Mirror box therapy added to cognitive behavioural therapy in three chronic complex regional pain syndrome type I patients: a pilot study

Y.I.G. Vladimir Tichelaar[a], Jan H.B. Geertzen[b,c], Doeke Keizer[d] and C. Paul van Wilgen[b,d]

Complex regional pain syndrome type I is a disorder of the extremities with disability and pain as the most prominent features. This paper describes the results of cognitive behavioural therapy combined with mirror box therapy in three patients with chronic complex regional pain syndrome type I. Before, during and at follow-up the following measurements were assessed: pain (visual analogue scale, 0–100), range of motion, muscle strength, and the areas of allodynia and of hyperalgesia. Furthermore, patients were asked for their feelings and thoughts about mirror box therapy and about the affected limb. Pain at rest, pain after measuring allodynia/hyperalgesia and pain after measuring strength decreased. Range of motion improved in two patients. Strength improved in one patient. The area of hyperalgesia increased for all three patients, whereas the area of allodynia remained stable in two patients and decreased in one patient. Two patients felt that their affected limb still belonged to them, one did not. Cognitive behavioural therapy combined with mirror box therapy for patients with chronic complex regional pain syndrome type I may facilitate rehabilitation. Measuring whether the affected limb still belongs in the patient's body scheme could be of prognostic value in the treatment of chronic complex regional pain syndrome type I patients. *International Journal of Rehabilitation Research* **30:181–188 © 2007 Lippincott Williams & Wilkins.**

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Introduction

Complex regional pain syndrome type I (CRPS-I) is a disorder of the extremities with disability and pain as the most prominent features, especially in chronic CRPS-I patients (Ribbers *et al.*, 1995, Geertzen *et al.*, 1990). CRPS-I is defined by diagnostic criteria proposed by the International Association for the Study of Pain. In CRPS-I there is no evidence of nerve damage, in contrast to CRPS-II (causalgia) (Stanton-Hicks *et al.*, 1995). Pain is usually located in the distal part of the limb, and has a tendency to spread proximally (Rommel *et al.*, 1999). Spreading of signs and symptoms beyond the site of initial trauma is characteristic of CRPS-I (Veldman, 1995). Women are more frequently affected than men (7:3) (Allen *et al.*, 1999).

In the acute phase, the five classical symptoms of inflammation (tumor, rubor, kalor, dolor and functio laesa) may all be present (Veldman, 1995). In the chronic phase of the syndrome (i.e. with features of CRPS-I for 6 months or longer), pain, sensory changes (allodynia and hyperalgesia) and trophic changes are more prominent, resulting in disuse and a painful, dystrophic or atrophic, dysfunctional limb (Veldman *et al.*, 1993). Allodynia is defined as pain due to a stimulus which does not normally provoke pain (Mersky and Bogduk, 1994). Hyperalgesia is defined as an increased response to a stimulus which is normally painful (Mersky and Bogduk, 1994).

Several theories are available, which may account for signs and symptoms in the chronic phase of CRPS-I. In the learned-nonuse theory, peripheral and central sensitization will lead to allodynia or hyperalgesia in CRPS-I. Immobility and disuse occur as a result of formerly received negative feedback (pain or failure) when trying to use the affected limb (Woolf *et al.*, 1994; Schürmann *et al.*, 1999). As a consequence, when disuse of the limb remains for a longer period of time this may lead to more atrophic changes, immobility and cortical reorganization of the somatosensory cortex (Bortz, 1984).

In the remapping hypothesis, in patients with chronic CRPS-I, absence of consistent proprioceptive feedback when giving motor commands to the affected limb may

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increase pain and changes in the primary somatosensory cortex in patients (Ramachandran and Hirstein, 1998). Evidence of underlying changes in the primary somatosensory cortex was found in patients with CRPS-I using a magneto encephalogram or functional magnetic resonance imaging (Maihofner *et al.*, 2003; McCabe *et al.*, 2003b).

Recently, studies have described that one could possibly break through the vicious circle of pain and disuse as a result of remapping the primary somatosensory cortex by providing visual feedback (Maihofner *et al.*, 2003, 2004). As described in patients with phantom pain and sensations, providing visual feedback as a substitute for missing proprioceptive feedback may reduce pain, enabling patients to experience a more 'vivid' phantom (Ramachandran, 2000). Also a central role for the premotor cortex could be present. When normal somatosensory feedback is missing, visual feedback restores the information flow from the posterior parietal cortex to the premotor cortex (Di Pelligrino *et al.*, 1992; Seitz *et al.*, 1998; Altschuler *et al.*, 1999). Recruiting the premotor cortex or rebuilding the motor programme in the premotor cortex by providing visual feedback could reduce pain and facilitate the limb movement (Rothgangel, 2004).

To achieve visual feedback, patients can be treated with mirror box therapy, in which their limbs are positioned in a box separated by a mirror placed saggitally. By looking in the mirror at the unaffected side, patients can be 'fooled' in believing that the affected limb is moving effortlessly (Ramachandran and Hirstein, 1998). In patients with hemiparesis after stroke, mirror therapy has been used in providing visual feedback to reduce pain and facilitate rehabilitation of the affected limb [Altschuler *et al.*, 1999, Rothgangel *et al.*, 2004]. Also in patients with phantom limbs and phantom pain, mirror box therapy has been used successfully (Ramachandran, 2000).

Concordantly, mirror box therapy in patients with CRPS-I existing for less than 2 years has shown to cause some regain of functionality and mobility, and to reduce pain (McCabe *et al.*, 2003a). Evidence of cortical reorganization of the primary somatosensory cortex was also found in parallel with clinical improvement of the patients (Maihofner *et al.*, 2004). In our hospital, patients with CRPS-I are treated with cognitive behavioural therapy (CBT).

Owing to the formerly mentioned results with mirror box therapy, we decided to add mirror box therapy to CBT in the treatment of three CRPS-I patients in our hospital. We tried to measure some outcomes to establish an idea whether mirror box therapy could be a useful add-on to CBT in the treatment of CRPS-I. We will describe three cases of patients with chronic CRPS-I, treated with CBT and mirror box therapy.

Methods

CBT consists of the following phases. In the preclinical phase, after multidisciplinary assessment, reconceptualization of the patients' cognitions about CRPS-I is established. The so-called sensitization model is used to explain signs and symptoms to the patients (van Wilgen and Keizer, 2004). The main goal of this reconceptualization of cognitions is to convince patients to no longer believe that actual tissue damage is responsible for their pain and dysfunction. In the clinical phase, operant, cognitive and respondent techniques are used by an experienced team consisting of a psychologist, physical therapist and a physician. A time-contingent detoxification protocol is implemented during the first week. After detoxification, mirror box therapy is introduced during the second week, as add-on to the desensitization therapy.

Three patients participated in a 4–6 weeks inpatient CBT combined with mirror box therapy aiming at regaining limb function and pain reduction. During the first week, all analgesics were gradually reduced or stopped (detoxification), as discussed with the patient in the preclinical phase. In the second week, mirror therapy was introduced three times a day for two cycles of 5 min. Patients exercised little movements of the nonaffected side, whereas they were instructed to imagine the movement was performed in both limbs. During this procedure patients looked at their unaffected limb in the mirror, so that it would appear as if both limbs were moving effortlessly. When the patient was able to perform little movements with the affected limb (with the toes) he or she was encouraged to exercise these movements with both limbs while looking in the mirror. In the third week mirror box therapy was performed five times a day, for two cycles of 5 min.

Measurements were performed by an investigator who was not involved in the treatment. The patients were evaluated before the clinical phase, once a week during therapy (the mean scores are presented as one) and at follow-up after the clinical phase. The first patient was evaluated at 14 weeks follow-up, the second at 8 weeks follow-up and the third at 5 weeks follow-up.

Quantitative aspects of pain were assessed using a visual analogue scale (VAS, range 0–100). The pain was measured at rest, and after testing range of motion (ROM), muscle strength, allodynia and hyperalgesia.

ROM was measured using a goniometer, to assess maximal hand (dorsal/palmar flexion) or feet (dorsal/plantar flexion) movements. In addition, the position of

the hand or foot in which the patient was most comfortable at rest was recorded. Muscle strength of the affected limb was measured with a hand-held dynamometer according to a standardized protocol using break tests (van Wilgen *et al.*, 2003). Allodynia and hyperalgesia were assessed using a brush and a Von Frey monofilament, respectively. With the brush Aβ-fibre-mediated allodynia was tested, whereas with the Von Frey monofilament (no. 4.98) Aδ-fibre nociceptors and thus hyperalgesia were tested. The upper borders of the areas with allodynia and hyperalgesia were measured. From each digit of the affected limb, a virtual line was extended proximally, passing an anatomical landmark (for the ankle, the malleoli; for the wrist, the styloid-proces of the radius), which was chosen as zero-point. The brush or Von Frey monofilament moved distally from an area where no allodynia or hyperalgesia was present, towards the affected area, along the five virtual lines of the five digits. When the patient perceived the stimulus to be painful, the distance between the anatomical zero-point and the judged painful stimulus, along each virtual line, was measured. The brush moved continuously at a speed of approximately 2 cm/s and the monofilament was pressed on the skin for 1 s, with intervals of 0.5 cm. At investigation, first the brush and thereafter the Von Frey monofilament was used.

Finally, patients were asked to write down their thoughts about mirror box therapy and about their affected limb, before, during and after CBT combined with mirror box therapy.

Results

Case 1

This patient was a 23-year-old man, who developed CRPS-I after a fracture of digit III in his right foot, 30 months before attending our hospital. Treatment with physical therapy, transcutaneous electrical nerve stimulation, a sympathetic block and medication did not improve complaints in the past.

Patient used two elbow-crutches for walking, carefully avoiding using the affected foot. He was not able to move his foot and did not exercise or touch his foot at all. Pain was always present. At the time of multidisciplinary assessment, the right foot was oedematous, allodynic and fixed in 45° plantar flexion at rest. Increased hair and nail growth was seen (Fig. A1 of the Appendix). In the preclinical stage he used vitamin C, nifedipine and acetylcystein.

Before mirror box therapy, the patient described his bodily sensations of his leg as if it was not responding, although he commanded it to move. During mirror box therapy, he initially reported an incongruent feeling seeing the affected foot moving in the mirror. Later, he described it to be more like a funny feeling. At the end of the treatment, the patient's foot felt like it was moving, but he did not see it moving in reality.

Table 1 Result of case 1 before, during and after CBT with mirror box therapy

	Before	Treatment	Follow-up
VAS at rest	43	41	37
VAS after allodynia and hyperalgesia	50	57	44
VAS after strength	74	68	54
ROM (dorsal plantar flexion, in degrees)	0–15–53	0–14–53	0–10–37
Position foot at rest, in degrees plantar flexion	45	24	5
Strength plantar flexion (Newton)	23	36	59
Strength dorsal flexion (Newton)	23	29	39

CBT, cognitive behavioural therapy; VAS, visual analogue scale; ROM, range of motion.

At follow-up the patient was able to walk very slowly, for little distances without using his elbow-crutches. He also stated that mirror box therapy improved his condition and experienced less pain without using medication.

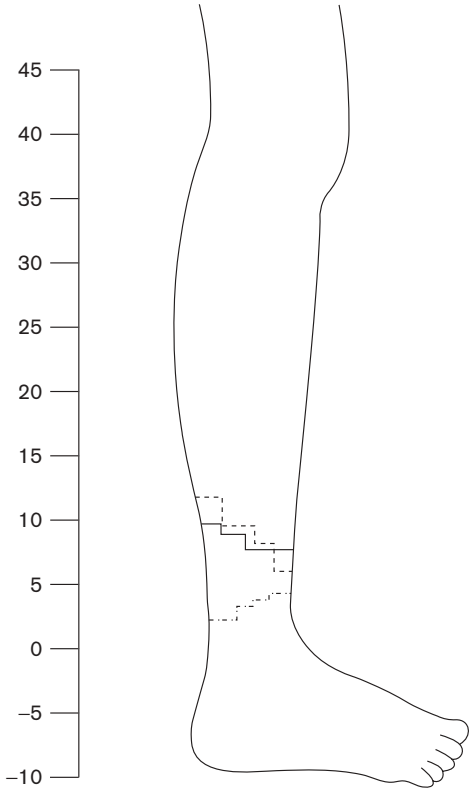
VAS scores, results of ROM and of strength tests are listed in Table 1. The course of the areas of allodynia and hyperalgesia is shown in Figs 1 and 2, respectively. Overall, pain decreased. ROM (dorsal flexion) increased and the position of the foot at rest turned from 45 towards 5° plantar flexion. This *pes equinus* restricted further progress of mobility; reconstruction surgery is currently considered. Strength improved, the area of allodynia decreased, but the area of hyperalgesia remained almost stable (Figs 1 and 2).

Case 2

This patient was a 42-year-old woman with CRPS-I of the left leg as a result of a minor trauma to the left knee 8 months earlier. For this condition she received pharmacotherapy and physical therapy. Pain was always there and was described as burning, descending from the left knee distal towards the toes. The patient sat in a wheelchair, and was unable to walk. The leg had a bluish colour, mostly distal. The knee was in 20° flexion position, with atrophic changes of the quadriceps muscle. The whole leg appeared sweaty, hyperpathic and allodynic. Hair growth was not visible on the distal part of the leg; there was a complete nonuse of the left leg (Fig. A2 of the Appendix). Daily medication at intake was tramadol, celecoxib and amitriptyline. Medication after detoxification was reduced to amitriptyline.

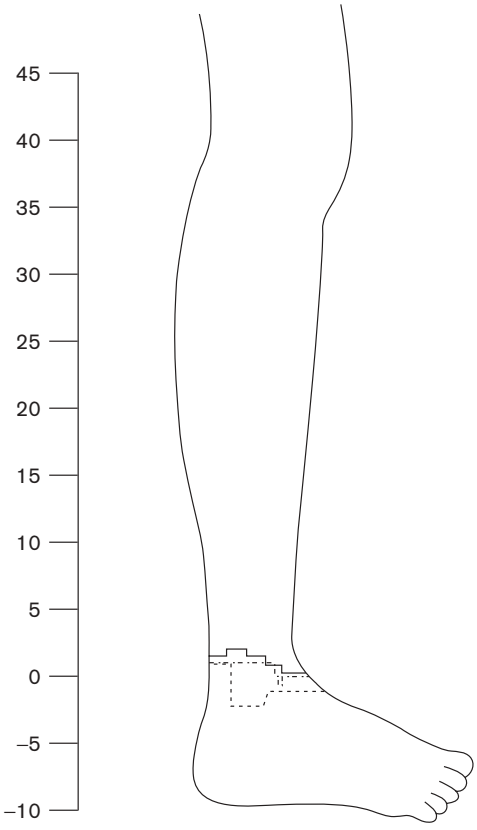
She described the affected leg as still belonging to her, but like it was not willing to move. Besides that, when

Fig. 1



Pain area of Aβ (allodynia) of case 1. Before treatment (·-·-·-·-·), during treatment (–) and follow-up (- - - -).

Fig. 2



Pain area of Aδ (hyperalgesia) of case 1. Before treatment (·-·-·-·-·), during treatment (–) and follow-up (- - - -).

trying to move or touching the leg, she experienced a lot of pain.

During mirror box therapy, she indicated repeatedly it felt like the leg was not responding to her commands. She never experienced the feeling of movement of the leg, and did not see it moving at all. After mirror box therapy, she was disappointed not making contact with the limb.

At follow-up, the situation did improve a little, i.e. the patient used less medication and experienced less pain, but was still not able to move the affected leg. The patient was disappointed by the results. It has to be noted that during therapy, she experienced a major life event, which decreased her motivation and interrupted treatment.

Table 2 shows the VAS scores and the result of ROM tests. Measuring strength was not possible, because the pressure of the hand-held dynamometer caused too much pain. In Figs 3 and 4 the course of the areas of allodynia and hyperalgesia is shown. Pain at rest and after testing allodynia and hyperalgesia decreased. Dorsal flexion increased a little, but plantar flexion decreased. Position of the foot at rest worsened a little, it was held more in plantar flexion at follow-up than before treatment. The area of allodynia decreased but the area of hyperalgesia increased.

Case 3

The third patient was a 46-year-old woman; 9 years ago she was involved in a car accident, which resulted in the development of CRPS-I in her left shoulder, nondominant arm and hand. Physical therapy and sympathetic blocks did not improve complaints. At the time of multidisciplinary assessment, flexion contractures in shoulder, elbow, wrist and fingers were present. Except little movements of the thumb and digit II, extension of the fingers was neither passively nor actively possible. The forearm was cold and atrophic (Fig. A3 of the Appendix). Pain was not always present; mostly it was provoked by trying to move the arm, or by contact with surroundings or cloth. Allodynia and hyperalgesia were present. She was not using any medication at intake.

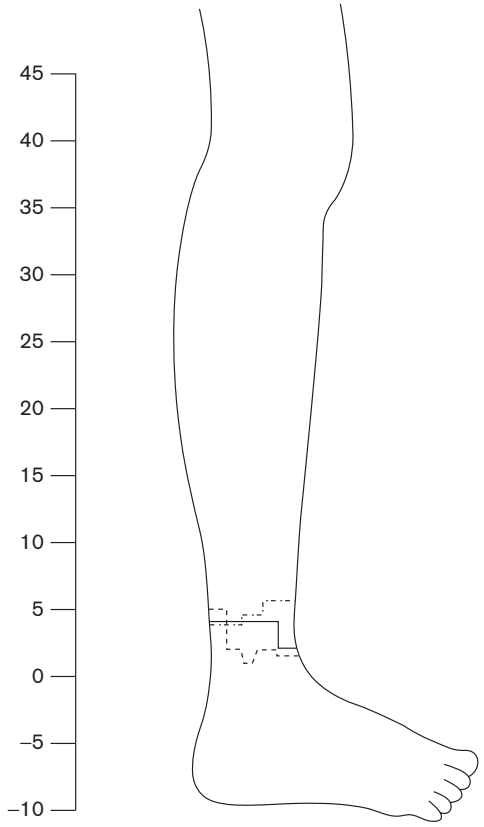
Before mirror box therapy she stated her arm did not belong to her anymore. She felt it was like something

Table 2 Result of case 2 before, during and after CBT with mirror box therapy

	Before	Treatment	Follow-up
VAS at rest	63	68	54
VAS after allodynia and hyperalgesia	67	72	57
ROM (dorsal plantar flexion, in degrees)	0–20–48	0–27–40	0–15–35
Position foot at rest, in degrees plantar flexion	24	31	30

CBT, cognitive behavioural therapy; VAS, visual analogue scale; ROM, range of motion.

Fig. 3

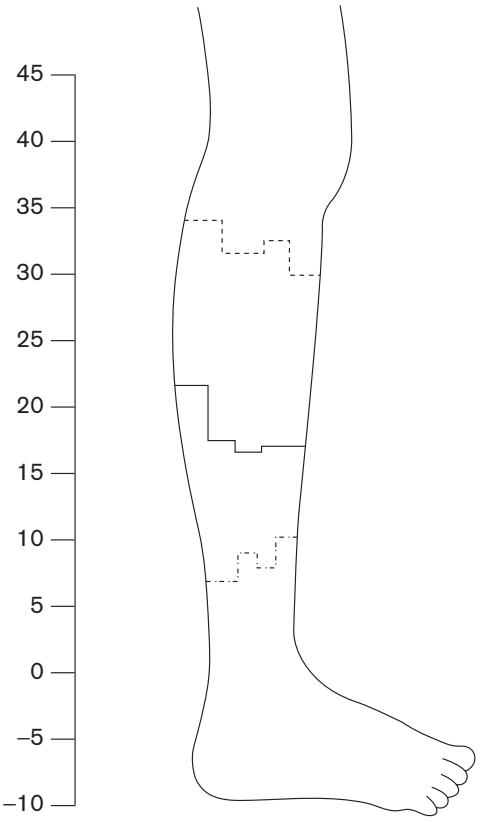


Pain area of Aβ (allodynia) of case 2. Before treatment (·-·-·-·-·), during treatment (–) and follow-up (- - - -).

strange to her, she even dreamed of herself without having a left arm. The patient was not even able to imagine her hand moving as it could before the accident.

During mirror box therapy the patient did not recognize the affected left arm as belonging to her. She also did not experience any feelings of making contact with her arm. After treatment, the patient did not improve on any outcome. She still could not move the arm and the pain attacks remained happening on movement or touching of the arm.

Fig. 4



Pain area of Aδ (hyperalgesia) of case 2. Before treatment (·-·-·-·-·), during treatment (–) and follow-up (- - - -).

Table 3 Result of case 3 before, during and after CBT with mirror box therapy

	Before	Treatment	Follow-up
VAS at rest	29	22	15
VAS after allodynia and hyperalgesia	58	23	22

CBT, cognitive behavioural therapy; VAS, visual analogue scale.

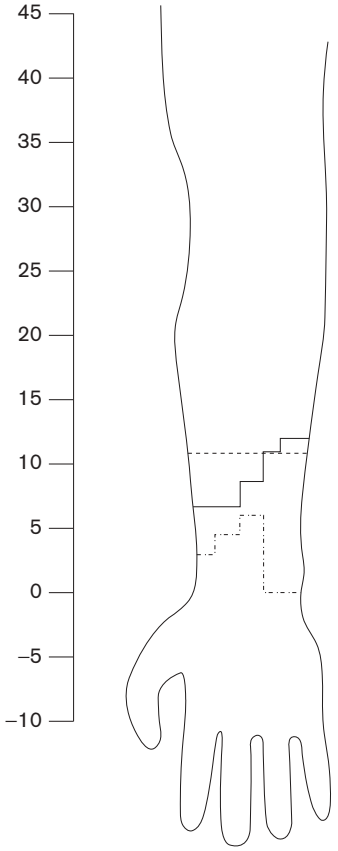
In Table 3 the results of VAS are shown. ROM and strength testing were not possible because of severe dystonia and contractures of the affected arm. The course of the area of allodynia and hyperalgesia is shown in Figs 5 and 6. Pain at rest and after testing allodynia and hyperalgesia decreased during treatment, but at follow-up she still experienced pain attacks.

Discussion

In this small group of CRPS-I patients with severe disuse and pain, mirror box therapy was added to CBT as treatment for CRPS-I.

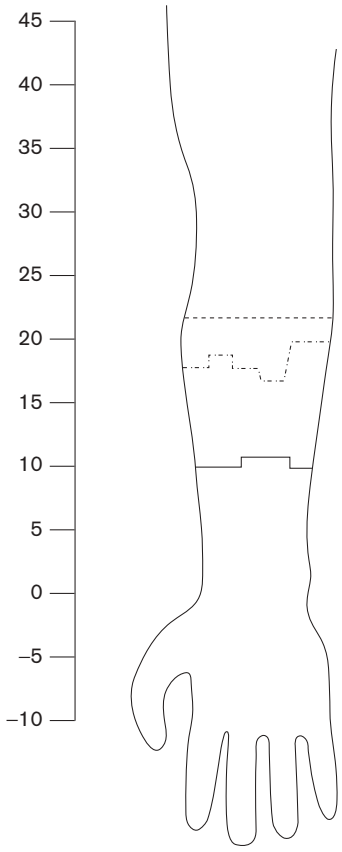
After treatment and follow-up we can conclude that case 1 improved, i.e. he experienced less pain without using any medication and could walk a little distance without

Fig. 5



Pain area of Aβ (allodynia) of case 3. Before treatment (·-·-·-·-·), during treatment (–) and follow-up (- - - -).

Fig. 6



Pain area of Aδ (hyperalgesia) of case 3. Before treatment (·-·-·-·-·), during treatment (–) and follow-up (- - - -).

using elbow-crutches. The patient in case 2 improved less. She experienced less pain, but mobility did not improve. In the last case, the patient did not improved at all.

Besides these results, it seems that the outcome of our combined treatment for CPRS-I could be predicted by some factors identified in these three case studies.

First, as mentioned earlier, the longer the CPRS-I diagnosed, the worse the disability and pain (Veldman *et al.*, 1993). In addition, mirror therapy alone does not seem to improve disability when CRPS-I exists longer than 2 years (Bortz, 1984). In our third case, CRPS-I was existing for almost 9 years, which had led to irreversible contractures and atrophy. Although in this case it may seem too obvious that such a subtle approach as CBT and mirror box therapy can do little about this major irreversible pathology, in other cases where CRPS-I does not exist that long, it may be less clear. In our first case, where CRPS-I existed for 2.5 years, the patient did improve, but this was also limited by contractures resulting from the duration of the CRPS-I. In addition, our second patient improved at least on pain. So chronic CRPS-I may not be susceptible to CBT and mirror box therapy. Whether this is caused by peripheral pathology (contractures, atrophy) alone or also by irreversible cortical changes of the primary somatosensory cortex has to be further investigated.

In addition, patients stating that their affected limb does not belong to them anymore (i.e. is not a part of their body scheme anymore), seem to have no benefit of CBT combined with mirror box therapy (case 3). In a lesser way, patients saying they cannot imagine or feel their affected limb moving in their mind, i.e. patients who are not able to make contact with their limb, also seem to have less benefit of our treatment (case 2). Alltogether, some chronic CRPS-I patients might have benefit from CBT and mirror box therapy, in making 'contact' with the affected side (Ramachandran and Hirstein, 1998; McCabe *et al.*, 2003a; Rothgangel *et al.*, 2004). An important prognostic value then might be the degree of 'foreignness' of the affected limb described by patients.

The more the patient describes their affected limb as foreign, the less the benefit of mirror box therapy may be expected. These descriptions of (feelings of foreignness) the affected limb might reflect (ir)reversible changes in the primary somatosensory cortex. In case of more definitive changes in the primary somatosensory cortex, as in long-standing CRPS-I, mirror box therapy seems less affective to 'train the brain', i.e. to remap the primary somatosensory cortex (Maihofner *et al.*, 2004). Therefore, asking for patients' subjective thoughts of the affected limb in the diagnostic process and during rehabilitation seems valuable in determining the possible outcomes of CBT combined with mirror box therapy.

Although pain levels at rest and after testing of allodynia and hyperalgesia decreased for all three patients, the hyperalgesic areas increased in all three patients, whereas allodynic areas decreased in two patients and remained stable in one patient. Therefore, CBT combined with mirror box therapy seems to establish desensitization of the Aβ-fibres but not of the Aδ-fibres. This could reflect the underlying, more central, pathophysiological mechanisms of allodynia, and the more peripheral pathophysiological mechanisms of hyperalgesia. Probably, mirror therapy establishes recruitment of peripheral nociceptors as a side effect. More research on this hypothesis is recommended.

Finally, medication intake was strongly reduced in two out of the three patients, combined with lower pain levels. This may reflect the noninflammatory, non-neuropathic aspect of pain. Probably, this pain is mediated by the central changes of the somatosensory cortex, as suggested for phantom limbs too (Ramachandran and Hirstein, 1998).

These three case reports suggest that mirror box therapy, combined with CBT, could have a positive role in the rehabilitation of some patients with CRPS-I. Positive outcomes of treatment seem to depend partially on the duration of the syndrome (less than 2 years), the absence of contractures and on whether the affected limb is still a part of patients body scheme. If so, CBT and mirror box therapy may reduce pain levels at rest and after stimulation, lower the medication intake, and improve the function of the affected limb a little. Although a placebo response seems highly unlikely, as stated in a study of patients with CRPS-I including a control group (McCabe *et al.*, 2003a), further research on this is strongly recommended.

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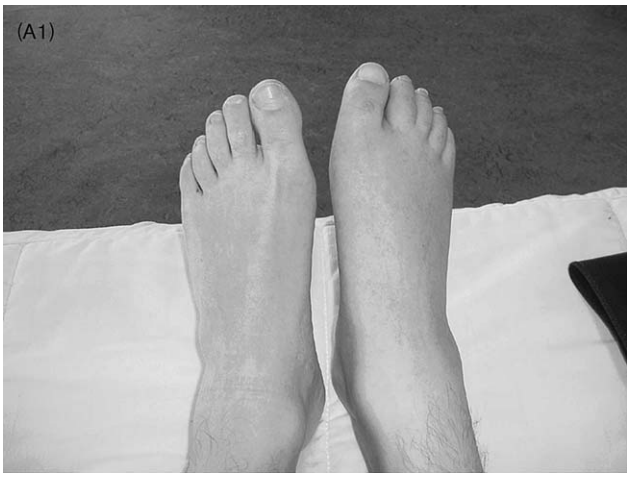
Appendix

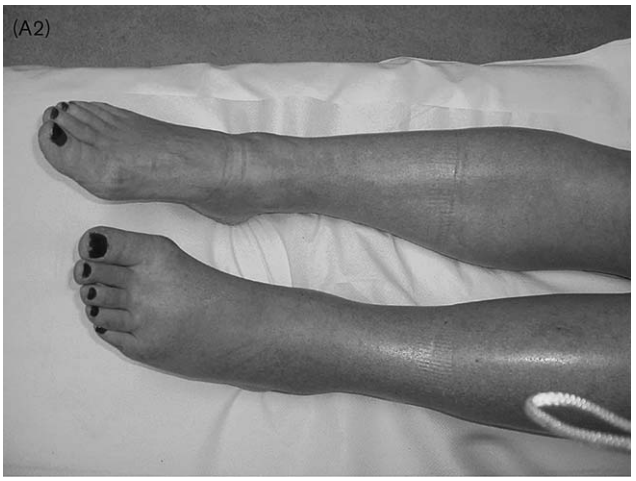
The results found in cases 1–3 are shown in Figs A1–A3.

Fig. A1



Case 1 at intake.

Fig. A2



Case 2 at intake.

Fig. A3

Case 3 at intake.