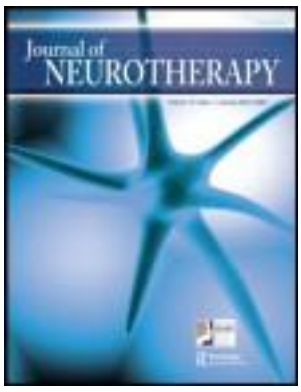

Journal of Neurotherapy: Investigations in Neuromodulation, Neurofeedback and Applied Neuroscience

Biofeedback for Movement Disorders (Dystonia with Parkinson's Disease): Theory and Preliminary Results

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Published online: 08 Sep 2008.

To cite this article: Michael Thompson MD & Lynda Thompson PhD (2002) Biofeedback for Movement Disorders (Dystonia with Parkinson's Disease): Theory and Preliminary Results, Journal of Neurotherapy: Investigations in Neuromodulation, Neurofeedback and Applied Neuroscience, 6:4, 51-70

To link to this article: http://dx.doi.org/10.1300/J184v06n04_06

Biofeedback for Movement Disorders (Dystonia with Parkinson's Disease): Theory and Preliminary Results

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ABSTRACT. *Background.* This paper presents a theoretical framework for using a combination of EEG biofeedback plus regular biofeedback with clients who have movement disorders.

Method. A case study is included that describes intervention and results with a 47-year-old woman with the dual diagnosis of Parkinson's disease and dystonia. The rational for adding biofeedback interventions to traditional medical treatment hinges on the fact that muscle spindles, which are involved in muscle movement and tone, have double innervations, cholinergic and sympathetic (Passatore, Grassi, & Filippi, 1985). Both of these systems can be operantly conditioned using biofeedback. There were two learning goals: (1) increase the production of 12 to 15 Hz activity since this sensor motor rhythm (SMR) is associated with decreased firing of the red nucleus and the red nucleus, in turn, has links to the muscle spindles (Sterman, 2000); (2) train for calm, relaxed autonomic nervous system functioning (decreased sympathetic drive and parasympathetic ascendance) because this may also have a beneficial effect on muscle tone by means of influencing muscle spindle activity (Banks, Jacobs, Gevirtz, & Hubbard, 1998). Training for balanced autonomic system functioning is facilitated by diaphragmatic breathing at a rate of about six breaths per minute. Diaphragmatic breathing results in respiration and heart rate variability, presented as a line graph, following

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Journal of Neurotherapy, Vol. 6(4) 2002

the same sinusoidal pattern when viewed on a biofeedback screen, a pattern termed respiratory sinus arrhythmia (RSA, Budzynski, 1989). This dual training of neurofeedback to enhance SMR activity and RSA biofeedback for relaxed autonomic nervous system (ANS) functioning was done for 30 sessions over a six-month period.

Results. Training was associated with significant reduction in dystonic movements. Additionally, the client became able to use diaphragmatic breathing to cue herself to turn on a mental state associated with increased SMR production and thus control incidents of freezing, a common problem in advanced Parkinson's disease. With twelve more sessions over the next 18 months, the improved quality of life has been maintained.

Discussion. This work is reported to put forth a theoretical model of why neurofeedback plus biofeedback is helpful in movement disorders and to encourage research in this area.

KEYWORDS. Biofeedback, neurofeedback, dystonia, Parkinson's disease, movement disorders, respiratory sinus arrhythmia (RSA), breathing, muscle spindles

INTRODUCTION

Three common movement disorders are Parkinson's disease (PD), Tourette's syndrome (TS) and dystonia. All three have symptoms which can wax and wane and all three can be debilitating in their effects if the symptoms are severe. There has been some promising work done using neurofeedback with people who have Tourette's syndrome (Tansey, 1986; Daly, 2001), though without a clear theoretical framework about why it should work. This article presents a hypothesis regarding why an intervention that used a combination of neurofeedback and biofeedback was successful when working with a client who had both dystonia and Parkinson's disease. The rationale for this combined intervention emanates from research that demonstrates that both neurofeedback and biofeedback can affect muscle spindle activity. Muscle spindles, which detect muscle length and are involved in muscle movement and tone, have a double enervation, cholinergic and sympathetic. Since both of these systems can be operantly conditioned, it was hy-

pothesized that neurofeedback plus biofeedback, through their effects on muscle spindle activity, could result in a decrease in the undesirable muscle movement and tone that characterizes movement disorders. This paper will briefly present information about dystonia, PD, and TS. Then the neurophysiology and neuro-anatomical connections that underlie the theoretical framework for combining neurofeedback and biofeedback as an intervention will be presented. Finally, a clinical case study of a 47-year-old woman with a severe case of dystonia combined with Parkinson's disease will be outlined.

In 1911, Hermann Oppenheim introduced the term dystonia for a condition, which demonstrated variations in muscle tone: hypotonic at times and in tonic muscle spasm at other times. He also described twisted postures associated with the muscle spasms and rapid, at times jerking, movements of limbs and trunk. It is a syndrome characterized by sustained muscle contractions. A recent definition states, "Dystonia consists of repetitive involuntary and usually twisting movements (i.e., dystonic movements) and postures caused by the co-contraction of agonist and antagonist muscles, with maximal displacement persisting for a second or longer" (Guberman, 1994). The classification of the different manifestations of dystonia is based on age of onset, parts of the body affected, and etiology. Primary dystonia is genetic with a fifty percent chance a child will have dystonia if one parent has the DYT1 gene. Approximately thirty percent of people with this gene manifest symptoms (Comella, 1999). Dystonia may be idiopathic or secondary to another abnormality such as stroke, tumor, head injury, or Wilson's disease. The secondary conditions appear to affect the putamen (Guberman, 1994). Since tardive dyskinesia can be induced by antipsychotic medication (phenothiazines) and because levodopa (L-dopa) can produce dystonia as a side effect, neurotransmitter disturbances are suspected as the cause. There are three common groupings. First, "focal" dystonia, the most common form, affects a single body area. Presentations may include cervical (spasmotic torticollis), blepharospasm (eye blinking or closing), spasmotic dysphonia (laryngeal with strangled, choked or whisper voice), writer's cramp, and limb dystonias. Second, "segmental" dystonia has similar symptoms but affects at least two body areas that are not next to each other. Third, "generalized" dystonia involves several body areas on both sides of the body (Guberman, 1994).

Dystonia has proven to be an extraordinarily disabling illness with no good single treatment. The objectives of current treatments are not to cure the illness but rather to decrease symptoms of pain and abnormal posturing and restore function. Approaches to treatment, as reviewed

by Fahn and Marsden (1987) and briefly summarized below, are pharmacological, surgical and electrical. There are unwanted side effects with each of these approaches.

The anticholinergic mediations (e.g., Artane, Cogentin, Parsitan) have central side effects of confusion, drowsiness, hallucinations, personality change, memory difficulties and peripheral side effects such as dry mouth, blurred vision, urinary retention and constipation.

Benzodiazepines (e.g., Valium and Ativan) block GABA-A receptors (CNS) and have side effects such as sedation, depression, and addiction. Medication to stimulate the GABA-B receptors has also been tried (Gordon, 1999). Also used are medications either to increase dopamine (L-dopa) or to decrease dopamine (antipsychotics such as clozapine). It should be noted, however, that with most patients physicians would avoid using dopamine blocking agents such as prochlorperazine, or haloperidol. One of the more helpful approaches has involved the use of botulinum toxin injections, which can relieve symptoms for weeks. This toxin binds to presynaptic cholinergic terminals, which decreases the release of acetylcholine. There may be troublesome side effects, including ptosis (eyelid laxity, Guberman, 1994). The time between injections is the time it takes for the nerves to sprout again. Although this has been successful it should be noted that thirty to forty percent of patients injected with placebo also experience improvement in symptoms (Jankovic & Brin, 1991). Tegretol (carbamazepine) and anticonvulsants for paroxysmal dystonia have also been tried. Surgical approaches have included thalamotomy and pallidotomy. Electrical treatments have used high frequency electrical stimulation in an attempt to turn off the pallidum (O'Brien, 1999). The range of treatments speaks to the lack of any single successful medical intervention.

Fortunately, dystonia may not progress relentlessly and the symptoms tend to plateau after an initial period of progression. Cognition, strength, senses (e.g., vision, hearing) remain normal. Depression however, can aggravate dystonia symptoms. As with Parkinson's disease and Tourette's syndrome, the symptoms may wax and wane. Both these factors make it difficult to evaluate treatment effectiveness without long-term follow-up.

Using PET scans and evoked potentials, it has been shown that the brain's response to somatosensory input is abnormal in dystonia. For example, sensory relay fields in the thalamus demonstrate expanded regions where cells all respond to the same passive movement. In addition, the muscle spindle system may be involved. Hallett (1999) stated, "There may be an important problem with the processing of muscle

spindle input." One proposed theory is a model of motor system dysfunction where there is excessive thalamo-cortical facilitation of accessory motor cortical areas (loss of thalamo-cortical selectivity) and a reduction of sensorimotor discrimination, leading to disordered motor planning (Grafton, 1999).

Parkinson's disease is associated with loss of dopamine-producing nerve cells from the substantia nigra, a nucleus in the midbrain. The neurons from the substantia nigra (dopaminergic) project to the corpus striatum. The result of depletion of these neurons is that the corpus striatum is deficient in dopaminergic stimulation yet still has normal innervation from other neurotransmitters such as acetylcholine. Acetylcholine appears to have the opposite effect to dopamine in this region and the resulting imbalance of neurotransmitters is posited to result in the symptoms of Parkinson's disease (Wilkinson, 1988). Symptoms include shaking of the head and limbs, muscular stiffness, an expressionless face, and an inability to control or initiate movement. The usual treatment is to administer levodopa, which can cross the blood-brain barrier and be converted to dopamine. It is usually given with a dopadecarboxylase inhibitor, which does not cross the blood-brain barrier thus preventing a rise in dopamine elsewhere in the body which might otherwise produce side effects such as nausea and vomiting. Nevertheless, an unwanted side effect of raising dopamine in the corpus striatum in some recipients can be choreo-athetoid involuntary movements. Other treatments include trials of anticholinergics to reduce the effect of acetylcholine and thereby address the problem of imbalance between acetylcholine and dopamine in the corpus striatum. Surgical treatments have also been tried and include stereotactic lesions in the thalamus or the globus pallidus (Wilkinson, 1988).

Another movement disorder, Tourette's syndrome (TS), usually begins before a child reaches 16 years of age. The symptoms of stereotyped, repetitive movements comprise motor and vocal tics. The vocal tics are usually involuntary grunts, barks or utterances (including swearing in advanced stages). These tics can be voluntarily suppressed, but often at the cost of discomfort. There is a subjective build-up of muscle tension and then a feeling of release when the person lets himself or herself tic. The symptoms wax and wane. It is generally felt to originate from an imbalance of neurotransmitters in the basal ganglia resulting in a relative excess of dopaminergic activity (Guberman, 1994). There may be dopamine receptor hypersensitivity (Dooley, 2000). It responds to dopamine blockers such as haloperidol or pimozide; however, a danger of long-term use of a drug such as haloperidol can be parkinsonian

symptoms or tardive dyskinesia. Obsessive-compulsive behaviours occur in about fifty percent of patients. Serotonin re-uptake inhibitors that are used when there is concurrent occurrence of obsessive-compulsive behaviours may be beneficial and this suggests a disturbance in the serotonin system (Dooley, 2000). Learning problems occur in about fifty percent of TS patients. Attention Deficit Hyperactivity Disorder (ADHD) may be seen in about fifty percent of TS patients. Neurofeedback enhancing SMR activity has been used for ADHD treatment and good results have also been reported concerning its effects on TS (Tansey, 1986; Daly, 2001).

These three movement disorders all involve involuntary muscle movements. In every case, the basal ganglia (caudate, putamen and globus pallidus) are involved. Interactions between the basal ganglia are probably responsible for smooth, stable movements. Damage to the globus pallidus or the ventral thalamus may cause deficiency of movement (akinesia). Thus, the globus pallidus and the ventral thalamus may be generally excitatory, whereas the caudate and the putamen appear to have inhibitory functions (Carlson, 1986). In each of these movement disorders, an imbalance of neurotransmitters is postulated to be at the core of the difficulty. In each case, increases and or decreases in dopamine are implicated.

Neuroanatomical and Neurophysiological Factors

Control of Muscle Tone–The Muscle Spindle

The muscle spindle is a stretch receptor, which monitors the length of skeletal muscles. It is essential to the maintenance of normal muscle tone. It contains modified, intrafusal muscle fibers, which, run in series with the muscle spindle and in parallel with the extrafusal muscle fibers. These are innervated by gamma motor efferent fibers from the ventral horn of the spinal cord. These motor fibers maintain the sensitivity or "gain" of the spindle. Contractions of the large muscle fibers are facilitated, smoothed out and refined by the action of the gamma motor efferent pathways to the intrafusal fibers of the spindle. Short latency rapid contraction and long latency smoother (afferent responses) contraction are governed by spindle activity (Marieb, 1998). It may be the case that, with top athletes, the effects of shorter latency fibers tends to decrease and the influence of longer latency fiber systems increase so that movements become smooth and not jerky. It is the long latency fibers that SMR training most likely affects. Without this smoothing effect, stretch reflex activity would result in jerky movements.

The brain's information concerning the length and rate of contraction or stretching of muscles comes from two sets of sensory fibers. Type Ia fibers surround the central portion of the intrafusal muscle filaments and are sensitive to the rate and amount of stretch, while the type II fibers surround the ends of the spindle and are only affected by the degree of stretch. The ability to smooth out movements and maintain appropriate muscle tone is dependent on the interplay of information from the sensory fibers and the balancing of the intrafusal muscle tension inside the spindle by the gamma motor efferent system. When the muscle is stretched the fibers of the spindle are also stretched, depolarizing the sensory neurons. This triggers action potentials that are transmitted back to the spinal cord. For example, when you suddenly stretch a muscle by hitting the anterior aspect of the knee with a reflex hammer to elicit a stretch reflex, the spindle is stretched and a signal goes from the muscle spindle to the spinal cord along a special sensory neuron pathway. This neuron synapses with a motor neuron that then conveys a signal to the quadriceps muscle to contract and the leg jerks forward. The sensory neuron also communicates to other motor neurons to inhibit muscles (flexors) which would oppose the contraction of the quadriceps and inhibit the reflex (Marieb, 1998). A more generalized reflexive reaction demonstrating a change in muscle tone occurs when one is startled by a loud noise.

The muscle spindle, however, also has other more complex connections beyond this elementary text-book description of the muscle reflexes. The brain is constantly informed of the position of a muscle through the sensory pathways. The intrafusal fibers of the muscle spindle receive input from connections to the red nucleus in the midbrain, and send sensory information back to pathways that eventually influence red nucleus activity (Sterman, 2000). This reflects the feedback loop organization of the CNS.

In addition to this gamma motor feedback loop, the muscle spindle also has innervation from the sympathetic nervous system (Grassi, Filippi, & Passatore, 1986).

Autonomic Activity

The sympathetic nerves are part of the autonomic nervous system. The latter is involved in running a large percentage of bodily functions and is intimately associated with the limbic system and our emotions. We are now well aware of how changing emotions changes functions within the autonomic system and we routinely monitor such aspects of

this as heart rate, heart rate variability, electrodermal responses, skin temperature, respiratory rate and respiratory sinus arrhythmia (Andreassi, 1995; Budzynski, 1989). We are also aware that, through biofeedback using these modalities, we can alter an individual's emotional state and change body functions in a positive direction. We observe a decrease in muscle tension in clients when they practice breathing from the diaphragm to produce good quality respiratory sinus arrhythmia (RSA) (Budzynski, 1989). Inspiration is associated with increased sympathetic drive and a corresponding increase in heart rate while with expiration, there is a release from the sympathetic drive and the parasympathetic (vagal) system slows the heart rate. Achieving synchrony between breathing and corresponding increases and decreases in heart rate usually occurs at a frequency of six breaths per minute. It is hypothesized that there is a direct effect on muscle spindle activity during these RSA training exercises.

Sensorimotor Rhythm (SMR)

Sensorimotor rhythm is the name given to a particular brain wave pattern that occurs within the lower frequencies of the beta range. SMR is a spindle-like, rhythmic activity in the frequency range 12 to15 Hz that is generated by recurrent bursting of the ventrobasal (VB) nucleus of the thalamus. This imposes a concurrent synchronous discharge on cortical neuronal pools, which is detected in the EEG over the central part of the brain across the sensory and motor (sensorimotor) cortex. When this rhythm is present there is attenuation (decrease) in the conduction of somatosensory information to the cortex and decreased motor excitability, which is why SMR training is used for seizure control (Sterman, 2000). Breathing and heart rate may be observed to slow down, as they do with relaxation, yet the person remains alert (Chase & Harper, 1971).

SMR relates behaviourally to sustained immobility. Being still is necessary but not sufficient. It has been shown that neck muscle activity is suppressed in strict association with the emergence of sensorimotor rhythm (Sterman, 2000; Wywricka & Sterman 1968; Chase & Harper, 1971). The suppression of the neck muscle activity was independent of motor quiescence since it occurred just prior to the appearance of SMR bursts. Sterman notes that because posture had not changed, the abrupt decrease in muscle discharge was attributed to a specific reduction in muscle tone rather than a change in length of the muscle. This observation implicated the gamma motor neuron (muscle spindle) pathway.

Further studies by Sterman demonstrated that the monosynaptic masseteric (jaw closing) reflex, which involves the masseter, which has muscle spindles, was significantly reduced during SMR activity. Activity in the reciprocal polysynaptic digastric (jaw opening) reflex, which involves no muscle spindles, was not affected. This suggests that the mechanisms regulating the gamma motor neuron/muscle spindle system are uniquely involved and related to alterations in SMR activity.

In summary, it has been demonstrated that there is a shift in motor excitability during SMR activity and this perhaps involves reduced output from pathways concerned with the execution and coordination of voluntary movement. In addition, there is a reduction in motor pathway cellular and reflex excitability, and in muscle tone, during SMR activity (Sterman, 2000).

Operant conditioning to increase the SMR can produce sustained changes in physiological regulation. This has been documented during sleep by increased EEG spindle density and stabilization of sleep states. It can affect the hyperexcitability of cortical cellular pools involved in seizure disorders and thereby result in a decrease in the frequency, duration and intensity of seizures (Sterman, 2000). It has also been associated with a decrease in impulsivity and hyperactivity in those with attention deficit disorder and an increased feeling of calm in anxious clients (Thompson & Thompson, 1998). Following on Sterman's work, it is hypoythesized that training for increased SMR activity (rhythmic, thalamically regulated spindle-like, sensorimotor rhythm) has a direct effect on muscle spindle activity, which correlates with a decrease in muscle tone and perhaps a more balanced gamma motor neuron involvement in muscle tension. This may reduce abnormal and unplanned movements.

Theoretical Basis for Biofeedback Interventions

Research carried out over the last 25 years has demonstrated that using an EEG signal to monitor a desired mental state and operant conditioning to prolong this state can be an effective method for altering cortical dynamics so that that desired mental state is sustained. Two clear examples of using EEG biofeedback to train individuals to alter their neurological states are the treatment of seizure disorders (reviewed by Sterman, 2000) and attention deficit disorder (reviewed by Lubar, 1997; Chabot, di Michele, Prichep, & John, 2001). The improvement seen with SMR training in clients with movement disorders may be analogous to the results with epilepsy and ADHD since all three

training paradigms involve trying to increase SMR activity as a means of decreasing unwanted motor activity.

If one critical factor in dystonia is, as suggested by Hallett (1999), that a problem exists with the processing of muscle spindle input, then encouraging a mental state which correlates with decreased motor excitability and decreased muscle tone should have a positive effect on dystonia. It may also explain some of the observed effects with Tourette's syndrome clients who have fewer tics after completing neurofeedback training to increase SMR (Daly, 2001; Tansey, 1986). Increases in SMR activity do correlate with decreased muscle tone and relaxation of muscle activity. SMR is merely a flag, which signals this mental state. Using this flag (SMR EEG activity) should enable us to train a patient using operant conditioning, to sustain the desired state wherein muscle tone is decreased in association with changes in the gamma motor neuron muscle spindle system. In addition, the autonomic nervous system can also have effects on muscle spindle. There are direct sympathetic connections to muscle spindles. There is also a large sympathetic output from the posterior hypothalamus to the tegmentum of the midbrain, which includes the red nucleus. Theoretically this would have a modulating effect on the activity of the red nucleus and thus on the activity of muscle spindles. The result would be less unplanned and unwanted movements.

If this hypothesis were borne out in clinical experience, it would provide a method of treatment that is free of side effects and that may provide the patient with some reasonable (not total) relief of symptoms and discomfort.

Case Study

Mary (fictitious name), age 47, has suffered from Parkinson's disease (PD) for 14 years and from dystonia for the last 5 years. There was no clear cause for her PD, though she had been in a car accident in 1975 and, despite wearing a seat belt, hit her forehead on the dashboard and was briefly unconscious. A second possible factor was exposure to paint fumes and varnish when she owned a craft store from 1982 until 1990. She has generalized dystonia, which affects her left side most. Mary's first episode of dystonia occurred when she was taken off all medication in 1996 to establish a baseline of symptoms for a research study involving the transplantation of fetal cells. While off medications her facial muscles suddenly tightened, her jaw clenched, her left leg muscles constricted and her left shoulder clenched up to meet her head

which pulled to the left. She described a typical dystonic contraction thus: "The top half of my jaw twists to the right and the lower part twists to the left with such force that it feels as if my mouth is going to rip apart." Her favourite exercise had been walking but she had to curtail this due to leg cramps.

It has not been possible for Mary to hold a regular job since 1994. She had worked for an accountant after selling her craft store but it became increasingly hard to write down phone messages and she started to have trouble with driving, becoming sleepy at the wheel. (Hobson, 2002 has written on extensive day-time sleepiness in PD.) She is now on a disability pension but she keeps busy with painting and also has a book at the publisher about her experience with Parkinson's. Mary is creative, writing and illustrating stories for relatives and friends.

Mary had noticed a decline in her ability to concentrate. She was finding it harder to stay focused and finish things. Her mind was always full of ideas, which she said made it hard to complete the task at hand. For a few years, she had not been able to finish reading a book, though she had previously been an avid reader. Sleep was another difficult area. Quality of sleep was poor with waking after a couple of hours, wake until about 6 AM, then sleeping for another two and one-half to three hours. During the day, she had one or two naps. In cars, she would often fall asleep and she was no longer driving herself. There was lack of appetite. She took a multivitamin/multimineral supplement and extra vitamin C and E as well as other antioxidants daily. She drank about three quarts of water a day as anti-Parkinson drugs produced thirst. Difficulty swallowing certain foods also affected her diet.

Other than the Parkinson's disease and dystonia, Mary enjoys good general health. A positive outlook has helped her face her challenges. Adjunctive approaches she had tried included massage, meditation and yoga, though she found it difficult to meditate due to difficulties sustaining her focus. Nevertheless, she found that attempts to meditate did help with pain management when she was facing surgery.

Mary has actively sought treatment and took part in a study involving the transplantation of fetal cells into the caudate. She had surgery December 1996 but a year later, it was revealed to her that she had been in the placebo group. She had been very positive about the treatment and initially felt some benefit but the results were not sustained and she began getting worse after about six months. She had the real surgery in December 1998 and this time there were real benefits. She had been almost immobilized prior to that second surgery–either very dyskinetic or just lying on the couch. After surgery, she regained her mobility. Mary

was the subject of a television episode on "The Nature of Things." She has been a spokesperson for better understanding of Parkinson's disease. When doing this public speaking, the adrenalin rush before she speaks increases her symptoms (both "freezing" and muscle spasms) but with her usual optimism, she notes that this helps people connect with her.

Regarding medications, Mary described those she was taking at the time she started training as follows. Sinemet (levodopa and carbidopa combined) is necessary to keep her mobile. She has been taking it for 14 years, since she was first diagnosed. Amantadine, which blocks the reuptake of dopamine by presynaptic terminals (Wilkinson, 1988) was taken to lessen the dyskinesia that accompanies the use of sinemet. She took one or two each day. Permax (pergolide) was being replaced with Requip, which is classified as an agonist and supplements the sinemet. Requip was the latest agonist and she had been on a number of different ones over the years. Ativan (lorazepam) was usually taken once or twice daily to help relieve her rigidity and feelings of tightness inside her body. General sleepiness and sudden sleep attacks may be related to its use. She stated that Prepulsid (cisapride) kept her "internal" muscles working smoothly. Otherwise, she got reflux. When she went to New York for her one-year post-surgical follow-up in January 2000, she was taken off all medications for some of the studies. Some, such as an MRI, could not be conducted because of her uncontrollable movements. It took a number of months thereafter to regain balance in terms of her medications.

Mary heard about neurofeedback when she was speaking in Winnipeg and, being open to alternative treatments, she did a couple of sessions with a practitioner there. She contacted our centre upon her return home to southern Ontario. She was assessed in April 2000 and began training in June 2000. She was informed that this would be an experimental application of neurofeedback and we emphasized the complete lack of studies and literature in this area. The distinct EEG profile of very low 13 to 15 Hz activity justified trying neurofeedback to attempt to normalize her pattern. She also demonstrated high 9 Hz activity. She and other Parkinson's patients had related to us problems with daytime sleepiness unrelated to medications. A similar observation has been recently reported (Hobson, 2002). Our initial objective was to obtain improved quality of sleep. We also wondered if the SMR training might result in somewhat reduced tremor. We hypothesized from her history that some of her cramping might be related to tension and that relaxation training might benefit those symptoms. It was also reasonable to

postulate that her ability to focus and concentrate might improve since we were using an EEG protocol similar to that used for clients with ADHD, namely, theta suppression and SMR increase (Lubar, 1991; Thompson & Thompson, 1998).

METHODS

When she presented at our centre for the initial interview the small jerking movements kept escalating into uncontrolled gross flailing of the limbs. The interview had to be discontinued. It was difficult to begin neurofeedback even though she tried to come during her "good" period of the day when the L-dopa was having maximum effect. Sequential placement at "FCz-CPz" was used initially in an attempt to reduce EMG and movement artifact. (This is taught by J. Lubar as the suggested placement of electrodes if bipolar placement is chosen when working with teenagers or adults with ADHD. It refers to sites half way between Fz and Cz and half way between Cz and Pz using the standard 10-20 placement system.) Neurofeedback, which encouraged an increase in 13 to 15 Hz activity, was combined with RSA training; that is, breathing with her diaphragm and watching the computer monitor to see if she could maintain synchrony in the rising and falling of two line graphs that showed her breathing and her heart rate. After completing a few sessions, she was able to control her movement sufficiently for us to begin using a referential placement with one active site at Cz referenced to the left ear lobe. The equipment used was usually the Procomp+/Biograph (Thought Technology), an instrument that allows simultaneous neurofeedback and up to six kinds of biofeedback. A description of screens used is found elsewhere (Thompson & Thompson, 2001). On some occasions she trained using F-1000 equipment (Focused Technology), beginning with feedback that encouraged breathing at six breaths per minute (bpm) and then doing neurofeedback to raise 14 Hz. (The F-1000 instrument has a feedback option called the Tansey screen that allows the client to focus on increasing 14 Hz activity.)

EEG assessment demonstrated both extremely low SMR and an unusually high alpha peak (eyes open) at 9 to10 Hz. (This finding was consistently observed in a dozen other clients with PD who volunteered for informal EEG assessments at a Parkinson's disease fund-raising picnic.) At the time of assessment, she was only able to print with great difficulty and only when not having a severe dystonic episode. She had difficulty reading, in part due to movement and in part due to a short at-

tention span. It was impossible, due to distance and volunteer driver availability, for Mary to continue to regularly attend two sessions a week. In her first four months of training, she made it to 30 sessions but then had to stop due to a driver not being available. She has recently returned to continue treatment on a once a week basis.

The first 10 minutes of her one-hour sessions were spent practicing deep, regular, diaphragmatic breathing at a rate of six bpm. She could observe the synchrony between inspiration and heart rate increase along with expiration and heart rate decrease on the screen. At the same time, she and the trainer working with her could track on the feedback screen her trapezius electromyogram (EMG), skin conductance (EDR) with sensors on index and ring fingers of one hand, and skin temperature on the little finger. Heart rate was measured with a plethysmograph on the thumb and breaths per minute were measured with a strap around the abdomen. While doing the breathing she practiced relaxing her neck and shoulder muscles and increasing her peripheral skin temperature. Mary learned these procedures rapidly and practiced them at home without feedback.

Neurofeedback combined with biofeedback was carried out for the next 50 minutes of each hour-long session. She was still able to monitor breathing rate, expressed as a number for bpm, and skin temperature while watching the neurofeedback screens. For the first month, she did manage to come in twice a week. Increasing the SMR activity (13 to15 Hz) was Mary's prime focus and all screen animations were directly attached to SMR production. The animations and sound feedback would not, however, register if either one of the two frequency bands set as inhibits was above threshold; that is, if the amplitude of those frequency bands were above a set threshold (derived from her baseline measures) then she would not receive the reward of hearing tones and seeing something happen on the screen. For Mary, one inhibit was placed on her dominant slow wave activity (6 to 10 Hz) and another was on high-beta activity (25 to 32 Hz). Her initial assessment had shown high 29 to 31 Hz activity which corresponded to her self-reports of worrying and ruminating. If she were distracted, it would decrease. She complained of dysphoria bordering on depression. The inhibit on high-beta (25 to 32 Hz) thus served a double purpose. It warned her of this tendency to ruminate, encouraged relaxation and acted as an EMG inhibit. A further inhibit on the EEG display (lines above and below the electroencephalogram that was shown at the top of the screen) showed when the amplitude of the EEG exceeded a set criterion. This also helped to decrease artefacts due to her movements since she would be instantly

aware of the problem and could try to modify her muscle activity and remain motorically calm.

As of February 2002, Mary had had 42 one-hour training sessions. She had 30 sessions on a regular basis (starting twice a week but decreasing due to travel logistical problems) from April to December 2000 and a further 12 sessions on an irregular basis thereafter.

RESULTS

One of the first benefits reported by Mary (after about 12 sessions) was being able to finish reading a book over a weekend. She had not been able to do this for three or four years prior to training. There has also been a reduction in dystonic movements and movements in general are smoother. She used to frequently walk with both a cane and sometimes another person to support her when she came for training and now she walks unassisted. When she went for her two-year post-surgical follow-up for the fetal cell implant study she again had all medication withdrawn but this time she was still able to manage. They could complete all her tests, including her staying still enough for an MRI. Because it is a research study, she was not told her results but her impression was that they were very pleased with her progress. Unlike her one-year follow-up when it took months to get her medications back in balance after they had been withdrawn, things were back to normal in a few days.

Eighteen months after her first training session Mary says that the main difference she finds in terms of quality of life is that she can "breathe her way out of freezing." Freezing (becoming frozen and unable to initiate movement) is a quite disabling symptom that is virtually inevitable in advanced cases of Parkinson's disease. Mary, for example, used to frequently find herself lying in bed unable to get up unless someone helped her start moving. (The differential diagnosis for the immobility in bed had to include narcolepsy, but Mary did not have either catalepsy or hypnagogic [falling asleep] or hypnopompic [waking up] experiences while in this state.) By the time she had done 30 sessions of training, she could control the freezing. When it occurs, she breathes diaphragmatically at a rate of six bpm, almost immediately regains motor control, and is able to initiate movement. An example was her description of getting up the morning of her 42nd session: upon awakening, she was frozen and unable to move so her husband hurried downstairs to fetch her medication. By the time he returned, she had gotten herself out

of bed with breathing (that is, breathing at six bpm, which had during training been paired with the production of SMR activity). This freezing occurs while walking on flat surfaces as well. Once, when doing public speaking for a Parkinson's organization, she found herself frozen and facing the wrong direction, away from the audience. She reported that she just told the audience to bear with her, started her breathing exercise and put herself into the "mental state" which she knows correlates with increased SMR activity. Almost immediately she was 'released' and could face the audience, take the microphone and speak. She wrote and illustrated a story about that experience. Mary recently described the process she uses as starting with diaphragmatic breathing and consciously putting her brain in the right zone. She feels the messages going from her brain to her body. She states, "I can feel my arms, legs and face relaxing, like they are sighing."

When she came to the centre for her first few sessions, she had to be helped into a chair. At times, the movements were so extreme that it was not possible to even sit still in a chair and certainly, no electrodes could be put in place. At other times, she could sit but it would take a while to get the electrodes on with acceptable impedance below 5Kohms because she was jerking so much and could not hold her head still. As noted above, only a sequential ("bipolar") placement could be made initially. Now she walks in without even using a cane, sits down (often perfectly still, sometimes with a little tremor evident) and the attachment of electrodes is simple.

Mary has decreased her daily medications. Sinemet (levodopa plus carbidopa) has been reduced significantly: initially she took two or three long release (Sinemet CR) and seven or eight regular tablets, now she takes just one-half of a CR tablet at bedtime and six or seven regular tablets in the course of a day. The amantadine usage remains the same. She has discontinued the use of Permax, Requip and Prepulsid. Comtan, a catechol-O methyltransferase (COMT) inhibitor, replaced the Requip eight months ago. An agonist assists the action of levodopa. (She has used a series of agonists over the years. They are necessary so that the dopamine effects of the L-dopa are limited to the brain and are blocked from affecting the rest of the body.) Ativan (lorazepam) is taken only occasionally on an as-needed basis rather than daily, as was previously the case. To have better functioning on less medication is particularly pleasing as it may extend the period over which the L-dopa is effective.

DISCUSSION

Training to control mechanisms that govern muscle movement and tone by means of neurofeedback (increasing SMR) plus biofeedback (synchrony between heart rate variability and respiration) appears to be a potentially important focus for intervention in movement disorders. The hypothesis is that both kinds of training have an effect on muscle spindle activity and this makes it possible to decrease unwanted movements and initiate desired movement. Since fibromyalgia is linked to muscle spindle activity and there is evidence of marked sympathetic hyperactivity in that disorder, the same rationale for using SMR enhancement and RSA training could apply to that condition (Martinez-Lavin, Hermosillo, Rosas, & Soto, 1998). With respect to this reported case, it is important to note that there is no cure for Mary's progressive Parkinson's disease, yet she is doing better than she has done in many years. She still has Parkinson's symptoms and occasional serious attacks of dystonia but she is regularly able to breathe her way out of freezing episodes and is much more independent and mobile. We postulate that this breathing may directly affect the muscle spindle system. However, it may also act as a behavioural cue to produce a mental state that is associated with an increase in SMR. Though she can become discouraged and depressed when symptoms increase, which occurs during times of stress, she reports that she generally feels much more in control of her body. The reduction in medication use, when the usual course of PD requires increasing medication over time, is particularly encouraging. Both of her disorders have symptoms that wax and wane, so this could be a period of time when the symptoms would have lessened even without biofeedback. On the other hand, the length of time with improved functioning (now approaching two years) suggests that neurofeedback combined with biofeedback has been helpful and is likely among the factors responsible for the observed improvements. It may also be that the fetal cell implants have played a role. It is interesting that her two-year follow-up (with 30 neurofeedback sessions done in the preceding six months) was easier for her than her one year follow-up in terms of fewer dystonic and dyskinetic problems and a quick return to normal (for her) functioning when medication was re-started, rather than taking months to get her equilibrium back. Nevertheless, her ability to "self-regulate" would point to biofeedback being an important contributor to her improved functioning. There has been considerable symptom relief and an increased quality of life; for example, being able to resume favourite activities like reading novels and producing crafts.

She wrote and illustrated a booklet about her experience, entitled "Neurofeedback and Me." There can be little doubt that Mary has achieved a degree of self-regulation and control that has led to enhanced feelings of self-efficacy.

Placebo effects might have contributed to the initial improvements. A physical basis for placebo effects (increased dopamine when placebo was injected) has recently been reported in Parkinson's disease (de la Fuente-Fernandez, Ruth, Sossi, Calne, Schulzer, Steossi, et al. 2001). Mary's positive outlook and enthusiasm for trying experimental procedures might enhance non-specific effects in her case. The length of time that results have been maintained, however, argues against this being the main cause of improvement, especially since the placebo effect after sham surgery wore off in a much shorter period with Mary.

Future research will have to investigate whether these results can be reliably replicated with a large number of clients. Long-term follow-up will be necessary to see if results are sustained after a certain period of training or if on-going, regular training is necessary for best management of symptoms. Double blind studies might be designed, though this is difficult since clients might readily detect that they were getting sham feedback. Research could also be done to assess the relative contribution of effects on the autonomic (sympathetic) nervous system as compared to skeletal-muscle innervation through cholinergic pathways and to assess the relative contributions of neurofeedback, RSA biofeedback and breathing training.

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RECEIVED: 10/15/01
REVISED: 03/09/02
ACCEPTED: 04/02/02