

Dystonia in complex regional pain syndrome : clinical, pathophysiological and therapeutic aspects Rijn, M.A. van

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Psychological Features of Patients with Complex Regional Pain Syndrome Related Dystonia

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Abstract

The objective of this study was to evaluate psychological features in severely affected patients with Complex Regional Pain Syndrome (CRPS) related dystonia. Personality traits, psychopathology, dissociative experiences, the number of traumatic experiences, and quality of life were studied in 46 patients. Findings were compared to two historical psychiatric control groups (54 patients with conversion disorder (CD) and 50 patients with affective disorders (AD)) and normative population data. The CRPS patients showed elevated scores on the measures for somatoform dissociation, traumatic experiences, general psychopathology and lower scores on quality of life compared to general population data, but had significantly lower total scores on the measures for personality traits, recent life events and general psychopathology compared to the CD and AD patients. Rates of early traumatic experiences were comparable to the CD and AD patients, and the level of somatoform dissociation was comparable to the CD patients but was elevated in comparison to the AD patients. Early traumatic experiences were reported in 87% of the CRPS patients and were found to be moderately related to somatoform dissociative experiences, indicating that early traumatic experiences might be a predisposing, though not necessary factor for the development of CRPS related dystonia. Although the psychological profile of the patients with CRPS related dystonia shows some elevations, there does not seem to be a unique disturbed psychological profile on a group level.

Introduction

Complex Regional Pain Syndrome (CRPS), formerly known as reflex sympathetic dystrophy (RSD) or Sudeck syndrome, 1 is commonly preceded by a minor to severe trauma to an extremity and occurs more frequently in women. 2 CRPS is characterized by various combinations of sensory, autonomic and trophic features, in absence of any evident nerve lesions. 1.2 Compelling evidence indicates that patients with CRPS may develop movement disorders (MDs), which may occur early in the disease course but generally tend to occur with a variable delay. 2-5 Fixed dystonia is among the most common MDs in CRPS and may spread to other extremities. 6

Although compelling evidence suggests a role for disinhibition of spinal and supraspinal neuronal circuits in dystonia of CRPS,^{5,7,8} the nature of CRPS and its associated MDs has since long been subject of debate. While some consider CRPS a somatic disorder,^{9,10} others have suggested that the MDs and other features of the disorder are psychogenic.^{11,12} From the latter perspective the symptoms of CRPS could be interpreted as a conversion reaction¹³ or malingering.¹⁴ Affective and anxiety disorders are often noted among CRPS patients,^{15,16} and similarities between CRPS and conversion disorder (CD) have also been documented.¹⁷ Many clinicians who treat CRPS patients feel there is a psychological aspect to the syndrome, either primary, as predisposing personality traits and premorbid psychiatric disorders, or secondary, as a result of the pain and disabilities.¹³ A specific CRPS-personality has been suggested in the literature,¹² but has not been confirmed.^{1,13} Studies of personality profiles and preand co-morbid psychiatric disorders in CRPS have primarily focused on patients with acute CRPS, and have yielded conflicting results.^{13,15,16}

To obtain more insight in the psychological features of patients with CRPS related dystonia, we assessed a chronic group of severely affected patients and compared them to two historical psychiatric control groups. Personality traits, psychiatric co-morbidity, dissociative experiences, and the number of traumatic experiences were studied. In addition, the experienced quality of life was assessed.

Methods

Patients

The department of Neurology of the Leiden University Medical Center (LUMC) is a national referral center for patients with CRPS related movement disorders. CRPS patients that suffer dystonia in at least one extremity are offered to participate in a trial which aims to evaluate the efficacy and safety of intrathecal administration of baclofen,

which is a specific γ -amino butyric acid (GABA) receptor agonist. Baclofen inhibits sensory input to the spinal cord,18 reducing muscle tone and stiffness. A total of 46 patients who participated in a screening for responsiveness to intrathecal baclofen between July 2003 and September 2005 were included in this study. All patients met the International Association for the Study of Pain (IASP) criteria of CRPS¹⁹ for their first affected extremity. Blood tests, nerve conduction and imaging studies of the spinal cord and brain were used to rule out other causes of dystonia. Dystonia generally affected the distal limb and was characterized by flexion postures. Medication used at the time of the screening falls into three categories: antidepressants (Selective Serotonin Re-uptake Inhibitors (used by 9% of the patients) and Tricyclic antidepressants (TCA) (15%), though the TCA's were mainly used as pain medication), muscle relaxing agents (baclofen (35%) and benzodiazepines (41%)), and pain medication (anticonvulsant drugs (15%), acetaminophen or NSAIDs (24%) and opiods (30%)). This study was approved by the medical ethical committee of the LUMC, and the patients gave their informed consent. Psychological features of the CRPS patients were assessed by means of self-report instruments before they entered the baclofen trial.

Two historical control groups were used which included 54 patients with CD and 50 patients with at least one affective disorder (AD). Both control groups were part of a previous study on the involvement of emotional traumas and dissociative features of CD.²⁰ Data collection of this study took place from 1997 until 2000 and all patients were seen by a psychiatrist to determine whether the diagnosis of CD or AD as stated in the fourth edition of the Diagnostic and Statistical Manual of Mental Disorders (DSM-IV)²¹ was applicable. The physical examination in the CD patients was performed by a neurologist. All patients gave their informed consent (for more detail, see the study of Roelofs et al.²⁰).

Instruments

The Personality Diagnostic Questionnaire-Revised (PDQ-R)²² can be used for screening for DSM-III-R²³ personality disorders. The PDQ-R consists of 133 true/false items. A high total score indicates severe personality pathology.

The *Traumatic Experiences Checklist* (TEC)²⁴ is a 25-item questionnaire that evaluates the presence or absence of emotional life events. Among these are emotional neglect, physical abuse, sexual harassment, sexual abuse, parentification and life threatening experiences.

The Dutch *Recent Life Event Questionnaire* ("Vragenlijst Recent Meegemaakte Gebeurtenissen", VRMG)²⁵ measures the perception of recent life events. The original instrument consists of 115 items, but in the current study a shortened version of the VRMG was used.²⁶ The addressed categories include health, pregnancy/birth, work,

relationships, and "other". For each event, patients had to indicate whether it had occurred in the twelve months preceding the symptom onset.

The *Dissociative Experiences Scale* (DES)²⁷ is a 28-item self-report questionnaire which assesses the frequency of various psychological or psychoform dissociative symptoms. The mean of all item scores ranges from 0 to 100 and is called the DES score. High DES scores indicate severe psychoform dissociative problems. The DES addresses disturbances in memory, awareness, identity and cognition, and feelings of depersonalization and derealisation.²⁷

To measure somatoform dissociation, the *Somatoform Dissociation Questionnaire-20* (SDQ-20) was used. The SDQ-20 consists of 20 items rated on a five-point scale with a total score range of 20-100. The items address medically unexplained analgesia, anesthesia, motor disturbances, alternating preferences for tastes and smells, pain, and loss of consciousness. A high total score is an indicator for many somatoform dissociative experiences.²⁸

To screen for the general level of psychopathology we used the *Symptom Checklist-90-Revised* (SCL-90-R),²⁹ consisting of 90 items rated on a five-point scale with a total score range of 90-450. High total scores indicate high levels of psychopathology.

The Research and Development-36 (RAND-36)³⁰ was administered to measure quality of life. The questionnaire consists of 36 items and assesses physical, psychological and social well-being on eight subscales. The maximum total score per subscale is 100. People with high scores view their health in a positive manner and report few psychical and emotional problems. The RAND-36 was not used in both control groups, yet was added to the current study to assess the quality of life of the CRPS patients.

Dutch versions of all instruments were used. All questionnaires were completed by the patients themselves, with the exception of patients in whom the severity of the dystonia would not allow this. In those cases the questionnaires were orally administered by a trained research nurse.

Statistics

SPSS for Windows, version 11.0, was used for data-analysis. As most of the data were not normally distributed, non-parametric test methods were used. The Mann Whitney U test was used when comparing two groups, whereas the Kruskall Wallis test was used for comparison of three groups. To compare the results with normative data available in the literature, mean scores and standard deviations were also calculated. To compare means of the CRPS group on the RAND-36 to norm data, an unpaired t-test was used. A 5% significance level was used.

Results

Characteristics of all patient groups are presented in table 1. There were no significant differences with respect to sex (χ^2 =4.64, df=2, p=0.10), marital status (χ^2 =7.68, df=6, p=0.26) and age (χ^2 =1.53, df=2, p=0.46). Table 2 reflects the characteristics and symptoms of the CRPS patients. In table 3 the results for PDQ-R, TEC, VRMG, DES, SDQ-20 and SCL-90-R for the three groups are presented.

Personality

No normative data for the PDQ-R were available in the literature. The total PDQ-R score of the CRPS patients is significantly lower than that of the CD patients (z=-3.97, p<0.001) and the AD patients (z=-5.26, p<0.001). Among the CRPS patients, most personality traits were observed in the schizoid, obsessive-compulsive, borderline, paranoid and schizotypical personality clusters.

Life events

No significant differences in the total number of traumatic experiences on the TEC were found between the CRPS patients and the CD patients (z=-1.34, p=0.18) or AD patients (z=-1.07, p=0.29). A total of 87% of the CRPS patients experienced at least one of the listed traumatic experiences. In specific they reported intense pain (67%); witnessing traumatic experiences of others (46%); emotional neglect (35%); emotional abuse (39%); physical abuse (28%); sexual traumas (35%); and incestuous acts (22%). A total of 52% of the CRPS patients reported at least one form of physical, emotional or sexual abuse.

The results on the VRMG showed that the CRPS patients experienced less life events in the year before the onset of their symptoms than both control groups (z=-4.72, p<0.001 compared to the CD patients; z=-5.02, p<0.001 compared to AD patients).

Dissociative experiences

The mean DES-score of the CRPS patients fell in the normal range (3.7-7.8), in contrast to both control groups. Fifteen CRPS patients (33%) obtained a score above 7.8. All three groups had lower scores than the mean scores of psychiatric patients (14.6-17.0), patients with dissociative identity disorder (49.5) and patients with other dissociative disorders (>25). 27,31 The score of the CRPS group was significantly lower than the CD group score (z=-2.95, p=0.003), but did not differ significantly from the AD group score (z=-1.82, p=0.07).

For the SDQ-20 no normative data are available in the literature. Compared to the CD patients, no significant differences were found (z=-0.30, p=0.76), indicating that both

groups had similar levels of somatoform dissociation. In comparison to AD patients, CRPS patients had significantly higher scores (z=-4.96, p<0.001).

Table 4 shows correlations between the total number of traumatic life events (TEC) on the one hand, and the DES and SDQ-20 scores on the other hand. The CD and AD groups showed moderate significant positive correlations between the number of traumatic life events and psychoform dissociative experiences. Only the CRPS patients demonstrated a moderate significant positive correlation between the number of traumatic life events and somatoform dissociative experiences¹.

General psychopathology

Compared to normative data for the general population, ²⁹ the CRPS patients obtained a high total score on the SCL-90-R, indicating an increased general level of psychopathology. However, the CRPS group had a significantly lower score compared to both control groups (CD group: z=-4.51, p<0.001; AD group: z=-4.90, p<0.001). For the CPRS-I patients, scores on the somatic and depression subscales were elevated. However, compared to the normative data²⁹ of chronic pain patients, no differences were found.

Quality of life

In comparison to normative data consisting of randomly chosen individuals from the general population,³² the quality of life of the CRPS patients was severely impaired, as measured by the RAND-36 (table 5). The patients reported severe limitations in physical activities, and indicated that this had a negative impact on work-related or other daily activities. Also, pain, fatigue and limitations in social functioning were reported more often by the CRPS patients.

^{1*} Previous investigations in the CD group using a structured trauma interview did show a significant correlation with SDQ-20 scores.²⁰ Here we only found a trend. This discrepancy is presumably due to the use of a different trauma measure.

Table 1. Characteristics of CRPS patients and control groups

	CRPS (N=46)	Conversion disorder (N=54)	Affective disorder (N=50)
Men/women (% female)	2/44 (96)	9/45 (83)	9/41 (82)
Median age in years (IQR)	41 (28-50)	36 (28-47)	36 (28-46)
Mean age in years (SD)	40 (12)	38 (12)	36 (11)
Marital status - Married (%) - Divorced (%) - Cohabiting (%) - Not married (%) Mean disease duration in years (SD)	23 (50) 3 (7) 7 (15) 13 (28) 10.1 (6.5)	28 (52) 7 (13) 7 (13) 12 (22) 5.1 (7.1)	26 (52) 0 (0) 8 (16) 16 (32) not available
No. of affected extremities with dystonia (%) - Two - Three - Four	8 (17) 12 (26) 26 (57)	not applicable	not applicable

IQR: interquartile range; SD: standard deviation

Table 2. Characteristics of the symptoms in CRPS patients

Anti- psychotics	1	1	1	ı	1	1	ı	1	ı	1	١	١	ı	1	ı	1	1	1	1	1	1	ı	1	١	1
SSRI	1	ı	1	1	ı	1	ı	1	ı	1	+	١	+	1	1	1	1	١	١	١	1	ı	١	ı	ı
TCA	+	+	ı	1	ı	ı	ı	١	ı	,	١	1	ı	1	1	1	1	1	ı	1	1	1	+	+	1
Autonomic Symptoms ²	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Hypoalgesia/ hypesthesia	+	+	+	+	+	+	+	+	+	ı	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Hyperalgesia/ hyperesthesia/ allodynia	+	+	+	ì	+	+	+	+	+	+	+	+	+	ì	ì	+	+	+	+	+	+	+	1	1	,
Pain	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Preceding Trauma ¹	fracture	soft tissue injury	soft tissue injury	soft tissue injury	fracture	none	soft tissue injury	fracture	facture	soft tissue injury	none	none	soft tissue injury	none	soft tissue injury	none	soft tissue injury	soft tissue injury							
No. affected extremities	8	3	4	8	4	4	4	4	4	4	4	7	2	8	4	4	4	4	4	4	2	2	4	4	4
Age	24	26	34	39	26	48	20	51	20	21	38	44	23	18	53	33	42	32	38	38	21	55	28	47	45
Sex	н	M	ц	Н	Ц	ц	Н	Н	Н	щ	Н	ц	Н	Ц	Н	Ц	ц	ц	ц	ц	ц	Н	Н	Н	Щ
		2	8	4	\sim	9		∞	6	10	11	12	13	14	15	16	17	18	19	20	21	22	23	24	25

Table 2. continued

Anti- psychotics	1	,	,	,	ı	,	ı	,	,	,	,	1	,	,	,	,	,	,	ı	1	1
SSRI	1	1	,	1	ı	1	ı	1	1	1	1	1	1	1	1	1	1	+	ı	+	1
TCA	+	1	1	ı	1	+	ı	ı	+	1	1	1	1	1	1	1	1	1	ı	1	1
Autonomic Symptoms ²	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Hypoalgesia/ hypesthesia	+	+	i	i	+	+	+	+	+	+	+	+	+	ì	+	+	+	+	+	+	+
Hyperalgesia/ hyperesthesia/ allodynia	+	+	1	ı	+	1	+	+	+	+	+	1	1	+	+	+	+	1	+	+	+
Pain	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+	+
Preceding Trauma¹	none	none	facture	none	soft tissue injury	fracture	soft tissue injury	soft tissue injury	soft tissue injury	none	soft tissue injury	fracture	none	soft tissue injury	soft tissue injury	soft tissue injury	fracture	none	fracture	soft tissue injury	soft tissue injury
No. affected extremities	2	4	4	2	8	8	4	8	2	4	8	8	3	3	4	4	4	4	2	4	3
Age	37	55	33	65	47	54	18	41	26	57	43	99	46	46	40	65	24	43	28	40	24
Sex	Н	Ц	Ц	Ц	Н	Ц	Ц	Ц	Ц	Ц	Ц	Ц	Ц	M	Ц	Ц	Ц	Ц	Ц	Ц	Щ
	56	27	28	59	30	31	32	33	34	35	36	37	38	39	40	41	45	43	44	45	46

¹ Preceding trauma of the first affected extremity

² Autonomic symptoms include edema, changes in skin blood flow (temperature or colour changes) or abnormal sudomotor activity. TCA = tricyclic antidepressants, always used as analgesics

SSRI = selective serotonine reuptake inhibitors, always used as antidepressants

Table 3. Mean total scores and medians

CRPS (N=46)	Conversion disorder (CD) (N=54)	Affective disorder (AD) (N=50)
19.3 (13), (N=45)#	29.2 (13.5)	35.2 (13.8)
14 (10.5-24)*CD, AD	26.5 (19-38.3)	37 (24.8-45)
4.5 (3.9)	5.4 (3.8)	3.7 (3.6)
4 (1.8-6.3)	5 (2-8)	2 (1-6)
1.2 (1.6) (N=45)#	4.2 (3.8; N=53)#	3.8 (3.1)
0 (0-2)*CD, AD	4 (1-6.5)	3.5 (1-5)
-1.9 (7.6)	-9.2 (15.8)	-8.2 (10.8)
0 (-5-0)*CD, AD	-6 (-15-0)	-6 (-15.3-0)
6.6 (5.8)	12 (10.9)	8.7 (7.1)
4.3 (2.1-9.8)*CD	8.5 (4.3-16.9)	7.9 (3.6-11.4)
30.86 (9.7)	30.7 (8.2)	23.6 (4.5)
28.5 (25-34.3)*AD	29.5 (24-36)	22 (20-26)
145.6 (39.8) (N=40)#	201.2 (66.5)	204.4 (59.9)
134 (119.3-166.5)*CD, AD	192.5 (147.8-241.5)	200 (157.5-233.5)
	(N=46) 19.3 (13), (N=45)# 14 (10.5-24)*CD, AD 4.5 (3.9) 4 (1.8-6.3) 1.2 (1.6) (N=45)# 0 (0-2)*CD, AD -1.9 (7.6) 0 (-5-0)*CD, AD 6.6 (5.8) 4.3 (2.1-9.8)*CD 30.86 (9.7) 28.5 (25-34.3)*AD 145.6 (39.8) (N=40)#	(N=46) (CD) (N=54) 19.3 (13), (N=45)# 14 (10.5-24)*CD, AD 26.5 (19-38.3) 4.5 (3.9) 4 (1.8-6.3) 5.4 (3.8) 5 (2-8) 1.2 (1.6) (N=45)# 0 (0-2)*CD, AD 4 (1-6.5) -1.9 (7.6) 0 (-5-0)*CD, AD -6 (-15-0) 6.6 (5.8) 4.3 (2.1-9.8)*CD 30.86 (9.7) 28.5 (25-34.3)*AD 201.2 (66.5) 145.6 (39.8) (N=40)# 201.2 (66.5)

^{#:} smaller sample size due to missing data

SD=standard deviation; IQR=interquartile range; PDQ-R=Personality Diagnostic Questionnaire-Revised; TEC=Traumatic Experiences Scale; VRMG=Recent Life Event Questionnaire ("Vragenlijst Recent Meegemaakte Gebeurtenissen"); DES=Dissociative Experiences Scale; SDQ-20=Somatoform Dissociation Questionnaire-20; SCL-90-R=Symptom Checklist-90-Revised.

The CRPS patients had significantly lower total scores on the PDQ-R, VRMG and SCL-90-R, compared to the conversion patients and patients with affective disorders. On the TEC, no significant differences were found. On the DES, the CRPS patients had a significantly lower score than the conversion patients only, and on the SDQ-20, both the CRPS and conversion patients had a significantly higher score than the patients with affective disorders.

 $^{^*}$: significantly different scores (p<0.05); only presented for medians, since non parametric tests were used to assess differences between groups

Table 4. Correlation values between life events (TEC) and psychoform dissociation (DES) on the one hand and somatoform dissociation (SDQ-20) on the other hand

	Spearman's rho DES	Spearman's rho SDQ-20
CRPS	0.22 (p=0.15)	0.30 (p=0.04)
Conversion disorder	0.28 (p=0.04)	0.25 (p=0.07)
Affective disorder	0.30 (p=0.03)	0.22 (p=0.13)

TEC=Traumatic Experiences Scale; DES=Dissociative Experiences Scale; SDQ-20=Somatoform Dissociation Questionnaire-20

Table 5. Mean (SD) RAND-36 scores

	CRPS (N=40)	General population (N=1063)	significance
PF	15.6 (16.9)	81.9 (23.2)	p<0.005
SF	55.3 (23.2)	86.9 (20.5)	p<0.005
PR	20.0 (30.6)	79.4 (35.5)	p<0.005
ER	69.2 (41.6)	81.1 (32.3)	p<0.005
МН	72.9 (18.1)	76.8 (18.4)	p=0.19
V	49.3 (17.5)	67.4 (19.9)	p<0.005
BP	35.1 (17.7)	79.5 (25.6)	p<0.005
GHP	46.6 (19.9)	72.7 (22.7)	p<0.005
PHC	24.3 (26.0)	52.4 (19.4)	p<0.005

RAND-36= Research and Development-36; SD=standard deviation; PF=physical functioning, SF=social functioning, PR=role limitations due to physical health problems, ER=role limitations due to emotional problems, MH=mental health, V=vitality, BP=bodily pain, GHP=general health perceptions, PHC=perceived health change An unpaired t-test was used to compare means of the CRPS group to the norm group.

Discussion

In accordance with other studies in CRPS patients ^{15,16,33} and chronic CRPS patients with dystonia³⁴ our study does not support the presence of a unique disturbed psychological profile. Compared to the general Dutch population, the general level of psychopathology was elevated in the CRPS patients in an extent similar to chronic pain patients²⁹ but this level was significantly lower than both psychiatric control groups. The total level of psychopathology is slightly higher as compared to earlier studies.^{34,35} However, these studies had smaller sample sizes and included patients with a shorter disease duration, and moreover these patients were less severely affected by CRPS. The relatively low scores with regard to affective, dissociative and anxiety features among patients with CRPS related dystonia contrast with the study of Schrag et al.,³⁶ who found affective disorders in 85%, dissociative symptoms in 42% and anxiety disorders in 58% of a group of 26 patients with fixed dystonia. However, one should be cautious comparing these results as the focus of the study of Schrag et al.³⁶ were patients with fixed dystonia of which a minority met the IASP-criteria of CRPS, while also different instruments were used.

CRPS patients reported only few relevant life events in the year preceding the symptom onset, but more than three quarters of the patients reported at least one traumatic experience in their early history. In more than half of the patients at least one form of physical, emotional or sexual abuse, or neglect had occurred. No official normative data of the TEC are available, but it has been administered in 73 Dutch students and in the general population (N=147).37 The total TEC scores of the CRPS, CD and AD groups we studied are much higher than those of the students and the general population. An interesting observation is that the total score of the CRPS patients is higher than the total score of the AD group and than the score of a group of various female psychiatric patients (eating disorders, substance abuse disorders and ADs, among others) in an earlier study.³⁸ Possibly, this is caused by the fact that one of the included traumas is the experience of intense pain, which is inherent to CRPS. However, when the data are corrected for this type of trauma, the total mean score for the CRPS patients is 3.89, which is still higher than the score of the AD patients of the current study. One may therefore conclude that in patients with CRPS related dystonia early traumatic experiences are more prevalent.

An association between somatoform dissociation and lifetime traumatic experiences has been suggested earlier, both in clinical^{20,24,39} and non-clinical populations.³⁷ In our study, we also found a significant, though moderate (r=0.30) association between the number of traumatic life events and levels of somatoform dissociation in CRPS patients. The elevated SDQ-20 scores that we found among the CRPS patients are

in the range of patients with somatoform disorders.⁴⁰ These results suggest similarity to the conversion disorder patients and may indicate that in chronic CRPS patients dissociative phenomena may be present. Though the association between traumatic experiences and somatoform dissociation in this study is only moderate, these results generate interesting hypotheses for further research. Also, a 0.3 correlation is considered meaningful in the social sciences.⁴¹ A relationship between psychological trauma and physical complaints, such as lung diseases,^{42,43,44} peptic ulcer,⁴³ diabetes,^{43,44} cardiac disease^{43,44} and headache⁴⁴ has been found, yet also a relation between trauma and the severity of 'medically unexplained symptoms', as chronic pelvic pain,^{45,46} irritable bowel syndrome,^{47,48,49,50} pseudo-epileptic seizures,⁴⁸ chronic fatigue⁵¹ and somatization disorder,⁵² has been found in previous studies. However, it should be noted that the SDQ-20 scores of CRPS patients are possibly inflated because some items of the SDQ-20, such as voiding symptoms and feelings of numbness, are features known to be associated with CRPS.^{4,53}

The current study clearly shows that the CRPS patients experience less personality pathology than both psychiatric control groups. Personality traits of the schizoid, obsessive-compulsive, borderline, paranoid and schizotypical personality disorders were most prevalent among the CRPS patients, which partly corresponds to results found by Monti et al.⁵⁴

In contrast to our study, Shiri et al., ¹⁷ found no significant differences in the psychological profiles of CRPS patients and CD patients. Possible explanations for these conflicting results include the use of different instruments and the smaller sample size (17 CRPS patients and 20 CD patients). The predominance of male CRPS patients (94%) in the study of Shiri and colleagues¹⁷ is conspicuous, but the difference in gender distributions between these studies is an unlikely explanation for the differences in results in view of the fact that in general both CRPS-1 and CD are more frequent among women.

The CRPS patients in our study reported poorer general health and quality of life as compared to the general population. The general health score of the CRPS patients in the current study, however, is similar to those reported for patients with other causes of chronic pain.⁵⁵

One of the strengths of this study is the applied extensive set of psychological instruments. Additionally, we were able to compare our patients with two psychiatric control groups. Some limitations of the present study should also be noted. Data was collected retrospectively and therefore no data on premorbid psychological symptoms and disorders are available. In this study self report instruments were used. Reported life events and other psychological symptoms were not confirmed by a clinical examination, and were not verified with third parties or authorities. Also, the mean disease duration of the CRPS patients was 10.1 years, much longer than in the conversion disorder

group, and it cannot be ruled out that some patients may have developed secondary psychological disorders during this period. Here recall bias could also have played a role since patients were asked about life events which occurred in the year preceding the first signs and symptoms. Also, the results of this study cannot be generalized to acute or milder forms of CRPS, as the CRPS group consisted of severely affected patients with a long mean disease duration. Next, due to the severity of their dystonia, some patients were not able to complete the questionnaires themselves. In these cases they were orally administered by a trained research nurse, which may have led to social desirable answers. Lastly, a normal control group was not used.

In summary, CRPS is a multifactorial condition where, aside biological factors, psychological and social factors may play a role in the onset or development of chronicity of the condition. Although in this study patients with CRPS related dystonia showed elevated scores on some of the scales we used, there does not seem to be a unique psychological profile on a group level, and only few similarities between the profiles of patients with CRPS and CD were found. Early traumatic lifetime experiences were frequently reported and may be a possible, though not necessary, predisposing factor for CRPS-related dystonia.

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References

- 1. Bruehl SP, Steger HG, Harden RN. Complex regional pain syndrome. In: Turk DC, Melzack R, eds. Handbook of pain assessment. New York: Guilford Press 2001: 549-66.
- 2. Veldman PHJM, Reynen HM, Arntz IE, Goris RJA. Signs and symptoms of reflex sympathetic dystrophy: Prospective study of 829 patients. Lancet 1993;342:1012-6.
- Schwartzman RJ, Kerrigan J. The movement disorder of reflex sympathetic dystrophy. Neurology 1990;40:57-61.
- 4. Birklein F, Riedl B, Sieweke N, Weber M, Neundorfer B. Neurological findings in complex regional pain syndromes: Analysis of 145 cases. Acta Neurol Scand 2000;101:262-9.
- 5. van Rijn MA, Marinus J, Putter H, van Hilten JJ. Onset and progression of dystonia in complex regional pain syndrome. Pain 2007; 130(3):287-293
- van Hilten JJ, van de Beek WJT, Vein AA, van Dijk JG, Middelkoop HAM. Clinical aspects of multifocal or generalized tonic dystonia in reflex sympathetic dystrophy. Neurology 2001;56:1762-5.
- 7. van Hilten JJ, Blumberg H, Schwartzman RJ. Factor IV: Movement disorders and dystrophy. Pathophysiology and measurement. In: Wilson P, Stanton-Hicks M, Harden RN, eds. CRPS: Current diagnosis and therapy. Seattle: IASP Press 2005: 119-37.
- Janig W, Baron R. Complex regional pain syndrome: mystery explained? Lancet Neurol. 2003;2:687-97.
- van der Laan L, ter Laak HJ, Gabreëls-Festen A, Gabreëls F, Goris RJA. Complex regional pain syndrome type I (RSD): Pathology of skeletal muscle and peripheral nerve. Neurology 1998;51:20-5.
- Oaklander AL, Rissmiller JG, Gelman LB, Zheng L, Chang Y, Gott R. Evidence of focal smallfiber axonal degeneration in complex regional pain syndrome-I (reflex sympathetic dystrophy). Pain 2006;120:235-43.
- Ochoa JL, Verdugo RJ. Reflex sympathetic dystrophy: A common clinical avenue for somatoform expression. Neurologic Clinics 1995;13:351-63.
- Egle UT, Hoffmann SO. Psychosomatische Zusammenhänge bei sympathischer Reflexdystrophie (Morbus Sudeck): Literaturübersicht und erste klinische Ergebnisse. Psychother Med Psychol 1990;40:123-35.
- Covington EC. Psychological issues in reflex sympathetic dystrophy. In: Jänig W, Stanton-Hicks M, eds. Reflex sympathetic dystrophy: A reappraisal. Seattle: IASP Press 1996: 191-215.
- Verdugo RJ, Ochoa JL. Abnormal movements in complex regional pain syndrome: Assessment of their nature. Muscle & Nerve 2000;23:198-205.
- Bruehl SP, Carlson CR. Predisposing psychological factors in the development of reflex sympathetic dystrophy: A review of the empirical evidence. Clin J Pain 1992;8:287-99.
- Lynch ME. Psychological aspects of reflex sympathetic dystrophy: A review of the adult and paediatric literature. Pain 1992;49:337-47.
- 17. Shiri S, Tsenter J, Livai R, Schwartz I, Vatine JJ. Similarities between the psychological profiles of complex regional pain syndrome and conversion disorder patients. J Clin Psychol Med Settings 2003;10:193-9.
- van Hilten JJ, van de Beek WJT, Hoff JI, Voormolen JHC, Delhaas EM. Intrathecal baclofen for the treatment of dystonia in patients with reflex sympathetic dystrophy. N Eng J Med 2000;343:625-30.
- 19. Merskey H, Bogduk N. Classification of chronic pain: Descriptions of chronic pain syndromes and definitions of pain terms. Seattle: IASP Press, 1994.
- Roelofs K, Keijsers GP, Hoogduin KA, Naring GW, Moene FC. Childhood abuse in patients with conversion disorder. Am J Psychiatry 2002;159:1908-13.
- 21. American Psychiatric Association. Diagnostic and statistical manual of mental disorders, fourth edition: Text revision (DSM-IV-TR). Washington: American Psychiatric Publishing, 2000.

- 22. Hyler SE, Skodol AE, Kellman HD, Oldham JM, Rosnick L. Validity of the Personality Diagnostic Questionnaire-Revised: Comparison with two structured interviews. Am J Psychiatry 1990;147:1043-8.
- 23. American Psychiatric Association. Diagnostic and statistical manual of mental disorders, third edition: Revised (DSM-III-R). Washington: American Psychiatric Publishing, 1987.
- 24. Nijenhuis ERS, Spinhoven P, van Dyck R, Van der Hart O, Vanderlinden J. Degree of somatoform and psychological dissociation in dissociative disorder is correlated with reported trauma. J Trauma Stress 1998;11:711-30.
- van de Willige G, Schreurs P, Tellegen B, Zwart F. Het meten van 'life events': De Vragenlijst Recent Meegemaakte Gebeurtenissen (VRMG). Ned Tijd Psychol 1985;40:1-19.
- 26. Roelofs K, Spinhoven P, Sandijck P, Moene FC, Hoogduin KA. The impact of early trauma and recent life-events on symptom severity in patients with conversion disorder. J Nerv Ment Dis 2005;193:508-14.
- 27. Bernstein EM, Putnam FW. Development, reliability, and validity of a dissociation scale. J Nerv Ment Dis 1986;174:727-35.
- 28. Nijenhuis ERS, Spinhoven P, van Dyck R, Van der Hart O, Vanderlinden J. The development and psychometric characteristics of the Somatoform Dissociation Questionnaire (SDQ-20). J Nerv Ment Dis 1996;184:688-94.
- 29. Arrindell WA, Ettema JHM. SCL-90: Herziene handleiding bij een multidimensionele psychopathologie-indicator. Lisse: Swets & Zeitlinger, 2003.
- 30. Hays RD, Morales LS. The RAND-36 measure of health-related quality of life. Ann Med 2001;33:350-7.
- 31. Boon S, Draijer N. Screening en diagnostiek van dissociatieve stoornissen. Lisse: Swets & Zeitlinger, 1995.
- 32. van der Zee KI, Sanderman R. Het meten van de algemene gezondheidstoestand met de RAND-36: Een handleiding. Groningen: Noorderlijk Centrum voor Gezondheidsvraagstukken, 1993.
- 33. Ciccone DS, Bandilla EB, Wu WH. Psychological dysfunction in patients with reflex sympathetic dystrophy. Pain 1997;71:323-33.
- 34. van der Laan L, van Spaendonck K, Horstink MW, Goris RJ. The Symptom Checklist-90 Revised questionnaire: No psychological profiles in complex regional pain syndrome-dystonia. J Pain Symptom Manage 1999;17:357-62.
- Geertzen JH, Bruijn-Kofman AT, de Bruijn HP, van de Wiel HB, Dijkstra PU. Stressful life events and psychological dysfunction in Complex Regional Pain Syndrome type I. Clin J Pain 1998;14:143-
- 36. Schrag A, Trimble M, Quinn N, Bhatia K. The syndrome of fixed dystonia: An evaluation of 103 patients. Brain 2004;127:2360-72.
- Näring G, Nijenhuis ERS. Relationships between self-reported potentially traumatizing events, psychoform and somatoform dissociation, and absorption, in two non-clinical populations. Aust N Z J Psychiatry 2005;39:982-8.
- Bracke D, van Leeuwen K, Verhofstadt-Denève L. Comorbiditeit van zelfverwondend gedrag en dissociatieve symptomen bij vrouwelijke psychiatrische patiënten. Tijdschr Psychiatr 2001;43:673-82.
- Waller, G, Hamilton, K., Elliott, P., Lewendon, J., Stopa, L., Waters, A., Kennedy, F., Lee, G., Pearson, D., Kennerley, H., Hargreaves, I., Bashford, V., Chalkley, J. Somatoform dissociation, psychological dissociation, and specific forms of trauma. J Trauma 2000:1:81-98.
- Nijenhuis ERS. The scoring and interpretation of the SDQ-20 and SDQ-5: Update 2003. http:// www.enijenhuis.nl/SDQ/sdq-update.doc
- 41. Cohen J. Statistical power analysis for the behavioral sciences. New York: Academic Press 1977.
- 42. Goodwin RD, Wamboldt MZ, Pine DS. Lung disease and internalizing factors: Is childhood abuse a shared etiologic factor? J Psychosom Res 2003;55:215-9.

- 43. Goodwin RD, Stein MB. Association between childhood trauma and physical disorders among adults in the United States. Psychol Med 2004;34:509-20.
- 44. Romans S, Belaise C, Martin J, Morris E, Raffi A. Childhood abuse and later medical disorders in women: An epidemiological study. Psychother Psychosom 2002;71:141-50.
- 45. Walker EA, Katon WJ, Neraas K, Jemelka RP, Massoth D. Am J Psychiatry 1992;149:534-7.
- 46. Reiter RC, Gambone DJC. Demographic and historic variables in women with idiopathic chronic pelvic pain. Obstet Gynecol 1990;75:428-32.
- 47. Hobbis ICA, Turpin G, Read, NW. A re-examination of the relationship between abuse experience and functional bowel disorders. Scand J Gastroenterol 2002;37:423-30.
- 48. Reilly J, Baker GA. Rhodes J, Salmon P. The association of sexual and physical abuse with somatization: Characteristics of patients presenting with irritable bowel syndrome and non-epileptic attack disorder. Psychol Med 1999;29:399-406
- 49. Salmon P, Skaife K, Rhodes J. Abuse, dissociation, and somatization in irritale bowel syndrome: Towards an explanatory model. J Behav Med 2003;26:1-18.
- 50. Walker EA, Katon WJ, Roy-Byrne PP, Jemelka RP, Russo J. Histories of sexual victimization in patients with irritable bowel syndrome or inflammatory bowel disease. Am J Psychiatry 1993;150:1502-6.
- 51. Heim C, Wagner D, Maloney E, Papanicolaou DA, Solomon L, Jones JF, Unger ER, Reeves WC. Early adverse experience and risk of chronic fatigue syndrome: Results from a population-based study. Arch Gen Psychiatry 2006;63:1258-66.
- 52. Brown RJ, Schrag A, Trimble MR. Dissociation, childhood interpersonal trauma, and family functioning in patients with somatization disorder. Am J Psychiatry 2005;162:899-905.
- 53. Chancellor MB, Shenot PJ, Rivas DA, Mandel S, Schwartzman R. Urological symptomatology in patients with reflex sympathetic dystrophy. J Urol 1996;155:634-7.
- 54. Monti DA, Herring CL, Schwartzman RJ, Marchese M. Personality assessment of patients with complex regional pain syndrome type I. Clin J Pain 1998;14:295-302.
- 55. Lamé IE, Peters ML, Vlaeyen JWS, Van Kleef M, Patijn J. Quality of life in chronic pain is more associated with beliefs about pain, than with pain intensity. Eur J Pain 2005;9:15-24.