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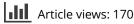
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The effects of a relaxation program featuring aquatic therapy and autogenic training among people with cervical dystonia (a pilot study)

Ana Isabel Useros-Olmo PhD, PT^{a,b}, David Martínez-Pernía, PhD, Psyc, PT^{c,d,e,f}, and David Huepe PhD^c

^aDepartment of Physiotherapy, Motion in Brains Research Group, Instituto de Neurociencias y Ciencias del Movimiento, Centro Superior de Estudios Universitarios La Salle, Universidad Autónoma de Madrid, Madrid, Spain; ^bUnidad de Daño Cerebral. Hospital Beata María Ana, Madrid, Spain; ^cCenter for Social and Cognitive Neuroscience(CSCN), School of Psychology, Universidad Adolfo Ibáñez, Santiago, Chile; ^dLaboratorio de Neuropsicología y Neurociencias Clínicas (LANNEC), Facultad de Medicina, Universidad de Chile, Santiago, Chile; ^eGeroscience Center for Brain Health and Metabolism (GERO), Santiago, Chile; ^fClínica de Memoria y Neuropsiquiatría (CMYN), Servicio de Neurología. Hospital del Salvador y Facultad de Medicina, Universidad de Chile, Santiago, Chile

ABSTRACT

Classic physical interventions for cervical dystonia (CD) have focused on treating motor components or, on motor components and relaxation programs. However, no CD treatment study has focused on a relaxation program alone. We developed a pilot study to assess whether a therapy completely based on a relaxation program could improve the physical and mental symptomatologies of patients with CD. Fifteen persons were included in the experimental group, which received individual sessions of aquatic (Watsu) therapy (WT) and autogenic training (AT). In addition, 12 persons were included in passive control group. We administered different questionnaires related to quality of life (SF-36), pain (Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) and Visual Ánalog Scale (VAS)) and mood (Beck Depression Inventory (BDI-II) and State-Trait Anxiety Inventory (STAI)). A significant interaction was observed between treatment and time with regard to the SF-36, VAS, and TWSTRS within the experimental group (p < 0.01). The BDI-II showed depression decrease as a simple effect (p < 0.05), and the STAI did not change. No effects were found with regard to the control group. In this exploratory study, we found that a therapy based on whole body relaxation improved the symptoms of patients with CD. This knowledge enables a disease-management strategy that uses a holistic perspective and moves beyond the dystonic focus.

Introduction

Cervical dystonia (CD), sometimes referred to as spasmodic torticollis, is a movement disorder characterized by involuntary contractions of the neck muscles, leading to a disabling abnormal head posture (Fahn, Marsden, and Calne, 1987). Although the etiology of idiopathic CD is unknown, it is likely caused by genetic and environmental factors (Ozelius, Lubarr, and Bressman, 2011; Phukan, Albanese, Gasser, and Warner, 2011) or exogenous causes (e.g., associated with heredodegenerative disorders or part of a dystonia-plus syndrome) (Dusek, Jankovic, and Le, 2012; Phukan, Albanese, Gasser, and Warner, 2011). This nonspecificity regarding the etiology of the disease leads to difficulty in the care management of these patients as well as with regard to their rehabilitation (Crowner, 2007). CD is a multidimensional disorder rather than a sensorimotor presentation in which

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physical, emotional, cognitive, and self-awareness aspects are affected (Wagle Shukla et al, 2016; Werle et al, 2014). Examples of these multidimensional factors include the previous findings showing that: (1) pain is associated with psychological aspects such as depression and anxiety (Ben-Shlomo, Camfield, Warner, and Epidemiological Study of Dystonia in Europe Collaborate Group, 2002); (2) pain associated with postural deviation is present in 75% of patients with CD, and it is the most common cause of disability (Kutvonen, Dastidar, and Nurmikko, 1997); (3) selfperceived nonmotor aspects (e.g., sleep, fatigue, mood, and cognitive processing) are key determinants of disability (Zetterberg, Lindmark, Söderlund, and Åsenlöf, 2012), and how they influence physical symptoms of CD (Jahanshahi, 2000; Soeder et al, 2009); and (4) patients with CD have higher rates of anxiety and depression and more frequent psychiatric comorbidities (Fabbrini et al, 2010; Gundel et al, 2001). In

CONTACT Ana Isabel Useros-Olmo anaiuseros@gmail.com Department of Physiotherapy, Motion in Brains Research Group, Instituto de Neurociencias y Ciencias del Movimiento, Centro Superior de Estudios Universitarios La Salle, Universidad Autónoma de Madrid, Madrid, Spain. Color versions of one or more of the figures in the article can be found online at www.tandfonline.com/iptp.

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summary, CD is an important health problem influenced by multiple factors that produce an overwhelming effect on all levels of daily living and the personal factors of individuals (De Pauw et al, 2014).

In relation to motor rehabilitation, physical therapy interventions used to treat CD are not standardized (Bleton, 2010), which hinders care management of physical therapists (De Pauw et al, 2014). Currently, this discipline includes a variety of therapeutic approaches that shift the basic assumptions of classic motor therapy to a methodology rooted in mind-body interventions. Despite the extensive literature on mindbody interventions regarding recovery from physical disorders, few studies have focused on CD. These limited studies included combined relaxation programs with conventional physiotherapy (Andrews and Gill, 1982; Boyce et al, 2013; Duddy and McLellan, 1995; Gildenberg, 1981; Jahanshahi, Sartory, and Marsden, 1991; Zetterberg et al, 2008); and a combined cognitive-behavioral intervention and mindfulness program (Sandhu et al, 2016). Although 73% of patients with CD can be involved in interventions based on complementary and alternative medicine (Junker, Oberwittler, Jackson, and Berger, 2004), no scientific research has specifically focused on mind-body approaches. All of the studies based on mind-body approaches have been implemented jointly with conventional physical treatment; thus, we do not know the efficacy of this therapeutic view regarding the treatment of CD because the outcomes are always confounded with conventional physical intervention. The aim of this study was to evaluate the effects of a mind-body therapy on pain, mood, and quality of life (QoL), in adult patients with CD. For this purpose, we designed a quasi-experimental study with experimental and control groups without random assignment. The experimental group received an aquatic therapy known as "Watsu" (Dull, 2004) and autogenic relaxation training (Schultz and Luthe, 1959) for 1 month; the control group did not receive treatment.

Methods

Participants

The participants for this study were recruited from the Asociación de Lucha contra la Distonía en España (ALDE). The recruitment procedure targeted all of the participants who fulfilled the inclusion criteria by sending a letter informing them about the research (Figure 1). The inclusion criteria were that the participants must have been diagnosed with CD by a neurologist at least 5 years before the beginning of the study.

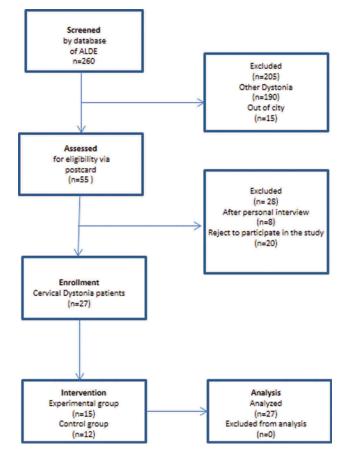


Figure 1. Flowchart of recruitment and participation via the Asociación de Lucha contra la Distonía en España.

Participants were excluded when they had secondary dystonia or had scheduled an infiltration of botulinum toxin during the clinical intervention. Given these criteria, 27 people were enrolled in the research. Of these people, 15 were interested in receiving the clinical interventions. The other 12 people did not participate because they were not interested or they were unable to attend all of the sessions; however, all participants completed the questionnaires at the beginning and end of the study. The participant demographics are shown in Table 1. All participants (experimental and control) signed an informed consent document prior to participation. The Ethical Committee of the ALDE approved this study.

Table 1. Participant demographic data.

	Gro	oups				
Variable	Control	(<i>N</i> = 12)	Experimer	ntal ($N = 15$)		
Age	54.08	+ 12.32	47.8 + 14.22			
Years of study	15.75 + 3.17		16.07 + 3.22			
Years with dystonia	11 + 4.53		14.2	14.2 + 9.5		
	Perce	entage	Perc	entage		
Sex	Men	Women	Men	Woman		
	33.3%	66.7%	6.7%	93.3%		

Intervention

The intervention group received four individual sessions (one per week) of an aquatic therapy known as Watsu therapy (WT) and autogenic training (AT) over 1 month. In addition, they practiced AT for 15 min, twice a day at home, with instructions provided via an audio compact disc. Participants in the control group did not receive the intervention.

WT is an aquatic intervention created by Dull (2004) in the 1980s that aims to engender deep physical and mental relaxation. WT has been applied to rehabilitate people with different neurological pathologies and physical conditions (Chon, Oh, and Shim, 2009; Faull, 2005; Leite et al, 2013; Schitter et al, 2015). The intervention in our research was managed by a physical therapist specialized in WT and certified by the Watsu Europe Institute. During each 50-min session, the physical therapist applied the same protocol to all participants (Table 2), which consisted of passive mobilizations following a basic sequence of movements adapted to patients with CD. Participants assumed a supine position in the water (33-35°C), held by the therapist and floats. They were instructed to close their eyes and focus their attention on their own body sensations while trying to increase their body relaxation through passive movements.

After each WT session, participants completed 15 min of AT in a room adjacent to a swimming pool. The standard procedure developed by the German psychiatrist Johannes Schultz was applied (Luthe, 1969; Schultz and Luthe, 1959). This technique has widely been applied for people with motor disorders (Ajimsha, Majeed, Chinnavan, and Thulasyammal, 2014; Kneebone, Walker-Samuel, Swanston, and Otto, 2014; Sugimoto, Theoharides, Kempuraj, and Conti, 2007; Sutherland, Andersen, and Morris, 2005) and for pain management (Gisèle Pickering et al., 2012; Kanji, 2000; Kanji, White, and Ernst, 2006; Kwekkeboom and Gretarsdottir, 2006; Sadigh and Montero, 2001). At the beginning of the session, participants assumed the supine position to reduce the tension in their cervical muscles. Then, they repeated a set of visualizations to induce a state of relaxation (e.g., "I am at peace," "My right/left arm is heavy," "My right/left arm is warm," "Breathing calm and regular," and "Heartbeat calm and regular"). At the end of the sessions, participants could ask questions of an experienced psychologist who managed their concerns.

In addition, patients performed AT twice a day at home, except on the days they received WT, when they practiced AT just one more time at home. With the aim of having control over the number of AT sessions participants performed, at the beginning of the study,

Table 2. The	sequence	of movements	applied	to	patients	with
CD based on	WT (Dull,	2004).				

Name of the technique	Description
Water breath	Stillness and gentle motion is coordinated with up
dance	and down shifts in buoyancy as the receiver breathes.
Slow offering	Gently pull alternately headward by the base of the
j	skull (occiput) and footward by the base of the spine
	(sacrum) to produce traction of the spine along with
	centripetal side force.
Free spine	Gently oscillate the spine. This freeing of the spine is
free spine	so important that it is the focus at the beginning of
	each Watsu session.
One-leg offering	Gently pull alternately headward by the base of the
one leg onening	skull (occiput) and footward by one leg to produce a
	complex pattern of turbulent drag with leg, torso, and
	spine stretches.
Two-leg offering	Gently pull alternately headward by the base of the
Two leg offering	skull (occiput) and footward by both legs to produce a
	complex pattern of turbulent drag with leg, torso, and
	spine stretches.
Accordion	Draw both knees toward and away from the chest in
Accordion	coordination with breathing while maintaining neck
	traction.
Rotating	Add a spiraling rotation to the accordion action of
accordion	moving both knees toward and away from the chest
accordion	while maintaining support of the occiput and allowing
	the head more freedom to roll.
Near-leg	Open and close the near leg toward and away from
rotation	the chest to produce an oscillating spine and hip
Totation	rotation.
Far-leg rotation	Open and close the far leg toward and away from the
rui ieg rotation	chest to produce an oscillating spine and hip rotation.
Stillness	Let the patient float perfectly still in front of you. Feel
Stimess	the stillness in your own body.
Follow	Listen with your hands under the head and sacrum for
movement	any tendency to move.
Seaweed	Slowly move the patient from side to side like
Scaweeu	seaweed. Permits mobilization and rotation of the
	spine and hips.
Explore	Listen for and slowly follow whatever movement or
movement	tendency to move appears in the patient.
Rock the heart	Sinking to the foot, lower your right shoulder and slip
nock the neur	the near leg over your shoulder with your hand. Place
	the right hand on the heart's center and shift forward
	with each exhaled breath.
Completion	Lean the patient back against the wall, supporting
compiction	them with the hands on each side of the chest and
	hold the heart's center until the patient is ready to
	stand on their own. It is important to prepare people
	beforehand so that when the wall is behind them or
	the steps underneath them; they will then know the
	Watsu session has ended.
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the participants were given a form to note the number of sessions they completed per day. In addition, a psychologist called the participants every week to collect the number of sessions they performed and encourage them to keep working on AT. All patients performed more than 90% of the relaxation sessions.

Outcome measures

To discover the changes produced by the intervention, we administered different questionnaires related to QoL, pain, and mood. These questionnaires were applied to both groups (experimental and control) prior to the beginning of the clinical interventions and when they concluded. The posttreatment assessment was realized during the first week after the end of the therapeutic intervention.

QoL was determined using the 36-Item Short-Form Health Survey (SF-36). We administered a Spanish version that was previously validated (Alonso, Prieto, and Antó, 1995). The SF-36 contains 36 questions that can be categorized into 8 subscales: physical functioning, role physical, bodily pain, role emotional, vitality, mental health, general health perception, and social functioning. Each item is scored from 1 to 100, with higher scores indicating better QoL. The reliabilities of all SF-36 dimensions were high in this study (Cronbach's $\alpha = 0.89$). Two dimensions were obtained through exploratory factor analysis: (1) "corporal and physical perception dimension" composed of: "physical function," "physical role," and "body pain," and (2) "subjective-social-emotional dimension" composed of: "mental health," "emotional role," "vitality," "general health," and "social function."

Pain was measured using two questionnaires. The first was the pain subscale of the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS), which is the most common standardized assessment of pain associated with spasmodic torticollis (Novak, Campbell, Boyce, Fung and Cerebral Palsy Institute, 2010)

Each item is scored from 0 to 5. The second pain questionnaire administered was the Visual Analog Scale (VAS), a popular pain rating scale (Williamson and Hoggart, 2005). The administration consisted of showing the participants a numbered scale from 0 (no pain) to 10 (worst imaginable pain). The participants scored the level of pain that corresponded to their feelings.

Finally, we administered the State-Trait Anxiety Inventory (STAI) (Spielberger, Gorsuch, and Lushene, 1970) and the Beck Depression Inventory (BDI-II) (Beck, Steer, Ball, and Ranieri, 1996) to measure mood changes. The STAI is a self-report questionnaire composed of two subscales: The State Anxiety Scale (20 items) and the Trait Anxiety Scale (20 items). STAI is scored from 0 to 3, with higher scores denoting greater levels of anxiety. The BDI-II is a 21-question inventory that measures the severity of depression. It is scored from 0 to 3, and higher total scores indicate more serious depressive symptomatologies.

Data analyses

This study followed a typical experimental design with a pretest, intervention, and posttest phases. The statistical analyses consisted of a factorial analysis of variance (ANOVA) for a mixed 2×2 design to test for differences between the two independent groups (experimental vs. control) over time (pretest vs. posttest). Particularly, this is the best statistical technique for evaluating two or more independent variables (as intervention groups, body pain) in which at least one of the independent variables is measured between subjects (i.e., different subjects serve under each level of that independent variable) and at least one of the independent variables is measured within subjects (i.e., the same subjects or matched sets of subjects serve under all levels of that independent variable) (Sheskin, 2003). Another great advantage of this approach is identifying interactions between the experimental conditions (between factors) and specific variables to measure the treatment effect (within factor).

Results

Descriptive statistics

Table 3 shows the descriptive results of the measures used. Almost all of the SF-36 dimensions showed mean differences between the experimental and control groups (with the exception of physical function) in the expected direction at time 2 (postintervention). Likewise, variations were observed in the averages for the additional scales used to assess physical pain (VAS and TWSRTS), the STAI and levels of depression (BDI-II). In addition, group differences were calculated by means of an independent *T*-test to show that both groups were comparable at baseline. Differences were found only in the emotional role (higher for the control group). The rest of the variables did not show statistically significant differences.

Intervention effects

Each SF-36 variable used in the hypothesis (i.e., "corporal and physical perception dimension" and "subjective-social-emotional dimension") test was previously tested for the normality of its distribution via the Kolmogorov-Smirnov test and the assumption was met. To evaluate the effect of the intervention on the function of each variable of interest, 2×2 ANOVAs were performed assuming equal or unequal covariance matrices where appropriate. In Table 4, the tested effects are shown.

In the experimental group, effects were found in the expected direction at time 2 (postintervention) for variables "corporal and physical perception dimension," "subjective-social-emotional dimension," VAS, TWSTRS, and BDI-II (Figure 2a–2e). Nevertheless, the STAI inventory did not

Table 3. Effects of relaxation	program	featuring	aquatic	therapy	autogenic	training.

$\begin{array}{c c c c c c c c c c c c c c c c c c c $	Experimental group	Pre	Post					
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	(<i>N</i> = 15)	(M ±SD)	(M ±SD)	Differences of means	SD from differ.	95% IC	d	р
Body pain 36.20 ± 24.25 54.80 ± 24.35 -18.60 14.23 $(-26.48 a - 10.72)$ 0.77 < 0.001 General health 37.20 ± 15.63 42.27 ± 19.89 -5.07 14.43 $(-13.06 a 2.93)$ 0.29 $ns.$ Vitality 38.20 ± 24.96 55.73 ± 21.34 -17.53 18.97 $(-28.04 a - 7.03)$ 0.76 < 0.01 Social function 47.50 ± 29.58 70.00 ± 25.35 -22.50 26.39 $(-37.11 a - 7.89)$ 0.82 < 0.01 Mental health 44.27 ± 23.54 61.87 ± 18.02 -17.60 17.81 $(-27.46 a - 7.74)$ 0.85 < 0.01 Pain scale (TWSTRS) $11.13 + 6.53$ $8.87 + 5.68$ 2.27 2.43 $(0.92 a 3.61)$ 0.37 < 0.01 Pain scale (TVAS) $4.80 + 3.23$ $2.27 + 2.19$ 2.53 2.36 $(1.23 a 3.84)$ 0.93 < 0.01 Pain scale (VAS) $4.80 + 3.23$ $2.27 + 2.19$ 2.53 2.36 $(1.23 a 3.84)$ 0.93 < 0.01 Pain scale (VAS) $4.80 + 3.23$ $2.27 + 2.19$ 2.53 2.36 $(1.23 a 3.84)$ 0.93 < 0.01 Pain scale (VAS) $4.80 + 3.23$ $2.27 + 2.19$ 2.53 2.36 $(1.23 a 3.84)$ 0.93 < 0.01 Pain scale (TVSTRS) $(1.43 + 3.20)$ $0.67 + 34.04$ 10.47 24.86 $(-3.30 a 24.24)$ 0.34 $ns.$ Control groupPrePotNotDifferences of meansSD from differ. 95% IC d p Mysical function<	Physical role	25.00 ± 41.19	43.33 ± 44.79	-18.33	33.36	(-36.81 a 0.14)	0.43	< 0.05
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $	Physical function	57.00 ± 29.57	57.33 ± 11.11	-0.33	11.87	(-6.91 a 6.24)	0.02	ns.
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	Body pain	36.20 ± 24.25	54.80 ± 24.35	-18.60	14.23	(-26.48 a - 10.72)	0.77	< 0.001
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	General health	37.20 ± 15.63	42.27 ± 19.89	-5.07	14.43	(-13.06 a 2.93)	0.29	ns.
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $	Vitality	38.20 ± 24.96	55.73 ± 21.34	-17.53	18.97	(-28.04 a - 7.03)	0.76	< 0.01
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $	Social function	47.50 ± 29.58	70.00 ± 25.35	-22.50	26.39	(-37.11 a - 7.89)	0.82	< 0.01
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	Mental health	44.27 ± 23.54	61.87 ± 18.02	-17.60	17.81	(–27.46 a – 7.74)	0.85	< 0.01
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	Emotional role	28.84 + 41.53	51.07 + 48.60	-22.22	43.00	(-46.04 a 1.58)	0.49	ns.
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	Pain scale (TWSTRS)	11.13 + 6.53	8.87 + 5.68	2.27	2.43	(0.92 a 3.61)	0.37	< 0.01
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	Pain scale (VAS)	4.80 + 3.23	2.27 + 2.19	2.53	2.36	(1.23 a 3.84	0.93	< 0.01
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	Depression (BDI-II)	17.40 + 9.82	13.33 + 7.74	4.07	8.16	(-0.45 a 8.59)	0.46	ns.
(N = 12)(M±SD)(M±SD)(M±SD)Physical role 50.00 ± 45.23 52.08 ± 40.53 -2.08 22.51 $(-16.38 a 12.22)$ 0.05 $ns.$ Physical function 63.33 ± 25.35 61.67 ± 23.09 1.67 18.50 $(-10.09 a 13.42)$ 0.07 $ns.$ Body pain 55.08 ± 30.13 55.50 ± 22.30 -0.42 18.88 $(-12.41 a 11.58)$ 0.02 $ns.$ General health 44.08 ± 16.82 43.00 ± 22.65 1.08 19.53 $(-11.32 a 13.49)$ 0.05 $ns.$ Vitality 41.75 ± 26.70 42.08 ± 20.94 -0.33 21.67 $(-14.10 a 13.43)$ 0.01 $ns.$ Social function 64.58 ± 23.13 71.88 ± 20.03 -7.29 13.55 $(-15.90 a 1.31)$ 0.34 $ns.$ Mental health 59.75 ± 19.34 59.83 ± 16.15 -0.08 10.13 $(-6.52 a 6.35)$ 0.00 $ns.$ Emotional role 86.08 ± 33.26 61.07 ± 42.26 25.02 53.44 $(8.94 a 58.97)$ 0.66 $ns.$ Pain scale (TWSTRS) 6.42 ± 6.22 8.00 ± 6.81 -1.58 3.00 $(-3.49 a 0.32)$ 0.24 $ns.$ Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 $(-0.87 a 0.70)$ 0.02 $ns.$ Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 $(-2.72 a 1.56)$ 0.08 $ns.$	Anxiety (STAI)	71.13 + 28.14	60.67 + 34.04	10.47	24.86	(-3.30 a 24.24)	0.34	ns.
Physical role 50.00 ± 45.23 52.08 ± 40.53 -2.08 22.51 $(-16.38 \text{ a } 12.22)$ 0.05 $ns.$ Physical function 63.33 ± 25.35 61.67 ± 23.09 1.67 18.50 $(-10.09 \text{ a } 13.42)$ 0.07 $ns.$ Body pain 55.08 ± 30.13 55.50 ± 22.30 -0.42 18.88 $(-12.41 \text{ a } 11.58)$ 0.02 $ns.$ General health 44.08 ± 16.82 43.00 ± 22.65 1.08 19.53 $(-11.32 \text{ a } 13.49)$ 0.05 $ns.$ Vitality 41.75 ± 26.70 42.08 ± 20.94 -0.33 21.67 $(-14.10 \text{ a } 13.43)$ 0.01 $ns.$ Social function 64.58 ± 23.13 71.88 ± 20.03 -7.29 13.55 $(-15.90 \text{ a } 1.31)$ 0.34 $ns.$ Mental health 59.75 ± 19.34 59.83 ± 16.15 -0.08 10.13 $(-6.52 \text{ a } 6.35)$ 0.00 $ns.$ Emotional role 86.08 ± 33.26 61.07 ± 42.26 25.02 53.44 $(8.94 \text{ a } 58.97)$ 0.66 $ns.$ Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 $(-0.87 \text{ a } 0.70)$ 0.02 $ns.$ Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 $(-2.72 \text{ a } 1.56)$ 0.08 $ns.$	Control group	Pre	Post	Differences of means	SD from differ.	95% IC	d	р
$\begin{array}{c c c c c c c c c c c c c c c c c c c $	(<i>N</i> = 12)	(M±SD)	(M±SD)					
Body pain 55.08 ± 30.13 55.50 ± 22.30 -0.42 18.88 $(-12.41 \ a \ 11.58)$ 0.02 $ns.$ General health 44.08 ± 16.82 43.00 ± 22.65 1.08 19.53 $(-11.32 \ a \ 13.49)$ 0.05 $ns.$ Vitality 41.75 ± 26.70 42.08 ± 20.94 -0.33 21.67 $(-14.10 \ a \ 13.43)$ 0.01 $ns.$ Social function 64.58 ± 23.13 71.88 ± 20.03 -7.29 13.55 $(-15.90 \ a \ 1.31)$ 0.34 $ns.$ Mental health 59.75 ± 19.34 59.83 ± 16.15 -0.08 10.13 $(-6.52 \ a \ 6.35)$ 0.00 $ns.$ Emotional role 86.08 ± 33.26 61.07 ± 42.26 25.02 53.44 $(8.94 \ a \ 58.97)$ 0.66 $ns.$ Pain scale (TWSTRS) 6.42 ± 6.22 8.00 ± 6.81 -1.58 3.00 $(-3.49 \ a \ 0.32)$ 0.24 $ns.$ Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 $(-0.87 \ a \ 0.70)$ 0.02 $ns.$ Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 $(-2.72 \ a \ 1.56)$ 0.08 $ns.$	Physical role	50.00 ± 45.23	52.08 ± 40.53	-2.08	22.51	(-16.38 a 12.22)	0.05	ns.
General health 44.08 ± 16.82 43.00 ± 22.65 1.08 19.53 $(-11.32 \text{ a } 13.49)$ 0.05 $ns.$ Vitality 41.75 ± 26.70 42.08 ± 20.94 -0.33 21.67 $(-14.10 \text{ a } 13.43)$ 0.01 $ns.$ Social function 64.58 ± 23.13 71.88 ± 20.03 -7.29 13.55 $(-15.90 \text{ a } 1.31)$ 0.34 $ns.$ Mental health 59.75 ± 19.34 59.83 ± 16.15 -0.08 10.13 $(-6.52 \text{ a } 6.35)$ 0.00 $ns.$ Emotional role 86.08 ± 33.26 61.07 ± 42.26 25.02 53.44 $(8.94 \text{ a } 58.97)$ 0.66 $ns.$ Pain scale (TWSTRS) 6.42 ± 6.22 8.00 ± 6.81 -1.58 3.00 $(-3.49 \text{ a } 0.32)$ 0.24 $ns.$ Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 $(-0.87 \text{ a } 0.70)$ 0.02 $ns.$ Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 $(-2.72 \text{ a } 1.56)$ 0.08 $ns.$	Physical function	63.33 ± 25.35	61.67 ± 23.09	1.67	18.50	(–10.09 a 13.42)	0.07	ns.
Vitality 41.75 ± 26.70 42.08 ± 20.94 -0.33 21.67 $(-14.10 \text{ a } 13.43)$ 0.01 $ns.$ Social function 64.58 ± 23.13 71.88 ± 20.03 -7.29 13.55 $(-15.90 \text{ a } 1.31)$ 0.34 $ns.$ Mental health 59.75 ± 19.34 59.83 ± 16.15 -0.08 10.13 $(-6.52 \text{ a } 6.35)$ 0.00 $ns.$ Emotional role 86.08 ± 33.26 61.07 ± 42.26 25.02 53.44 $(8.94 \text{ a } 58.97)$ 0.66 $ns.$ Pain scale (TWSTRS) 6.42 ± 6.22 8.00 ± 6.81 -1.58 3.00 $(-3.49 \text{ a } 0.32)$ 0.24 $ns.$ Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 $(-0.87 \text{ a } 0.70)$ 0.02 $ns.$ Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 $(-2.72 \text{ a } 1.56)$ 0.08 $ns.$	Body pain	55.08 ± 30.13	55.50 ± 22.30	-0.42	18.88	(-12.41 a 11.58)	0.02	ns.
Social function 64.58 ± 23.13 71.88 ± 20.03 -7.29 13.55 $(-15.90 \text{ a } 1.31)$ 0.34 $ns.$ Mental health 59.75 ± 19.34 59.83 ± 16.15 -0.08 10.13 $(-6.52 \text{ a } 6.35)$ 0.00 $ns.$ Emotional role 86.08 ± 33.26 61.07 ± 42.26 25.02 53.44 $(8.94 \text{ a } 58.97)$ 0.66 $ns.$ Pain scale (TWSTRS) 6.42 ± 6.22 8.00 ± 6.81 -1.58 3.00 $(-3.49 \text{ a } 0.32)$ 0.24 $ns.$ Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 $(-0.87 \text{ a } 0.70)$ 0.02 $ns.$ Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 $(-2.72 \text{ a } 1.56)$ 0.08 $ns.$	General health	44.08 ± 16.82	43.00 ± 22.65	1.08	19.53	(–11.32 a 13.49)	0.05	ns.
Mental health 59.75 ± 19.34 59.83 ± 16.15 -0.08 10.13 $(-6.52 \ a \ 6.35)$ 0.00 $ns.$ Emotional role 86.08 ± 33.26 61.07 ± 42.26 25.02 53.44 $(8.94 \ a \ 58.97)$ 0.66 $ns.$ Pain scale (TWSTRS) 6.42 ± 6.22 8.00 ± 6.81 -1.58 3.00 $(-3.49 \ a \ 0.32)$ 0.24 $ns.$ Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 $(-0.87 \ a \ 0.70)$ 0.02 $ns.$ Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 $(-2.72 \ a \ 1.56)$ 0.08 $ns.$	Vitality	41.75 ± 26.70	42.08 ± 20.94	-0.33	21.67	(–14.10 a 13.43)	0.01	ns.
Emotional role 86.08 ± 33.26 61.07 ± 42.26 25.02 53.44 (8.94 a 58.97) 0.66 ns. Pain scale (TWSTRS) 6.42 ± 6.22 8.00 ± 6.81 -1.58 3.00 (-3.49 a 0.32) 0.24 ns. Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 (-0.87 a 0.70) 0.02 ns. Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 (-2.72 a 1.56) 0.08 ns.	Social function	64.58 ± 23.13	71.88 ± 20.03	-7.29	13.55	(–15.90 a 1.31)	0.34	ns.
Pain scale (TWSTRS) 6.42 ± 6.22 8.00 ± 6.81 -1.58 3.00 (-3.49 a 0.32) 0.24 ns. Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 (-0.87 a 0.70) 0.02 ns. Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 (-2.72 a 1.56) 0.08 ns.	Mental health	59.75 ± 19.34	59.83 ± 16.15	-0.08	10.13	(-6.52 a 6.35)	0.00	ns.
Pain scale (VAS) 3.33 ± 3.75 3.42 ± 3.37 -0.08 1.24 (-0.87 a 0.70) 0.02 ns. Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 (-2.72 a 1.56) 0.08 ns.	Emotional role	86.08 ± 33.26	61.07 ± 42.26	25.02	53.44	(8.94 a 58.97)	0.66	ns.
Depression (BDI-II) 11.75 ± 7.25 12.33 ± 7.35 -0.58 3.37 (-2.72 a 1.56) 0.08 ns.	Pain scale (TWSTRS)	6.42 ± 6.22	8.00 ± 6.81	-1.58	3.00	(-3.49 a 0.32)	0.24	ns.
	Pain scale (VAS)	3.33 ± 3.75	3.42 ± 3.37	-0.08	1.24	(-0.87 a 0.70)	0.02	ns.
	Depression (BDI-II)	11.75 ± 7.25	12.33 ± 7.35	-0.58	3.37	(–2.72 a 1.56)	0.08	ns.
Anxiety (STAI) 60.67 ± 28.02 62.08 ± 28.35 -1.42 21.06 (-14.80 a 11.96) 0.05 ns.								

Table 4. Interaction effects in variables related to QoL, pain, and mood.

	Interaction (treatment \times time)		Simple effects/effect size			
Variable	F	р	Control group	Experimental group		
Corporal and physical perception dimension	F(1, 25) = 5.32	p < 0.01	F(1, 25) = 0.005, ns	F(1, 25) = 12.53, p < 0.01 (d = 0.43)		
Subjective-social-emotional dimension	F(1, 25) = 10.71	p < 0.01	F(1, 25) = 0.61, ns	F(1, 25) = 16.28, p < 0.001 (d = 0.77)		
Visual Analog Scale (VAS)	F(1, 25) = 12.08	p < 0.01	F(1, 25) = 0.022, ns.	F(1, 25) = 25.43, p < 0.001 (d = 0.93)		
Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS)	F(1, 25) = 13.59	p < 0.01	F(1, 25) = 4.14, p = 0.053 (d = 0.24)	F(1, 25) = 10.60, p < 0.01 (d = 0.37)		
Beck Depression Inventory (BDI-II)	F(1, 25) = 3.41	<i>p</i> = 0.078	F(1, 25) = 0.10, ns.	F(1, 25) = 5.86, p < 0.05 (d = 0.47)		
State-Trait Anxiety Inventory (STAI)	F(1, 25) = 1.74	ns.	ns.	ns.		

show a significant interaction of treatment by time (Figure 2f). No effect was observed in the control group.

Finally, with the aim of controlling for possible interaction effects between the botulinum toxin and our program, we performed the statistical analysis again. Thus, we removed the four participants treated with botulinum toxin in the experimental group from the analysis. These new results showed no significant differences from the previous results, with the exception that in this new analysis, the BDI-III depression inventory exhibited interaction effects and significant differences with regard to the experimental condition.

Discussion

The purpose of this study was to show that a clinical intervention based on a mind-body approach alone could improve different aspects of health in people with CD. In this exploratory study, we demonstrated that WT and AT significantly improve QoL, pain, and symptoms of depression.

Traditionally, rehabilitation of dystonia has been focused on two major physical rehabilitation therapeutic perspectives: mechanism and functionalism (Martínez-Pernía, González-Castán, and Huepe, 2017). Examples of these therapies include: sensorial training (Schramm, Reiners, and Naumann, 2004); electromyography biofeedback training (Jahanshahi, Sartory, and Marsden, 1991); visual and auditory feedback (Brudny, Grynbaum, and Korein, 1974); hydrotherapy (Useros-Olmo and Collado Vazquez, 2010); and multimodal physical programs (El-Bahrawy, El-Tamawy, Shalaby, and Abdel-Alim, 2009; Queiroz, Chien, Sekeff-Sallem, and Barbosa, 2012; Tassorelli et al, 2006). In addition, only a few studies in CD were based on mind-body therapies but were always combined with conventional physiotherapy

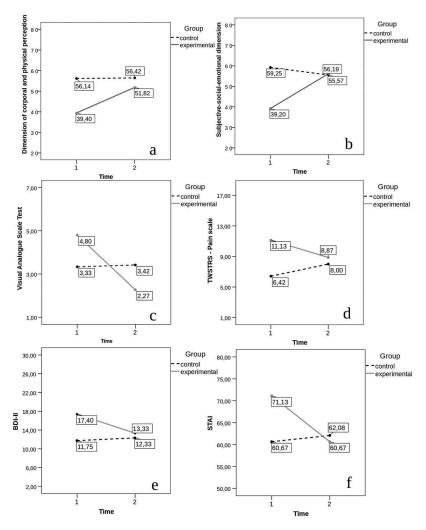


Figure 2. Graphics of all of the study variables. (a) Corporal and physical perception dimension, SF-36; (b) subjective-socialemotional dimension, SF-36; (c) VAS; (d) TWSTRS; (e) BDI-II; and (f) STAI.

(Andrews and Gill, 1982; Boyce et al, 2013; Duddy and McLellan, 1995; Gildenberg, 1981; Jahanshahi, Sartory, and Marsden, 1991; Zetterberg et al, 2008). Unlike these previous studies, this research is the first that shows that a therapy fully focused on a relaxation program can improve physical and mental symptomatology in patients with CD.

One of the key goals of rehabilitating patients with CD is improving their pain because these patients suffer from more pain than those with other types of focal dystonia (Pekmezovic et al, 2009; Soeder et al, 2009). The outcomes of our study demonstrated that an intervention based on a relaxation program improved pain similarly to other studies that reported pain score on the VAS (Tassorelli et al, 2006; Zetterberg et al, 2008) and the pain subscale of the TWSTRS (Queiroz, Chien, Sekeff-Sallem, and Barbosa, 2012).

Our results showed that the experimental group improved QoL as measured by the SF-36. This finding is consistent with other studies that implemented the same questionnaire among patients with CD (Jen et al, 2014; Queiroz, Chien, Sekeff-Sallem, and Barbosa, 2012) as well as those that applied different questionnaires to assess QoL (e.g., the Craniocervical Dystonia Questionnaire (CDQ-24)) (Zetterberg et al, 2008). Finally, participants with depression partially improved their symptomatology in our study, although other studies found clearer improvements (Jahanshahi, Sartory, and Marsden, 1991) or no significant changes in participants (Boyce et al, 2013).

We are aware that this research has three limitations: (1) the sample size was small; (2) a passive control group was used for comparison with the experimental group; and (3) no follow-up period was included. Nevertheless, it is important to highlight that we developed a pilot study to assess whether a therapy completely based on a relaxation program could improve the physical and mental symptomatologies of patients with CD. The findings show that this aim was achieved. In addition, the experimental design was sensitive enough to observe significant differences in the sample because the effect sizes of certain variables were medium and high.

The injection of botulinum toxin in patients with chronic CD has been shown to provide effective symptomatic relief (Ramirez-Castaneda and Jankovic, 2013). In addition, some studies of physical therapy have shown that clinical interventions enhance the effects of these injections, reducing pain, and disability (Queiroz, Chien, Sekeff-Sallem, and Barbosa, 2012; Tassorelli et al, 2006). Nevertheless, based on our data, we cannot claim there was an interaction effect between the botulinum toxin treatment and the relaxation program in those participants of the experimental group who were injected with the drug (N = 4). To support this evidence, further studies with larger samples are necessary.

Similar to this relaxation program (WT and AT) study, physical therapy disciplines have implemented other mind-body approaches for motor rehabilitation such as: yoga (Saper et al, 2014); Tai Chi (Tousignant et al, 2013); Qigong (Liu et al, 2012); experiential neurorehabilitation (Martínez-Pernía and Ceric, 2011; Martínez-Pernía et al, 2016); Feldenkrais Method (Hillier and Worley, 2015); and the Alexander Technique (Cacciatore et al, 2011). Although all of these approaches have important methodological differences from one another, they all seek to enhance body awareness in clinical intervention (Mehling, DiBlasi, and Hecht, 2015). This goal has been described as a key mechanism for providing health benefits (Hölzel et al, 2011; Mehling et al, 2009). Body awareness has been defined as "the subjective, phenomenological aspect of proprioception and interoception that enters conscious awareness" (Mehling et al, 2011) or the ability to recognize subtle body cues (Baas et al, 2004). Given these assumptions and bearing in mind that the patients in this study were instructed to focus on their internal bodily sensations, we presume that the experimental group increased their body awareness, which might be the key mechanism for improving health. In accordance with the work developed by Payne, Levine, and Crane-Godreau (2015), the therapeutic effect of this intervention is the activation of the core response network, which comprises the: autonomic nervous system; limbic system (Heimer and Van Hoesen, 2006); emotional motor system (Holstege, Bandlerz, and Saper, 1996); and the reticular arousal system (Krout, Belzer, and Loewy, 2002; Strominger, Demarest, and Laemle, 2012). The activation of these regions through the perception of internal bodily sensations allows muscle tone changes,

emotional state modulation and cognitive activity modification (Norman, Berntson, and Cacioppo, 2014).

A significant number of studies on CD chose to rehabilitate patients with physical symptoms through the implementation of an extensive multimodal physical program that included stretching, strengthening, massage, isometric and dynamic exercises, balance training, and body perception (El-Bahrawy, El-Tamawy, Shalaby, and Abdel-Alim, 2009; Queiroz, Chien, Sekeff-Sallem, and Barbosa, 2012; Tassorelli et al, 2006; Zetterberg et al, 2008). Unlike this therapeutic view focused on physical symptoms, we assumed that CD is a multidimensional disorder affected by physical and mental stressors that worsen the symptoms of CD (Ben-Shlomo, Camfield, and Warner, ; Jahanshahi, 2000; Soeder et al, 2009; Zetterberg, Lindmark, Söderlund, and Åsenlöf, 2012). Based on this assumption, this study showed a high improvement among patients with CD using an intervention of whole-body relaxation alone and not an extensive physical intervention.

Following the idea of the previous paragraph, we highlight that CD is a physical disease affected by multidimensional factors (i.e., physical, emotional, cognitive, social, lifestyle, mood, and self-awareness) that directly or indirectly influence the health of patients. Thus, future researchers could address CD recovery by addressing more than physical factors. This approach would allow the physical therapy field to surpass the physical component of the disease, which is the most common treatment in the discipline (Bleton, 2010), and address the disease in a more holistic way.

Conclusion

This exploratory study shows that a therapy based on whole-body relaxation alone benefits patients with CD. In contrast with most physical therapy interventions that focus on treating motor component, we created an intervention that addresses physical and mental stressors. This view enabled a disease-management strategy that applied a more holistic perspective to work beyond the dystonic focus.

Declaration of Interest

The authors report no conflicts of interest.

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