Disability and Rehabilitation

The long term effect of Complex Regional Pain Syndrome type 1 on disability and quality of life after foot injury

Dominique Mouraux[a,b] , Cédric Lenoir[c,d], Turgay Tuna[e], Eric Brassinne[a,b], Stéphane Sobczak[f,g]

[a] Department of Physical Therapy and Rehabilitation, Hôpital Erasme, Université libre de Bruxelles (ULB), Brussels, Belgium.
[b] Faculté des Sciences de la Motricité, Université Libre de Bruxelles (ULB), Brussels, Belgium.
[c] Department of Neuroscience, Physiology and Pharmacology, University College, London, UK.
[d] Institut of Neuroscience, Université catholique de Louvain (UCLouvain), Brussels, Belgium.
[e] Department of Anaesthesiology, Pain Clinic, Hôpital Erasme, Université libre de Bruxelles (ULB), Brussels, Belgium.
[f] Département d'anatomie, Université du Québec à Trois-Rivières (UQTR), Trois-Rivières, Québec, Canada.
[g] Groupe de Recherche sur les Affections Neuro-musculo-squelettiques (GRAN), Université du Québec à Trois-Rivières (UQTR), Trois-Rivières, Québec, Canada.

Implications for rehabilitation

- The long-term evolution of patients suffering from lower-limb Complex Regional Pain Syndrome is associated with persistent disability, pain and impacts the quality of life.
- Strength, proprioceptive, functional and subjective assessments are necessary to better identify deficits.
- Rehabilitation should focus on the overall deficit of the affected and contralateral limb.

Abstract

Purpose. To study the long-term evolution of patients with lower-limb Complex Regional Pain Syndrome (CRPS), focusing on functional and proprioceptive aspects and quality of life.

Methods: In 20 patients suffering from chronic distal lower-limb CRPS diagnosed using Budapest criteria, we assessed joint position sense and strength of the knee muscles at the CRPS and unaffected leg, functional exercise capacity, pain, CRPS severity score, quality of life and kinesiophobia. Similar assessments were performed in 20 age-matched controls.

Results: The joint position performance (at 45°) was significantly lower for the CRPS leg as compared to controls. The knee extensor strength of the CRPS leg was significantly reduced as compared to the unaffected leg (-27%) and controls (-42%). CRPS patients showed significantly reduced performance at the 6 minute-walk test as compared to their age group predicted value and controls. Patients suffering from CRPS for 3.8 year in average still exhibit high pain, severity and kinesiophobia scores.

Conclusions: Long-term deficits in strength and proprioceptive impairments are observed at the knee joint of the CRPS leg. This persistent functional disability has significant repercussions on the quality of life. We highlight the importance of including strength and proprioceptive exercises in the therapeutic approaches for CPRS patients.

Keywords

Complex Regional Pain Syndrome, isokinetic strength, joint position sense, proprioception, recovery, quality of live

Introduction

Complex Regional Pain Syndrome (CRPS) is characterized by continuous pain that is disproportionate in duration and intensity to the trauma that triggered it. Usually the painful area is limited to the limb that has been injured, nonspecific to a nerve territory or dermatome, predominantly distal and accompanied by sensory, motor, sudomotor, vasomotor and/or trophic alterations [1]. CRPS has been subdivised in two subtypes CRPS-I and CRPS-II, when focused on injury to the musculoskeletal system and when peripheral nerve damage is found electrophysiologically respectively [2]. The diagnosis is essentially clinical using the validated Budapest criteria [3]. The most common initiating traumatic events are fractures (45%), sprains (18%) and elective surgeries (12%), while a small proportion of CRPS (<10%) is spontaneous [4]. The prevalence of post-fracture CRPS varies from 0.03% to 37% [5]. Findings of different studies on the long-term symptoms and consequences seem contradictory. Sandroni et al. [6] reported 74% resolution at one year. According to Zyluk et al. [7] only 2% of patients had still CRPS at one year. Veldman et al. [8] and Galer et al. [9] showed persistent problems in the majority of patients. After 3.3 years, the affected limb is reported as weak or perceived as disconnected from the body respectively in 96.8% and 74.2%. All patients in this study reported moderate to severe pain intensity associated with substantial disability. More recently, de Mos et al. [10] highlighted that after 5 to 8 years 31% of patients with upper-limb CRPS are unable to work while 30% are cured. For some patients, symptoms can persist and lead to a long-term disability. According to Bean et al. [11], only 5.4% of patients were symptom-free at 12 months. The results are difficult to compare given the heterogeneity of the populations studied as well as the evolution of the clinical criteria used and the lack of consensus on what really defines healing and recovery [4]. Liewellyn et al. [12] showed that patients considered that they have recovered when their CRPS-related pain, movement difficulties (muscle weakness, decreased range of motion) and reliance on medication have been addressed. In more than 60%, the injury is located at the upper-limb and the clinical sign are more distal [6]. This suggests that an injury affecting a given joint or part of a limb might trigger CRPS symptoms also on remote regions of the affected limb. To date, some studies have examined the consequences of CRPS at the lower-limb [13,14], or upper-limb [7,15] or both together [6,9,10-12,14,16] in terms of persistence of clinical signs. The symptoms at the lower-limb are more refractory to intervention than those involving the upper-limb [17]. Lower-limb CRPS could lead to a severe disability preventing the patients from walking, altering their functional capabilities, autonomy and in turn the quality of life. Several studies explored and quantified the impact of CRPS on proprioception and strength of the upper-limbs [18-20]. They reported bilateral proprioceptive impairment for patients suffering of a unilateral CRPS characterized predominantly by a bilateral reduced accuracy in upper limbs re-positioning tasks. These authors suggested that central mechanisms, such as altered processing of afferent information contributing to motor dysfunction, might be responsible for proprioceptive deficits in CRPS. In contrast, to our knowledge, no study reported the assessment of proprioception and strength in patients suffering from lower-limb CRPS, while these measures are part of the routine evaluation of patients in traumatology and orthopedics physiotherapy.

In view of this observation, the purpose of this study was to expand the knowledge of the long-term evolution of patients with lower-limb CRPS, focusing on the functional and proprioceptive aspects and more globally on the quality of life. In this context, the assessment was not limited to the injured region but took into consideration the entire injured limb in a more global approach.

Materials and Methods

Population

From 2007 to 2015, 36 patients who had been treated for lower extremity (foot and ankle injuries) CRPS type 1 in the pain clinic department of the Hôpital Erasme (Université libre de Bruxelles, Belgium) were asked to participate. CRPS diagnosis was based on both, the clinical Budapest criteria and bone scintigraphy. Of the 36 patients eligible, 7 could not be reached, 9 refused to participate for health-related reasons (n=2) or lack of availability (n=7). Finally, 20 patients (17 women; age: 46.3±12.7 (mean±SD); range 22 - 76 years) were included and compared to a matched 20 healthy control population (5.6 women for one man). The follow-up was 3.8±2.2 years (mean±SD); (range 1 – 8 years). We assessed 8 subjects with CRPS at the right foot and 12 subjects with CRPS at the left foot. Exclusion criteria were any other pathology or disorder that might affect the lower limb and other conditions which might influence the results of the tests (vestibular disorders and psychiatric, neurological or cognitive disorders). CRPS patients and the control group were matched by age, height, weight, and gender. The control group was recruited by word of mouth in the neighborhood area. The demographic variables of both groups are presented in Table 1. CRPS was triggered by ankle or foot fracture in twelve patients and by ankle sprain in eight patients as reported in the demographic and clinical characteristics of individual CRPS patients in Table 2.

Table 1 and 2 should be inserted here

All patients and matched control individuals completed a consent form approved by the Institutional Ethic Committee Board of Hôpital Erasme (ref P2014/421 B406201422513) according to the declaration of Helsinki.

Functional Assessment

Joint position sense testing

Before starting the test, each subject performed a standardized warm-up protocol consisting of four minutes on a non-loaded ergometric bicycle (Monark 828E). This warm-up aimed at improving the accuracy of the assessments of the joint position test [21]. The joint position sense test consisted of passive knee joint positioning in a seated position, followed by passive repositioning at a pre-determined angle on an isokinetic dynamometer (Humac Norm, Stoughton, Massachusetts, USA). Subjects were seated and stabilized on the device with the hips and knees flexed at 90° with support for the back. The knee extension-flexion axis was approximated by aligning the dynamometer rotational axis with the lateral femoral condyle. The lower leg was attached to the resistance arm of the isokinetic dynamometer by means of a shin pad attached two finger-widths above the lateral malleolus. The target angles established for the tests were 15° and 45° for knee flexion. The order of the tests was counterbalanced across subjects. During the test, subjects were blindfolded to prevent any visual cue [22] (Figure 1). The tests were conducted in a quiet room, by the same researcher, who systematically used standardized verbal commands to guide the participants during the testing procedure. The subjects were asked to be as relaxed as possible in order to avoid any active contraction [23]. Before starting the test, the 0° angle of the resistance arm was defined for each participant as the angle reached when the participant extended actively the leg as far as possible. During the test, the resistance arm of the dynamometer passively extended the subject's lower leg at an angular velocity of 3°/s from the initial position of 90° of knee flexion until the target angle was reached. The position was maintained for 6 seconds to allow the subject to memorize it [23]. The lower leg was then brought back at 3°/s to the starting position (90°) which was held for 5 seconds. Then, the resistance arm extended the lower leg at a speed of 2°/s until the subject judged that the target position was reached and verbally stop the movement of the resistance arm. A different speed was used for repositioning to eliminate the possible cue due to the time interval needed to reach the target position [24]. Low velocities were used to avoid reflex contraction [23]. Three repetitions were carried out for each target angle (15° and 45°) and for each leg. The angle absolute error value was used and defined as the absolute difference between the target angle and the actual angle covered by the participant. Each test included the complete sequence of positioning-memorization-repositioning. At the "stop" signal of the participant, the reached angulation was recorded and the movement of the isokinetic dynamometer was stopped allowing the limb to resume the initial position under the action of gravity. The proprioceptive acuity tests were performed before the muscular performance measures to avoid fatigue, which could have affected the subject's performance during the joint position sense and the kinesthetic tests [25].

Strength

The setup was the same as for the position sense testing. Quadriceps and hamstring strengths were assessed at the velocities of 60°/s and the range of motion was 0° to 100° of knee flexion (0° was defined as full active extension). Before data collection started, the subjects were familiarized with the testing apparatus with three training trials at different angular velocities. After one minute of rest, subjects performed three maximal effort trials with verbal encouragement to perform the tests as fast and as forcefully as possible. The uninjured limb was tested first and then the same procedure was conducted for the injured limb. Quadriceps and hamstring isokinetic peak torques of the injured limb were expressed as a percentage of those of the uninjured limb. Meireles [26] et al. and Adsuar et al. [27] showed that the isokinetic test is reliable and valid for patients suffering from chronic pain.

Six-minute walk test (6MWT)

The 6MWT was performed using the methodology set out by Enright [28]. Subjects were asked to walk from one end of a 24-meters track to the other during six minutes, as fast as possible and without running. Subjects were not encouraged during the test but were informed each minute of the remaining time. The distance in meters was recorded for analysis.

Questionnaires

Pain

Patients were asked to rate their average pain intensity level for the last week on a Visual Analog Scale from 0 to 100 (0 corresponding to no pain and 100 to the worst imaginable pain).

Tampa scale for Kinesiophobia (TSK)

TSK is a 17-items questionnaire (with score ranging from 17 (absence of fear) to 68 (highest fear)) that measures fear related to movement and (re)injury. TSK has been shown to be reliable and valid for chronic pain patients [29].

CRPS Severity Score

The CRPS severity score is a measure of CRPS severity that corresponds to the sum of the signs and symptoms described in the Budapest criteria. It is based on 16 CRPS diagnostic features. The resulting severity score ranged potentially from 0 to 16, with higher scores indicating greater CRPS severity. Harden et al. has demonstrated its reliability [3] and validity [30], indicating that combining all items into a single summary score is justified and useful in clinical monitoring and outcomes research.

The Short Form-36

The Short Form-36 [31] is the most widely used survey to assess overall health-related quality of life [32]. Results from the Short Form-36 can be reported as two summary scores: the mental component score and the physical component score. These component scores are constructed using normative values so that the optimal mean score is 50 with a standard deviation of 10 [31]. The lower the Short Form-36 score is the more quality of life is affected. The mental component and the physical component scores are used in these analyses.

Statistical Analysis

Descriptive statistics were calculated (mean and standard deviation) for each data set. The Kolmogorov Smirnov test was applied to assess the normality of each variable and the Levene test was applied to assess the homogeneity of variances of compared variable. According to the normality of the data and homogeneity of variances, we applied either a *t*-test or a Mann-Withney U test. To assess the relationship between variables Pearson correlation coefficients were computed. All above statistical analysis were performed using Statistica 13.5.0 (TIBCO Software Inc, Palo Alto, USA) with a confidence interval at 95%. Significance threshold was set at $p<0.05$.

Results

Functional Assessment

Joint position sense

No significant difference in the angular absolute error was observed for the joint position sense between the left and right legs in the control group for the target angle of 15° (-0.9°±3.8; 95%CI [-2.5,0.8], t=2.807 p=0.155) and for 45° (0.3°±3.7; 95%CI [-1.3,1.9], t=0.324, p=0.749). The data of the two legs of the control group were then averaged and compared to the data obtained for the unaffected leg and the affected leg (CRPS leg) of the CRPS group. We observed a significant difference in the angular absolute error between the control legs and the CRPS leg only at the target angle of 45° (2.8±4.7°; 95%CI [0.7,4.8], U= 318.5, p=0.013).This error is greater for the CRPS leg (Figure 2). No significant difference in the angular absolute error was observed between the CRPS leg and unaffected leg within the CRPS group for both the target angle of 45° (1.3±1.6°; 95%CI [-0.3,2.9], 1.097 ,p=0.279) and 15° (1.9±5.9°; 95%CI [-0.7,4.5], t=-1.066, p=0.293) (Figure 2).

Strength

Within the CRPS group, we observed a significant reduction of the peak torque of the quadriceps of the CRPS leg as compared to the unaffected leg, (25.3±24.3 Nm; 95%CI [10.3,40.3], t=2.531, p=0.015). As compared to the control group, the CRPS leg and the unaffected leg exhibited a significant deficit (respectively 49.8±40.1 Nm; 95%CI [32.2,67.3], t=-4.341, p=0.0001 and 24.5±37.3 Nm; 95%CI [8.1,40.8], t=-2.224, p= 0.032). A significant deficit of the peak torque of the hamstrings was also observed for the CRPS leg as compared to the unaffected leg (11.7±15.8 Nm; 95%CI [4.7,18.6], t=2.661, p=0.011) and as compared to the controls (20.2±24 Nm; 95%CI [9.7,30.8], U=93.5, p=0.003). There was no significant difference between the unaffected leg in the CRPS group and the control group (8.6±22.9 Nm; 95%CI [-1.5,18.6], U=162, p=0.30) (Figure 3).

6MWT

We observed a significant deficit of the 6MWT for the CRPS group amounted to 421.7 ± 106.1 m, while the predicted value of the test for the same age group is 588.8 ± 91.6 m [27] and lower than the control group 557.5± 69 m. There were no significant differences between the predicted value and the control group (31.3±108.3 m; 95%CI [-16.2,78.8],t=1.523, p=0.136) Compared to the control group, the CRPS group showed a significant reduction of the distance (135.8±111.5 m; 95%CI [111.1,223.2], U=49, p=0.00004). The distance covered during the 6MWT in the CRPS group and the strength deficit of the CRPS Leg were significantly and positively correlated for the quadriceps deficit (r=0.769; n=20; p=0.0001) and the hamstrings deficit (r=0.740; n=20; p=0.0002) (Figure 4).

Questionnaires

Pain, kinesiophobia and CRPS severity score

Pain intensity ratings on the visual analogue scale (51±15mm) and the CRPS Severity Score (9.1±2.8) showed results that were still high. The TSK score (45.8±8.7) showed a high level of kinesiophobia.

The Short Form-36

Compared to control group, both physical and mental score components of the Short Form-36 were significantly lower (physical component score: 14.2±8.2; 95%CI [10.6,17.8], t=-5.787, p= 0.00001 and mental component score: 7.8±12.9; 95%CI [2.1,13.5], t=-2.233, p=0.03) and the physical component was predominantly reduced .

Relationship between the different outcomes

To test whether there was a relationship between functional parameters such as joint position sense, isokinetic strength and distance at the 6MWT and subjective parameters such as pain, quality of life and level of kinesiophobia, Pearson correlations were performed. Pain ratings and physical component of the Short Form-36 were significantly and negatively correlated (r=-0.762; n =17; p=0.0001). This meant that a high level of pain is associated with a low physical component. Physical component components exhibited a significant positive correlation with the 6MWT distances (r=0.674; n =17; p=0.003).

Kinesiophobia and joint position test were not significantly correlated with any other parameters.

Discussion

Functional assessment

The aim of this study was to assess the overall functional and subjective outcomes of patients suffering from distal lower-limb CRPS on the long term. The results suggested that after a mean follow-up of 3.8 years, the consequences of CRPS, are not limited to the affected joint or body part but extend to the adjacent joint with global functional consequences. We observed a significant deficit in the knee joint position sense, a overall significant strength deficit of the quadriceps (-42%) and hamstrings (-37%) as compared to the control group. A significant relationship was present between these deficits and the decreased functional exercise capacity revealed by the 6-minutes walking test. Subjective parameters such as persistent high level of pain was related to a decreased quality of life.

Joint position sense

We observed a highly significant difference between control subjects and those with CRPS in terms of passively repositioning the leg to a target angle of 45° knee flexion. This result is in line with the studies of Lewis et al. [18] and Bank et al. [19] who observed a significant difference between the proprioceptive performance of subjects with CRPS and healthy subjects at the level of the upper limb. In the study of Lewis et al. [18], the patients were asked to actively move the forearm in a horizontal plane to point a series of clock positions with eyes closed. Based on the observation that the performances were significantly poorer for the CRPS limb and also for the unaffected limb of the patients with CRPS as compared to healthy subjects. The authors concluded that this bilateral impairment would be related to central processes (cortical reorganization of regions associated with body schema and deviation of the median line) responsible of the proprioception alteration. Although the mean value of the repositioning error was greater for the unaffected leg in the CRPS group than in the control group, the difference did not reach significance, therefore our results do not fully support a bilateralization of the impairment. Since pain has been associated in several studies to the disturbance of body perception and the degree of cortical reorganization in [5,33], it seemed worthwhile to examine whether there was a link between proprioceptive performance and pain intensity. We did not observed any significant correlation between pain intensity and proprioceptive performance for the repositioning at 15° and 45°. Bank et al. [20] did not find any correlation between the pain and the repositioning neither for active and passive wrist mobilization with eyes closed. The unilateral impairment of proprioception could be explained, apart from central mechanisms, by elements such as vasomotor changes, muscular weakness, mechanical restrictions due to inactivity, and alterations in sensory and nociceptive transmission. All of these factors could be responsible for disturbing proprioceptive afferents by modifying nerve structures and deep tissues [18, 33]. It should be noted that our assessment protocol was focused on the knee and not the CRPS-affected foot. These regional (or local) factors cannot therefore entirely explain the impairment found at distance from the affected region. It has been shown that the CRPS joint could lead to proprioceptive deficits on an adjacent joint [18]. In a significant proportion of patients with CRPS, it was also observed that the pathology could spontaneous spread either ipsilaterally (34%), contralaterally (63%) or at a different segmental level (3%) compared to the initially affected joint [34]. The significant difference observed at 45° but not at 15° in our study could possibly be explained by the fact that the 15° position is close to the extreme amplitude of the movement in which joint receptors are recruited [34]. Proske [36] reported that joint receptors (Ruffini-like endings, and Pacinian corpuscles) are recruited in the extreme amplitudes. Nevertheless, it is known that joint proprioceptors provide input throughout the entire range of motion of a given joint.. Nevertheless, the patients in our study had in average a higher absolute error with a higher variability than the controls for the joint positioning test at 15°. Further investigations are needed to evaluate an angle-specific alteration of the sense of repositioning.

Strength

Our study confirmed that the majority of patients exhibited a reduction of the strength of the flexors and extensors of the knee at the affected limb and even bilaterally for the strength of the quadriceps. These alterations might be a consequence of foot CRPS. Isokinetic torque production capability is commonly used as a clinical indicator of strength as well as a correlate of the functional ability of the patient. The quadriceps weakness may result from either muscle atrophy or neuromuscular inhibition [37,38]. Mizner et al. [38] reported that failure of voluntary contraction (neuromuscular inhibition), that the loss of muscle cross-section area (atrophy) was not the primary determinant in the impairment of quadriceps strength. Muscle inhibition has been attributed to altered afferent input from the injured structures or diseased joint structures resulting in altered efferent motor neuron stimulation of the quadriceps [39]. Sedory et al. [39] showed that individuals suffering from chronic ankle instability exhibit altered motoneuron pool excitability of the quadriceps and hamstring muscles as compared to a control group. Gribble et al. [40] compared the force production capabilities of the ankle, knee, and hip in the sagittal plane among those with and without unilateral chronic ankle instability. In chronic ankle instability, the authors reported, in addition to deficits in ankle plantar flexion torque, deficits in knee flexor and extensor torque, suggesting that distal joint instability may lead to knee joint neuromuscular adaptations. The authors [39,40] recommended to take into account the motor control deficit to treat the lateral ankle sprains and chronic ankle instability. It suggests that a rehabilitation program should take into account adjacent joints, i.e. the knee in case of rehabilitation for ankle instability. In our study, deficit in strength of the extensors muscles is also present in the contralateral limb. This could be explained by the chronic state of our CRPS patient. Indeed, in chronic CRPS patient the cortical reorganization associated with shrinkage of the representation of the affected hand or foot in the primary somatosensory cortex (S1) and altered function of the primary motor cortex [41]. These persistent changes in the central nervous system might contribute to central sensitization [42], which in turn could play a role in the maintenance of chronic pain, allodynia, and the development of wide spread pain to adjacent non-injured areas [34], as well as motor dysfunctions such as dystonia [43], body perception disturbances [44,45], neglect like syndrome [46], i.e. affected ability of the patients to mentally represent, perceive and use their affected limb [47]. Those results could be attributed to “learned nonuse” [48] because of fear-avoidance behavior that reduced attempts to move. This pattern of behavior could maintain at least in part the quadriceps amyotrophy of the affected side but also the contralateral side.

Llewellyn et al. [12] reported on 242 chronic CRPS that muscle weakness and decreased range of motion were the most frequent symptoms. These findings underlined the importance to improve motor function and reduce stiffness as priorities in rehabilitation. In this context the rehabilitative program should aim at increasing the strength of the knee extensors and flexors bilaterally, to encourage the patients to engage movement and promote activities without triggering pain symptoms.

Finally the repercussions of this chronic knee extensor muscles weakness could later induce knee pain. Citaker et al. [49] found a direct relationship between quadriceps strength and patellofemoral pain . Caetano et al. [50] showed that a weakness of the quadriceps may lead to increased fall risk with aging. Therefore, it seems imperative to improve rehabilitation strategies to better target this lingering weakness.

6MWT

We showed a significant decrease in the walking distance between the 2 populations. This demonstrates a decrease in walking activity for CRPS patients. This parameter is highly correlated with the strength of the thigh. Impaired gait adaptability, particularly stepping errors and reduced gait speed, was associated with high risk of falls; reduced executive function, increased concern about falling and weaker quadriceps strength contributed to this relationship [50].

We think that the relationship observed between quadriceps muscle force production and performance during gait should not be ignored but self-selected walking speed is influenced not only by lower limb strength but also by balance, reaction time, vision, pain and emotional well-being [51].

As other has noted [45,46,52] patients may remain far from their premorbid health state.

Questionnaire

Pain and Kinesiophobia

Our results demonstrated after a follow-up of 3.8 years that the level of pain, severity of CRPS and kinesiophobia continue to be high. Vlayen et al. [53] considers a score of 40/68 to be a significant level of kinesiophobia. Pain-related fear of movement has been identified as an important factor contributing to the maintenance of chronic pain [29]. The kinesiophobia score evaluated by the TSK in our sample was in average of 45 points out of 68 (ranging from 25 to 63). These results correspond to those of de Jong et al. [54] who obtained an average of 48 (20 to 63) for patients with acute CRPS (1 to 6 months) and an average of 38 (19 to 59) for patients with chronic CRPS (approximately 8 years), for lower and upper extremities combined. Marinus et al. [55] obtained a mean score of 38 for patients with chronic lower limb CRPS for 1.6 years. The TSK score of our patients was not correlated with their pain. However, Moseley et al. [56] showed for upper limb CRPS that pain was associated with a higher TSK score. We expected an association between pain and fear of movement based on the fact that patients no longer use their limb because of pain, which in turn might lead to deconditioning and reduced sensitivity. However, we did not observe a significant correlation between TSK and patient performance such as muscular strength and walking distance. These results could be explained by the fact that the patients included in the present study kept on walking without a cane and that disuse of the affected limb was less present than with upper limb injury. de Jong et al. [54] suggested that TSK was not a reliable predictor of functional limitations for CRPS because it is not specific enough for the type of activity or movements feared or avoided. Some studies [29,57,58] demonstrated a significant association between functional disability and TSK score in patients with chronic low back pain. In a population of fibromyalgia patients, Roelofs et al. [29] reported an average of TSK score of 28.2, which was also associated with physical performance. Nevertheless Ramond et al. [59] stated that half the existing studies on this topic did not demonstrate this relationship. However, in the study of de Jong et al. [54] the average of TSK scores in CRPS subjects were not significantly different from those of patients with chronic low back pain for whom TSK is associated with the level of disability. They maintained that the TSK would not be suitable for neuropathic pain such as CRPS type 1.

CRPS Severity Score

Other studies [12,16] assessing pain and severity scores confirmed that pain remained present in the long term showed that CRPS patients consider themselves recovered when they were relieved from local or generalized pain and discomfort. For the majority, CRPS resolves within a year, but prospective studies have indicated severe pain remaining in 13% of the patients at one year or more after the diagnosis [59]. Summary of longer term retrospective review reported the persistence of symptoms in 22% to 64% of patients three years or more after diagnosis [16].

The Short Form-36

Our patients were more affected by the physical complaints than psychological components. The literature remains unclear about the fact that altered psychological functioning is a specific psychological profile characteristic of CRPS patients rather than a natural outcome of chronic pain [61]. Our results showed that pain, strength and the walking distance are directly linked to the physical recovery. Llewellyn et al. [12] showed altered subjective and objective outcomes (pain, physical performance, quality of life and energy/fatigue) in patients suffering from lower limb CRPS, than those with CRPS of the upper limb. Van Welzen et al. [62] demonstrated a lower self-reported quality of live in the physical domain of the Short Form-36. Unremitting symptoms in CRPS are associated with long-term disability, poor psychological health and reduced quality of life [61,63,64].

Limitations and future directions

Since we know that traumas such as sprains or fractures represent most of the traumas that trigger CRPS and are responsible for alterations in proprioception and balance [65], it seems crucial to complement our observations with further studies comparing our results with a population having presented an ankle trauma that did not complicate in CRPS in order to define more precisely the consequences of CRPS. Considering the design of our study, additional interpretations on the causes and consequences of the deficits we observed might be too speculative. Finally, the comparison with the literature is limited by the preponderance of studies on the upper limb. Therefore, further investigations appear crucial to deepen our knowledge on the functional consequences of CRPS affecting the lower limb.

Conclusions

This study showed persistent functional disability and significant long-term pain, which have significant repercussions on the quality of life of patients suffering from CRPS 1 at the foot level. This syndrome has also an impact on strength and proprioceptive abilities in the adjacent joint to the affected one. These observations highlight the importance of a therapeutic approach that focuses not only on the affected joint but on the whole leg concerned as well as the contralateral side in order to improve the quality of life of the patient and avoid the long term consequences that these deficits could generate.

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Tables

Table 1. Population characteristic

	CRPS	Control
Gender ♀/♂ (%)	85/15	85/15
Age (years)	46.3±12.7	45.9±13.3
Height (cm)	165.9±7.1	165.3±7.2
Weight (kg)	73.7±18.1	70.9±18.1
fractures/sprains (%)	60/40	-----
follow-up (years)	3.8±2.2	-----

♀: female, ♂: male. SD: standard deviation; CRPS : Complex Regional Pain Syndrome. Data are presented as mean ± SD unless otherwise indicated.

Table 2 : Demographic and clinical characteristics of individual patients suffering from CRPS.

Patient	Age	Gender	CRPS duration (years)	Initial traumatism	Orthopaedic treatment	Affected foot
1	76	♀	5	ankle sprain LAL lesion	immobilisation	R
2	38	♀	6	ankle sprain LAL lesion	immobilisation	R
3	53	♀	2	V metatarsal fracture	immobilisation	L
4	56	♀	4	II metatarsal stress fracture	immobilisation	L
5	26	♀	4	ankle sprain, LAL lesion	immobilisation	L
6	57	♀	8	bimalleolar fracture	immobilisation	R
7	66	♀	3	Medial malleolar fracture	immobilisation	L
8	37	♂	2	ankle sprain LAL lesion	immobilisation	L
9	54	♂	6	bimalleolar fracture	surgery+ immobilisation	L
10	50	♀	2	V metatarsal fracture	immobilisation	L
11	45	♀	2	II-III metatarsal fracture	immobilisation	R
12	32	♀	6	calcaneum fracture	immobilisation	R
13	43	♀	5	IV-V metatarsal fracture	immobilisation	L
14	40	♀	3	ankle sprain, LAL lesion	immobilisation	L
15	58	♀	2	talus fracture	surgery+ immobilisation	L
16	45	♀	8	calcaneum fracture	surgery+ immobilisation	L
17	22	♀	4	talus fracture	surgery+ immobilisation	R
18	53	♀	2	Lisfranc sprain	immobilisation	R
19	35	♂	1	ankle sprain LAL lesion	immobilisation	R
20	40	♀	1	ankle sprain LAL lesion	immobilisation	L

Legends : ♀ : woman, ♂ : man, R : right, L : left, LAL : Lateral Ankle Ligament

Figures

Figure 1. Initial position for knee joint position sense assessment

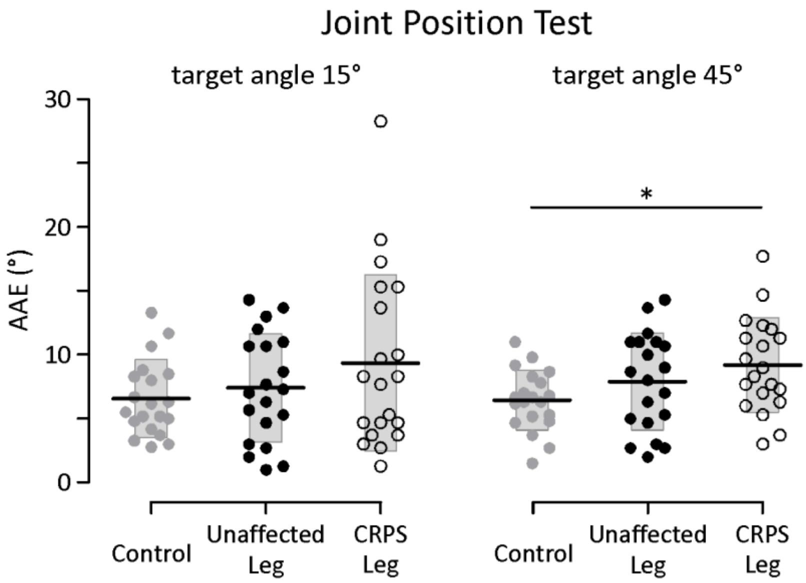

Figure 2. Individual Absolute Angle Error (AAE) for the Joint Position Test at 15° and 45° target angles in the control group (average of both legs) and in the CRPS group for the unaffected and CRPS leg. Box plots represent the group-level average ± standard deviation. Note the significant difference between the AEE obtained for the CRPS leg and those obtained in the controls. *p<0.05

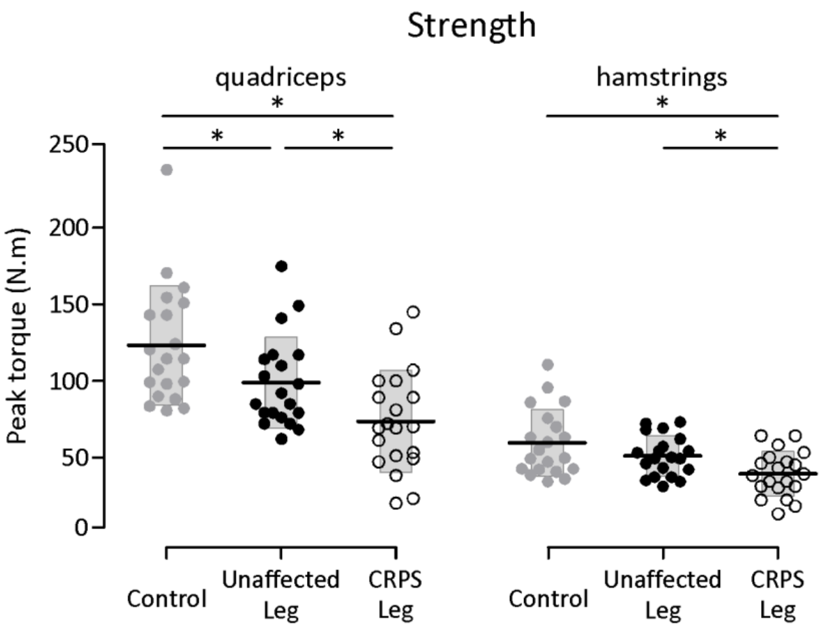

Figure 3. Individual peak torque of the quadriceps and hamstrings (isokinetic strength test at velocity 60°/s) for the control group and the unaffected leg and CRPS leg of the CRPS group. Box plots represent the group-level average ± standard deviation. *p<0.05

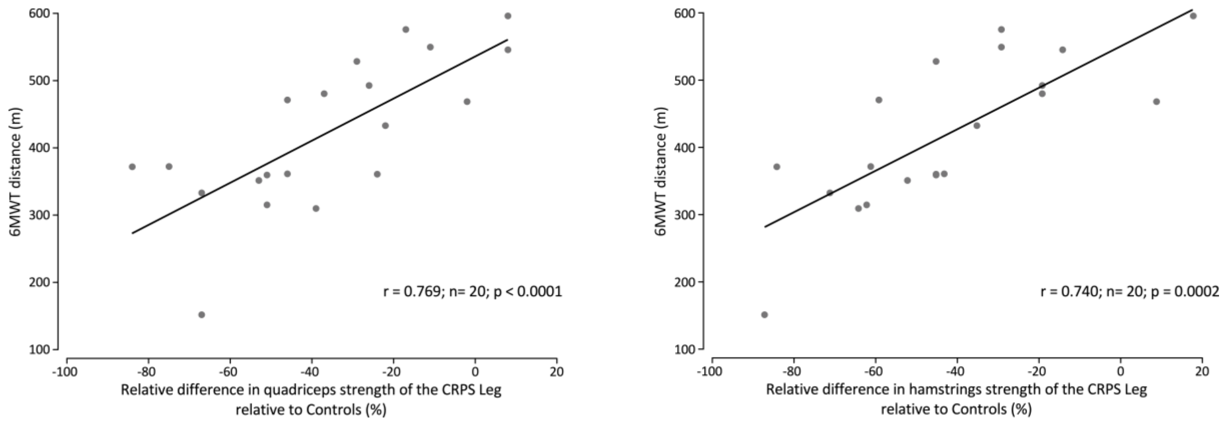

Figure 4. Linear regression between the distances (meters) covered during the 6MWT in the CRPS group and the relative difference in strength of the CRPS Leg (%) for the quadriceps and the hamstrings as compared to the control group. Negative values represent a deficit in strength at the CRPS Leg as compared to controls.