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Complex regional pain syndrome, alexithymia, and psychological distress

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ABSTRACT

Objective: This study aims to elucidate the relationships between alexithymia, psychological distress, and pain in persons with complex regional pain syndrome (CRPS).
Methods: Participants were 60 Israeli adults ages 19–65. This is a cross sectional study with a comparison group. Alexithymia, psychological distress, and pain were assessed in 30 individuals with CRPS in comparison to 30 gender- and age-matched persons with lower back pain (LBP). Assessments included the Toronto Alexithymia Scale, Hospital Anxiety and Depression Scale, and two subscales of the McGill Pain Questionnaire.
Results: Persons with CRPS had significantly higher ratings of psychological distress and of alexithymia when compared to LBP controls. Pain severity was significantly associated with higher levels of alexithymia and psychological distress among persons with CRPS, but not among controls. Alexithymia and pain severity correlations were significantly different between the two groups. In persons with CRPS, the relationships between alexithymia and pain severity and between difficulty identifying feelings and pain were not confounded by psychological distress.
Conclusions: To our knowledge, this is the first cross sectional study providing empirical evidence on the relationship between alexithymia and CRPS. From the perspective of conceptualizing alexithymia as an outcome of CRPS, findings highlight the importance of early CRPS diagnosis and support the provision of care that addresses pain-related psychological distress and alexithymia among CRPS patients. Also, findings underscore the need to generate alternative, non-physical avenues, such as learning to identify feelings for processing pain, in order to reduce pain among persons with CRPS.

Introduction

Complex regional pain syndrome (CRPS) is a chronic pain condition characterized by a continuing (spontaneous and/or evoked) regional pain that is seemingly disproportionate in length of time or degree of pain after trauma or other lesions. The pain usually has a distal predominance of abnormal sensory, motor, sudomotor, vasomotor edema, and/or trophic findings [1]. The evolution of CRPS varies, but it typically results in functional impairment in the affected limb, which adversely affects one's quality of life [2,3]. While the etiology of CRPS is still unclear, several underlying patho-physiological mechanisms have been identified. These include aberrant changes in vasomotor function, abnormalities in regulating inflammatory responses on a local, regional and/or central level, and problems with regulation of neuroplasticity [4,5].

Psychological factors such as stressful life events and psychological states such as anxiety, depression, and anger may alter catecholamine activity. Thus, they may directly affect pain intensity and in this way impact patho-physiological mechanisms of CRPS and contribute to its perpetuation [6,7]. Therefore, pain in persons with CRPS may be affected by factors relating to emotional regulation. The findings on the relationships between psychological distress, emotional regulation, and pain in persons with CRPS are inconclusive. Understanding these relationships may elucidate effective coping styles and beneficial rehabilitation efforts for persons with CRPS [8].

One indicator of emotional regulation is alexithymia, a multidimensional psychological structure that includes difficulty identifying, verbalizing, or describing feelings and an externally-oriented thinking style [9]. High levels of alexithymia have been reported among persons with various types of pain, including lower back pain (LBP) [10], fibromyalgia [11], and myofascial pain [12,13]. Conversely, others have found no association between alexithymia and pain among chronic pain patients [e.g., [14]]. The relationship between alexithymia and CRPS has been scantly examined. In a psychiatric evaluation of 34 persons with CRPS it was found that 88% met the clinical criteria of alexithymia [15]. On the other hand, in a prospective study of patients

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with a fracture in the distal radius, no differences in alexithymia scores were found among patients who subsequently developed CRPS when compared to patients who did not develop CRPS [16].

The relationship between CRPS, anxiety, and depression is inconclusive [see review by 6]. With regards to depression, higher [17,18], comparable [19,20], and lower [21,22] levels of depression have been found in persons with CRPS when compared to other groups. In a similar manner, findings on anxiety and CRPS diverge, with reports of higher [18], comparable [19], and lower [23] anxiety levels in persons with CRPS in comparison to other groups.

Psychological distress may confound the association between pain and alexithymia in persons with CRPS. It has been suggested that alexithymia hinders a successful regulation of negative feelings, which in turn leads to increased negative affect or distress, chronic overactivation of the sympathetic system, and decreased capacity of the immune system. These psychological and physical outcomes may contribute to CRPS exacerbation and increased pain [24,25]. Accordingly, distress has been associated with pain, demonstrating a more potent association among persons with CRPS in comparison to persons with other chronic pain [17]. In addition, alexithymia has been associated with psychological distress [26], depression [27], anxiety, and higher internalized anger expression [13]. Studies of persons with non-CRPS chronic pain have shown that when controlling for distress factors, the association found between alexithymia and pain did not reach significance [28] or was significant for affective pain but not sensory pain [12].

The objective of the current study is to elucidate the relationships between alexithymia, psychological distress, and pain in persons with CRPS in order to promote accurate assessment of pain and effective rehabilitation initiatives for this population. Accordingly, we examined levels of alexithymia, psychological distress, and pain in persons with CRPS in comparison to gender- and age-matched persons with chronic LBP. A measurement of psychological distress, a multidimensional assessment of pain, and an empirical assessment of alexithymia were utilized.

Methods

Participants

Ninety-three Israeli adults aged 19–65 were approached, of whom 46.2% had a diagnosis of CRPS, and 53.7% reported chronic LBP. Inclusion criteria were: 1. aged 18 or older; 2. experiencing pain for duration of at least 3 months; and 3. Hebrew proficiency. Exclusion criteria were: 1. pain intensity lower than 3/10 on the Visual Analogue Scale; 2. suffering simultaneously from CRPS and LBP; 3. history of CRPS in persons with LBP; and 4. history of mental illness or brain injury. CRPS was diagnosed according to the clinical Budapest Criteria [1], by physicians specialized in physical medicine. Inclusion in the comparison group was based on a subjective self-report of chronic LBP that was diagnosed by pain specialist physicians as nonspecific low back pain or back pain potentially associated with radiculopathy or spinal stenosis. Sixty participants met inclusion criteria and participated in the study.

Participants had a mean age (SD) of 38.2 (13.30). Of participants with CRPS, most were males, married, born in Israel, with a high school education, and of poor to adequate socio-economic status. No significant differences were found in background variables, duration of disease, or duration of treatment between persons with CRPS and persons with LBP (Table 1). Pain was experienced as continuous by most of the persons with CRPS (83.3%) as well as by those with chronic LBP (73.3%). Of the medications in use, non-narcotic medications were most frequently used by both participants with CRPS (76.7%) and by controls (56.7%). Rates of use of narcotic medication, mood stabilizers, and nonsteroidal anti-inflammatory drugs (NSAIDs) were (%) 53.3, 40.0, and 3.3 for the former, and 30.0, 20.0, and 10 for the latter. No significant differences in type of pain or medication use between the two groups were found.

Table 1
Participants' characteristics by group ($N = 60$).

	CRPS $n = 30$		LBP $n = 30$	
	M (SD)	*n* (%)	*M* (SD)	*n* (%)
Age	38.25 (14.29)		38.2 (12.46)	
Gender (female)		12 (40)		12 (40)
Marital status				
Single		9 (30)		14 (46.7)
Married		17 (56.7)		13 (43.3)
Divorced		4 (13.3)		3 (10)
Place of birth				
Israel		24 (80)		24 (80)
East Europe		3 (10)		4 (13.3)
West Europe		1 (3.3)		2 (6.7)
Asia\Africa		2 (6.7)		0 (0)
No. of children				
0		11 (36.7)		13 (43.3)
1–2		10 (33.3)		9 (30)
3 or more		9 (30)		8 (26.7)
Education				
Elementary		0 (0)		2 (6.7)
High-school		21 (70)		16 (53.3)
Higher education		9 (30)		12 (40)
Financial status				
Poor		11 (36.7)		5 (16.7)
Adequate		12 (40)		12 (40)
Good to excellent		7 (20)		13 (43.3)
Duration of disease	3.33 (3.27)		6.00 (6.64)	
Duration of treatment	2.43 (2.17)		1.90 (2.75)	

CRPS = complex regional pain syndrome; LBP = lower back pain.

Procedure

The study was approved by the institutional review boards of Reuth Rehabilitation Hospital, Tel Aviv Sourasky Medical Center and Ariel University. The CRPS patients were recruited at the Israeli CRPS center in Reuth Rehabilitation Hospital. Among 43 patients diagnosed with CRPS according to the clinical Budapest Criteria, 30 patients (69.7%) met inclusion criteria and agreed to participate in the study. Control participants that matched the CRPS sample in terms of age and gender were sought and recruited from the pain clinic at Tel Aviv Sourasky Medical Center. Fifty patients suffering from chronic LBP were approached. Of these 50 patients, 30 patients (60%) met inclusion criteria and agreed to participate. All the participants signed informed consent forms. The data was collected via structured self-report questionnaires that were completed in the presence of the investigator (LBH) providing clarifications as needed. The duration of medical condition was calculated as time elapsed since the pain-initiating event. The duration of treatment was calculated as time passed since the beginning of the first pain treatment after experiencing pain.

Measurements

Socio-demographics and medical background were recorded and included age, place of birth, marital status, education, and economic status.

Alexithymia was assessed by the Hebrew translated version [29] of the Toronto Alexithymia Scale-20 (TAS 20; [30]). The questionnaire evaluates three factors: difficulty identifying feelings (seven items; e.g., I am often confused about what emotion I am feeling), difficulty describing feelings (five items; e.g., I find it hard to describe how I feel about people), and externally-oriented thinking (eight items; e.g., I prefer talking to people about their daily activities rather than their feelings). Items are rated on a 5-point scale ranging from 1 (total disagreement) to 5 (total agreement). The total score on each subscale is the

mean rating of all items. The total alexithymia score is the sum of ratings on the 20 items, ranging 20–100. Some items are reversed and were scored on a reversed scale. Alpha Cronbach in this study was 0.88 for the total scale, 0.86 for the subscale of difficulty identifying feelings, 0.73 for the subscale of difficulty describing feelings, and 0.51 for the subscale of externally-oriented thinking.

Psychological distress was assessed by the Hospital Anxiety and Depression Scale (HADS; [31]). The scale includes two sub-scales of 7 items each. One sub-scale evaluates anxiety (e.g., I get a sort of frightened feeling as if something awful is about to happen) and the second sub-scale evaluates depression (e.g., I feel as if I am slowed down). Each item is rated on a scale ranging from 0 (not at all) to 3 (most of the time or its equivalent). A higher rating represents a higher frequency of anxiety or depression. The total score is calculated by the mean rating of the 14 items. A score of 11 or higher in each of the subscales indicates significant psychological distress, 8–10 borderline distress, and 0–7 lack of distress. Good psychometrics have been reported [31–33]. Alpha Cronbach in this study was 0.93.

Pain was assessed by the validated Hebrew version [34] of the McGill Pain Questionnaire (MPQ; [35]). The MPQ is a validated and reliable multidimensional pain assessment evaluating medication, location of pain, description of pain in words, variability of pain, and pain intensity [36]. Two subscales were used. On *The Visual Analogue Scale* (*VAS*), participants were asked to rate the point that best represents their pain intensity on a 10 cm long vertical line with one end labeled "no pain" and the other end labeled "pain as bad as it could be". *The Pain rating Index Scale* (*PRIS*) assesses pain severity by summing the numeral value of sensory (e.g., pulsing), affective (e.g., exhausting), evaluative (a single item—miserable), and miscellaneous (e.g., piercing) descriptors.

Statistical analysis

Statistical analyses were conducted using SPSS 16. In order to examine whether research and comparison groups differ on background variables, pain, psychological distress and alexithymia, *t*-tests and χ^2 comparisons were conducted. Pearson correlations were used to examine the association between psychological distress, alexithymia, and pain. In order to compare the potency of correlations between the two groups, Fisher's Z analysis was conducted [37]. We conducted multiple comparisons using Bonferroni adjusted alpha levels of 0.01 per test. First, Pearson correlations of pain (PRIS, VAS), alexithymia (total), and psychological distress were compared. Second, Pearson correlations of pain (PRIS) and alexithymia (subscales) were compared. Finally, in order to examine if the relationship between alexithymia and pain is confounded by psychological distress, we calculated two-tailed partial correlations between alexithymia (total score and sub-scales) and pain (PRIS, VAS) after controlling for psychological distress among persons with CRPS.

Results

A comparison of pain, psychological distress and alexithymia

Alexithymia total scores as well as sub-scale scores (identifying feelings, describing feelings, externally-oriented thinking) were significantly higher in persons with CRPS when compared to controls, $t(58) = 5.23, 4.85, 4.93, 3.62$, $p = 0.01$, respectively. Persons with CRPS had significantly higher ratings of psychological distress when compared to those with chronic LBP, $t(58) = 4.35$, $p = 0.01$. No statistically significant differences in pain indicators (PRIS, VAS) were found between persons with CRPS in comparison to controls (Table 2).

Table 2
A comparison of measures of pain, psychological distress and alexithymia ($N = 60$).

	CRPS $n = 30$	LBP $n = 30$	$t_{(58)}$
	M (SD)		
VAS	7.24 (2.24)	7.02 (1.99)	0.39
PRIS	51.03 (13.93)	44.21 (16.3)	1.74, $p = .09$
Psychological distress	1.73 (0.69)	0.98 (0.64)	4.35[a]
Alexithymia (total)	66.83 (13.64)	48.53 (13.44)	5.23[a]
Difficulty identifying feelings	3.70 (0.99)	2.49 (0.93)	4.85[a]
Difficulty describing feelings	3.45(1.00)	2.27(0.84)	4.93[a]
Externally-oriented thinking	2.88 (0.57)	2.34 (0.59)	3.62[a]

VAS = Visual Analogue Scale; PRIS = Pain Rating Index-Scale; CRPS = complex regional pain syndrome; LBP = lower back pain.
[a] $p < 0.01$.

Associations between pain, psychological distress, and alexithymia

In persons with CRPS, significant associations were found between psychological distress and pain (PRIS, $r = 0.65$, $p = 0.00$; VAS, $r = 0.43$, $p = 0.01$), alexithymia (total) and pain (PRIS, $r = 0.77$, $p < 0.001$; VAS, $r = 0.42$, $p = 0.02$), and alexithymia (total) and psychological distress ($r = 0.71$, $p < .01$). Pain severity as measured by the PRIS was significantly associated with each of the sub-scales of alexithymia (difficulty identifying feelings ($r = 0.74$, $p < 0.001$), difficulty describing feelings ($r = 0.61$, $p < 0.001$), and externally-oriented thinking ($r = 0.42$, $p = 0.01$) among persons with CRPS. Pain, as measured by the VAS, was significantly associated with difficulty identifying feelings (VAS, $r = 0.55$, $p < 0.001$) among persons with CRPS. The associations between VAS, difficulty describing feelings and externally-oriented thinking were not statistically significant. Psychological distress was significantly associated with each of the sub-scales of alexithymia (difficulty identifying feelings ($r = 0.62$), difficulty describing feelings ($r = 0.65$), and externally-oriented thinking ($r = 0.50$, $p < .01$), in persons with CRPS (Table 3).

A significant difference between alexithymia and PRIS correlations was found among persons with CRPS when compared to those with LBP ($Z = 2.49$, $p = 0.01$). The two groups did not show significant differences between the correlations of alexithymia and VAS, psychological distress and VAS, or psychological distress and PRIS after adjusting for multiple comparisons. Subsequent analyses revealed a significant difference between difficulty identifying feelings (alexithymia subscale) and PRIS correlations among persons with CRPS when compared to those with LBP ($Z = 2.23$, $p = 0.01$). The two groups did not show significant differences between the correlations of difficulty describing feelings and PRIS, and externally-oriented thinking and PRIS after adjusting for multiple comparisons.

Partial correlations between alexithymia and pain

When controlling for psychological distress, the partial correlations between alexithymia (total) and PRIS scores was statistically significant ($pr = 0.58$, $p \leq 0.001$) as well as the partial correlations between difficulty identifying feelings and pain scores (PRIS; $pr = 0.53$, $p < 0.01$; VAS, $pr = 0.38$, $p < .05$). The partial correlation between difficulty describing feelings and PRIS scores approached a statistical level of significance ($pr = 0.33$, $p = .08$). The partial correlations between alexithymia (total) and VAS, and between externally-oriented thinking and PRIS were not statistically significant ($pr = 0.18, 0.15$, respectively).

Discussion

This study examined the relationships between alexithymia, psychological distress, and pain in 30 persons with CRPS in comparison to 30 gender- and age-matched persons with chronic LBP. Persons with CRPS showed higher levels of alexithymia when compared to persons with chronic LBP. To our knowledge, this is the first cross sectional study with a comparison group providing empirical evidence on the relationship between alexithymia and CRPS. In addition, persons with CRPS had higher levels of psychological distress when compared to persons with chronic LBP. Pain was significantly associated with alexithymia and difficulty identifying feelings among persons with CRPS, but not among those with LBP. Finally, among persons with CRPS, the relationships between alexithymia and pain severity and between difficulty identifying feelings and pain were not confounded by psychological distress.

Persons with CRPS showed higher levels of alexithymia including symptoms of difficulty identifying feelings, difficulty describing feelings, and externally-oriented thinking, when compared to persons with chronic LBP. These findings corroborate previous evidence of high alexithymia prevalence found among 34 persons with CRPS by means of a clinical diagnosis [15]. Due to the cross-sectional design of the current study causality could not be inferred, and alexithymia could be conceptualized either as an outcome or a determinant of CRPS. In support of the former, in a prospective study of patients with a fracture in the distal radius, it was found that patients who developed CRPS did not differ

Table 3
Pearson correlations between pain, distress, and alexithymia in persons with CRPS and persons with LBP.

	Pain				Psychological distress		Alexithymia							
	VAS		PRIS				Total		Difficulty identifying feelings		Difficulty describing feelings		Externally-oriented thinking	
	CRPS	LBP	CRPS	LBP	CRPS	LBP	CRPS	LBP	CRPS	LBP	CRPS	LBP	CRPS	LBP
Pain														
VAS														
PRIS	0.51*	0.43*												
Psychological distress	0.43*	0.19	0.65**	0.30										
Alexithymia (total)	0.42*	0.09	0.77**	0.33	0.71**	0.63**								
Difficulty identifying feelings	0.55*	0.21	0.74**	0.33	0.62**	0.70**	0.92**	0.87**						
Difficulty describing feelings	0.33	0.20	0.61**	0.29	0.65**	0.52**	0.89**	0.80**	0.77**	0.61**				
Externally-oriented thinking	−0.04	−0.15	0.42*	0.24	0.50**	0.27	0.55**	0.75**	0.38*	0.53**	0.31	0.40**		

CRPS = complex regional pain syndrome, LBP = lower back pain, VAS = Visual Analogue Scale, and PRIS = Pain Rating Index-Scale
* $p < 0.05$.
** $p < 0.01$.

from those who did not develop CRPS in alexithymia scores taken prior to the onset of CRPS [16]. When considered alongside the above finding, it seems that higher levels of alexithymia may be a possible outcome of CRPS rather than its determinant. This notion is in accordance with the conceptualization of alexithymia as a state-dependent phenomenon [e.g., [38,39]] and as a protective coping mechanism against psychological distress at times of high vulnerability [40], such as that caused by CRPS. From this perspective, current findings highlight the importance of early CRPS diagnosis, and the provision of care that addresses pain-related psychological distress and alexithymia among CRPS patients. For example, practicing emotional self-awareness may result in reduction of alexithymia [41]. From the perspective of family system models, alexithymia is shaped by the family's attitudes towards and acceptance of emotional expressions and may develop due to parental cognitive or emotional dysfunction in childhood. In line with this view, family expressiveness has been associated with alexithymia [42], and specific alexithymia deficits have been associated with indicators of family dysfunction such as emotional involvement [43]. Viewed from this perspective, alexithymia in persons with CRPS may be the outcome of early family experiences. In line with this, the personal histories of persons with CRPS included adversities such as alcoholic parents, early death of parents, rape, and child abuse [15], which suggests that alexithymia in persons with CRPS may be caused by childhood trauma. It would be interesting to examine the relationship between family emotional dysfunction in childhood and alexithymia in future studies of persons with CRPS.

Higher levels of psychological distress were found among persons with CRPS when compared to those with chronic LBP. This finding corroborates previous evidence [17,18,23]. For example, persons with CRPS had significantly higher anxiety, depression, interpersonal sensitivity and somatization when compared to hand-injured persons [18]. In another study, it was found that after a distal radius fracture, patients with high anxiety trait scores had a higher risk of developing CRPS [16]. Nonetheless, findings regarding anxiety and depression among persons with CRPS are inconclusive [see [6]]. Higher levels of psychological distress among persons with CRPS may be due to a lower pain threshold in comparison to other populations that experience pain [18]. It may also be that adapting to and coping with CRPS and its adverse symptoms engenders psychological distress among persons with CRPS [44].

Pain ratings as measured by the MPQ (PRIS and VAS) were significantly associated with alexithymia and difficulty identifying feelings among persons with CRPS, but not among those with LBP. This association may be mediated by increased arousal due to physiological markers that may lead to bodily changes, such as increased sympathetic activity and tensed muscles, which in turn induce pain [45]. For example, alexithymia has been associated with high noradrenaline levels, which affects pain mechanisms [46]. In addition, it has been suggested that psychological factors that alter catecholamine activity and relate to emotional regulation may directly affect pain intensity and thus contribute to the maintenance of CRPS symptoms [47]. Thus, the above finding may be due to a higher sensitivity of persons with CRPS to catecholamine. Finally, findings support the notion that impaired affective consciousness is associated with somatosensory amplification, which is manifested in increased attention to bodily processes and increased physical sensations and pain [26].

We utilized a multidimensional assessment of pain, with the PRIS evaluating sensory, affective, and evaluative dimensions of pain and the VAS evaluating sensory pain. Significant associations between pain as assessed by the PRIS, but not VAS, and difficulty describing feelings and externally-oriented thinking were found among persons with CRPS, but not among those with LBP. These findings support the need for utilizing multidimensional assessments of pain in persons with CRPS. Two important pain dimensions that were not examined in the current study are pain threshold and tolerance [48]. Future studies of the relationship between pain and alexithymia should examine these pain dimensions in persons with CRPS, as well as the possibility that increased pain tolerance may substitute as expression of emotional distress. In addition, there was a significant difference between alexithymia (total), difficulty identifying feelings, and PRIS (but not VAS) correlations among persons with CRPS when compared to those with LBP. Thus, it may be that alexithymia and difficulty identifying feelings in persons with CRPS mainly involve affective and cognitive aspects of pain. This is in line with the notion that in the absence of symbolic functioning, a characteristic of persons with alexithymia, one has to use physical avenues to release inner tension [49,50]. Indeed, associations between alexithymia and development of functional somatic symptoms have been reported [14,51,52]. However, other studies did not detect such connections [53,54]. Current findings suggest that pain among persons with CRPS may be construed as a physical avenue to release pain or emotional tension that is not verbally expressed. Accordingly, in order to reduce pain among persons with CRPS, findings support the provision of care that focuses on generating alternative avenues for processing pain, such as promoting emotional self-awareness and learning to identify feelings. Finally, we found that the relationship between difficulty identifying feelings and pain as assessed by the PRIS and VAS was not confounded by psychological distress. This is consistent with research in pain populations that reported this scale to be the most consistent predictor of pain and pain-related dysfunction [55].

The current sample was composed mostly of men. Since CRPS is more common among women [56], generalizability may be limited. The number of persons with CRPS was small and replication studies using larger samples are warranted. These limitations notwithstanding, this study provides valuable information on the relationship between

alexithymia, psychological distress, and pain among persons with CRPS and is the first to report an association between alexithymia and pain in this population. From the perspective of conceptualizing alexithymia as an outcome of CRPS, findings bear practical implications with regards to rehabilitation efforts in this population. However, future longitudinal studies are needed in order to clarify the direction of causality between alexithymia and CRPS, and the value of alexithymia in predicting pain among persons with CRPS. Future neuro-imaging studies should examine if emotion-related brain regions operate differently among persons with CRPS and alexithymia.

Conflict of interests

The authors have no competing interests to report.

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