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MYOFASCIAL PAIN AND TREATMENT: Observational Study

Proximal myofascial pain in patients with distal complex regional pain syndrome of the upper limb

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ABSTRACT

Background: Patients suffering from complex regional pain syndrome (CRPS) endure myofascial-related pain in at least 50% of cases.
Aims: To evaluate the association of upper limb CRPS with myofascial pain in muscles that might influence arm or hand pain, and to evaluate whether the paraspinal skin and subcutaneous layers' tenderness and allodynia are associated with CRPS.
Methods: A case-control study comprising 20 patients presenting with upper limb CRPS, and 20 healthy controls matched for sex and age, were evaluated in the thoracic paraspinal area and myofascial trigger points (MTrPs) (infraspinatus, rhomboids, subclavius, serratus posterior superior and pectoralis minor) via a skin rolling test.
Results: The prevalence of MTrPs in the affected extremity of the subjects was significantly higher than in the right limb of the controls: 45% exhibited active and latent MTrPs in the infraspinatus muscle ($\chi^2 = 11.613$, $p = 0.001$); 60% in active and latent MTrPs in the subclavius muscle ($\chi^2 = 17.143$, $p < 0.001$); and in the pectoralis minor muscle ($\chi 2 = 13.786$, $p < 0.001$). In addition, 55% of the cases exhibited active and latent MTrPs in the serratus posterior superior muscle ($\chi^2 = 15.172$, $p < 0.001$). Significant differences between the groups in skin texture and pain levels ($p = 0.01$, $p < 0.001$, respectively) demonstrated that CRPS patients felt more pain, and their skin and subcutaneous layers were much tighter than in the healthy controls.
Conclusion: There is a high prevalence of MTrPs in the shoulder and upper thoracic area muscles in subjects who suffer from CRPS. We recommend adding an MTrPs evaluation to the standardized examination of these patients.

1. Introduction

Complex regional pain syndrome (CRPS) is characterized by continuing regional pain, disproportionate in time or degree compared to the usual course of a trauma or a lesion. The pain does not occur in a specific nerve territory or dermatome and usually presents with a distal predominance of abnormal sensory, motor, sudomotor, vasomotor, and/or trophic findings (IASP, 2011). CRPS is divided into two types: type 1 - no evidence of nerve injury; type 2 - electrodiagnostic or physical evidence of nerve lesions (Birklein et al., 2015; Wasner et al., 2003). The most recent retrospective cohort study carried out in the Netherlands reported an incidence of 26.2 new cases per 100,000 persons each year (de Mos et al., 2007). The upper limb is more frequently affected than the lower limb in adults, and women are more frequently affected than men (ratio of 3–4:1)(Sandroni et al., 2003; de Mos et al., 2007).

Differential diagnostic possibilities may include bone or soft tissue injury, arthritis, brachial neuritis, Reynaud's disease, thoracic outlet syndrome, Gardner-Diamond syndrome, lymphatic/venous obstruction, peripheral nerve injury, neuropathic pain, small fiber neuropathy, infections, compartment syndrome, and arterial insufficiency (Turner-Stokes and Goebel, 2011; Goebel et al., 2012). There are no diagnostic tests to support a positive CRPS diagnosis i.e., no MRI indications for CRPS, while plain x-rays are insensitive, and there are no serum markers. 3-phase bone scintigraphy may

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demonstrate an increased bone metabolism in the mineralization phase with moderate specificity and is sometimes considered a specific diagnostic test for diagnosing CRPS I in an upper limb (Wüppenhorst et al., 2010). However, the diagnostic value of 3-phase bone scintigraphy as a screening test or for confirmation of the CRPS I diagnosis formulated by the use of the Budapest criteria is lower (Moon et al., 2012; Wertli et al., 2017). Dynamic temperature differences (affected vs unaffected side) of 2 °C support the diagnosis of CRPS (Krumova et al., 2008).

According to the clinical guidelines, CRPS treatment should be interdisciplinary and include pain management, nursing, physical therapy, occupational therapy, psychotherapy and additional therapies as the situation requires (art therapy, recreational therapy, etc.) (Harden et al., 2006, 2013; Perez et al., 2010). The aim of the rehabilitation treatment program is to restore patient functions. Almost all patients with advanced CRPS will present with myofascial pain of a supporting joint (Harden et al., 2013).

Myofascial pain is "the complex of the sensory, motor and autonomic symptoms caused by myofascial trigger points (MTrPs)" (Simons et al., 1999). A MTrP is defined as a hyperirritable spot within a palpable taut band of a skeletal muscle that is painful on compression, stretch, or overload, and this gives rise to a referred pain. Furthermore, the spot is painful on compression and may produce characteristic referred pain and tenderness, motor dysfunction and autonomic phenomena. Two major types of MTrPs have been described: active and latent. Active MTrPs are associated with spontaneous complaints of pain. In contrast, latent MTrPs do not cause spontaneous pain, but pain may be elicited by manual pressure or by needling the MTrP. Despite not being spontaneously painful, latent MTrPs have been assumed to restrict ROM and alter motor recruitment patterns (Bron et al., 2007).

One recent review found that MTrPs in shoulder muscles is a common condition amongst patients with shoulder complaints and can be reliably diagnosed by palpation (Sergienko and Kalichman, 2014). Others have found a prevalence of myofascial dysfunction in more than 50% of CRPS patients (Rashiq and Galer, 1999; Allen et al., 1999). Treatment of myofascial pain may be an important component of a successful CRPS treatment (Allen et al., 1999; Harden et al., 2006, 2013).

The aims of our study were to evaluate the association of upper limb CRPS with myofascial pain in muscles that might influence arm or hand pain (infraspinatus, rhomboids, subclavius, serratus posterior superior and pectoralis minor) and to evaluate whether the paraspinal soft tissue (skin and subcutaneous layers) tenderness and allodynia are associated with CRPS in the upper limb.

2. Methods

Design: A case-control analytical study.

Setting: Department of Physical Therapy, Center for Rehabilitation of Pain Syndromes, Reuth Rehabilitation Hospital, Tel Aviv, Israel.

Sample: All CRPS subjects were selected from patients treated at the Center for Rehabilitation of Pain Syndromes, Reuth Rehabilitation Hospital. Included in the study were 20 patients (cases) diagnosed with CRPS of the upper limb (according to the "Budapest" clinical criteria), ≥6 months prior to the trial and were suitable according to the inclusion and exclusion criteria. Twenty healthy subjects comparable in sex and age to the CRPS group, without any symptoms present in the upper limb, were recruited from the hospital staff (controls) from April 2016 until August 2017.

Inclusions Criteria for "cases":

- Ages 18-70
- CRPS diagnosed by a board-certified specialist in rehabilitation or pain medicine according to the "Budapest" clinical criteria
- Constant chronic pain for ≥6 months preceding the trial
- Spontaneous pain on the day of the first evaluation (numeric pain rating scale (NPRS) ≥4/10)
- Stimulated pain was not attributable to other causes, such as peripheral inflammation

Inclusion Criteria for "controls":

- Ages between 18 and 70
- Did not suffer from any medical issues or chronic pain.

Exclusion criteria for all:

- Tumors
- Participation in another interventional clinical trial
- History of major psychiatric or neurological illness (i.e., stroke)
- Rheumatoid arthritis or other major rheumatologic disorders
- Fibromyalgia or any other systemic illnesses
- Pregnancy

Ethical considerations: Participation in the study was voluntary. Subjects were informed of the aims and procedures of the study and signed informed consent prior to inclusion in the study. The study was approved by the Ben Gurion University thesis committee and the ethical (Helsinki) committee of the Reuth Rehabilitation Hospital.

Allocation: The subjects were recruited into two groups: upper limb CRPS patients (cases) and age and sex-matched healthy subjects with no symptoms in the upper limbs (controls).

2.1. Procedure

Each subject filled out two questionnaires: Demographic data, and the Disabilities of the Arm, Shoulder, and Hand (DASH). CRPS patients were examined by a board-certified specialist in rehabilitation medicine to rate their CRPS severity score. Subsequently, the examiner performed a skin rolling test in the thoracic paraspinal area, and examined the relevant muscles for MTrPs.

2.2. Outcome measures

Demographic data: age, sex, occupation, leisure physical activity, health history, data on the present disease (onset of symptoms, previous treatments, etc.) and medications were collected on the initial examination.

DASH questionnaire: a 30-item questionnaire in which the patients reported their abilities to perform certain upper extremity activities. The patients rated the difficulty and interference with daily life on a 5-point Likert scale. Though not CRPS specific, this questionnaire is, however, frequently used in studies involving CRPS patients (Grieve et al., 2015). The questionnaire has been found to be reproducible, valid and reliable for proximal and distal disorders in the upper limb (ICC = 0.96, r > 0.69) (Beaton et al., 2001a,b). The cross-cultural adaptation of the questionnaire to Hebrew was previously performed by Efrat Ziv, Ety Shiwmer, Yael Kaufman and Dr. Hagar Patish (with the authorization of the authors).

The CRPS severity scale (CSS): The patients were evaluated by a physician, including an interview and physical examination (Harden et al., 2010a,b). The scale measured the CRPS markers according to the diagnostic "Budapest Criteria" which has been found reliable (alpha = 0.88) (Harden et al., 2017).

Numerical Pain Rating Scale (NPRS): This scale is a one-

dimensional measure of pain intensity in adults and a valid and reliable tool to assess pain (Childs et al., 2005; Downie et al., 1978; Ferreira-valente et al., 2011; Jensen and McFarland, 1993). This pain scale is not CRPS specific, but is very often used in studies involving CRPS patients (Grieve et al., 2015).

Skin rolling test: Normally, the skin rolls over the back freely and painlessly. When a patient is subjected to paravertebral skin rolling, the examiner may feel a tissue texture change in the form of tightness (the skin is less pliable, more difficult to raise, and resists rolling) or autonomic dysfunction (such as localized warming or increased sweating). The patient may experience pain (allodynia) in the area associated with pathological changes. The skin rolling test has been found reliable by Taylor (Taylor et al., 1990). The examination procedure was as follows: the side and spinal levels of skin tightness or pain were recorded (Banssevicius et al., 1997). The thoracic area was divided into 3 areas: upper (T1-T3), middle (T4-T7), and lower thorax (T8-T12). The examiner noted if the patient reported pain or tenderness during the test, and whether the skin texture was free or tight. Pain was recorded on a 4-level scale; 0 = no pain, 1 = tenderness, 2 = medium and 3 = strong pain. Skin texture was recorded as normal, hard, or very hard ("lemon peel").

MTrPs evaluation: The following muscles were examined for MTrPs: infraspinatus, rhomboids, subclavius, serratus posterior superior, and the pectoralis minor. The examiner ascertained the presence of active, latent, or no MTrPs in the aforementioned muscles.

Statistical analysis: All statistical analysis was performed using the SPSS statistical package (Version 17). Significance levels were set at $P < 0.05$. Descriptive statistics were used to characterize the study sample. The comparison of prevalence of MTrPs, paraspinal soft tissue tightness, and tenderness or painfulness (ordinal variables) was performed using the Chi-square and McNemar tests.

3. Results

3.1. Subject demographics

Our subjects were mostly male (65%), average age 35.35 ± 9.58 in the case group, 35.15 ± 10.36 in the controls, with no significant difference in age, sex, and dominant hand (Table 1). There was a significant difference between the groups in hand pain score (the case group suffered from hand pain while the controls did not). The DASH score indicated that the controls' hand function was good, whereas the case group had difficulties with hand functions. There was also a significant difference in the years of education between groups (cases - 12.4 ± 3.63; controls - 15.85 ± 3.77. The case group exhibited 50% injuries to the right hand (90% of the patients had CRPS type 1).

3.2. Association of paraspinal soft tissue tenderness and allodynia with upper limb CRPS

Significant differences in all thoracic areas when comparing pain levels between CRPS patients and healthy controls ($p < 0.001$; Table 2) were revealed in the skin rolling test. When comparing the skin texture between the CRPS and the controls, we found statistically significant differences in the upper, middle and lower thorax ($p = 0.01$, $p = 0.001$, $p = 0.021$ respectively), indicating that patients with upper limb CRPS not only felt more pain, but their skin and subcutaneous layers were much tighter than those of the healthy controls.

3.3. Association of upper limb CRPS with myofascial pain in muscles that might influence arm or hand pain

MTrPs evaluations are detailed in Tables 3–5. Table 3 shows the results of MTrPs evaluation in the control group when comparing right and left side muscles. The controls did not exhibit any active or latent MTrPs except for one healthy subject with a latent MTrP in the pectoralis minor muscle and the rhomboids. However, the subjects in the control group did have tender muscular points in all examined muscles with no significant difference between the sides of the body.

Table 4 compares the MTrPs in the case group when comparing the affected and unaffected sides. A significant difference in the total sum of MTrPs between sides in the infraspinatus ($p = 0.016$), subclavius ($p = 0.002$) and pectoralis minor muscles ($p = 0.004$) was found. There was also a significant difference in the tender points of the rhomboids ($p = 0.002$). All other comparisons were not significant.

Table 5 compares the MTrP prevalence between the right side in the controls and the affected side in the cases showing that 45% of the cases had active and latent MTrPs in the infraspinatus muscle ($\chi^2 = 11.613$, $p = 0.001$); 60% had active and latent MTrPs in the subclavius muscle ($\chi^2 = 17.143$, $p < 0.001$) and in the pectoralis minor muscle ($\chi 2 = 13.786$, $p < 0.001$). In addition, 55% of cases had active and latent MTrPs in the serratus posterior superior muscle ($\chi^2 = 15.172$, $p < 0.001$).

Table 1
Descriptive statistics of the studied sample.

	Cases (N = 20)	Controls (N = 20)	Comparison
	mean ± SD	mean ± SD	*t*-test
Age (years)	35.35 ± 9.58	35.15 ± 10.36	t = 0.63, d.f. = 38. P = 0.950
DASH*	73.65 ± 12.72	1.00 ± 1.92	**t = 25.26, d.f. = 38. P < 0.001**
NPRS*	8.00 ± 3.13	0.00 ± 0.00	**t = 11.43, d.f. = 38. P < 0.001**
CSS*	11.60 ± 2.06	–	–
Duration of symptoms (yrs)	1.61 ± 1.81	–	–
Education (yrs)	12.4 ± 3.63	15.85 ± 3.77	**t = -2.945, d.f. = 38. P = 0.005**
	N (%)	**N (%)**	**χ^2-test**
Gender (males)	13 (65.0%)	13 (65.0%)	$\chi^2 = 0.00$, d.f. = 1, p = 1.000
Dominant hand-right	17 (85%)	20 (100%)	p = 0.231 (Fisher's exact test)
Injured hand - right	10 (50%)	–	–
CRPS type 1*	18 (90%)	–	–
CRPS type 2*	2 (10%)	–	–

* DASH- disabilities of arm shoulder and hand questionnaire; NPRS- numerical pain rating scale; CSS- CRPS severity scale; CRPS type 1- without nerve lesions, type 2- with nerve lesions; d.f. = degrees of freedom; statistically significant ($p < 0.05$) differences marked bold.

Table 2
Results of the skin rolling test.

Variable	Thorax area	Grade	Cases N (%)	Controls N (%)	Comparison χ^2-test
Pain*	Upper (T1-T3)	0	3 (15%)	17 (85%)	**$\chi^2 = 22.30$, d.f. = 3, $p < 0.001$**
		1	5 (25%)	3 (15%)	
		2	9 (45%)	0 (0%)	
		3	3 (15%)	0 (0%)	
	Middle (T4-T7)	0	4 (20%)	19 (95%)	**$\chi^2 = 23.35$, d.f. = 3, $p < 0.001$**
		1	6 (30%)	1 (5%)	
		2	9 (45%)	0 (0%)	
		3	1 (5%)	0 (0%)	
	Lower (T8-T12)	0	7 (35%)	20 (100%)	**$\chi^2 = 19.26$, d.f. = 3, $p < 0.001$**
		1	8 (40%)	0 (0%)	
		2	4 (20%)	0 (0%)	
		3	1 (5%)	0 (0%)	
Texture*	Upper (T1-T3)	0	5 (25%)	14 (70%)	**$\chi^2 = 9.26$, d.f. = 2, $p = 0.010$**
		1	12 (60%)	6 (30%)	
		2	3 (15%)	0 (0%)	
	Middle (T4-T7)	0	7 (35%)	17 (85%)	**$\chi^2 = 10.42$, d.f. = 1, $p = 0.001$**
		1	13 (65%)	3 (15%)	
		2	0 (0%)	0 (0%)	
	Lower (T8-T12)	0	10 (50%)	18 (90%)	**$\chi^2 = 7.74$, d.f. = 2, $p = 0.021$**
		1	9 (45%)	2 (10%)	
		2	1 (5%)	0 (0%)	

* Grades of pain: 0- no pain, 1-tenderness, 2-medium pain, 3-strong pain; Grades of texture: 0-normal, 1-hard, 2-very hard ("lemon peel"); d.f.- degrees of freedom; statistically significant ($p < 0.05$) differences marked in bold.

Table 3
Results of MTrPs evaluation in subjects without CRPS (control group, N = 20).

MTrPs	Right	Left	Right vs. Left (McNemar's Test)
Latent MTrP Infraspinatus	0	0	
Active MTrP Infraspinatus	0	0	
Total MTrP Infraspinatus	0	0	
Tender point Infraspinatus	1 (5%)	4 (20%)	p = 0.250
Latent MTrP Subclavius	0	0	
Active MTrP Subclavius	0	0	
Total MTrP Subclavius	0	0	
Tender point Subclavius	7 (35%)	8 (40%)	p = 1.000
Latent MTrP Pectoralis minor	1 (5%)	0	
Active MTrP Pectoralis minor	0	0	
Total MTrP Pectoralis minor	1 (5%)	0	
Tender point Pectoralis minor	8 (40%)	8 (40%)	p = 1.000
Latent MTrP Rhomboids	1 (5%)	0	
Active MTrP Rhomboids	0	0	
Total MTrP Rhomboids	1 (5%)	0	
Tender point Rhomboids	5 (25%)	6 (30%)	p = 1.000
Latent MTrP SPS	0	0	
Active MTrP SPS	0	0	
Total MTrPSPS	0	0	
Tender point SPS	10 (50%)	6 (30%)	p = 0.219

* Latent MTrP = latent myofascial trigger point; Active MTrP = active myofascial trigger point; Total MTrP = summation of active and latent MTrPs; Tender point = localized muscular tender point; statistically significant ($p < 0.05$) differences marked in bold. SPS = serratus posterior superior.

4. Discussion

Our results confirm those of previous studies (Allen et al., 1999; Harden et al., 2013; Kharkar et al., 2011; Rashiq and Galer, 1999; Safarpour et al., 2010) that found a higher prevalence of MTrPs in the thoracic area in patients with CRPS in the upper limb. In another study, the presence or absence of a myofascial component was determined by palpating for MTrPs in the proximal musculature, such that when MTrPs were present, they reproduced some or all of the patient's reported symptoms. In one of these studies, the authors found that 40/58 (69%) of the upper extremity CRPS patients exhibited myofascial dysfunction (Allen et al., 1999). They also suggested that the duration of CRPS symptoms and the involvement of the upper extremity was significantly associated with the presence of myofascial dysfunction. There was no description of the MTrPs evaluation and in which muscles they were found, except for a general statement that they examined the shoulder girdle muscles; hence, it is not possible to compare their results to ours.

In 2013, Harden et al. (2013) postulated that virtually all patients with advanced CRPS will present with myofascial pain syndrome of the supporting joint. The authors claimed that aggressive treatment of this myofascial pain is a critical component of successful treatment. Moreover, it has been suggested that hands-on techniques such as massage and myofascial release can offer effective relief from the pain (Simons et al., 1999). Kharkar et al. (2011) treated upper limb CRPS patients with botulinum toxin injections into the following muscles: trapezius, splenius capitis, levator scapulae, longissimus capitis, obliqus capitis, scalene, semispinalis, sternocleidomastoid, and the paraspinal muscles. They asserted that after the injections, the patients reported pain relief, thus concluding that intramuscular injections of botulinum toxin into the upper limb girdle muscles was beneficial for short-term relief of pain in

Table 4
Results of MTrPs evaluation in subjects with CRPS (case group, N = 20*).

MTrPs	Affected side	Unaffected side	Affected vs. unaffected (McNemar's Test)
Latent MTrP Infraspinatus	4 (20%)	2 (10%)	p = 0.687
Active MTrP Infraspinatus	5 (25%)	0	
Total MTrP Infraspinatus	9 (45%)	2 (10%)	**p = 0.016**
Tender point Infraspinatus	6 (30%)	4 (20%)	p = 0.727
Latent MTrP Subclavius	5 (25%)	2 (10%)	p = 0.453
Active MTrP Subclavius	7 (35%)	0	
Total MTrP Subclavius	12 (60%)	2 (10%)	**p = 0.002**
Tender point Subclavius	2 (10%)	5 (25%)	p = 0.453
Latent MTrP Pectoralis minor	6 (30%)	3 (15%)	p = 0.453
Active MTrP Pectoralis minor	6 (30%)	0	
Total MTrP Pectoralis minor	12 (60%)	3 (15%)	**p = 0.004**
Tender point Pectoralis minor	2 (10%)	4 (20%)	p = 0.625
Latent MTrP Rhomboids	1 (5%)	1 (5%)	p = 1.000
Active MTrP Rhomboids	3 (15%)	0	
Total MTrP Rhomboids	4 (20%)	1 (5%)	p = 0.250
Tender point Rhomboids	12 (60%)	2 (10%)	**p = 0.002**
Latent MTrP SPS	5 (25%)	0	
Active MTrP SPS	6 (30%)	0	
Total MTrP SPS	11 (55%)	0	
Tender point SPS	5 (25%)	8 (40%)	p = 0.508

* The table is divided into the affected and unaffected hand (only patients with CRPS were included in this table, therefore 20 cases are presented).
** Latent MTrP = latent myofascial trigger point; Active MTrP = active myofascial trigger point; Total MTrP = summation of active and latent MTrPs; Tender point = localized muscular tender point; statistically significant ($p < 0.05$) differences marked in bold. SPS = serratus posterior superior.

Table 5
Comparison of MTrPs prevalence between the right side in the control group (N = 20) and the affected side in the case group (N = 20*).

Muscle		Right limb controls (N = 20)	Affected limb cases (N = 20)	Comparison Right vs. affected*
Infraspinatus	Latent MTrP	0	4 (20%)	**$\chi^2 = 4.444$, d.f. = 1, p = 0.035**
	Active MTrP	0	5 (25%)	**$\chi^2 = 5.714$, d.f. = 1, p = 0.017**
	Total MTrP	0	9 (45%)	**$\chi^2 = 11.613$, d.f. = 1, p = 0.001**
	Tender point	1 (5%)	6 (30%)	**$\chi^2 = 4.329$, d.f. = 1, p = 0.037**
Subclavius	Latent MTrP	0	5 (25%)	**$\chi^2 = 5.714$, d.f. = 1, p = 0.017**
	Active MTrP	0	7 (35%)	**$\chi^2 = 8.485$, d.f. = 1, p = 0.004**
	Total MTrP	0	12 (60%)	**$\chi^2 = 17.143$, d.f. = , p < 0.001**
	Tender point	7 (35%)	2 (10%)	$\chi^2 = 3.584$, d.f. = 1, p = 0.058
Pectoralis minor	Latent MTrP	1 (5%)	6 (30%)	**$\chi^2 = 4.329$, d.f. = 1, p = 0.037**
	Active MTrP	0	6 (30%)	**$\chi^2 = 7.059$, d.f. = 1, p = 0.008**
	Total MTrP	1 (5%)	12 (60%)	**$\chi^2 = 13.789$, d.f. = 1, p < 0.001**
	Tender point	8 (40%)	2 (10%)	**$\chi^2 = 4.800$, d.f. = 1, p = 0.028**
Rhomboids	Latent MTrP	1 (5%)	1 (5%)	$\chi^2 = 0.000$, d.f. = 1, p = 1.000
	Active MTrP	0	3 (15%)	$\chi^2 = 3.243$, d.f. = 1, p = 0.072
	Total MTrP	1 (5%)	4 (20%)	$\chi^2 = 2.057$, d.f. = 1, p = 0.151
	Tender point	5 (25%)	12 (60%)	**$\chi^2 = 5.013$, d.f. = 1, p = 0.025**
Serratus posterior superior	Latent MTrP	0	5 (25%)	**$\chi^2 = 5.714$, d.f. = 1, p = 0.017**
	Active MTrP	0	6 (30%)	**$\chi^2 = 7.059$, d.f. = 1, p = 0.008**
	Total MTrP	0	11 (55%)	**$\chi^2 = 15.172$, d.f. = 1, p < 0.001**
	Tender point	10 (50%)	5 (25%)	$\chi^2 = 2.667$, d.f. = 1, p = 0.102

* Results of chi-square test; Latent MTrP = latent myofascial trigger point; Active MTrP-active myofascial trigger point; Total MTrP = summation of active and latent MTrPs; Tender point = localized muscular tender point; statistically significant ($p < 0.05$) differences marked in bold.

patients with CRPS who presented with spasm or dystonia in the neck and shoulder girdle muscles, secondary to CRPS.

Rashiq and Galer (1999) examined the MTrPs in the proximal muscles of CRPS patients. Muscles examined in the upper extremity were the scalene, deltoids, infraspinatus, supraspinatus, latissimus dorsi, subscapularis, serratus anterior and posterior and trapezius. The diagnostic criteria for myofascial dysfunction included the presence of a palpable, tender, firm nodule or band in the affected muscle (MTrP), tenderness in the taut muscle, exacerbation of the patient's pain complaint or altered sensation in the (distal) area of the CRPS caused by palpation and restricted ROM. The muscles were palpated with the index finger. The authors examined only patients with CRPS of the hand or foot and assessed the prevalence of clinically evident myofascial dysfunction and its relationship to motor neglect in CRPS. Myofascial dysfunction was detected in 61% of the CRPS patients and was more prevalent in the upper limb (70%) than in the lower (47%). One of their recommendations was to examine a comparative control group of normal individuals for MTrPs. To the best of our knowledge, there are no studies in the literature that have compared MTrPs prevalence in these two populations.

Safarpour et al. (2010) published a report of two cases: a patient with hand CRPS was reported as presenting with CRPS signs and symptoms, along with significant right neck and shoulder pain with many tender spots and MTrPs over the left trapezius and upper back muscles (developed gradually, months after the development of allodynia on the same side), and intermittent painful flexor spasms of the three middle fingers of the right hand. She was injected with botulinum toxin into the MTrPs located in the right trapezius, rhomboid, levator scapulae, and right flexor digitorum superficialis. The second patient exhibited several tender areas and MTrPs over the left trapezius muscle, the left side of the neck and

the upper rhomboid muscles. Upon pressing the MTrPs, a tight muscle band was felt under the finger and the pain referred to distant areas from the pressure point. Botulinum toxin was injected into the MTrPs of trapezius, splenius capitis, and rhomboid muscles. The number of injected MTrPs were 4, 3, and 5 for the aforementioned muscles, respectively. Both patients reported a decrease in pain and were able to fully participate in activities of daily living. In these two cases, proximal myofascial pain syndrome developed ipsilaterally to the distal painful limb in patients with CRPS type 1. The administration of botulinum toxin-A into the affected proximal muscles alleviated both the MTrPs and the distal allodynia, discoloration, and tissue swelling. These results confirm our assumption that a more focused MTrP treatment would be more beneficial for CRPS patients than the general soft tissue manipulation treatment.

In his 2002 review, Argoff described 11 patients with CRPS type 1 and proximal myofascial pain syndrome who received an intramuscular injection of botulinum toxin-A into the thoracic and neck muscles, relieving myofascial pain and distal symptoms (pain, swelling, tissue discoloration). The patients continued the treatment with consistent relief of their symptoms for approximately four years (Argoff, 2002). Several articles have suggested that there is a myofascial component in the proximal muscles of CRPS patients, but only some have explained how the MTrPs were examined, or in which muscles, making it impossible to compare their results to ours. We chose the examined muscles since they affect the patients' posture. According to Simons et al. (1999), active MTrPs will refer the pain into the hand area. We suggest that additional research in a larger sample should be performed to examine these muscles.

In our study, we found the presence of myofascial pain syndrome in patients with upper limb CRPS. However, it is unclear which syndrome preceded the other. Other studies have suggested that myofascial pain develops secondary to CRPS due to pain and disuse of the involved limb (Allen et al., 1999; Harden et al., 2013); however, they cannot rule out the possibility that myofascial pain syndrome was the primary syndrome in certain patients. It is reasonable to assume that CRPS precedes myofascial pain syndrome when considering that in many CRPS patients' disuse of the limb and severe guarding may contribute to secondary proximal myofascial pain that can mimic the spread of the disorder (and further increase fear). Albeit that myofascial pain is secondary to CRPS, it can cause some of the relevant pain and/or serve as a perpetuating factor (Allen et al., 1999) and as such, obstruct recovery.

On the other hand, it is theoretically possible that existing myofascial pain syndrome serves as a facilitating factor or even a trigger for the development of CRPS. Hong (2000) presented a case study of a 25-year-old man after a traumatic supraspinatus tendon tear in his left shoulder. He underwent physical therapy, but his symptoms worsened and spread distally. His left hand swelled and he experienced episodic cold sensations with skin discoloration. He was sensitive to touch and was unable to move his upper limb due to the pain. After 6 months of various treatments and medications, he received focused MTrPs treatment, first by gentle massage as a desensitization technique, then with modalities (hot packs, ultrasound, electrotherapy for muscle stimulation) and mobilizations. The treatment was performed from distal to proximal. His symptoms began to decrease, and subsequently he received a steroid injection to the shoulder. Coupled with MTrPs treatment 2–3 times a week for 2 months and a gradual activation at home, the patient was virtually pain-free with nearly full ROM (Hong, 2000). It is possible that in this case study, CRPS was secondary to myofascial pain syndrome. However, it is important to note that most CRPS patients experience trauma in the distal part of the limb, not the proximal part, and for most, the symptoms start distally and gradually progress to the proximal part of the limb, not the reverse.

Regardless of whether myofascial pain is primary or secondary, it is important to be aware of early and gradual mobilization following soft-tissue injury, especially following surgery or trauma. Moreover, it is important to examine CRPS patients for MTrPs and treat appropriately. In our opinion it is essential to examine whether myofascial pain syndrome is secondary to CRPS. This will help us to fully understand the pathological picture of CRPS and formulate a more effective treatment of this syndrome.

When comparing CRPS patients and healthy subjects, we found significant differences in the rate of pain and texture of the skin in the thoracic area. The patients reported more pain and less flexible skin. Oaklander et al. identified significantly lower densities of epidermal neurites (up to 29% lower) in CRPS-affected limbs in relation to contralateral, unaffected limbs, with these changes primarily affecting nociceptive fibers. Similar differences in neurite density were not noted when comparing the affected and unaffected limbs of patients with painful unilateral non-CRPS conditions such as osteoarthritis (Oaklander et al., 2006). A difference in nerve density in the skin could affect skin pain and texture.

Another explanation for thoracic skin pain during a skin rolling test could be linked to the assumption that central sensitization by inflammatory cytokines might contribute to pain in CRPS. This is supported by findings that show increased interleukin-1β and interleukin-6 cytokine concentrations in the spinal fluid of patients with chronic CRPS (Alexander et al., 2005, 2007). Central sensitization results in exaggerated responses to nociceptive stimuli (hyperalgesia) and enables normally non-painful stimuli such as a light touch or cold to activate nociceptive pathways (allodynia)(Bruehl, 2017). These phenomena gradually develop from the affected region to other, proximal parts.

Endothelial changes may also influence peripheral circulation in chronic CRPS. Endothelial dysfunction is linked to a decreased ability to release endothelial nitric oxide, leading to continuous vasoconstriction. Reduced concentrations of nitric oxide have been observed in CRPS, both directly (impaired concentrations of nitric oxide in suction blister fluid of the affected side compared with the unaffected side)(Dayan et al., 2008; Groeneweg et al., 2006) and indirectly (impaired acetylcholine-induced vasodilatation of the impaired side compared with the unaffected side and compared with control individuals)(Schattschneider et al., 2006). It is unclear whether these changes contribute to the development of CRPS or are a result of the trophic alterations affecting the skin, muscles, and bones in individuals with CRPS.

All these studies describe, to a certain extent, the differences in skin texture and pain and sensitivity in the thoracic area of the upper limb in CRPS patients. It is essential to continue investigating whether more factors are involved in these changes and whether desensitization and MTrPs treatment to the area can alter the differences in skin formation in these patients.

It is important to note that the pathophysiological mechanism of CRPS appears to be attributable to multiple factors, that may include peripheral and central sensitization, inflammation, altered sympathetic and catecholaminergic function, decreased representation of the affected limb in the somatosensory cortex, genetic factors, and psychophysiological interactions. Furthermore, the degree of contribution of specific mechanisms to CRPS through time may vary from one patient to another and even within a single patient. Processes influence each other and contribute to varied clinical symptoms (Birklein et al., 2015; Bruehl, 2017; Bussa et al., 2015). Therefore, an interdisciplinary treatment plan is essential for CRPS patients. We examined the prevalence of MTrPs in upper limb CRPS patients, as a means of addressing some of the impairments the patients suffer from, such as decreased ROM in the

proximal regions, and incorrect posture.

5. Limitations

This study has a few limitations. Firstly, our study encompassed a relatively small number of subjects. Even though CRPS is not a common syndrome, it would have further enhanced the study if we had recruited more subjects. Furthermore, we did not document the allodynic areas of each subject. Some of our subjects developed allodynia only in the palm area, while others developed proximal allodynia in the forearm, arm, and shoulder. This is important to mention since it could have affected thoracic tightness and pain. In addition, our outcome measures were mostly subjective; some were reliant upon the examiner's skill and experience. However, pain is subjective and there is no objective way to measure it in a clinic, hence, we chose to assess the patient with commonly available tools accessible in every clinic.

6. Conclusions

The current study found that there is a higher prevalence of MTrPs in the shoulder and upper thoracic area muscles in those who suffer from CRPS compared to healthy subjects. This confirms the conclusions of previous studies (Allen et al., 1999; Harden et al., 2013; Kharkar et al., 2011; Rashiq and Galer, 1999; Safarpour et al., 2010) regarding the high prevalence of myofascial pain syndrome in CRPS patients. It is necessary to corroborate this conclusion using larger samples, however, in our opinion, it can potentially contribute to the understanding of the etiology and pathogenesis of CRPS and subsequently improve the treatment strategies. Therefore, we recommend that further studies examine the relationship between myofascial pain and CRPS and the prevalence of MTrPs in other neck and thoracic muscles in CRPS patients. We also recommend adding an MTrPs evaluation to the routine examination of CRPS patients.

Conflicts of interest

None.

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