobics, postural exercises, stress free hand use), and to carry out supervised, attended, individualized, repetitive sensorimotor training activities at least once week for 12 weeks and reinforced daily at home.

Main Outcome Measures: Standard tests documenting somatosensory hand representation, target-specific hand control, and clinical function.

Results: On the affected side, the 3 subjects improved an average of 86.8% on somatosensory hand representation, 117% on target-specific performance, 23.9% on fine motor skills, 22.7% on sensory discrimination, 31.9% on musculoskeletal skills, and 32.3% on independence. All 3 subjects improved 10% or more on 90% of the subtests with 20% improvement on 50% of the subtests.

Conclusion: Individuals with focal hand dystonia who have a history of repetitive hand use can improve cortical somatosensory responses and clinical motor function after individualized sensorimotor training consistent with the principles of neural adaptation.

Key Words: Dystonia; Focal dystonia; Rehabilitation; Somatosensory disorders.

OCCUPATIONAL HAND CRAMPS, also referred to as focal hand dystonia, involve loss of inhibition between agonist and antagonist muscles of the hand, disrupting fine motor control at a target task.[1-9] This condition can develop in performing artists, athletes, business executives, and assembly line workers who perform high levels of attended, stereotypical, repetitive movements.[1,4,5,9]

There are no specific clinical laboratory tests to confirm the diagnosis of focal hand dystonia, however, loss of inhibition, excessive muscle firing, and inability to release muscle contractions are documented with electromyography.[2] Patients with simple target-specific dystonia usually have a normal neurologic examination when performing functional activities.[7,9] Observing the onset of abnormal movements when performing the target task is critical to making the diagnosis of focal hand dystonia. It is not uncommon for patients with focal hand dystonia to report a history of stressful, excessive overuse of the hands,[1,3-5] neuromusculoskeletal trauma (eg, head trauma, radial fractures),[10] degenerative disk disease or cervical injury,[11] biomechanical limitations (eg, poor posture, limited finger spread, decreased forearm and shoulder rotation),[12-15] or peripheral nerve entrapments.[16,17] Researchers and clinicians have documented a loss of inhibition between agonists and antagonists,[18-20] and abnormal neuronal firing patterns in the motor cortex,[21-26] the basal ganglia,[27-29] the spinal cord,[30] and the somatosensory cortex[31-41] in patients with focal hand dystonia.

Focal dystonia is challenging to treat. Traditional intervention usually includes peripheral injections of botulinum toxin,[42-47] sometimes also electric stimulation (for cervical dystonia torticollis).[48] More recently, innovative therapeutic programs emphasizing the principles of neuroplasticity appear to be promising.[49,50]

Over the last 10 years, research[51-58] has clearly established that the central nervous system is adaptable. Goal-directed, repeated, and rewarded sensory and motor behaviors can drive changes in neural structure and function.[54-57] However, adaptation of the nervous system is not infinite. If behaviors become stereotypical and nearly simultaneous, the brain may not be able to distinguish and represent the overused part distinctly and precisely.[58] Evidence supporting aberrant learning has been documented in studies with naive nonhuman primates trained to perform daily, highly repetitive, stereotypical, near simultaneous movements of the hand.[34] Ultimately, the monkeys lost the ability to perform the target task. Electrophysiologic mapping revealed a degradation of the somatosensory representation of the hand, including enlarged receptive fields extending across multiple digits and/or across glabrous and dorsal surfaces.[33,34] In healthy musicians, intensive practice of articulated hand movements is usually associated with an increase in the somatotopic representation of the hand.[59] However, when dystonic movements develop after excessive overuse, the cortical hand representation may shrink in size and the digits can become dedifferentiated (clumped together at the same location without normal sequencing).[33,34,36,60,61]

If focal hand dystonia results from a dedifferentiation of somatosensory structure, then improvement in task-specific

From the Department of Physical Therapy and Rehabilitation Science, School of Medicine, UCSF/SFSU Graduate Program in Physical Therapy, University of California, San Francisco, CA (Byl, McKenzie), Department of Radiology, University of California, San Francisco, CA; and School of Physical Therapy, Chapman University, Orange, CA (McKenzie).

Presented as a poster the Society of Neuroscience's annual meeting, November 2001, New Orleans, LA.

Supported in part by the School of Medicine, UCSF, REAC Dubois Funds, and the National Institutes of Health (grant no. PO1-NS34835).

No commercial party having a direct financial interest in the results of the research supporting this article has or will confer a benefit upon the author(s) or upon any organization with which the author(s) is/are associated.

Reprint requests to Nancy Byl, PhD, PT, FAPTA, UCSF Dept of Physical Therapy and Rehabilitation Medicine, 1320 7th Ave, Box 0736, San Francisco, CA 94143-0736, e-mail: *byl@itsa.ucsf.edu.*

0003-9993/03/8410-7737$30.00/0
doi:10.1053/S0003-9993(03)00276-4

fine motor performance must be contingent on a positive learning paradigm where precise and distinct representations of the digits are restored. By using a case-study approach, repeated with 3 musicians with focal hand dystonia, the present report describes changes in somatosensory evoked responses and clinical function after a sensorimotor training program based on the principles of neuroplasticity. Compared with controls, subjects with focal hand dystonia were expected to show dedifferentiation of the somatosensory cortex and diminished performance on clinical, sensory, motor, musculoskeletal, and functional independence. Focal hand dystonia subjects were also expected to improve on all structural and clinical variables after sensorimotor training.

METHODS

Participants

Three musicians (2 flutists, 1 bagpipe player) with focal hand dystonia of the left hand were referred to the Peter Ostwald Health Program for Performing Artists, University of California, San Francisco (UCSF), CA. Two subjects came to San Francisco to participate in a supervised period of rehabilitation (subject 1 [flutist] was from New Zealand, subject 2 [bagpipe players], from Australia) and 1 subject (subject 3) lived in the San Francisco Bay Area. All flutists had been diagnosed with focal hand dystonia by a neurologist between 1 and 2 years prior to admission to the study. During instrumental play, all 3 subjects presented with complaints of painless, uncontrollable curling of digits 4 (D4) and 5 (D5) on the left hand. All 3 subjects reported difficulty controlling D4 and D5 when D3 was pressing down.

The subjects were a sample of convenience—patients consecutively admitted to the health program who were able to participate in sensorimotor training for a minimum of 10 visits (1–2 sessions/wk), carry out a fitness program 3 days per week, integrate a sensorimotor training program at home, and participate in extensive testing before and immediately after treatment. At the same time, a small grant was awarded for a pilot study to evaluate the feasibility and responsiveness of using magnetoencephalography to document changes in somatosensory-evoked field responses before and after intervention.

The 3 subjects had no known systemic disease. Subject 1 had a previous episode of tendonitis on the involved side and complaints of cervical tension. Subject 3 complained of a resting tremor (since birth) that varied in severity in both hands depending on her level of stress. All subjects were completely independent in personal care and household management. All subjects had to put musical performance on hold because of their hand dystonia. At the time of the study, 1 subject was on medical disability from the symphony (second flutist), the second subject had just returned from intense performance with a traveling bagpipe group, and the third subject had been a full-time music student at a conservatory. The testing procedures were explained to each subject and before starting the study each gave signed consent according to the protocol approved by the Committee on Human Research. Ten healthy age-matched controls served as historical reference norms for magnetoencephalography and 30 healthy subjects (21 women, 9 men) served as the historical reference norms for clinical performance parameters.

Assessment Procedures

All subjects participated in before and after treatment testing using standardized magnetoencephalography[62,63] and clinical sensory and motor tests.[64-73] Scores on standardized tests were summed into 5 dependent variables: (1) somatosensory structure (amplitude, area of representation, sequential order of digits), (2) motor control (task-specific motor control, fine motor control [Purdue Pegboard test, digital reaction time, line tracing accuracy and time]), (3) sensory discrimination (localization, 2-point discrimination, graphesthesia, stereognosis), (4) musculoskeletal performance (posture, neural tension, flexibility [finger spread, forearm rotation, shoulder external rotation], strength of intrinsics to flexor digitorum), and (5) independence (functional independence, work status). The details of test administration for the instruments have been described previously by the test distributors and Byl et al.[64-74]

The Bioimaging Laboratory of the UCSF Department of Radiology examined the primary sensory cortex of each subject using magnetic source imaging (MSI). The somatosensory-evoked potentials (SEPs) of the somatosensory cortex were measured with magnetoencephalophy after 250 air puffs (each 15–20psi) were delivered to each segment of each finger.[62,63] A 37-channel biomagnetometer[a] was used to measure the somatosensory representation of the hand. This test is considered a reliable and valid tool to determine the somatotopic representation of the digits of the hand as well as plotting the location of tumors or epileptic foci.[62,63] Stimulus-related fields are recorded under a circular sensory area 14.4cm over the primary sensory cortex. The MSI data were fit into a model that assumed that the magnetic field was arising from a single equivalent-current dipole. The model included selecting a peak response within 20 to 70ms poststimulus (400–500ms inter-stimulus interval), with a signal to noise ratio greater than 4, a goodness of fit greater than .95, and a minimal confidence volume less than $300mm^3$. Latency, amplitude, and location of the digits on the *x, y,* and *z* axes were quantified from each evoked response. Amplitude was plotted over time. The area of the hand representation was calculated (formula based on volume of an ellipsoid), and the sequential order of the digits on the *z* axis was noted.

Baseline Differences for Controls and Patients

In the healthy control subjects, no significant differences existed between the parameters of the somatosensory-evoked responses on the right compared with the left side. The average latency was 50 to 60ms, and the amplitude averaged 50 to 70fT with the digits sequentially organized from inferior to superior on the *z* axis. Compared with the unaffected side, the amplitude of the early phase of the somatosensory-evoked response was reduced for focal hand dystonia subjects, the area of the hand representation was smaller, and digits 1 to 5 were not sequentially organized from inferior to superior on the *z* axis for either hand. Compared with controls, the amplitude was lower in the early phase for those with focal hand dystonia, and the area of the hand representation was larger on both sides for the focal subjects with hand dystonia.

On the clinical tasks, the control subjects performed similarly on sensory and fine motor tasks on both sides with digital motor reaction time slightly slower for D4 and D5 (bilaterally). Subjects with focal hand dystonia performed motor tasks more accurately and efficiently with the unaffected side, but digital reaction time was similar on both sides. Compared with controls, fine motor accuracy and performance speed was slower for subjects with focal hand dystonia. Digital reaction time varied for subjects with focal hand dystonia compared with controls (with 1 focal hand dystonia subject performing more quickly, one the same, and one more slowly).

On sensory discrimination tasks, control subjects performed similarly on both sides, whereas the subjects with focal hand dystonia performed better on the unaffected side. Compared

Table 1: Summary of Outcomes as Percentage and Probability of Change After Treatment

Dependent Variable (no. of subtests)	Improvement (%) 1	2	3	Avg All FHD Subjects	Probability (*P*) Subjects Had 20% Improvement on Subtest >20%	<20%	Average *P* Subjects All Subtests	Average Posttest Scores vs Controls (nominal)
MSI (2)				86.8%	Overall		<.625	
Area of representation	210.0%	33.0%	64.0%		<.125			Better
Order of digits: *z* axis*	2/3 improved				NA			Similar
SEP amplitude	40.0% increase when amplitude plotted over response time				<.50			Similar
Motor control of target task							<.125	83%–95% of 100%
Fine motor control (4)				23.9%			<.0625	
Digital motor reaction time	14.0%	0.0%	−8.0%		0.0	<.125		Similar
Motor accuracy %	31.3%	1.3%	124.0%		<.25			Similar
Motor accuracy time	30.0%	9.7%	62.6%		<.25			2 similar; 1 slower
Purdue time	4.0%	9.2%	9.1%		0.0	<.125		Slower
Sensory (6)				22.7%			<.002	
2-point discrimination	11.0%	29.0%	32.0%		<.25			Better
Localization (glabrous)	17.0%	15.0%	25.0%		<.50			Better
Graphesthesia	33.0%	49.0%	26.0%		<.125			Better
Kinesthesia	7.0%	15.0%	90.0%		<0.5			Better
Stereognosis %	45.0%	43.0%	20.0%		<.125			Better
Stereognosis time	5.0%	4.0%	24.0%		<0.5			Slower
Musculoskeletal (7)				31.9%			<.0005	
ROM: finger spread	23.0%	20.0%	32.0%		<.125			Similar
ROM: supination/pronation	12.5%	14.1%	0.0%		0.0	<.125		Similar
ROM: external rotation	33.3%	0.0%	20.0%		<.125			Similar
Lumbricals: extensors ratio	148.0%	79.0%	40.0%		<.125			Similar
Posture	6.0%	6.0%	5.0%		0.0	<.125		Similar
Neural tension	60.0%	80.0%	80.0%		<.125			Similar
Independence (2)				32.3%			<.0625	
Independent function	6.9%	3.2%	40.9%		<.50			Similar
Work	60.0%	50.0%	33.3%		<.125			Lower

NOTE. The probability of 0 to 3 subjects improving more than 20% on a subtest ranged from 0 to $P<.125$. The probability that 0 to 3 subjects would improve more than 20% on each subtest within each dependent variable ranged from 0 (no subjects improving on any of the subtests) to $P<.00195$ (all 3 subjects improving >20% on 3 subtests) to $P<.0000038$ (all 3 subjects improving on 6 subtests).
Abbreviations: Avg, average; FHD, focal hand dystonia; NA, not appropriate.
*The *z* axis is inferior to superior on the somatosensory cotex.

with controls, 2 of the subjects with focal hand dystonia performed with similar accuracy on the tasks of 2-point discrimination, localization, and kinesthesia. The subjects with focal hand dystonia performed with similar accuracy as controls on graphesthesia and stereognosis but performed the stereognosis task more slowly.

For control subjects, strength of the intrinsic muscles, range of motion (ROM), and signs of adverse neural tension were within normal limits and similar on the right and left sides. Strength and flexibility were better on the unaffected side for subjects with focal hand dystonia. Compared with controls, subjects with focal hand dystonia had a lower ratio of strength in the lumbricals compared with the flexor profundus, signs of neurovascular entrapment at the thoracic outlet, and decreased postural alignment. Focal hand dystonia subjects 1 and 3 had limited finger spread between D3-D4 and D4-D5 on the affected side (25° on the affected side vs 35°–45° on the unaffected side). Subject 1 also had limited shoulder external rotation, and subject 3 had decreased supination.

The healthy subjects were completely independent in activities of daily living (ADLs), even though they reported that physical complaints interfered with maximum quality of life (rating functional independence an average of 87% out of 100%). The subjects with focal hand dystonia were also independent in ADLs with self-rated functional independence rated on an ordinal scale as 63%, 78%, and 87%, respectively, subjects 1 through 3.

Medical Diagnosis and Functional Problems

The medical diagnosis was focal hand dystonia involving digits 3 through 5 of the left hand. The handicap was the involuntary, uncontrollable movements of digits 4 and 5 on the affected side primarily while playing their instrument. Subject 1 experienced increased tension and curling when resting D4 and D5 on any surface in the pronated position (dystonic dystonia), and the other 2 subjects only had difficulty performing the target task (simple dystonia). The disability for all 3 subjects was the inability to perform on their instruments. The severity of the dystonia for all 3 subjects was rated 2 (able to play the instrument for short periods with compensatory strategies to control for dystonic digits).

Prognosis

Given the intractable nature of focal hand dystonia, the prognosis for all 3 subjects was guarded. All 3 subjects were committed to participating in an intense training program. However, all were worried it might not be possible to return to professional performance. In addition, neither the exact amount of sensory stimulation nor the precise time required to modify the cortical somatosensory hand representation is known. Also,

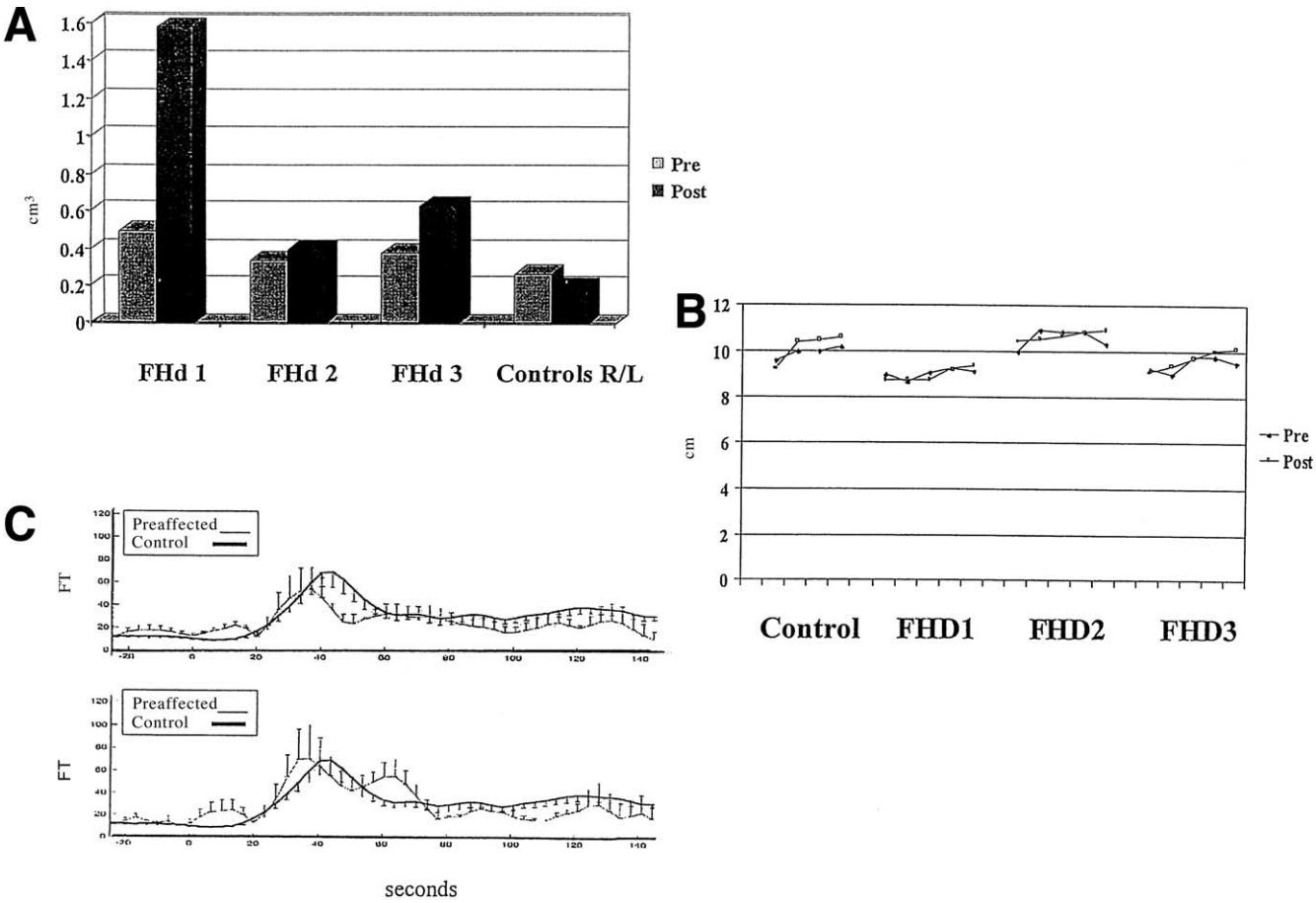

Fig 1. Pre-post changes in somatosensory structure: affected side for with focal hand dystonia subjects. After learning-based sensorimotor training, there were objective improvements in neural structure, as measured by magnetic source imaging: (A) Increased area of somatosensory representation of the hand, (B) improved sequencing of the digits from D1 to D5 in 2 of the 3 subjects, (C) similar amplitude and latency of the somatosensory-evoked field potential between patients with focal hand dystonia and controls.

it is not clear whether complete sensory reorganization must be achieved before normal fine motor control can be restored.

Rehabilitation Plan

To create an environment for positive learning, all subjects were educated about the theory of aberrant learning as 1 etiology of focal hand dystonia and the potential for recovery based on capacity for adaptation in the nervous system. The patients were asked to stop all activities that caused abnormal finger movements (ie, the target task as well as other work related tasks or ADLs that triggered abnormal movements) and to implement a health and wellness program by: (1) evaluating stress and outlining a plan to manage the stress, (2) participating in a fitness program 3 times a week, and (3) practicing stress-free hand movements at target and nontarget tasks.[74] Musculoskeletal problems (eg, decreased ROM) were initially evaluated and addressed by the therapist in terms of soft tissue and joint mobilization reinforced with a flexibility program in the gym. Two patients elected to see a massage therapist.

Sensorimotor training behaviors included attended, goal-oriented, rewarded activities, performed normally and accurately even in limited range (80%), repeated at regular intervals, and progressing in complexity over time. These activities, performed under supervision 1.5 to 2 hours a week, were reinforced with a daily home program (1h/d). Limited skin surfaces were engaged in sensory tasks involving active (localization, stereognosis, kinesthesia, stereognosis) and passive (graphesthesia) stimuli. To facilitate controlled hand shaping without excessive gripping, rough surfaces were placed on all objects that were being manipulated, including the target instrument. To assure broad-based sensory information, patients worked on hand activities in the prone, supine, sitting, and standing position. Positive reinforcement was provided by verbal, visual, tactile, or auditory feedback. Subjects also mentally rehearsed normal task performance. A detailed description of the intervention program can be reviewed in Byl et al.[74]

Subject 1 participated in supervised treatment, twice weekly for 12 weeks (two 6-wk sessions), subject 2 participated daily for 2 weeks, and subject 3 participated in the program for 17 weeks (1 session weekly). Consequently, the total number of visits with a physical therapist varied: 23 visits for subject 1, 19 visits for subject 2, and 23 visits for subject 3.

Research Design and Data Analysis

This study was a single-case design repeated with 3 sequential subjects with focal hand dystonia of the left hand and a history of repetitive injury. The following dependent variables were included in the analysis: somatosensory hand representation, target-specific task performance, fine motor skills, sensory discrimination, musculoskeletal performance, and independence.

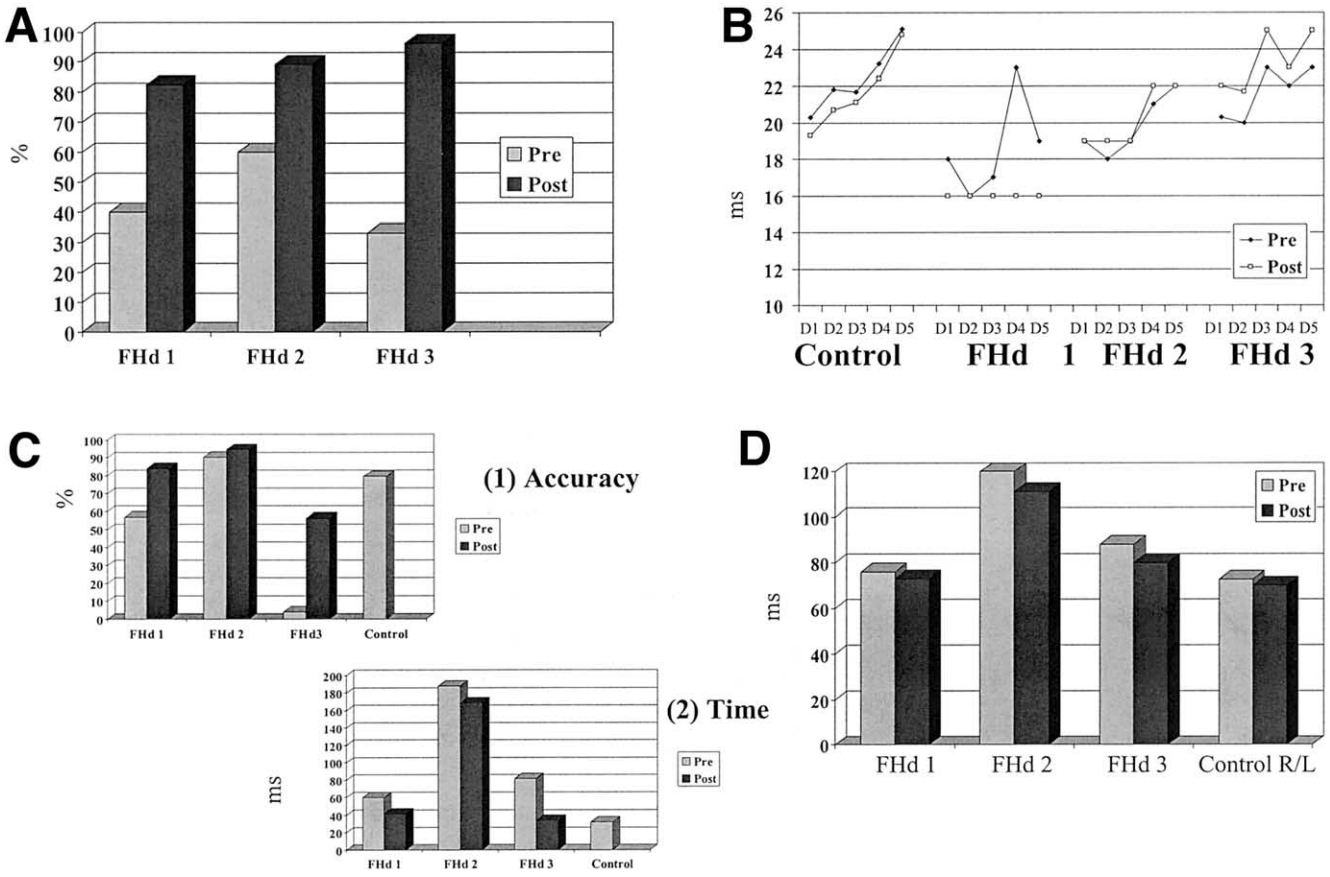

Fig 2. Change in motor control: (A) on the target task, (B) in digital reaction time, (C) for motor tracing, and (D) on the Purdue Pegboard test. Abbreviations: L, left; R, right.

The dependent variables were not correlated,[74] consequently, each dependent variable was considered an independent family.[75] Changes in performance were calculated on each subtest as a percentage score and plotted for visual analysis. For descriptive purposes, performance levels of subjects with focal hand dystonia were compared with reference controls. Inferences were made based on probability theory.[75] On each dependent variable, there was a 50:50 chance that each subject would improve or not improve on each subtest. Improvement was defined as a gain of 20% or more. Digit sequencing could not be subjected to the 20% rule. Each dependent variable consisted of 3 to 6 subtests. Thus, the probability that all 3 subjects would improve 20% or more ranged from P less than .0156 (all 3 subjects improving $>$20% on all 3 subtests) to P less than .00195 (all 3 subjects improving $>$20% on all 6 subtests). This analysis was applied to determine the significance of the changes related to sensorimotor training.

RESULTS

Somatosensory-Evoked Potentials

After training, the somatosensory-dependent variables improved an average of 86.8% across the 3 subjects (table 1). All 3 subjects with focal hand dystonia increased the area of the hand representation by more than 20% and the amplitude of the SEPs by an average of 40%; 2 improved the sequential ordering of the digits (fig 1). Only 2 parameters could be evaluated on the 20% rule. The probability that all 3 subjects would improve on all 6 measurements was P less than .0156. After the training, the area of the hand representations was similar on the affected and unaffected sides, and was larger for subjects with focal hand dystonia than for controls. Integrated across time and across subjects with focal hand dystonia, the average SEP amplitude was similar to that of the controls.

Clinical Performance Parameters

Motor control. All 3 subjects with focal hand dystonia improved their performance on the target task by more than 20%, with the average improvement 117% (table 1). Subjects 1, 2, and 3 performed at 83%, 89%, and 95% accuracy, respectively ($P<.125$) (fig 2A). On fine motor control, the average improvement for the 3 subjects was 23.9%. None of the subjects improved 20% on digital reaction time or performance time on the Purdue Pegboard test. After training, 2 of the 3 subjects with focal hand dystonia improved more than 20% on motor tracing accuracy (performing better than controls), and 2 of the 3 subjects improved performance time less than 20%; only 1 performed the test within time comparable to that of controls (figs 2B–D). The probability of improving more than 20% on 5 of the 12 measurements (4 measurements per subject) was P less than .0625.

Sensory discrimination. At the end of training, the 3 subjects improved an average of 22.7% on sensory discrimination. None improved more than 20% on kinesthesia and only 1 improved more than 20% on speed of performance on the

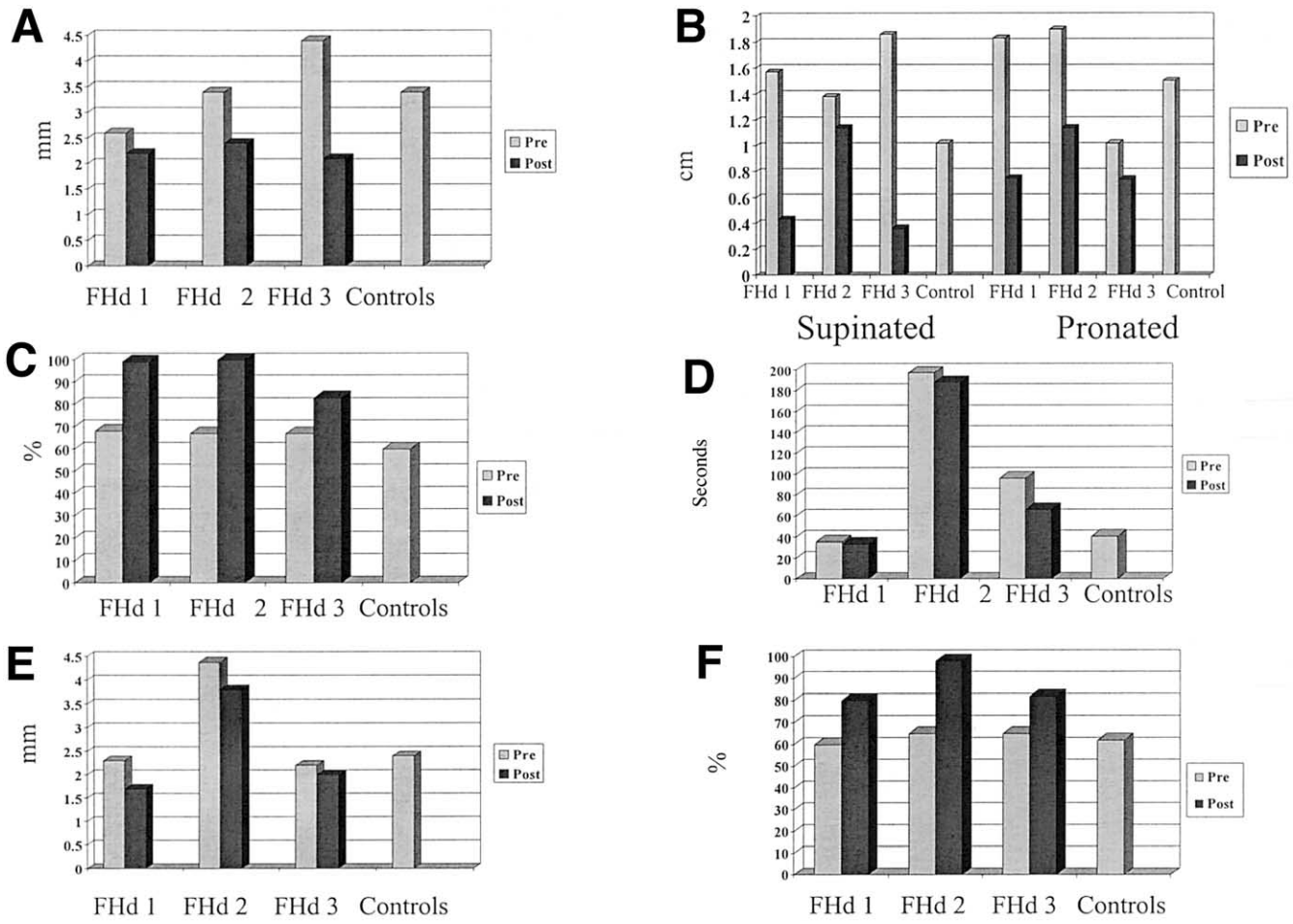

Fig 3. Change in sensory discrimination: (A) 2-point discrimination, (B) localization, (C) stereognosis accuracy, (D) stereognosis speed, (E) kinesthesia, and (F) grapesthesia.

stereognosis test. The subjects with focal hand dystonia performed more accurately than controls on 2-point discrimination, localization, graphesthsia, and stereognosis, but performance time on the stereognosis task took longer than controls, still requiring approximately twice as much time (figs 3A–D). Over the 6 measurements performed by the 3 focal hand dystonia subjects (ie, 18 measures), 11 measurements improved more than 20% ($P<.0005$).

Musculoskeletal performance parameters. At the end of training, the 3 subjects with focal hand dystonia improved 31.9% in musculoskeletal performance. Two subjects improved more than 20% on all musculoskeletal parameters except posture and forearm rotation. The ratio of strength of the lumbricals to flexor profundus was higher for 2 of the subjects with focal hand dystonia compared with controls, and all flexibility measurements became similar for focal hand dystonia subjects and controls (fig 4). The probability that the 3 subjects would improve more than 20% on 11 of the 18 measurements was *P* less than .0005.

Physical independence and work. The average improvement in work and independence for the 3 subjects with focal hand dystonia was 30.7%. These subjects were functioning at 88%, 89%, and 92% of maximum independence and quality of life—a level comparable to healthy subjects. Only 1 subject with focal hand dystonia improved more than 20% on independence. All 3 improved more than 20% in work status. Two subjects returned to their previous work (one to performance but on a modified schedule, one to finish studies at the conservatory) (fig 5). The probability that the subjects would improve more than 20% on 4 of the 6 measurements was .0625.

DISCUSSION

The present results add to the evidence that (1) patients with focal hand dystonia with a history of repetitive overuse can have measurable degradation of the hand representation in the somatosensory cortex, (2) they have associated dysfunction in sensory processing and fine motor control, and (3) dysfunction can be modified with an intervention program based on the principles of neuroplasticity. Sensorimotor training had to be tailored to each subject, with appropriate progression of task difficulty. All subjects made significant progress performing the target task, but only 2 returned to their musical careers, with a modified performance schedule.

On the affected side, the improvements in SEPs included increased amplitude, expanded representation of the hand, and improved digit order on the *z* axis. Mean differences in amplitude of the SEPs have not been reported in other studies.[1,36] Unique to the present study, the amplitude of the SEPs was integrated across time and across subjects, revealing noticeable

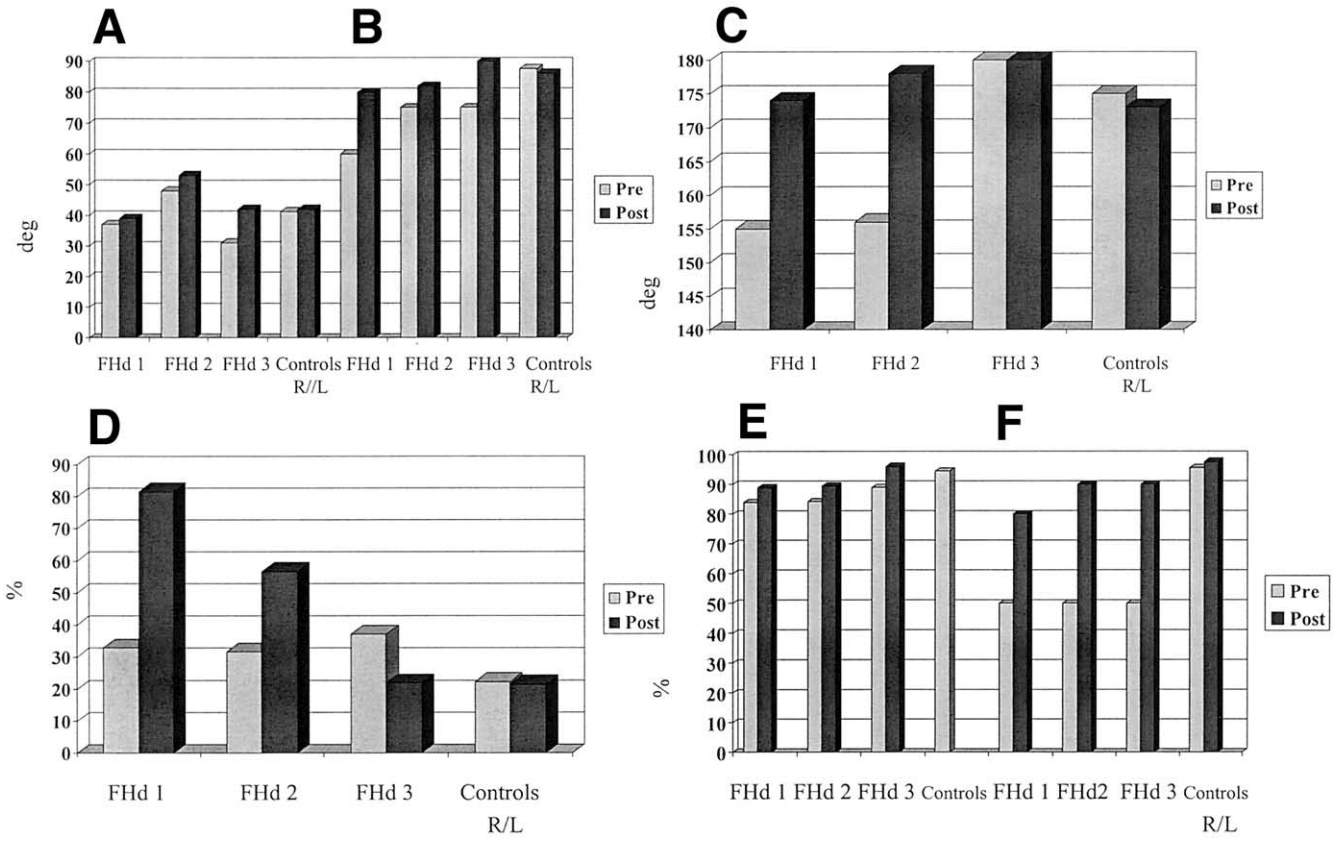

Fig 4. Change in muskuloskeletal performance: (A) finger spread (abduction), (B) shoulder external rotation, (C) supination and promotion, (D) lumbricales: extensor digitorum ratio, (E) posture, and (F) neural tension.

variations in the early and late phases. Averaging amplitudes across subjects would have concealed these differences.

Similar to other clinical studies, all 3 subjects had problems with some elements of sensory discrimination,[36-42] however, before the study they were unaware of these sensory problems. They were also surprised that increasing the cutaneous input to their affected digits (eg, using tape) had a powerful effect on improving fine motor control. Increased sensory cues can improve the ability of the nervous system to differentiate sensory information and organize a motor response. Interestingly, 1 subject was hyposensitive to light touch, and 2 were hypersensitive. In the first case, sensorimotor training had to begin with intensive tactile stimuli to enable adequate discrimination. In the latter cases, tactile stimuli had to be lightly delivered with the subjects positioned where sensory discrimination could be performed without triggering abnormal muscle contractions (eg, supine with shoulder elevation and forearm pronation). Ultimately, sensorimotor training activities must be integrated into the position in which the subject performs the target task.

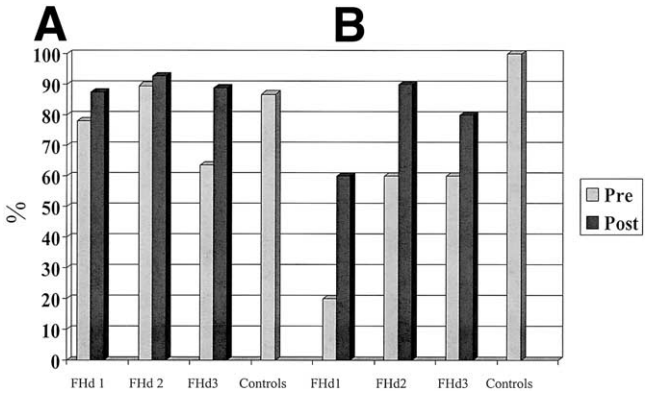

Fig 5. Change in independence measures: (A) functional independence and (B) work status.

As reported in other studies of hand dystonia,[12-15] we also measured limitations in flexibility in the hand and forearm in our subjects. Congenitally or traumatically induced restrictions in joint or soft-tissue movements can create imbalances in muscle performance, excessive shortening in 1 muscle and lengthening in a related muscle, as well as abnormal patterns of movement with excessive end range loading. The question is whether these variations in mobility actually cause focal hand dystonia[10] or simply increase the risk of developing focal hand dystonia when an individual is subjected to stressful, excessive, repetitive hand use. In 1 animal study,[33,34] nonhuman primates were trained to perform attended, repetitive, stressful, hand opening and closing until they developed dystonic posturing that interfered with the performance of the target task. At autopsy, Topp and Byl[13] described an anatomic defect of the flexor profundus tendon on D4 of the trained side and D3 on the untrained side in 1 of the 6 monkeys trained. There were no signs of motor dysfunction on the untrained side, suggesting that restricted mobility was a risk factor but not a cause of focal hand dystonia.

Fine motor movements of the hand require sequenced, individuated, fractionated movements.[16] Normal motor reaction time does not ensure skilled fine motor control. Because voluntary, fine motor digital movements are controlled by various cortical and subcortical pathways with accurate sensorimotor feedback, it should not be surprising to find normal motor reaction time in subjects with focal hand dystonia. However, decreased fine motor accuracy and prolonged performance time should be expected.

There are significant constraints in a case-study design[75,76]: small sample size, lack of controls, no random selection or assignment, learning with retesting, the Hawthorne effect, bias in measurement because of lack of blinding, limited statistical analysis, and inability to generalize findings to a larger population. Some of these constraints were minimized in the present study. An independent evaluator, unaware of study objectives, performed MSI; research assistants blinded to group assignment performed the clinical measurements; 2 control groups were included for reference; and probability theory was applied to provide insight regarding the significance of the measured changes. Because responses are individualized, single-subject design repeated over multiple subjects can provide strong arguments in support of a theory. Averaging data in large groups can be seriously biased by variability in measurement, skewed distribution, and heterogeneity.

The present intervention study included a comprehensive approach to sensorimotor training. This type of multitask intervention does not isolate the effects of training on a single sensory task (eg, Braille reading vs graphesthesia). In 1 study (including patients with writer's cramp),[77] Braille reading alone was associated with improved motor performance and a reduction in the severity of cramping, but recovery of target-specific motor control was not 100%. Sensory training may have to drive changes not only in cortical area 3b but also in other sensory areas, including basal ganglia-thalamic-cortical pathways.

Multisite, controlled, randomized clinical trials are needed to confirm that sensorimotor retraining normalizes somatosensory structure, improves sensory discrimination, and restores normal fine motor control at target and nontarget tasks for patients with focal hand dystonia. These studies should also include genetic testing to determine what proportion of subjects with focal hand dystonia have the DYT1 or other known gene,[78-80] and should combine treatment approaches based on crossover designs such as pairing botulinum toxin or limb immobilization with sensorimotor retraining. Future studies also must include more rigorous strategies for assuring patient compliance for home training activities. Studies are also needed to detail more specifically training parameters (repetitions needed, spacing of practice, reward, progression of difficulty, length of training).

CONCLUSION

Aberrant learning may explain the development of focal hand dystonia in some patients who perform highly repetitive, stressful, stereotypical hand tasks. A conservative intervention strategy based on the principles of neuroplasticity can improve somatosensory structure and clinical function. However, 3 to 6 months of supervised training, once a week, reinforced with a self-directed home program may not be sufficient to return musicians to professional levels of performance. Musicians may benefit from behavioral programs designed to help them meet these objectives: maintain accurate somatosensory feedback and healthy biomechanical movement strategies, incorporate mental practice as 1 way to reduce the intensity and strain of physical practice, and avoid long periods of stressful, repetitive, alternating, stereotypical, near-simultaneous digital movements.

References

1. Chen R, Hallett M. Focal dystonia and repetitive motion disorders. Clin Orthop 1998;Jun(351):102-6.
2. Cohen L, Hallett M. Hand cramps: Clinical features and electromyographic patterns in focal dystonia. Neurology 1988;38:1005-12.
3. Fry H. Overuse syndromes in musicians 100 years ago: an historical review. Med J Aust 1986;146:620-5.
4. Hochberg F, Harris S, Blartert T. Occupational hand cramps: professional disorders of motor control. Hand Inj Sports Perform Arts 1990;6:427-8.
5. Jankovic J, Shale H. Dystonia in musicians. Semin Neurol 1989; 9:131-5.
6. Rhoad R, Stern P. Writer's cramp–a focal dystonia: etiology, diagnosis, and treatment. J Hand Surg [Am] 1993;18:541-4.
7. Rothwell J, Oveso J, Day B, Marsden C. Pathophysiology of dystonias. Adv Neurol 1983;39:851-63.
8. Sheehy M, Marsden C. Writer's cramp: a focal dystonia. Brain 1982;105:461-80.
9. Tubiana R. Incidence: classification of severity and results of therapy, In: Winspur I, Parry CB, editors. The musician's hand. London: Martin Dunitz; 1998. p 164-7.
10. Jancovic J. Can peripheral trauma induce dystonia and other movement disorders? Yes. Mov Disord 2001;16:7-11.
11. Katz R, Williams C. Focal dystonia following soft tissue injury: three case reports with long-term outcomes. Arch Phys Med Rehabil 1990;71:345-9.
12. Leijnse JN. Anatomical factors predisposing to focal dystonia in the musician's hand: principles, theoretical examples, clinical significance. J Biomech 1996;30:659-69.
13. Topp KS, Byl NN. Movement dysfunction following repetitive hand opening and closing: anatomic analysis in owl monkeys. Mov Disord 1999;14:295-306.
14. Wilson F, Wagner C, Homberg V, Noth J. Interaction of biomechanical and training factors in musicians with occupational cramps/focal dystonia [abstract]. Neurology 1991;41(3 Suppl 1): 291.
15. Wilson F, Wagner C, Homberg V. Biomechanical abnormalities in musicians with occupational cramp/focal dystonia. J Hand Ther 1993;6:298-307.
16. Quartarone A, Girlanda P, Risitano G. Focal hand dystonia in a patient with thoracic outlet syndrome. J Neurol Neurosurg Psychiatry 1998;65:272-4.
17. Charness ME. Unique upper extremity disorders of musicians. In: Millender LH, Louis DS, Simmons BP, editors. Occupational disorders of the upper extremity. New York: Churchill Livingstone; 1992. p 117-51.
18. Chen R, Tsai C, Lu C. Reciprocal inhibition in writer's cramp. Mov Disord 1995;10:556-61.
19. Nakashima K, Rothwell J, Day B, Thompson P, Shannon K, Marsden C. Reciprocal inhibition between forearm muscles in patients with writer's cramp and other occupational cramps, symptomatic hemidystonia and hemiparesis due to stroke. Brain 1989;112:681-97.
20. Ridding M, Sheean G, Rothwell J, Inzelberg R, Kujirai T. Changes in the balance between motor cortical excitation and inhibition in focal, task specific dystonia. J Neurol Neurosurg Psychiatry 1995;59:493-8.
21. Chase T, Tamminga C, Burrows H. Positron emission tomographic studies of regional cerebral glucose metabolism in idiopathic dystonia. Adv Neurol 1988;50:237-41.
22. Defendini R, Fahn S. Magnetic resonance imaging of dystonic states. Adv Neurol 1988;50:265-75.
23. Deuschl G, Toro C, Matsumoto J, Hallett M. Movement-related cortical potentials in writer's cramp. Ann Neurol 1995;38:861-8.
24. Gilman S, Junck L, Young A. Cerebral metabolic activity in idiopathic dystonia studied with positron emission tomography. Adv Neurol 1988;50:231-6.

25. Mavroudais N, Caroyer J, Brunko E, Zegers de Beyl D. Abnormal motor evoked responses to transcranial magnetic stimulation in focal dystonia. Neurology 1995;45:1671-7.
26. Tempel L, Perlmutter J. Abnormal cortical responses in patients with writer's cramp. Neurology 1993;43:2252-7.
27. Black KJ, Ongur D, Perlmutter JS. Putamen volume in idiopathic focal dystonia. Neurology 1998;51:819-24.
28. Karbe H, Holfhof V, Rudolf J. Positron emission tomography demonstrates frontal cortex and basal ganglia hypometabolism in dystonia. Neurology 1992;42:1540-4.
29. Perlmutter JS, Stambuk MK, Markham J. Decreased [18F] piperone binding in putamen in idiopathic focal dystonia. J Neurosci 1997;17:843-50.
30. Kaji R, Rothwell J, Katayama M, et al. Tonic vibration reflex and muscle afferent block in writer's cramp. Ann Neurol 1995;38:155-62.
31. Odergren T, Iwasaki N, Borg J, Forssberg H. Impaired sensory motor integration during grasping in writer's cramp. Brain 1996; 119(Pt 2):569-83.
32. Hallett M. Is dystonia a sensory disorder? Ann Neurol 1995;38: 139-40.
33. Byl NN, Merzenich M, Jenkins WM. A primate genesis model of focal dystonia and repetitive strain injury: I. Learning-induced de-differentiation of the representation of the hand in the primary somatosensory cortex in adult monkeys. Ann Neurol 1996;47: 508-20.
34. Byl NN, Merzenich M, Cheung S, Bedenbaugh P, Nagarajan S, Jenkins W. A primate model for studying focal dystonia and repetitive strain injury: effects on the primary sensory cortex. Phys Ther 1997;77:269-84.
35. Sanger TD, Pascual-Leone A, Tarsy D, Schlaug G. fMRI evidence of sensory receptive fields spanning multiple fingers in writer's cramp. Mov Disord 2001;16:94-9.
36. Bara-Jiminez W, Catalan M, Hallett M. Abnormal somatosensory homunculus in dystonia of the hand. Ann Neurol 1998;44:828-31.
37. Byl NN, Wilson F, Scott P, Oakes A. Sensory dysfunction associated with repetitive strain injuries of tendinitis and focal hand dystonia: a comparative study. J Ortho Sports Phys Ther 1996; 23:234-44.
38. Byl NN, Hamati D, Melnick M, Wilson F, McKenzie A. The sensory consequences of repetitive strain injury in musicians: focal dystonia of the hand. J Back Musculoskeletal Rehabil 1996; 7:27-39.
39. Sanger TD, Tarsy D, Pascual-Leone A. Abnormalities of spatial and temporal sensory discrimination in writer's cramp. Mov Disord 2001;16:94-9.
40. Tinazzi M, Frasson E, Bertolasi L, Fiaschi A, Aglioti S. Temporal discrimination of somesthetic stimuli is impaired in dystonic patients. Neuroreport 1999;20:1547-50.
41. Topp KS, Byl NN. Focal hand dystonia. Phys Ther Case Rep 1997;1:39-52.
42. Brin M, Fahn S, Moskowitz C, et al. Localized injections of botulinum toxin for the treatment of focal dystonia and hemifacial spasm. Mov Disord 1987;2:237-54.
43. Cole R, Hallett M, Cohen L. Double-blind trial of botulinum toxin for treatment of focal hand dystonia. Mov Disord 1995;10:466-71.
44. Karp B, Cole R, Cohen L, Grill S, Lou JS, Hallet M. Long-term botulinum toxin treatment of focal hand dystonia. Neurology 1994;44:70-6.
45. Pullman S, Greene P, Fahn S, Pedersen S. Approach to the treatment of limb disorders with botulinum toxin A. Arch Neurol 1996;53:617-24.
46. Tsui J, Bhatt M, Calne S, Calne D. Botulinum toxin in the treatment of writer's cramp: a double-blind study. Neurology 1993;43:183-5.
47. Van den Bergh P, Francart J, Mourin S, Kollmann P, Laterre E. Five-year experience in the treatment of focal movement disorders with low-dose Dysport botulinum toxin. Muscle Nerve 1995;18: 720-9.
48. Goetz C, Penn R, Tanner C. Efficacy of cervical cord stimulation in dystonia. Adv Neurol 1988;50:645-9.
49. Byl NN, Nagarajan S, McKenzie AM. Treatment effectiveness of patients with a history of repetitive strain injury and focal hand dystonia: a planned, prospective follow-up study. J Hand Ther 2000;13:289-301.
50. Candia V, Elbert T, Altenmuller E, Rau H, Schafer T, Taub E. Constraint-induced movement therapy for focal hand dystonia in musicians [letter]. Lancet 1999;253:42-3.
51. Jenkins WM, Merzenich M. Reorganization of neocortical representations after brain injury: a neurophysiological model of the bases of recovery from stroke. Prog Brain Res 1987;71:249-66.
52. Jenkins WM, Allard T, Nudo R. Cortical representational plasticity. In: Raskic P, Singer W, editors. Neurobiology of the neocortex. New York: John Wiley & Sons; 1988. p 41-67.
53. Jenkins W, Merzenich M, Ochs M, Allard T, Guic-Robles E. Functional reorganization of primary somatosensory cortex in adult owl monkeys after behaviorally controlled tactile stimulation. J Neurophysiol 1990;63:82-104.
54. Nudo R, Wise BS, Fuentes F, Milliken G. Neural substrates for the effects of rehabilitative training on motor recovery after ischemic infarct. Science 1996;272:1791-5.
55. Recanzone G, Jenkins W, Hradek G, Merzenich MM. Progressive improvement in discriminative abilities in adult owl monkeys performing a tactile frequency discrimination task. J Neurophysiol 1992;67:1015-30.
56. Xerri C, Coq JO, Merzenich MM, Jenkins WM. Experience-induced plasticity of cutaneous maps in the primary somatosensory cortex of adult monkeys and rats. J Physiol (Paris) 1996;90: 277-87.
57. Byl NN, Merzenich MM. Principles of neuroplasticity: implications for neurorehabilitation and learning. In: Gonzalez EG, Myers SJ, Edelstein JE, Lieberman JS, Downey JA. Downey and Darling's physiological basis of rehabilitation medicine. Boston: Butterworth-Heinemann; 2001. p 609-28.
58. Wang X, Merzenich MM, Somethethesa K, Jenkins WM. Remodeling of hand representation in adult cortex determined by timing of tactile stimulation. Nature 1993;378:71-5.
59. Elbert T, Pantev C, Wienbruch C, Rockstroh B, Taub E. Increased cortical representation of the fingers of the left hand in string players. Science 1995;270:305-7.
60. McKenzie AM, Nagaragan S, Roberts T, Merzenich MM, Byl NN. Somatosensory changes and clinical correlates in patients with focal hand dystonia. Poster presented at: Society for Neuroscience Annual Meeting; 2001 Nov 10-15; San Diego (CA).
61. Byl NN, Merzenick MM, Roberts T, Nagarajan S, McKenzie AM. The correlation of clinical neuromusculoskeletal and central somatosensory parameters in patients with focal hand dystonia. Poster presented at: Society for Neuroscience Annual Meeting; 1998 Nov 7-12; Los Angeles (CA).
62. Roberts T, Poeppel D, Rowley HA. Magnetoencephalography and magnetic source imaging. Neuropsychiatry Neuropsychol Behav Neurol 1998;11:49-64.
63. Rowley HA, Roberts T. Functional localization by magnetoencephalography. Neuroimaging Clin North Am 1995;5:695-710.
64. Ayres A. Sensory integration and praxis tests: SIPT manual. Los Angeles: Western Psychological Assoc; 1989.
65. Bohannon RW. Stopwatch for measuring thumb movement time. Percept Mot Skills 1995;81:122-6.
66. Carlsson AM. Assessment of chronic pain. I. Aspects of the reliability and validity of the visual analogue scale. Pain 1983;16: 87-101.
67. Edgelow P, Feinberg S. Controversies and questions: thoracic outlet syndrome. Calif Workers Compensation Enquirer 1995;7: 10-3.
68. Fung S, Byl NN, Melnick M, Callahan P, et al. Functional outcomes: the development of a new instrument to monitor the effectiveness of physical therapy. Eur J Phys Med Rehabil 1997; 7:31-41.
69. Kendall FP, McCreary EK, Provance PG. Muscles: testing and function. 4th ed. Philadelphia: Williams & Wilkins; 1993.
70. Instructions and normative data: Purdue Pegboard. Lafayette (IN): Lafayette Instrument Co; 1996.
71. Norkin CC, White DJ. Measurement of joint motiona guide to goniometry. Philadelphia: FA Davis; 1995.

72. Schmidt RT, Toews JV. Grip strength as measured by the Jamar dynamometer. Arch Phys Med Rehabil 1970;51:321-7.
73. Shimokata H, Kuzuya F. Two-point discrimination test of the skin as an index of sensory aging. Gerontology 1995;42:267-72.
74. Byl NN, Nagarajan S, McKenzie AM. Effectiveness of sensorimotor training on structure and function in musicians with focal hand dystonia: three case studies. Poster presented at: Society for Neuroscience Annual Meeting; 2000 Nov 7; New Orleans (LA).
75. Maraschuilo LA, Serlin RC. Statistical methods for social and behavioral sciences. New York: WH Freeman; 1988.
76. Portney LG, Watkins MP. Foundations of clinical research: applications to practice. Upper Saddle River: Prentice Hall; 2000. p 223-58.
77. Gerloff C, Corwell B, Chen R, Hallett M, Cohen LG. The role of the human motor cortex in the control of complex and simple finger movement sequences. Brain 1998;121(Pt 9):1695-709.
78. Zeuner KE, Bara-Jimenez W, Noguchi PS, Goldstein SR, Ambrosia JM, Hallett M. Sensory training for patients with focal hand dystonia. Ann Neurol 2002;451:593-8.
79. Leube B, Rudnicki D, Ratzlaff T, Kessler K, Benecke R, Auburger G. Idiopathic torsion dystonia: assignment of a gene to chromosome 18p in a German family with adult onset, autosomal dominant inheritance and purely focal distribution. Hum Mol Genet 1996;5:1673-7.
80. Gasser T, Bove C, Ozelius L, et al. Haplotype analysis at the DYT1 locus in Ashkenazi Jewish patients with occupational hand dystonia. Mov Disord 1996;11:163-6.

Supplier

a. Magnes®; Biomagnetic Technologies Inc, 9727 Pacific Heights Blvd, San Diego, CA 92121-3719.