Article



The impact of mirror therapy on body schema perception in patients with complex regional pain syndrome after distal radius fractures

British Journal of Pain 2019, Vol 13(1) 35–42 © The British Pain Society 2018 Article reuse guidelines: sagepub.com/journals-permissions DOI: 10.1177/2049463718782544 journals.sagepub.com/home/bjp



Viktor Kotiuk², Olexander Burianov¹, Olexander Kostrub², Ludmila Khimion³ and Ivan Zasadnyuk²

Abstract

Introduction: Mirror therapy requires a minimum of equipment, is relatively simple to perform and effective for various pathological conditions. The effect of mirror therapy on body schema disturbances registered in complex regional pain syndrome type I (CRPS I) patients has not yet been determined.

Methods: The study is based on the analysis of the treatment results of 30 patients with CRPS I, developed as a result of the distal radius fractures, with help of mirror therapy together with exercise therapy and medications. The control group consisted of 20 patients with CRPS I developed as a result of the distal radius fractures treated only with exercise therapy and medications. We evaluated the results before the treatment, after 3 days and after 6 weeks of treatment according to The Bath CRPS Body Perception Disturbance Scale.

Results: A total of 83.33% patients experienced the positive effect of mirror therapy on the perception of the 'body schema' after 6 weeks of treatment, and 35% underwent standard treatment without mirror therapy. The positive effect was statistically significant for the first five points of the Bath scale after 6 weeks of treatment compared to the control group. Improvements observed in some patients after 3 days of treatment were less pronounced and statistically insignificant.

Conclusion: Mirror therapy can improve the perception of the body schema as an element of integrated treatment of CRPS I developed after fractures of the distal radius less than 3 years duration.

Keywords

Musculoskeletal pain, pain perception, complex regional pain syndromes, somatoform disorders, somatosensory disorders, reflex sympathetic dystrophy, body perception

Introduction

Mirror therapy is a kind of biofeedback therapy. It requires minimum equipment and is relatively simple to practise. The effectiveness of mirror therapy for treatment of various pathological conditions has been demonstrated in several studies.^{1,2} Among published studies, only few involved patients with complex regional pain syndrome type I (CRPS I). They enrolled small and heterogeneous patients groups or consisted

¹Bogomolets National Medical University, Kyiv, Ukraine ²SI 'Institute of Traumatology and Orthopedics of the Academy of Medical Sciences of Ukraine', Kyiv, Ukraine ³Department of Family Medicine, Shupyk National Medical Academy of Postgraduate Education, Kyiv, Ukraine

Corresponding author:

Viktor Kotiuk, 13, Salutna str, ap. 57, Kyiv 04111, Ukraine. Email: kotyuk_v@ukr.net

of case reports; researches included early-disease stages and cases of CRPS I caused by stroke.³⁻⁶ In post-stroke CRPS I patients, Pervane et al.⁵ and Cacchio et al.³ demonstrated the efficacy of the mirror therapy for pain decrease and function improvement. We have found no studies about efficacy of the mirror therapy in typical posttraumatic CRPS I with a sufficient number of patients and uniform trigger. Moreover, there were very few original studies on mirror therapy in CRPS I patients the last years. Most articles on this subject during the last several years were reviews. Studies suggested the relationship between pain, body perception disturbances and remapping of the brain. Treatment modalities that target cortical areas may reduce body perception disturbance and pain.7 According to the one of many theories, CRPS I pain may be caused by mismatch between muscles movements and proprioceptive feedback.

The body schema is a central representation of the body's parts that includes the length of limb segments, their hierarchical arrangement, the configuration of the segments in space and the shape of the body surface.8 There are studies that confirm the reduction of the central representation area of the injured extremity in the contralateral hemisphere's primary and secondary somatosensory cortex in CRPS I patients9-12 and studies pointing to the responsibility of these zones for the so-called 'body schema perception'.8,13 Therefore, the body schema disturbances in CRPS I patients must be expected. These disorders are typical for CRPS I but often overlooked by doctors. Patients also often do not complain because they cannot explain them or are reluctant to discuss the problem in fear of misunderstanding and suspicion of mental illness.¹⁴ Selfperception of the hand is impaired in many patients with CRPS. It is thought to be the consequence of the higher central nervous system processing alteration.¹⁵

The primary motor cortex of the brain may not differentiate between the actual and reflected in the mirror limbs. Mirror therapy 'fools' the brain, makes it to take the affected limb without pain and limits of movements. In addition, the mirror therapy may replace inadequate proprioceptive feedback with a visual feedback signal. This activates cortical areas, leading to cortical reorganization.¹⁶

In spite of many theories and theoretical explanations, the effect of mirror therapy on the body schema perception, which is often seen in CRPS I patients, has not yet been determined.

The aim of the study

To evaluate the effectiveness of mirror therapy in treatment of the 'body schema' perception in CRPS I patients.

Materials and methods

The study is based on the analysis of the results of treatment of 30 patients with CRPS I (developed as a result of the distal radius fractures, up to 3 years long), by use of therapeutic complex consisted of mirror therapy, exercises and medications (gabapentin 300 mg three times a day, dexketoprofen on demand, combined vasoactive drug (capillary stabilizing and venotonic) per os and compresses with dimethyl sulfoxide and dexamethasone) - treatment group. The control group consisted of 20 patients with CRPS I (same etiology and duration with the treatment group) who were treated with combination of exercises and medications (gabapentin 300 mg three times a day, dexketoprofen on demand, combined vasoactive drug per o and compresses with dimethyl sulfoxide and dexamethasone). It was technically impossible to monitor the study with placebo because it was impossible to replace the mirror with something else to hoodwink the patient. Cacchio et al.³ tried to replace placebo by imaginary physiotherapy (patients performed imaginary limb movements) and mirror therapy with a covered mirror. However, the first method is not a placebo, but is another therapeutic technique. A mirror therapy with a covered mirror also does not meet the requirements for placebo, because it cannot make the patient to believe in it. Another limitation of the study was the unblinded assessor. The investigator was not blinded because of the internal regulations of the Research Institute which is also a hospital. The internal regulations oblige doctors to document every kind of treatment in medical records. The assessor was the employee of the same institution. So, he could see the medical records of the patients. The study continued for a rather long time and all the CRPS patients were assessed one by one for a long period of time. The study had not been sponsored financially, so we could not invite the assessor from the other institution.

We used a randomized study design. Patients were randomly assigned to the mirror therapy group or the control group using computer-generated random numbers. According to Helsinki Declaration, participants signed written informed consent forms. There were more patients in each group. But several patients were excluded because of their absence on checkups usually because of their remote residence.

Patients from both groups received the complex treatment for 6 weeks. The results were analysed at the third day and just after the sixth week visit. The mirror therapy technique was explained and demonstrated to the patient by trained specialist and was controlled and corrected as needed on further visits (the third day and the sixth week). Changes in the intervals between visit time of the second visit for 1 day (i.e. the second day or the fourth day) and the third visit up to 1 week was not encouraged, however allowed and was not exclusion criterion. The results in such cases were calculated for the third day and 6 weeks of treatment. The mirror therapy was recommended for 10 minutes two times a day, but a one-time administration was allowed, as well as omission for up to seven inconsequent days of mirror therapy during 6 weeks, except for the first 3 days.

The mirror therapy was performed for the injured upper limb only (forearm and hand). Patients were asked to perform simultaneous movements by both hands (injured and healthy). These movements included finger flexion and extension (all fingers simultaneously and in sequence), abduction and adduction, wrist flexion, extension, abduction, adduction and forearm rotation. The speed of movements was selfselected. We used a commercially available folding black square box $36 \times 36 \times 47$ cm, in which one of the external sides was covered by a mirror. The patient placed the affected hand inside the mirror box and could move the injured hand freely but without visual control. The healthy hand was placed outside of the box and the patient could visually control the movements looking in the mirror. The patient was asked to perform the same movements by both hands, to concentrate on these movements and to accept the reflection of the healthy hand as their injured hand. Each mirror therapy session lasted for 10 min because most patients could not keep concentration for longer time. After training with physician, the folding mirror box was provided for the patients to take home.

The exclusion criteria were the duration of the disease for more than 3 years (calculated from the moment of injury), the potential causes/triggers of CRPS I, which could require surgical treatment, lack of adequate co-operation with the patient, lack of satisfactory effect from the prescribed treatment regimen (gabapentin, dexketoprofen on demand occasionally, combined capillary stabilizing and venotonic agent per os and compresses with dimethyl sulfoxide and dexamethasone) requiring the use of other treatment methods, and thus violating homogeneity of the sample. The duration of the disease for more than 3 years was an exclusion criterion, both for the purpose of increasing homogeneity of the sample and because of the potentially less effect of mirror therapy due to the usually significant dystrophic changes in the affected limb which make restoration of the sensory-motor pathways less likely. Lewis and McCabe¹⁴ reported a decrease in the mirror therapy effectiveness with an increase in the duration of the disease.

We evaluated the results before the treatment, after 3 days and after 6 weeks of treatment according to The Bath CRPS Body Perception Disturbance Scale.¹⁴ Probably, some patients could expect some effect from the first procedure (and in some cases it was). However, most patients need several days to master the method to a sufficient degree and to learn to concentrate on the exercise. The assessments were performed by the same investigator, who was not blinded to group allocation.

Results

In our study, the results of treatment of CRPS I patients with and without mirror therapy are summarized in Table 1. Out of 30 (83.33%) patients in treatment group, 25 patients demonstrated significant improvement in 'body schema' perception after 6 weeks of treatment; the results in control group were significantly weaker (only 35% had evident improvement). It is important to point that worsening in body schema perception after 6 weeks of treatment was observed in only 1 patient (3.33%) at the treatment group and in 11 of 20 patients (55%) at the control group. We have not analysed the changes in the body schema perception at the first day after the first mirror therapy session. However, several patients noted a decrease in pain and improvement in various aspects of pain immediately after the first mirror therapy session or even during practice. We have doubts about such an immediate effect of this type of treatment on the body schema perception. However, we cannot exclude it. Furthermore, during the treatment course, the improvement was found in 19 of 30 patients (63.33%) after 3 days and in 25 out of 30 (83.33%) after 6 weeks of mirror therapy. The improvement was on average by 1.6 points of Bath scale (11.88%, p = 0.508) and 6.64 points (49.29%, p = 0.003), respectively. Thus, after 6 weeks, the improvement was statistically significant. Some positive changes were also observed in patients treated in control group -8 out of 20 (40%) in 3 days and in 7 out of 20 (35%) in 6 weeks; however, these changes were less pronounced as it is shown in Table 1.

We also assessed the effect of mirror therapy on specific symptoms of body schema perception disorder. The difference was statistically significant for the first five points of the Bath scale after 6 weeks of treatment in treatment group comparing to the control group (p (t - c) < 0.05; p (t - c) is the statistical significance of difference between levels of variables in the treatment and control groups (the Mann-Whitney U test). After 6 weeks of mirror therapy, the feeling of dissociation from the limb improved by 58.57% (p (t - c) = 0.044), the awareness of the position of the affected limb in space has improved by 52.68% (p (t - c) = 0.0006), the attention deficit to the affected limb decreased by 47.37% (p (t - c) = 0.0005), the emotional feelings about the affected limb shifted to positive from negative by 47.18% (p (t - c) = 0.047) and the difference in perception of how the affected limb looks or how it

Table 1. The results of CR	PS I patients' t	reatment with and	without mirror the	rapy.					
Symptoms of body	Groups	Statistical	Treatment period			Difference			
perception distantiante			Baseline data	The third day	Sixth week	Baseline –	third day	Baseline –	sixth week
						Absolute value	%	Absolute value	%
Intensity of feeling of	Treatment	M (SD)	2.1 (1.56)	1.97 [1.63]	0.87 (1.17)	-0.13	-6.19%	-1.23	-58.57%
the detachment of the	group	Me [25%–75%]	2 [1–3]	2 [1–3]	0 (0–2)				
affected limb	Control	M (SD)	1.8 (1.58)	1.6 [1.67]	1.9 (1.83)	-0.20	-11.11%	0.10	5.56%
	group	Me (25%–75%)	2 (0–3)	1 (0–2.5)	2 (0–3.5)				
p [t – c]			ρ = 0.512	p = 0.348	ρ = 0.044				
Awareness about the	Treatment	M (SD)	3.17 (2.39)	2.6 (2.43)	1.5 (1.78)	-0.57	-17.98%	-1.67	-52.68%
physical position of the	group	Me (25%–75%)	3 [1–5]	2 (0–5)	1 (0–2)				
limb	Control	M (SD)	2.95 (2.16)	2.75 (2.07)	3.25 [1.68]	-0.20	-6.78%	0.30	10.17%
	group	Me [25%–75%]	3 (1.5–5)	2.5 [1.5-4.5]	3 [2–4]				
p [t – c]			ρ = 0.771	ρ = 0.717	ρ = 0.0006				
Attention paid to the	Treatment	M (SD)	1.33 (1.24)	0.8 (1.19)	0.7 [1.64]	-0.53	-39.85%	-0.63	-47.37%
affected limb in terms of	group	Me (25%–75%)	1 [0–2]	0 (0–1)	0 (0–1)				
looking at it and thinking	Control	M (SD)	0.9 (1.07)	0.85 (0.93)	2.2 (1.85)	-0.05	-5.56%	1.30	144.44%
about it	group	Me (25%–75%)	0.5 (0–2)	1 (0–1.5)	2 (0.5–4)				
p [t-c]			ρ = 0,207	ρ = 0,560	ρ = 0,0005				
Intensity of the negative	treatment	M (SD)	3.73 (2.99)	3.3 (2.73)	1.97 [2.4]	-0.43	-11.53%	-1.76	-47.18%
emotional feelings about	group	Me (25%–75%)	4 [0–5]	3 (0–5)	1 [0–4]				
the affected limb	control	M (SD)	3.8 (2.65)	3.75 (2.61)	3.35 (2.39)	-0.05	-1.32%	-0.45	-11.84%
	group	Me (25%–75%)	4.5 [1–6]	4.5 [1–6]	4 [1–5.5]				
p [t – c]			ρ = 0.763	ρ = 0.449	ρ = 0.047				
The difference between	Treatment	M (SD)	2.47 [1.53]	2.47 [1.53]	1.43 [1.25]	0.00	%00.0	-1.04	-42.11%
how the affected limb	group	Me (25%–75%)	3 [1–4]	3 [1–4]	1 (0–2)				
looks or is on touch	Control	M (SD)	2.2 (1.51)	2.3 (1.53)	2.3 (1.26)	0.10	4.55%	0.10	4.55%
compared to how it feels	group	Me [25%–75%]	2.5 (1–3.5)	2.5 [1-4]	2 (1–3.5)				

$ \begin{array}{c cccc} Symptoms of body \\ \mbox{perception disturbance} $										
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $	Symptoms of body	Groups	Statistical	Treatment period			Difference			
$ \begin{array}{c c c c c c c c c c c c c c c c c c c $				Baseline data	The third day	Sixth week	Baseline –	third day	Baseline –	sixth week
$ \begin{array}{ccccc} p \ t - c \ \\ \mbox{Treatment} & \ M(SD) & 0.13 (0.35) & 0.17 (0.38) & 0.04 & 30.77\% & 0.04 \\ \mbox{the fersive to amputate} & \ Treatment & \ M(SD) & 0.13 (0.21) & 0.05 (0.22) & 0.06 & 0.00 \\ \mbox{group} & \ Me (25\%-75\%) & 0.06 (0.22) & 0.05 (0.22) & 0.00 & 0.00\% & 0.00 \\ \mbox{group} & \ Me (25\%-75\%) & 0.0-0 & 0 & 0-0 & 0 & 0-0 & 0 \\ \mbox{group} & \ Me (25\%-75\%) & 0.0-0 & 0 & 0-0 & 0 & 0-0 & 0 & 0-0 & 0 \\ \mbox{group} & \ Me (25\%-75\%) & 0.0-0 & 0 & 0-0 & 0 & 0-0 & 0 & 0 & 0 & 0 $							Absolute value	%	Absolute value	%
$ \begin{array}{cccc} \mbox{The desire to amputate} & \mbox{Treatment} & \mbox{M}(SD) & 0.13 (0.35) & 0.17 (0.38) & 0.04 & 30.77\% & 0.04 \\ \mbox{the limb} & \mbox{group} & \mbox{M}(SD) & 0.05 (0.22) & 0.06 (0.22) & 0.06 (0.22) & 0.00 & 0.00\% & 0.00 \\ \mbox{group} & \mbox{M}(SD) & \mbox{M}(SD) & 0.05 (0.22) & 0.06 (0.22) & 0.00 & 0.00\% & 0.00 \\ \mbox{group} & \mbox{M}(SD) & \mbox{M}(SD) & 0.15 (0.67) & 0.13 (0.51) & 0.13 (0.51) & 0.07 & -17.50\% & -0.27 \\ \mbox{amputate the limb} & \mbox{group} & \mbox{M}(SD) & 0.15 (0.67) & 0.15 (0.67) & 0.13 (0.51) & 0.00 & 0.00\% & 0.00 \\ \mbox{group} & \mbox{M}(SD) & \mbox{M}(SD) & 0.15 (0.67) & 0.15 (0.67) & 0.13 (0.51) & 0.00 & 0.00\% & 0.00 \\ \mbox{group} & \mbox{M}(SD) & \mbox{M}(SD) & \mbox{0} & 0.00 & 0.00\% & 0.00 & 0.00\% & 0.00 \\ \mbox{group} & \mbox{M}(SD) & \mbox{M}(SD) & \mbox{0} & 0.00 & 0.00\% & 0.00 & 0.00\% & 0.00 & 0.00\% & 0.00 & 0.00\% & 0.00 & 0.00\% & 0.00 & 0.00\% & 0.00 & 0.00\% & 0.00\% & 0.00 & 0.00\% & 0.00$	p (t – c)			ρ = 0.475	ρ = 0.653	ρ = 0.021				
$ \begin{array}{cccc} \mbox{the limb} & group & \mbox{M} (25\%-75\%) & 0 (0-0) & 0 (0-0) & 0 (0-0) \\ \mbox{Ccontrol} & \mbox{M} (SD) & 0.05 (0.22) & 0.05 (0.22) & 0.00 & 0.00\% & 0.00 \\ \mbox{group} & \mbox{M} (SD) & 0 (0-0) & 0 (0-0) & 0 (0-0) & 0 (0-0) \\ \mbox{group} & \mbox{M} (SD) & 0.415 (1.52) & 0.33 (1.27) & 0.13 (0.51) & -0.07 & -17.50\% & -0.27 \\ \mbox{m} putate the limb & \mbox{group} & \mbox{M} (SD) & 0 (0-0) & 0 (0-0) & 0 (0-0) & 0 (0-0) & 0 (0-0) \\ \mbox{group} & \mbox{M} (SD) & 0.15 (0.67) & 0.15 (0.67) & 0.13 (0.51) & -0.07 & -17.50\% & -0.27 \\ \mbox{group} & \mbox{M} (SD) & 0 (0-0) & 0 (0-$	The desire to amputate	Treatment	M (SD)	0.13 (0.35)	0.17 (0.38)	0.17 (0.38)	0.04	30.77%	0.04	30.77%
$ \begin{array}{cccc} \mbox{Ccontrol} & \mbox{M}(SD) & 0.05 (0.22) & 0.05 (0.22) & 0.05 (0.22) & 0.00 & 0.00\% & 0.00 \\ \mbox{proup} & \mbox{M} & (25\%-75\%) & 0 (0-0) & 0 (0-0) & 0 (0-0) & 0 (0-0) \\ \mbox{proup} & \mbox{M} & (SD) & 0.4 (1.52) & 0.31 (1.27) & 0.13 (0.51) & -0.07 & -17.50\% & -0.27 \\ \mbox{amputate the limb} & \mbox{group} & \mbox{M} & (SD) & 0 (0-0) & 0 (0-0) & 0 (0-0) & 0 (0-0) \\ \mbox{group} & \mbox{M} & (SD) & 0.4 (1.52) & 0.3 (1.27) & 0.13 (0.51) & -0.07 & -17.50\% & -0.27 \\ \mbox{amputate the limb} & \mbox{group} & \mbox{M} & (SD) & 0 (0-0) & 0 (0-0) & 0 (0-0) & 0 (0-0) \\ \mbox{group} & \mbox{M} & (SD) & 0 (0-0) & 0 (0-0) & 0 (0-0) & 0 (0-0) \\ \mbox{group} & \mbox{M} & (SD) & 0 (0-0) & 0 (0-0) & 0 (0-0) & 0 (0-0) \\ \mbox{affected and unaffected} & \mbox{control} & \mbox{M} & \mbox{M} & \mbox{affected and unaffected} & \mbox{amplex} & \mbox{m} & \mbox{affected and unaffected} & \mbox{amplex} & \mbox{affected and unaffected} & \mbox{affected} & $	the limb	group	Me (25%–75%)	0-0) 0	0-0) 0	0-0] 0				
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$		Ccontrol	M (SD)	0.05 (0.22)	0.05 (0.22)	0.05 (0.22)	0.00	0.00%	0.00	0.00%
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$		group	Me [25%–75%]	0-0] 0	0-0) 0	0-0) 0				
$ \begin{array}{c ccccccccccccccccccccccccccccccccccc$	p (t – c)			p = 0.341	ρ = 0.218	ρ = 0.218				
$ \begin{array}{c ccccccccccccccccccccccccccccccccccc$	The intensity of desire to	Treatment	M (SD)	0.4 [1.52]	0.33 (1.27)	0.13 (0.51)	-0.07	-17.50%	-0.27	-67.50%
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$	amputate the limb	group	Me [25%–75%]	0-0] 0	0-0) 0	0-0] 0				
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$		Control	M (SD)	0.15 (0.67)	0.15 (0.67)	0.15 (0.67)	0.00	%00.0	0.00	0.00%
$ \begin{array}{cccccccccccccccccccccccccccccccccccc$		group	Me (25%–75%)	0-0) 0	0-0) 0	0-0) 0				
$ \begin{array}{llllllllllllllllllllllllllllllllllll$	p [t-c]			ρ = 0.773	ρ = 0.773	ρ = 0.847				
of mental image of the group Me $(25\%-75\%)$ 0 $(0-0)$ 0 $(0-0)$ 0 $(0-0)$ 0 $(0-0)$ affected and unaffected Control M (SD) 0.4 (0.68) 0.35 (0.67) 0.2 (0.41) -0.05 -12.50% -0.20 body parts group Me $(25\%-75\%)$ 0 $(0-1)$ 0 $(0-0.5)$ 0 $(0-0)$ p $(1-c)$ Treatment M (SD) 13.47 (9.26) 11.87 (9.3) 6.83 (7.11) -1.60 -11.88% -6.64 group Me $(25\%-75\%)$ 13.5 $(5-20)$ 9.5 $(4-18)$ 4.5 $(2-9)$ -1.60 -4.08% 1.20 control M (SD) 12.5 (8.56) 11.75 (8.23) 13.45 (7.8) -0.50 -4.08% 1.20 group Me $(25\%-75\%)$ 12.5 $(4.5-18.5)$ 11.5 $(4.5-18)$ 12.5 $(8-17.5)$ $p^2 = 0.851$	The severity of distortion	Treatment	M (SD)	0.3 (0.6)	0.27 (0.58)	0.1 (0.31)	-0.03	-10.00%	-0.20	-66.67%
affected and unaffectedControlM (SD)0.4 (0.68)0.35 (0.67)0.2 (0.41)-0.05-12.50%-0.20body partsgroupMe (25%-75%)0 (0-1)0 (0-0.5)0 (0-0)-12.50%-0.20 $p (1 - c)$ TreatmentM (SD) $p = 0.588$ $p = 0.661$ $p = 0.323$ -11.88%-6.64 $p (1 - c)$ TreatmentM (SD)13.47 (9.26)11.87 (9.3)6.83 (7.11)-11.60-11.88%-6.64 $p (1 - c)$ TreatmentM (SD)13.5 (5-20)9.5 (4-18)4.5 (2-9) $p^2 = 0.508$ $p (1 - c)$ groupMe (25%-75%)12.25 (8.56)11.75 (8.23)13.45 (7.8)-0.50 $p (1 - c)$ TotalTreatmentM (SD) $12.25 (8.56)$ $11.75 (8.23)$ $13.45 (7.8)$ -0.50 -4.08% $p (1 - c)$ Total $p (1 - c)$ $p (1 $	of mental image of the	group	Me (25%–75%)	0-0) 0	0-0) 0	0-0) 0				
groupMe $(25\%-75\%)$ 0 $(0-1)$ 0 $(0-0.5)$ 0 $(0-0)$ $p(t-c)$ TreatmentM (SD) $p=0.588$ $p=0.661$ $p=0.323$ TotalTreatmentM (SD) 13.47 (9.26) 11.87 (9.3) $6.83 (7.11)$ -1.60 TotalTotal13.65 (9.26) 11.87 (9.3) $6.83 (7.11)$ -1.60 -11.88% CoupMe $(25\%-75\%)$ 13.5 $(5-20)$ $9.5 (4-18)$ $4.5 (2-9)$ $p^2 = 0.508$ ControlM (SD) 12.25 (8.56) $11.75 (8.23)$ $13.45 (7.8)$ -0.50 groupMe $(25\%-75\%)$ $12.5 (4.5-18.5)$ $11.5 (4.5-18)$ $12.5 (8-17.5)$ $p^2 = 0.851$	affected and unaffected	Control	M (SD)	0.4 (0.68)	0.35 (0.67)	0.2 (0.41)	-0.05	-12.50%	-0.20	-50.00%
$ \begin{array}{llllllllllllllllllllllllllllllllllll$	body parts	group	Me (25%–75%)	0 (0–1)	0 (0–0.5)	0-0] 0				
Total Treatment M (SD) 13.47 (9.26) 11.87 (9.3) 6.83 (7.11) -1.60 -11.88% -6.64 group Me (25%-75%) 13.5 (5-20) 9.5 (4-18) 4.5 (2-9) p² = 0.508 Control M (SD) 12.25 (8.56) 11.75 (8.23) 13.45 (7.8) -0.50 -4.08% 1.20 group Me (25%-75%) 12.5 (4.5-18.5) 11.5 (4.5-18) 12.5 (8-17.5) p² = 0.851	p [t – c]			ρ = 0.588	p = 0.661	ρ = 0.323				
group Me (25%–75%) 13.5 (5–20) 9.5 (4–18) 4.5 (2–9) p² = 0.508 Control M (SD) 12.25 (8.56) 11.75 (8.23) 13.45 (7.8) –0.50 –4.08% 1.20 group Me (25%–75%) 12.5 (4.5–18.5) 11.5 (4.5–18) 12.5 (8–17.5) p² = 0.851	Total	Treatment	M (SD)	13.47 (9.26)	11.87 (9.3)	6.83 (7.11)	-1.60	-11.88%	-6.64	-49.29%
Control M (SD) 12.25 (8.56) 11.75 (8.23) 13.45 (7.8) –0.50 –4.08% 1.20 group Me (25%–75%) 12.5 (4.5–18.5) 11.5 (4.5–18) 12.5 (8–17.5) p² = 0.851		group	Me (25%–75%)	13.5 (5–20)	9.5 (4–18)	4.5 [2–9]		$p^2 = 0.508$		$p^2 = 0.003$
group Me (25%–75%) 12.5 (4.5–18.5) 11.5 (4.5–18) 12.5 (8–17.5) p² = 0.851		Control	M (SD)	12.25 (8.56)	11.75 (8.23)	13.45 [7.8]	-0.50	-4.08%	1.20	9.80%
		group	Me (25%–75%)	12.5 (4.5–18.5)	11.5 (4.5–18)	12.5 [8–17.5]		$p^2 = 0.851$		p ² = 0.646
p (t - c) $\rho = 0.751$ p = 0.796 p = 0.0014 p ¹ = 0.080	p [t – c]			ρ = 0.751	ρ = 0.796	ρ = 0.0014		p ¹ = 0.080		p ¹ = 0.0001

control groups [the Mann–Whitney U test]; p1: statistical significance of difference between the treatment and control groups in trend data over the corresponding periods [the Mann– Whitney U test]; p2: statistical significance of difference in obtained data over time for the each group of patients [Wilcoxon test].

is on touch comparing to how it feels (point 5 of the Bath scale) decreased by 42.11% ((t - c) = 0.021). These improvements were observed in some patients already after 3 days of treatment. However, they were less pronounced and statistically insignificant. The least often CRPS I patients developed symptoms of the body schema disorder in a form of desire to amputate the affected limb (8% in both groups together) and in a form of incorrect graphic representation of the injured limb (26% in both groups together). Mirror therapy had shown a positive influence on the desire for amputation of the affected limb only in one from four those patients (25%), and others had not showed the improvement in that symptom.

Discussion

The experiment on healthy volunteers, which was about the development of incongruent proprioceptive input through active and passive movements in the radial-wrist joints, and vibrations of the tendons, conducted by Moseley et al.,17 demonstrated the possibility to induce other illusory sensations, in particular oedema or foreignness of the limb and nausea. This may explain the fact that CRPS I patients often feel their affected limb alien or more swollen than it actually is. This happens due to disruption of the internal proprioceptive representation of the injured limb in the cerebral cortex.17 The scientists considered that the mentioned mechanism might be insufficient for pain generation. But on our opinion, the results of the study that Moseley et al.¹⁷ do not deny completely indicated mechanism, because in case of CRPS, pain develops in predisposed organism, with the presence of additional pathologic factors. However, McCabe et al.¹⁸ argued with the mentioned theory, presenting the confirmation that increasing pain and sensory impairment is a result of sensory-motor conflict in healthy subjects (volunteers performed movements which were reverse to those they've seen in the mirror¹⁸) and in patients with CRPS I and fibromyalgia,¹⁹ as well as evidence of the pain and sensory impairment development in the contralateral healthy limb. These data point at the possible mechanisms of biofeedback therapy influence on the pain generation and body schema perception.

Our results are to some extent correlated with the results of McCabe et al.,²⁰ who showed that mirror therapy is highly effective and leads to an immediate reduction in pain in patients with CRPS duration less than 8 weeks. Same investigators confirmed the continuing efficacy of the mirror therapy in reducing joint stiffness and thermal asymmetry (within 6 weeks) in case of longer duration of the disease – up to 1 year, but the use of this method later in disease flow was

considered ineffective. However, the number of observations in this study was not significant, so the results cannot be considered final.²⁰ In our study, we demonstrated good results of the mirror therapy as a part of the complex treatment course in rather homogeneous patient group with longer CRPS symptoms caused by same trauma locus. To contrast with other studies we were aimed to investigate the effect of the mirror therapy specifically on the body schema perception, we did not evaluate thoroughly its influence on other symptoms. But during the study, we admitted some positive effects of such treatment on other certain symptoms. Thus, some of our patients reported decrease in pain and increase in movement capacity and/or muscle strength. In majority of patients, the effect was not stable and significantly decreased in days or even hours - both after one session and after course of treatment. However, for such cases, even a short-term improvement in selected parameters of movements and muscles strength makes the mirror therapy worth to use. This prevents tissue atrophy and other irreversible changes in affected limb until a more effective treatment can be applied.

On the basis of results of mirror therapy and prism adaptation use in CRPS patients, investigators Bultitude and Rafal²¹ proposed several hypotheses of the CRPS pathogenesis in some patients. First, in some patients with CRPS I, symptoms of image disturbance of the affected limb are primary to the other symptoms of the disease. Thus, at least in some patients, pain can be the consequence, but not the cause, of the body representation disturbances. Second, the immobilization, but not the pain, may precipitate and stimulate this reorganization of somatomotor circuits in susceptible individuals. Third, limitations in voluntary movements are neither due to pain nor due to weakness but, rather, because of the derangement of body representation which renders certain postures from the repertoire of hand movements inaccessible.²¹ In this study, we could neither confirm nor deny these hypotheses.

Recently, the kind of the modern analogue of mirror therapy is in practice for the treatment of such patients – it is so-called 'virtual reality' technology. Today, there is no evidence of the efficacy of this treatment in patients with CRPS I, but there are some reports of successful use of this method in patients with phantom pains after the limb amputation. Today, this therapy direction is promising and requires further researches.²² Another potentially effective method of CRPS I treatment is the neuromodulation technique, in particular, stimulation of the motor cortex, which proved its effectiveness in reducing the pain and correcting sympathetic disorders in CRPS I²³ and CRPS II (several observations) patients²⁴ and potentially (not yet proven) can also positively affect the body schema perception. We also assume that mirror therapy positively affects restoration of the body schema perception not only directly but also indirectly due to the reduction of other disease symptoms.

Conclusion

Mirror therapy as part of complex integrative treatment is effective for the correction of the body schema perception disturbances in patients suffering from CRPS I, developed after fractures of the distal radius less than the duration of 3 years. However, the treatment of CRPS I always demands to use complex approach to the patient, more than one treatment method to get better results and also sufficient number of patients to analyse the data. Further researches are required to investigate the long-term efficacy of such therapy and precise influence of the mirror therapy on different CRPS I symptoms.

Acknowledgements

All authors studied literature and planned the study. O.B., O.K. and V.K. were involved in protocol development, getting ethical approval, patient recruitment and data analysis. V.K. and L.K. wrote and translated the manuscript. I.Z. and L.K. were involved also in patient recruitment and data analysis. All authors reviewed and edited the manuscript and approved the final version.

Conflict of interest

The author(s) declared no potential conflicts of interest with respect to the research, authorship and/or publication of this article.

Ethical approval

The ethics committee of SI 'Institute of Traumatology and Orthopedics of the Academy of Medical Sciences of Ukraine' approved this study (REC number: 1/2017).

Funding

The author(s) received no financial support for the research, authorship and/or publication of this article.

Guarantor

V.K. is the study guarantor.

Informed consent

Written informed consent was obtained from all subjects before the study and also for their anonymized information to be published in this article.

ORCID iD

Viktor Kotiuk ២ https://orcid.org/0000-0001-8837-8603

References

- Yucha C and Montgomery D. Evidence-based practice in biofeedback and neurofeedback. Wheat Ridge, CO: Association for Applied Psychophysiology and Biofeedback, 2008.
- Ezendam D, Bongers RM and Jannin MJA. Systematic review of the effectiveness of mirror therapy in upper extremity function. *Disabil Rehabil* 2009; 31(26): 2135– 2149.
- Cacchio A, De Blasis E, Necozione S, et al. Mirror therapy for chronic complex regional pain syndrome type 1 and stroke. N Engl J Med 2009; 361(6): 634–636.
- Selles RW, Schreuders TAR and Stam HJ. Mirror therapy in patients with causalgia (complex regional pain syndrome type II) following peripheral nerve injury: two cases. *J Rehabil Med* 2008; 40: 312–314.
- Pervane VS, Nakipoglu YGF, Sezgin OD, et al. Effects of mirror therapy in stroke patients with complex regional pain syndrome type 1: a randomized controlled study. *Arch Phys Med Rehabil* 2016; 97(4): 575–581.
- Al Sayegh S, Filén T and Johansson M. Mirror therapy for complex regional pain syndrome (CRPS) – a literature review and an illustrative case report. *Scand J Pain* 2013; 4(4): 200–207.
- Lewis JS, Kersten P, McCabe CS, et al. Body perception disturbance: a contribution to pain in complex regional pain syndrome (CRPS). *Pain* 2007; 133(1–3): 111–119.
- Haggard P and Wolpert DM. Disorders of body schema. In: Leiguarda R (ed.) *Higher-order motor disorders: from neuroanatomy and neurobiology to clinical neurology*. Oxford: Oxford University Press, 2005, pp. 261–271.
- Maihöfner C, Handwerker HO, Neundörfer B, et al. Cortical reorganization during recovery from complex regional pain syndrome. *Neurology* 2004; 63(4): 693– 701.
- Pleger B, Tegenthoff M, Ragert P, et al. Sensorimotor retuning in complex regional pain syndrome parallels pain reduction. *Ann Neurol* 2005; 57(3): 425–429.
- 11. Juottonen K, Gockel M, Silen T, et al. Altered central sensorimotor processing in patients with complex regional pain syndrome. *Pain* 2002; 98: 315–323.
- Pleger B, Rager P, Schwenkreis P, et al. Patterns of cortical reorganization parallel impaired tactile discrimination and pain intensity in complex regional pain syndrome. *Neuroimage* 2006; 32(2): 503–510.
- Graziano MSA and Botvinick MM. How the brain represents the body: insights from neurophysiology and psychology. In: Prinz W and Hommel B (eds) *Common mechanisms in perception and action*. Oxford: Oxford University Press, 2002, pp. 136–157.
- 14. Lewis JS and McCabe CS. Body perception disturbance (BPD) in CRPS: current and emerging therapeutic approaches including desensitization techniques and mirror visual feedback, together with the introduction of a new clinical tool for the early identification of BPD. *Pract Pain Manag* 2010; 10(3): 60–66.
- 15. Forderreuther S, Sailer U and Straube A. Impaired selfperception of the hand in complex regional pain syndrome (CRPS). *Pain* 2004; 110: 756–761.

- Priganc VW and Stralka SW. Graded motor imagery. J Hand Ther 2011; 24(2): 164–168.
- Moseley GL, McCormick K, Hudson M, et al. Disrupted cortical proprioceptive representation evokes symptoms of peculiarity, foreignness and swelling, but not pain. *Rheumatology* 2006; 45(2): 196– 200.
- McCabe CS, Haigh RC, Halligan PW, et al. Simulating sensory-motor incongruence in healthy volunteers: implications for a cortical model of pain. *Rheumatology* 2005; 44(4): 509–516.
- McCabe CS, Haigh RC, Halligan PW, et al. Distorting proprioception in chronic pain patients exacerbates sensory disturbances – implications for pathology. *Rheumatology* 2003; 42(4): 145.
- 20. McCabe CS, Haigh RC, Ring EF, et al. A controlled pilot study of the utility of mirror visual feedback in the

treatment of complex regional pain syndrome (type 1). *Rheumatology* 2003; 42(1): 97–101.

- 21. Bultitude JH and Rafal RD. Derangement of body representation in complex regional pain syndrome: report of a case treated with mirror and prisms. *Exp Brain Res* 2010; 204(3): 409–418.
- 22. Giraux P and Sirigu A. Illusory movements of the paralyzed limb restore motor cortex activity. *Neuroimage* 2003; 20: S107–S111.
- Velasco F, Carrillo-Ruiz JD, Castro G, et al. Motor cortex electrical stimulation applied to patients with complex regional pain syndrome. *Pain* 2009; 147(1–3): 91–98.
- Son UC, Kim MC, Moon DE, et al. Motor cortex stimulation in a patient with intractable complex regional pain syndrome type II with hemibody involvement. *J Neurosurg* 2003; 98(1): 175–179.