Resilience in patients with amputation because of Complex Regional Pain Syndrome type I

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Abstract

Purpose: Although controversial, an amputation for longstanding and therapy-resistant Complex Regional Pain Syndrome Type I (CRPS-I) may improve quality of life and pain intensity. Resilience, the way people deal with adversity in a positive way may be related to these positive outcomes. This study focused on the relationship between resilience and post-amputation outcomes, i.e. quality of life, pain and recurrence of CRPS-I and psychological distress. Method: Twenty-six patients with an amputation related to CRPS-I filled in the Connor-Davidson Resilience Scale (CD-RISC), World Health Organisation – Quality of life Assessment (WHOQOL-Bref) and the Symptom Checklist-90 Revised (SCL-90-R). An interview was conducted and a physical examination performed. Results were compared with reference groups from literature and a control group from the outpatient rehabilitation clinic at our medical center. Results: Resilience correlated significantly with all domains of the WHOQOL-Bref ($r$ ranged from 0.41 to 0.72) and negatively with all domains of the SCL-90-R ($r$ ranged from $-0.39$ to $-0.68$). Patients with an amputation because of CRPS-I have higher scores on resilience and quality of life than the control group. Resilience was lower in patients who reported CRPS-I symptoms compared to those who did not. Conclusions: The results confirmed our hypothesis that patients with an amputation because of CRPS-I who have a higher resilience also have a higher quality of life and experience lower psychological distress. The prognostic value of resilience in this patient group requires further research.

Keywords

Amputation, complex regional pain syndrome, prognostic relevance, resilience

History

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Introduction

Pain and swelling following a seemingly minor injury of wrist or ankle, do not recover in some patients within a normal timeframe. When pain intensifies and other symptoms occur and worsen (e.g. changes in sweating, color or nail and hair growth) Complex Regional Pain Syndrome type I (CRPS-I) is likely to be present [1].

Guidelines offer evidence based treatment options for CRPS-I such as medication, physical therapy and occupational therapy [2]. However, not all patients respond to these therapies and in some patients CRPS-I may further develop into a dysfunctional limb with uncontrollable pain or life-threatening infection [3–5]. Sometimes a patient requests an amputation of the affected limb as a last resort [6,7].

Amputation for longstanding and therapy-resistant CRPS-I is controversial and a rare intervention [8]. Primarily, the aim of the amputation is to increase quality of life and mobility of the patient but also to decrease pain intensity. Outcome variables after an amputation such as quality of life have been infrequently reported [8]. Previously, there was insufficient evidence that amputation positively contributes to the treatment of CRPS-I, with just a few published case studies with positive outcomes [2,7,9,10]. Guidelines warn against amputation because of the risk of recurrence of the syndrome due to their referral to one or two larger studies with predominantly negative outcomes [5,11–13]. A systematic review on CRPS-I and amputation could not find enough evidence for or against amputation [8]. Results from our
recent study in a group of 21 patients who had an amputation because of longstanding and therapy-resistant CRPS-I did show an overall improvement of life in general and improvements in pain intensity, quality of life, mobility, use of a prosthesis and job or study enrolment [6]. It is unknown why patients from this study have better results than most other patients described in literature [6,8]. Patients faced physical disability and severe pain often several years prior to the amputation. After the amputation they seem to “bounce back” beyond what could be expected according to literature.

The ability to bounce back in times of adversity, including physical stress, is called resilience [14]. Resilience is defined as “the process of adapting well in the face of adversity, trauma, tragedy, threats or even significant sources of stress – such as family and relationship problems, serious health problems or workplace and financial stressors” [15]. It represents a person’s qualities that enable that person to thrive in the face of adversity [16]. In patients with traumatic amputations psychological recovery and acceptance of limb loss were positively influenced, not only by social support or medical care, but also by higher resilience [17]. Resilience may, in part, explain why patients are able to increase quality of life after amputation. Insight in resilience of patients with a limb amputation because of CRPS-I could guide patient selection and give reason for offering patients a program to increase their resilience before and after amputation.

Based on the results from our previous study on quality of life, we hypothesized that higher scores on quality of life and participation in daily life may be correlated with higher resilience. The aim of this study was to analyze resilience and post amputation outcome (CRPS-I symptoms, quality of life, psychological distress and participation in daily life) and to analyze how resilience relates to these outcome variables in patients with an amputation because of CRPS-I.

**Methods**

**Participants and procedures**

Patients with a request for amputation were referred to our outpatient clinic by their consultant in rehabilitation medicine, their general practitioner, or they came on their own initiative. Our rehabilitation medicine outpatient clinic is situated in a university-based medical center which serves as one of the referral clinics for people with longstanding and therapy-resistant CRPS-I in our country. Upon referral the patient was independently assessed by a consultant in rehabilitation medicine, vascular surgeon, physical therapist and psychiatrist or psychologist. CRPS-I was diagnosed according to the criteria of the International Association for the Study of Pain (IASP) and the criteria of Bruehl [18,19]. Patients were considered eligible for amputation if other diagnoses were ruled out, if (all) therapies for CRPS-I advised in guidelines were tried but failed (including infection and wound therapy), if quality of life was experienced as poor and participation in daily life activities was hindered excessively. In a multidisciplinary meeting, the health care professionals discussed the pros and cons of an amputation together, and then later discussed these with the patient.

All patients (n=27) who underwent elective amputation because of CRPS-I at our centre between 2000 and 2011 were contacted to participate in this cross-sectional explorative study. After agreement on participation, patients were sent information about the study, questionnaires and an informed consent form. Patients with insufficient knowledge of the Dutch language or younger than 18 years were excluded from the study. The study included several questionnaires, a semi-structured interview and a physical examination. The medical ethical committee approved the research (METc 2009/117).

**Questionnaires**

Resilience was assessed with a Dutch version of the Connor-Davidson Resilience Scale (CD-RISC), a 25-item self-report measure that was developed to quantify current resilience [16,20]. The score ranges from 0 to 100 with higher scores indicating a better resilience.

Quality of life was evaluated with the World Health Organization – Quality of life Assessment (WHOQOL-Bref), a 26-item questionnaire covering four domains: physical health, psychological health, social relationships and environment [21]. The scores range in each domain from 4 to 20; higher scores indicate better quality of life in a certain domain. The results of the WHOQOL-Bref of 21 patients included in this study have been described previously [6].

Psychological distress was assessed with the Symptom Checklist-90 Revised (SCL-90-R) [22]. The SCL-90-R assesses self-reported psychological distress and multiple aspects of psychopathology. It consists of 90 questions in eight dimensions of psychological distress: anxiety, agoraphobia, depression, somatisation, insufficiency, sensitivity, hostility and insomnia. Patients report to which extent the symptoms of the checklist were present in the week preceding the completion of the questionnaire. Higher scores in the SCL-90-R indicate more problems. It can be used with single dimensions but also as a total psycho-neuroticism. All questionnaires have five point Likert scales, scoring from 0 to 4 (CD-RISC) or 1 to 5 (WHOQOL-Bref and SCL-90-R).

**Interview and physical examination**

A visit to the patient for an interview and physical examination by a psychologist and a physician was scheduled in a hospital close to or at the patient’s home. Main results from these interviews have been published [6]. Patients were asked if they still experienced CRPS-I related symptoms, stump pain and phantom pain in the two weeks before the visit. Stump pain and phantom pain were recorded on a visual analogue scale (VAS) in millimetres (mm).

After the interview, the physician performed a physical examination of the limbs for (recurrence of) CRPS-I [19] and the psychologist checked all questionnaires for missing answers and asked patients to fill in the missing answers.

**Analysis**

The results of the CD-RISC and WHOQOL-Bref questionnaires were compared with results from a control group from our outpatient rehabilitation clinic. The control group exists of chronic pain patients selected from patients seen by the psychologist from our rehabilitation clinic between 2008 and 2013 (n=111; male 34%, mean age 45.9 years SD 13.4 years, female 66%, mean age 40.0 years SD 13.2 years). Patients in this control group experienced chronic pain (>6 weeks) and social and psychological factors played a considerable role in maintaining the health related complaints.

The results of the CD-RISC were also compared with those of a non help-seeking general population sample (n=577) and primary care outpatients (n=139) in the United States of America [16]. WHOQOL-Bref scores were additionally compared with scores found in the general Dutch population (n=218, male 41%, mean age 37.5 years SD 7.6, female 59%, mean age 37.4 SD 8.2) [23]. SCL-90-R scores were compared with norm values for the Dutch population (n=2394, male:female 50%:50%, mean age 41.1 years SD 14.5) and for patients with chronic pain (n=2461, male:female 32%:68%, mean age 46.2 years SD 15.4) [22]. Comparisons were made using Confidence Interval Analysis (CIA 2.2.0 University of Southampton) [24]. Associations between
resilience and the other outcome variables were analyzed. Non-parametric correlations (Spearman’s ρ) and Mann–Whitney U tests were used.

PASW Statistics version 18 for Windows was used for data analysis. Results are significant at $p \leq 0.05$.

Results

Patient characteristics

Of the 27 contacted patients, 26 agreed to participate: 23 women and three men, median age 44 years (Interquartile range (IQR): 34; 48). Patients underwent amputation between May 2000 and May 2010. Median duration of CRPS-I was 5.5 years (IQR: 3; 10). Median interval between amputation and study was 56 months (IQR: 25; 69). Twenty patients underwent amputation of a lower extremity (LE) and six patients of an upper extremity (UE). No patients were excluded. Previous failed therapies included combinations of, e.g. physical therapy including pain exposure physical therapy [25], occupational therapy, manipulation, sympathetic blocks or sympathectomy, medication such as morphine, anti-anxiety agents and dimethylsulfoxide cream (50%) [12]. Before amputation, patients generally experienced their quality of life as poor and often referred to their affected limb as “paw”, “canon” or “obstacle”.

Measures

CD-RISC

The mean CD-RISC was significantly higher than that of the control group at our outpatient rehabilitation clinic (Table 1). CD-RISC scores were significantly lower compared to values for a USA non help-seeking general population sample and similar to patients seeking primary care (Table 1) [16].

WHOQOL-Bref

Sixteen patients (62%) reported a good or very good quality of life; four patients (15%) reported good nor bad and six patients (23%) reported a poor or very poor quality of life. Patients scored significantly higher (=better) on the physical and psychosocial domain compared to patients in our control group (Table 1). Patients scored significantly lower on the physical and environmental domain compared to Dutch norm values.

Table 1. Mean and standard deviation for CD-RISC and WHOQOL-Bref domain scores of patients who had a limb amputation because of longstanding therapy-resistant Complex Regional Pain Syndrome type I (CRPS-I) compared to reference and control groups.

<table>
<thead>
<tr>
<th></th>
<th>CRPS-I</th>
<th>Reference and control groups</th>
<th>Difference (95% CI)</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>CD-RISC</strong></td>
<td>73.3 (11.7)</td>
<td>Non help seeking [16]</td>
<td>7.1 (2.1; 12.1)*</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Primary care [16]</td>
<td>71.8 (18.4)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Outpatient rehabilitation clinic</td>
<td>60.2 (12.3)</td>
</tr>
<tr>
<td><strong>WHOQOL-Bref Domains</strong></td>
<td></td>
<td>Dutch norm values [23]</td>
<td></td>
</tr>
<tr>
<td>Physical</td>
<td>12.7 (3.6)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Psychosocial</td>
<td>14.4 (2.7)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Social</td>
<td>15.1 (3.7)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Environment</td>
<td>13.9 (2.8)</td>
<td></td>
<td></td>
</tr>
<tr>
<td></td>
<td></td>
<td>Outpatient rehabilitation clinic</td>
<td>9.8 (2.4)</td>
</tr>
<tr>
<td>Physical</td>
<td>12.8 (2.3)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Psychosocial</td>
<td>13.7 (3.5)</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Social</td>
<td>13.6 (2.2)</td>
<td></td>
<td></td>
</tr>
</tbody>
</table>

CD-RISC: Connor-Davidson Resilience Scale; reference values taken from Development of a new resilience scale: the Connor-Davidson Resilience Scale (CD-RISC) [16]. WHOQOL-Bref: World Health Organization Quality of Life-BREF questionnaire; reference values taken from Quality of life and psychopathology: Investigations into their relationship [23]. Control group: outpatient rehabilitation clinic: results from patients with chronic pain (>6 weeks duration). CI: Confidence interval; *$p \leq 0.05$. 

SCL-90-R

Patients scored significantly higher (=worse) on depression, somatization, insomnia and psycho neuroticism compared to the Dutch norm values (Table 2) [22]. However, they scored similar to Dutch norm values for chronic pain patients [22].

Interview and physical examination

Fifteen patients (56%) reported recurrence of CRPS-I-like symptoms. Twenty-three patients (88%) reported stump pain (median VAS score 31 mm; IQR: 6; 63) and 20 patients (77%) reported phantom pain (median VAS score 25 mm; IQR: 2; 51). Five patients (19%) met Bruehl’s criteria [19] for recurrence of the syndrome in the stump and two patients (8%) for recurrence in another limb.

Associations

The CD-RISC correlated positively with all domains of the WHOQOL-Bref ($p$ ranged from 0.41 to 0.72) and negatively with all domains of the SCL-90-R ($p$ ranged from −0.39 to −0.68) (Table 3). A positive, though not significant association ($p = 0.457$, $p = 0.065$) was found between CD-RISC score and frequency of prosthesis use for patients with a prosthesis ($n = 17$).

CD-RISC scores in patients who did not report persistence of CRPS-I related symptoms ($n = 11$) (median: 81, IQR: 76; 83) was higher compared to patients who did report these symptoms ($n = 15$) (median: 71, IQR: 64; 78) (Mann–Whitney $U$: $p = 0.032$). CD-RISC scores were significantly lower in patients reporting more stump pain ($p = 0.508$, $p = 0.008$). For phantom pain such an association was not found ($p = 0.297$, $p = 0.14$). CD-RISC scores did not differ significantly between patients with or without objectified recurrence of CRPS-I (Mann–Whitney $U$: $p = 0.53$).

Discussion

This research focused on resilience (the ability to bounce back from adversity) in a group of patients with an amputation because of longstanding therapy-resistant CRPS-I. Resilience is an interactive concept concerning the combination of serious risk experiences and a relatively positive psychological outcome
events have a higher chance of developing CRPS-I [30]. Amputation because of CRPS-I is controversial due to clinicians’ opinions on the negative outcome. Literature on amputation because of CRPS-I also focuses on reasons (risk factors) for amputation [8]. Case studies on amputation due to longstanding therapy-resistant CRPS-I are characterized by predominantly negative reporting on topics such as pain, quality of life, mobility and use of a prosthesis [8]. Recurrence of the syndrome underlies most opinions about not to amputate in case of longstanding therapy-resistant CRPS-I. However, recurrence is often not (clearly) described in those case reports [8]. Our clinical experience with these patients led us to believe in a more positive outcome after amputation regarding quality of life [6]. Shifting the focus of research from identification of risk factors to this more positive approach on patients’ competencies and strengths, offers a new perspective.

We are aware of the limitations of this study. Clinical relevance of differences in CD-RISC scores is not yet clear. A 7-point difference between our group and a non-help seeking population on a 0–100 scale (in which the upper and lower boundaries never occur) seems to be meaningful (Table 1). Another limitation is that we do not have pre- and post-test measurements. This is also applicable for the results of the control group with chronic pain. Patients from this control group seek medical care for their (pain) problem, which is not necessarily the case for the CRPS-I and amputation population. Measurements presented from this control group are scores at the beginning or during the rehabilitation process and not after the rehabilitation process which makes comparing the results difficult. We do believe that this control group is more or less comparable to our CRPS-I population since both groups have been dealing with pain for a longer period.

Several explanations for relatively high resilience scores can be thought of. First, the high resilience scores in our study may be related to patient selection. It is possible that the specialists who made the decision to amputate unknowingly selected patients on the basis of resilience; the patient’s previous ability to bounce back from adversity. According to this explanation our patients were more likely to have better outcome than could be expected based on literature. Whether this phenomenon occurred is unclear since we have no information about the patients who were denied amputation. It may also be that only the most resilient patients with CRPS-I do not give up on looking for a solution in the face of repeated treatment failures. Another explanation for relatively high scores on questionnaires in general for this specific population years after amputation could be a phenomenon (clearly) described in those case reports [8]. Our clinical experience with these patients led us to believe in a more positive outcome after amputation regarding quality of life [6]. Shifting the focus of research from identification of risk factors to this more positive approach on patients’ competencies and strengths, offers a new perspective.

Table 3. Correlations between CD-RISC and WHOQOL-Bref scores and between CD-RISC and SCL-90-R in patients with amputation because of longstanding therapy-resistant CRPS-I.

<table>
<thead>
<tr>
<th></th>
<th>Correlation coefficient</th>
<th>p</th>
</tr>
</thead>
<tbody>
<tr>
<td>WHO-QOL-Bref$^{21}$</td>
<td>0.549</td>
<td>0.004</td>
</tr>
<tr>
<td>Physical</td>
<td>0.454</td>
<td>0.020</td>
</tr>
<tr>
<td>Psychosocial</td>
<td>0.721</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Social</td>
<td>0.448</td>
<td>0.022</td>
</tr>
<tr>
<td>Environmental</td>
<td>0.407</td>
<td>0.039</td>
</tr>
<tr>
<td>SCL-90-R$^{22}$</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Anxiety</td>
<td>−0.586</td>
<td>0.002</td>
</tr>
<tr>
<td>Agoraphobia</td>
<td>−0.405</td>
<td>0.040</td>
</tr>
<tr>
<td>Depression</td>
<td>−0.680</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Somatization</td>
<td>−0.439</td>
<td>0.025</td>
</tr>
<tr>
<td>Insufficiency</td>
<td>−0.543</td>
<td>0.004</td>
</tr>
<tr>
<td>Sensitivity</td>
<td>−0.539</td>
<td>0.005</td>
</tr>
<tr>
<td>Hostility</td>
<td>−0.660</td>
<td>&lt;0.001</td>
</tr>
<tr>
<td>Insomnia</td>
<td>−0.391</td>
<td>0.048</td>
</tr>
<tr>
<td>Psychoneuroticism</td>
<td>−0.668</td>
<td>&lt;0.001</td>
</tr>
</tbody>
</table>

WHOQOL-Bref: World Health Organization Quality of Life-Bref questionnaire. Resilience was measured with Connor-Davidson Resilience Scale (CD-RISC). Correlation Coefficient: between CD-RISC and SCL-90-R or CD-RISC and WHOQOL-Bref scores, calculated with Spearman’s Rho.

despite those experiences [26]. Higher resilience is positively related to better physical functioning, higher quality of life and lower pain scores among patients with chronic conditions [27–29].

In a previous publication, we showed relatively high quality of life scores in a group of patients with amputation due to longstanding therapy-resistant CRPS-I [6]. Based on the findings in literature and the results of our study [6], we hypothesized that patients with a CRPS-I related amputation who have relatively good results also score high on resilience. We found a positive association between resilience and quality of life, especially within the psychosocial domain. Despite living with CRPS-I for many years and experiencing an amputation, scores on the psychosocial domain are significantly better than patients with chronic pain who visit the psychologist at a rehabilitation outpatient clinic and similar to Dutch norm values [20]. Even on the physical domain they score significantly better than the chronic pain patients.

The focus of most previous research on CRPS-I has been on risk factors. With an unknown cause of the CRPS-I, it is frequently assumed that psychological factors play an important role in the development of the syndrome. However, a systematic review showed that life events appear to be the only factor related to the development of CRPS-I; patients who experience more life events have a higher chance of developing CRPS-I [30].
Another factor that should be considered in explaining our results is the cognition of the patients. It is not unreasonable to assume that patients respond positively to their ‘last resort’; an amputation of their limb affected by longstanding therapy-resistant CRPS-I. Additionally, patients may feel understood or feel that their problems are being taken seriously when, at last a team of medical specialists is found willing to deliberate amputation. Although the mechanism is poorly understood, the positive effect of clinician–patient communication on outcomes has been found repeatedly in other pathologies [33].

Another explanation of the score may lie in the intervening period between amputation and our study. Life experiences between these two points may also have given a raise in resilience scores and accounts for one of the limitations of this study.

Finally, cognitive dissonance could explain the relatively good results. Cognitive dissonance is the discomfort caused by holding conflicting cognitions. Based on that theory, the patient will try to minimize regret of their irrevocable choice [34]. These explanations should be taken into account in future research in this field.

The domain scores of the SCL-90-R correlated negatively with resilience. These findings indicate that participants with a better resilience experience less psychological distress which is in line with our hypothesis. This negative correlation between resilience and psychological distress was found previously in women with fertility problems [35]. Not all associations were in line with our hypothesis. We expected that patients with a higher resilience score would improve in a larger number of topics. However, the association between resilience and the amount of topics patients improved upon was weak and not significant. Another ‘logical’ hypothesis would be that those patients with a higher resilience score would use their prosthesis more often. The association between resilience and frequency of prosthesis use was not significant either ($p = 0.065$). This lack of significance could be attributed to lack of power due to the small sample size. However, it is very well possible that resilient patients find ways of participating without the use of a prosthesis.

The direction of the association between resilience and quality of life remains unclear because of the study design. It is possible that the relatively good results encourage the patients to feel resilient rather than resilience leading to better results and the competency to restore parts of life. Programs for improving resilience are currently being developed and studied for effectiveness. The results of these programs substantiate that training can improve resilience [36]. Resiliency training may indirectly lead to improvement in quality of life [37,38]. When patients ask for an amputation for their therapy-resistant CRPS-I a training to improve resilience prior to the amputation might be considered.

Medical care is known to influence a patient’s quality of life, therefore rehabilitation after amputation plays an important role in the final results. Rehabilitation in our patient group, however, took place near patients’ homes in different centres for rehabilitation in all parts of the country. Therefore, we cannot estimate the effect of it on the outcome. Despite our relatively positive results, amputation for CRPS-I remains controversial. Screening for psychopathology and assessment of resilience should be performed prior to amputation.

We think that resilience might be a key factor in helping patients to accept and adapt to their new situation. Longitudinal studies are needed to analyze the strength of resilience over time and to analyze its prognostic value. Exploring competencies offers a new perspective on why some patients report positive outcomes after amputation. We conclude that the results of this explorative study confirm our hypotheses.

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Declaration of interest

All authors report no conflicts of interest.

References

20. Giesbrecht T, Abidi K, Smeets T, et al. Adversity does not always explain the relatively good results. Cognitive dissonance could explain the relatively good results. Cognitive dissonance is the discomfort caused by holding conflicting cognitions. Based on that theory, the patient will try to minimize regret of their irrevocable choice [34]. These explanations should be taken into account in future research in this field.

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