

Adults with Complex Regional Pain Syndrome Type 1 Involving the Upper Extremity: A Novel Biokinesio-Psychosocial Movement System Diagnostic Classification Approach

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Abstract

Complex regional pain syndrome type 1 (CRPS Type 1) is a painful and often disabling condition that encompasses an array of complex pathophysiological mechanisms. Along with an assortment of various diagnostic symptoms, CRPS Type 1 generally encompasses pathological centrally and/or peripherally based motor symptoms, including dystonia, bradykinesia, tremor, perceptual and neglect-like symptoms, deconditioning, reduced range of motion, along with others. These motor symptoms can commonly lead their way to movement system dysfunctions. Although irretractable pain is the most frequent and intense symptom experienced in patients with CRPS Type 1, motor symptoms and the movement system are the primary content explored within this paper. Notably, the following review focuses its attention on CRPS Type 1 in the upper extremity of the adult population. The author of this paper has introduced the novel term of the biokinesio-psychosocial model, and is suggested to embrace the movement system in a richer and more inclusive light. The main distinguishing factor between the biopsychosocial model and the biokinesio-psychosocial framework is the introduction of a kinesiological approach towards examining and treating a dysfunctional movement system, specifically in patients with CRPS Type 1. In addition, the review pays attention to biological/physiological, psychological and social variables that importantly contribute to the diagnosis and treatment of CRPS Type 1. A description of the movement system in relation to CRPS Type 1 is also provided along with an example and critique of a few current existing measures that may be used to assess various motor impairments in patients with CRPS Type 1. Whilst concentrating on the central nervous system as a key perpetrator towards various motor impairments, the review also focuses on psychosocial elements that contribute to the overall experience of a defective movement system in patients with CRPS Type 1. Finally, the review describes the notion of movement system diagnoses or classification systems in relation to CRPS Type 1. With an emphasis on the biokinesio-psychosocial paradigm, 3 newly established movement system diagnoses or classification schemes in relation to adult patients with CRPS Type 1 of the upper extremity, are produced and described in terms of how and when to use them.

Keywords: Complex Regional Pain Syndrome Type 1 (CRPS Type 1), Chronic Pain (CP), Movement System Examination, Kinesiology, Movement System Diagnoses (MSDxs), Biopsychosocial Model, Biokinesio-psychosocial Model

Introduction

Complex regional pain syndrome type 1 (CRPS Type 1) is a painful and often disabling syndrome of complex multifaceted pathophysiology that can occur unexpectedly or after trauma or surgery, without identifiable nerve damage, generally to a limb [1,2]. The condition is characterized by various combinations of sensory, autonomic and motor abnormalities [1-3]. These changes occur resulting from a myriad of dysfunctional effects on the central nervous system (CNS), and/or the peripheral nervous system (PNS) and/or the autonomic nervous system (ANS), as well as a conglomeration of pathophysiological processes occurring in other intricate physiological and anatomical systems. In addition, we cannot ignore the connection between the above pathophysiological processes ensuing in patients with CRPS Type 1 and the numerous imperative variables associated with

patients' psychosocial mechanisms impacting on their CRPS Type 1 experience as a whole; the particularly popular biopsychosocial model. However, the aim of this review is not to dissect each pathological process mentioned above. The paper, nonetheless, does take a slightly deeper dive into some of the central mechanisms and psychosocial variables at play within a patient with CRPS Type 1, specifically when analyzing the movement system (the motor and movement system being the central topic in this review). A medical diagnosis of CRPS Type 1, according to the International Association of the Study of Pain (IASP), is made on the foundation of on-going pain and dysfunction in numerous body systems, including the motor system based on reorganization of the primary motor cortex (M1) [5].

Table 1 reveals the most up to date signs and symptoms required for

a clinical diagnosis of CRPS Type 1 [3]. The motor and movement disorders forming part of the overall diagnosis for CRPS Type 1, as revealed in Table 1, will be the focus of this paper, highlighting the symptom of dystonia. Approximately 25% of patients with CRPS Type 1 suffer from movement disorders including loss of voluntary control, bradykinesia, dystonia, myoclonus and tremor [4]. In comparison to the percentage of 25% of patients with CRPS Type 1 suffering from some movement disorder [4], Di Pietro (2013) refers to the prevalence of movement dysfunction in CRPS Type 1 being even higher [5]. One study reports movement disorders being present in 97% of their sample [6], and another study revealed movement disorders being evident in 65% of their sample experiencing CRPS Type 1 [7]. In addition to the above movement disorders, an individual may present with changes in localized (segmental) or global strength, range of motion and overall mobility. This may be due to both primary mechanisms and secondary mechanisms, as well as deconditioning resulting from the pathology. Furthermore, due to the presenting condition, musculoskeletal and cardiopulmonary endurance may also be compromised.

Sahrman (2014) states that ideal movement is the result of the relationship between the skeletal, muscular, nervous, cardiovascular, pulmonary, integumentary, immune, endocrine and other physiological systems (based on the 2015 American Physical Therapy Association-APTA- white paper) [8]. Van Hilten (2010) goes on to state that movement disorders, as described above, are tremendously difficult to manage and contribute substantially due to the weight of the condition [4]. Therefore, when reviewing the quantity and quality of the signs and symptoms of CRPS Type 1, specifically paying reference to the movement system for the purpose of this review, it may be critical to incorporate a movement system-based examination with definitive validated and reliable movement system assessment tools or classification schemes for adult patients with CRPS Type 1.

Irrespective of the attention paid by this review on the movement system, it is still of the utmost importance to embrace APTA's vision, as well as many other medical organizations' missions, being that rehabilitation clinicians should treat all factors that

hamper movement whether it's pain, neuromuscular dysfunction, psychosocial factors and comorbid medical conditions [9]. Thus, a biopsychosocial model (biokinesio-psychosocial model as will be described further on) surrounding patient evaluation and treatment should always be implemented.

The following review will pay attention to analyses of the movement system pertaining to CRPS Type 1 involving particularly the upper extremity in the adult population. Various articles surrounding the movement system and analysis with regards to pathology of CRPS Type 1 will be presented. Importantly, the author of this paper introduces a novel term, that being a 'biokinesio-psychosocial' model that is suggested to be incorporated into the assessment and management of patients with CRPS Type 1 and potentially other chronic pain (CP), complex pain and other medical conditions. As such, the paper will also describe what a clinician should analyze during the examination of each essential activity as well as consider the criteria that are utilized to measure and report the results of the movement system examination. The review will look at an array of existing standardized measures which may be appropriate to utilize within such an examination. In addition, the author suggests how certain tools may be adjusted to be directed more exclusively towards movement within patients presenting with CRPS Type 1. In turn, this paper attempts to create movement system based diagnostic classification systems for CRPS Type 1, again paying specific attention to adults with CRPS Type 1 of the upper limb. For the purpose of this paper, the focus will be on the classic movement system disorders, particularly dystonia, which is regularly seen in CRPS Type 1. In addition, with reference to the developed classification systems, the review will provide movement system diagnostic names. The newly introduced diagnostic names produced through this review will try to describe the problem in a way that will direct evidence-based treatment possibilities, particularly sticking within the boundaries of Physical Therapy (PT) and other similar movement-based health care professions. Finally, as mentioned, the current review paper specifically focuses on CRPS Type 1 in the adult population, however, very intermittently provides passing information regarding the pediatric population.

A. The patient has continuing pain which is disproportionate to any inciting event			<input type="checkbox"/>
B. The patient reports at least one symptom in 3 or more of the categories			<input type="checkbox"/>
C. The patient displays at least one sign in 2 or more of the categories			<input type="checkbox"/>
D. No other diagnosis can better explain the signs and symptoms			<input type="checkbox"/>
Category		Symptom (the patient reports a problem)	Sign (you can see or feel a problem on examination)
1 "Sensory"	<i>Allodynia</i> (to light touch/brush stroke and/or temperature sensation and/or deep somatic pressure and/or joint movement), and/or <i>hyperalgesia</i> (to pinprick)	Reported hyperesthesia also qualifies as a symptom <input type="checkbox"/>	<input type="checkbox"/>
2 "Vasomotor"	Temperature asymmetry and/or skin colour changes and/or skin colour asymmetry	<input type="checkbox"/>	<input type="checkbox"/>
3 "Sudomotor/oedema"	Oedema and/or sweating changes and/or sweating asymmetry	<input type="checkbox"/>	<input type="checkbox"/>
4 "Motor/trophic"	Decreased range of motion and/or motor dysfunction (weakness, tremor, dystonia) and/or trophic changes (hair/nail/skin)	<input type="checkbox"/>	<input type="checkbox"/>

Abbreviations: IASP (International Association for the Study of Pain).

Table 1: New IASP Diagnostic Criteria for CRPS Type 1 ("Budapest Criteria") (A to D must apply).

Motor and Movement System Dysfunctions in Patients with CRPS Type 1

Brun et al, (2019) explored the relationships between altered body perception, sense of limb position, and limb movement in individuals with CRPS Type 1 [10]. The 2 central aims of this study were to firstly assess patients' sense of abnormal limb movement during active and no intentional movement and secondly to investigate possible connections between body perception, the senses of limb movement and limb position, and pain in the CRPS Type 1 affected limb. Previous investigations into that of Brun et al (2019) focused their attention specifically on body perception and the sense of limb position [11]. Therefore, Brun et al's (2019) research seems to embrace the movement system aspect when examining patients with CRPS Type 1. The results to this study indicated that the sense of limb position and limb movement were altered in the painful limb of patients with CRPS Type 1 compared to the control group within the research [10]. These findings are in accordance with the theory that body representations are distorted in the presence of pain, as identified by behavioral and neuroimaging studies commenting on changes in motor and sensory cortical regions in CRPS Type 1 [11]. Importantly, research has concluded that alterations in kinesthesia were not correlated with the subjects' reported perceptions of the painful limb [10]. Thus, clinically, it may be relevant to evaluate alterations in movement (movement system examination) separately to body image perception (perceptions of the painful limb) in patients with CRPS Type 1. However, this is not to suggest excluding one from

the other in terms of symptomology within patients with CRPS Type 1, but rather to acknowledge a biokinesio-psychosocial approach when assessing and treating these patients. Therefore, understanding how all facets of this condition potentially interact or lack direct interaction and is thus of the utmost importance when screening and treating patients with CRPS Type 1.

Motor dysfunction in CRPS Type 1 is commonly considered a functional movement disorder [12]. Earlier research in individuals with functional movement disorders found evidence of cortical inhibition during explicit but not implicit motor activities [12]. Van Velzen et al (2015) analyzed whether active inhibition in cortical regions occurs in patients with CRPS Type 1 [12]. The researchers used transcranial magnetic stimulation to evaluate patients with CRPS Type 1 of the hand versus two control groups [12]. Van Helzen et al, 2015, included measuring corticospinal excitability at rest and during motor imagery (explicit motor task) and motor observation (implicit motor task) [12]. Secondary movement and functional outcome measurements through using various assessment tools (to be further reviewed in this paper) were also assessed [12]. Additional examination tools included the Radboud Skills Questionnaire (measuring manual activity of the upper extremity) and the Vividness of Movement Imagery Questionnaire-2- VMIQ-2 scale (the ability to perform imagined movements) [12]. In addition, the researchers collected data around other movement impairments including dystonia (Burk-Fahn-Marsden Scale), reduced strength, decreased active range

of motion and reduced velocity of movement.[12]. Importantly to note, patients with CRPS Type 1 involving the upper limb (and lower limb) often present with the above movement pathologies or insufficiencies which frequently impinge on activities including all activities of daily living (ADLs) i.e., bathing, eating, dressing as well as other functional activities involving the upper limb and lower limb. Therefore, when examining the movement system of patients with upper extremity CRPS Type 1 (analyzing only upper limb for the purpose of this review), it is critical to assess all activities involving the affected upper limb and thus should also pay attention to the above-mentioned possible impairments that may be contributing to the individual's function and social engagements. It is key to note, that although specifically focusing on upper limb movement dysfunction in patient with CRPS Type 1 in this review, lower extremity movement disorders or biomechanics abnormalities, may potentially affect an upper limb movement disorder/s and vice versa. Therefore, although this review is highlighting the upper limb, the author emphasizes not ignoring a lower limb and full body assessment in order to address how that may be functioning and potentially contributing to the movement system dysfunction in patients with CRPS Type 1 in the upper limb. Ultimately, the Van Velzen et al (2015) study concluded that the quality of motor dysfunction in patients with CRPS Type 1 appears to be unlike what is evident in patients with motor/functional paresis or in situations when a limb is immobilized such as a hand being immobilized to treat a scaphoid fracture (one of the control groups) [12]. Therefore, once again when analyzing the movement system in patients with CRPS Type 1, examination needs to appreciate that movement dysfunctions in patients with CRPS Type 1 differs, physiologically and often empirically, from movement dysfunctions in patients who have other pathological movement disorders and vice versa.

Introducing Movement System Diagnoses and a Newly Prescribed Biokinesio-Psychosocial Paradigm

Meaningful movement system diagnoses (MSDXs), described by Van Sant (2017), should allow PTs/Physiotherapists and other movement-based clinicians to select evidence-based treatment modalities for the identified MSDx [13]. For example, the necessity to forecast likely outcomes from occupations based on movement interventions is one of the most crucial arguments around the development of MSDx in the profession of PT [13]. Van Sant (2017) continues to assert that to the greater extent, presently, PTs proceed to use pathoanatomical and/or pathophysiological diagnoses provided by physicians to distinguish groups of patients for and from research on treatment efficacy [13]. It is suggested that a kinesio-pathological model of evaluation and a potential diagnosis of a patient, produces a diagnostic classification which speaks to the characteristic movement impairments, that are the cause of or consequence of the individual's pain and/or dysfunction [14]. Ludewig et al (2017) continues to assert that this classification lends itself to a PT or other movement-based profession treatment approach [14]. This would entail treating specific movement impairments found through an assessment of the patient and in turn may be defined as a movement system model.

Although the movement system still embraces the importance of a medical diagnosis (ie pathoanatomical), diagnosing according to a movement system classification scheme, as PTs or other, may be more beneficial for both the clinician and patient to attain better short, medium and long term outcomes. Notably, an MSDx should not exclude psychosocial factors that may be contributing to the patient's condition. These factors, contribute greatly to the patients' overall pain perception [24-41]. Although CRPS Type 1 requires a medical diagnosis, the argument the author intends to make in this paper is that this condition (and many other pain conditions) may require a multitude of diagnoses to be effectively managed rather through a new term developed by the author of this paper, as alluded to earlier, a 'biokinesio-psychosocial' approach, versus the well-versed 'biopsychosocial model'. With regards to evaluation and treatment of patients with CRPS Type 1, the 'bio' component of the newly derived 'biokinesio-psychosocial' expression incorporates the physiological and/or anatomical elements surrounding the condition. Kinesiology refers to the scientific analysis of human or non-human movement. More so it reflects "...an extensive scope of multidisciplinary study of human movement, regular physical activity of different duration, intensity, purpose and content, as well as its effect on the body and life of the individual and society as a whole..." (Sporis, et al. 2013, p7) [31]. Therefore, although the 'bio' incorporates many important physiological and anatomical aspects that make up human and non-human movement, the term 'kinesiology' appears to incorporate a clearer reflection of what makes up the entirety of the movement system including psychosocial elements. Therefore, the element of 'kinesio' under the umbrella term of 'biokinesio-psychosocial' is suggested in this paper to reflect, in a deeper manner, the notion of the movement system analysis. Although psychological and social factors are described above under the practice of 'kinesiology', the 'psychosocial' component in the label of 'biokinesio-psychosocial' emphasizes the requirement to pay specific attention to psychological and social factors that may be having an impact on the patient's wellbeing.

Examples of Movement System Dysfunction Questionnaires

When completing movement analyses with patients who have CRPS Type 1 involving either the upper limb or lower limb, a clinician should pay attention to all forms of movement and activity involving the affected body part and other uncompromised body regions. In addition, assessment of secondary/compensatory movement complications, as described previously, should be examined. A description of only some of the possible appropriate movement examination instruments that may be viable to use in patients with CRPS Type 1 of the upper limb will now be introduced. An analysis of some of the scoring criteria used to quantify and qualify the examination findings will also be explained. Furthermore, when appropriate, factors specific to the instrument being described such as for example, its appropriateness for application in an outpatient setting as well as its usage for patients with CRPS Type 1 of the hand, specifically, will be explored.

The Radboud Skills Questionnaire (RASQ)

The reliability of the RASQ in patients with CRPS Type 1 of one upper extremity has been investigated in the past with positive results [32]. The RASQ (see Appendix 1), a patient self-administered instrument, was initially developed to score a patient's ability to perform ADLs in a 'normal' way. The tool is divided into 3 broad categories namely personal care, domestic activities, and other activities. These major categories are further divided into 11 subcategories: personal hygiene, toilet hygiene, dressing, eating, and drinking, housekeeping, meal preparation, taking care of clothes, recreational activities, social activities, other items, and work. The 11 subcategories are further divided into more sub-items.

The RASQ makes comprehensive use of assessing many distinct upper limb functional tasks, however as a limiting factor, a significant amount of these activities may only be subjectively accounted for by the patients themselves, for example toilet hygiene activities, underwear undressing and dressing. Although the RASQ is subjectively filled in by the patient, the clinician may also have the opportunity to view and score many of the tasks/activities with reference to a movement system examination in the outpatient setting or even in the patient's home and daily life. These may include activities such as different aspects of dressing, recreational activities, home chores and more. The RASQ incorporates a clear scoring system that quantifies between '1' (able to complete the task normally with no effort) and 5 (unable to do the task anymore) [11]. This tool incorporates various other strengths including the a) ability to administer the tool in various settings such as outpatient, patient home, in-patient settings, and b) appears suitable for most age groups and population types. The tool also provides a value of '9' when the activity is 'not applicable' [32]. Therefore, although this tool was initially designed to be completed by the patient, there may be room to further modify it and reassess its reliability and validity in terms of dividing it into sections that are completed by the patient solely and other sections that are assessed and scored by the clinician as well as the patient. The above leaves room for future research around using this instrument for a more thorough movement examination of patients with CRPS Type 1 of the upper limb.

Through clinical assessment and treatment of patients with CRPS Type 1, it has been increasingly recognized that imagined movements (motor imagery) can result in increased pain and swelling of the specific body part [12,13]. Moseley (2004) states that an increase in pain and swelling as a response to imagined movements is identified without muscle activity or movement of the limb [34]. Furthermore, in a previous study, an increase in pain and swelling based on motor imagery, was further proven specifically in the affected upper limb in patients with CRPS Type 1 more so than in patients with generalized CP of the upper extremity [36]. To add, the increase in pain and swelling is suggested to correlate with the duration of signs and symptoms and appears to be moderated by autonomic arousal and patient beliefs about pain and movement [36]. The neurophysiological mechanisms are still not clearly understood, however this response to motor imagery points towards

cortical mechanisms at play which are associated with movement of the affected body part, and therefore has implications for treatment [34-36]. Graded Motor Imagery (GMI), a key treatment protocol for patients with CRPS Type 1, utilizes visualization, and imagined movements as an integral component of the therapeutic process [13,14,38]. Visualization and imagined movements has been identified, through evidence-based practice and research, as particularly significant in reducing pain and restoring movement in patients with CRPS Type 1 [13,14,38]. GMI appears to target clinical and neurophysiological biomarker effects of CRPS Type 1 via an incremental progression through lateralization exercises, implicit and explicit imagined movement training, mirror therapy and finally functional tasks [38]. A clinician, although initially requesting the patient to imagine various movements, is unable to view, assess and objectively describe the movement that a patient may be visualizing. However, based on the patient's descriptions and experiences whilst visualizing the affected limb or movement, this still produces clinical insight and information pertaining to how the patient's movement system may be functioning in a potentially systemic pathological manner. Furthermore, imagined movements may form an integral component of the initial movement system examination. Within this preliminary assessment of the movement system, clinicians may attempt to reduce fear and anxiety surrounding movement of the affected and commonly unaffected body part/s through evaluating how a patient responds to imagined movements prior to instructing a patient to physically initiate and attempt to complete an actual physical movement. The following instrument is designed to assess imagined movements, however it is not specifically developed for patients with CRPS Type 1.

The Vividness of Movement Imagery Questionnaire-2 (VMIQ-2) (Roberts et al, 2008) [39]

The VMIQ-2 scale (see Appendix 2), specifically designed for sports players, provides 12 items/skills that a patient is asked to imagine in their mind. The patient is first asked to imagine watching themselves perform the movement (external visual imagery) [39]. Secondly, they are asked to imagine looking through their own eyes whilst performing the movement (internal visual imagery) [39]. Thirdly, the patient is asked to imagine feeling themselves do the movement (kinesthetic imagery) [39]. They are required to describe and score the movement on a scale from 1 to 5: A score of 1=perfectly clear and vivid as normal vision, a score of 2=clear and reasonably vivid, a score of 3=moderately clear and vivid, a score of 4=vague and dim and a score of 5=no image at all- however the patient knows that they are thinking of the skill [39].

The first downside to the VMIQ-2 is clearly that this is a subjective questionnaire filled in by the patient in which the clinician, as previously mentioned, is unable to grade the movements objectively and visually. However, once again, this limitation may speak to the instrument's strength in assessing movement through initially reducing fear and anxiety of literal physical movement. Additionally, responses to imagined movements provides the examiner with crucial information regarding cortical mechanisms underlying the formation of movement or lack thereof, and in turn assists in directing treatment modalities for CRPS Type 1 such

as GMI. The VMIQ-2 is specifically designed for individuals involved in sports at different levels [39], and thus the items, as they currently exist, would not provide suitable information regarding a population that is not involved in sporting activities. Finally, the current VMIQ-2 combines lower limb and upper limb imagined movements and thus is not specific for only upper limb or only lower limb limitations [39].

Recommendations for future research may be to design further valid and reliable VMIQ instruments where the items listed within each specific VMIQ aim to support not only a sporting/athletic population group, but other population ensembles as well. These may therefore include separate VMIQs tested for reliability and validity on different populations such as for example, geriatrics, pediatrics, chronic and complex pain conditions such as CRPS Type 1 and other pathological conditions that may include movement disruption/s. In addition, moving forward, designing separate VMIQ's or a single VMIQ that has subcategories of items that focus on upper extremity movements and others that focus on lower extremity movements (and other body structures as well) may be a necessary task. Developing a VMIQ as such would assist clinicians and patients in firstly isolating how different body parts are perceived through imagined movements and potentially how these regions may interact with each other through visualization tasks. Therefore, ultimately, through adopting the above approach, an initial evaluation of the movement system through imagined movements would benefit in analyzing patients' overall movement systems more precisely. With reference to patients with CRPS Type 1, this method in assessment may potentially provide further insight into the patient's perceptions of their affected limb/s and thus paint a clearer picture into potential cortical and other mechanisms that may be altered. Therefore, clinicians may be able to further navigate their treatment directly towards pathophysiological and pathoanatomical mechanisms at play in patients with CRPS Type 1, thus navigating patient-centered treatment approaches.

Dystonia Scales: Burke-Fahn-Marsden Scale (BFM), Unified Dystonia Rating Scale (UDRS) and Global Dystonia Rating Scale (GDS)

Dystonia, a significant and frequent movement disorder in patients with various neurological conditions including CRPS Type 1, "is defined as a syndrome consisting of involuntary movements characterized by twisting or sustained movements" (Comella, et al, 2003. P.303) [40]. Significantly, dystonia has been found to occur in patients with CRPS Type 1 more frequently later in the disease than in the initial stages of the condition [7]. This piece of knowledge will become a key piece of information further on in this review when novel MSDx classification schemes are further discussed.

The BFM (see Appendix 3) was first published in 1985 and has been broadly used in clinical and epidemiological studies (cited in Krystkowiak et al, 2007) [41]. Although this scale has been widely used to assess dystonia in both adults and children, it has not been suitably tested across multiple centers and investigators. It has also not specifically been tested for reliability and validity in a CRPS

Type 1 population. In addition, the BFM has its limitations when testing dystonia. The instrument pays attention to 6 body regions. However, the tool provides for variable definitions of body areas [40]. In terms of a CRPS Type 1 diagnosis in the upper extremity, for example, the BFM is not specific enough pertaining to the actual upper limb diagnostic location i.e., hand, elbow, shoulder and other. In terms of scoring criteria, each item is scored according to 2 measures: 1. The provoking factor and 2. The severity factor. Each factor is rated between 0 and 4 ('0' being no dystonia and '4' being the maximum) [40]. Although the BFM lacks precise body part location, it's strength appears to lie in a relatively simple scoring system. As an additional note, making use of the BFM in a non-adult population with dystonia resulting from CRPS Type 1, such as within children with CRPS Type 1, the instrument's scales are influenced by the age of the child [42]. Thus, it is advised to consider the pediatric age of dystonic children when interpreting the results of the measure [42], and even more so when using this measure with children who have CRPS Type 1. The aim is to provide further in-depth analysis of CRPS Type 1 in a pediatric population in a future paper.

The UDRS (see Appendix 4), which incorporates 10 items (10 different body parts) was created to include a more detailed examination of dystonia in individual body areas including separate ratings for proximal and distal limbs [40]. This scale, however, was also not developed specifically to examine dystonia in patients with CRPS Type 1. However, in terms of the movement system when examining patients with CRPS Type 1, the UDRS seems to be a more useful tool to use than the BFM as it identifies, to a greater degree of accuracy, where on the extremity (i.e., distally, or proximally) the dystonia may be occurring and thus being examined. In addition, in contrast to the BFM, Comella et al (2003) adds that the UDRS has no weighting factors applied for any area of the body [40]. Finally, to the UDRS's advantage, its scoring criteria which ranges from none to extreme (5 scales) are defined further through various percentages in which the duration of the dystonia takes place for that particular body part [40]. For example, a severity score of mild equates to dystonia being present for less than and equal to 25% of the time during movement of the distal arm or hand. Therefore, although more time consuming, the UDRS scoring criteria allows for the clinician to precisely assess the dystonic movement of the CRPS Type 1 hand, for example, with making note of the severity level according to length of time this movement disorder takes place during the overall movement of the affected limb.

Finally, The GDS (see Appendix 5), as with the UDRS and BFM, is a general scale used to measure dystonia not specifically related to a diagnosis of CRPS Type 1. The GDS examines dystonia in 10 distinct body regions (eg proximal and distal portions of the extremities) and thus may also be applicable to examining dystonic movement in a CRPS Type 1 affected limb. This tool however, in terms of scoring criteria, is simpler but less descriptive than the UDRS. The GDS has a numeric rating scale ranging from no dystonia (a score of '0') to most severe dystonia (a score of '10'), with minimal dystonia (score of '1') and moderate dystonia (score

of '5') [40]. Although simpler and quicker to use, it may be that the UDRS provides more detailed information regarding the quantity and quality of the dystonia as previously mentioned. In addition, as described above, the UDRS utilizes percentages correlating with length of time that the dystonia is present during the movement, whereas the GDS does not do so. Therefore, the UDRS possibly adopts greater objectivity in assessing the movement system in comparison to the GDS, however the GDS may be simpler and quicker to use.

Furthermore, Comella et al's (2003) paper evaluated the UDRS, BFM and GDS for internal consistency and reliability. The results suggested that the UDRS, BFM and GDS all revealed excellent internal consistency and good to excellent intra-rater correlation [40]. Interrater agreement was fair to excellent [40]. Although reliability and agreement displayed equivalence amongst all 3 scales, the simplicity of application of the GDS testified by the majority of raters, suggests that this tool appears to be the most practical to implement in a number of environments, in order to rate the severity of dystonia [40]. As suggested previously, this conclusion is somewhat supported by the current review. However, the BFM, GDS and UDRS should be further tested in future research, specifically concerning their validity and reliability in a sample of CRPS Type 1 patients displaying dystonic features. Furthermore, this may be taken one step further where each one of these scales may be reproduced into numerous separate forms, where specific body parts are evaluated in one instrument instead of a more global dystonic assessment through a single and possibly diluted instrument. For example, the GDS may be revisited with multiple versions of this instrument being produced i.e., 1 version only assessing the hand, another version only assessing the foot and so forth. Through adopting this process, it potentially may allow clinicians to gain a fuller and richer perspective of the affected dystonic limb, for example in patients with CRPS Type 1 of the hand. In addition, this may then be incorporated, through using other versions of the instrument (pertaining to other parts of the affected upper limb, for example), to paint a more extensive evaluation of the movement system of the the affected upper extremity, as an entirety by putting all the detailed pieces of the 'puzzle' together.

Yes, patients with CRPS Type 1 may present with more than 1 extremity or other anatomical regions affected by dystonia. In this case, one of 2 possibilities are presented: 1. It may be useful to use the original more global instruments that have been retested for validity and reliability on a CRPS Type 1 population, as mentioned previously, and 2. Perhaps, recommended more by the author of this review, is using the modified instrument, as recommended above, in terms of having multiple versions of each instrument assessing merely a single body part. For example, a patient presenting with dystonia in the hand and the foot due to CRPS Type 1, may be examined using a newly developed 'GDS-hand version' and a 'BDS-Foot version'.

Notably, this paper has only touched on a few outcome measures

and instruments regarding a movement analysis of CRPS Type 1 of the upper limb describing scales measuring dystonia and other movement-related variables (the RASQ, VMIQ-2, BFM, UDRS and GDS). Importantly, further instruments should certainly also be taken into consideration when adopting a biopsychosocial movement system assessment of a condition such as CRPS Type 1. Instruments, for example, may include, but are not limited to: The Pain Catastrophizing Scale (PCS), the Tampa Scale for Kinesiophobia (TSK), The Pain Self Efficacy Questionnaire (PSEQ), the Disabilities of the Arm, Shoulder and Hand (the DASH), the Perceived Injustice Questionnaire (PIQ) and many more. There is currently no internationally approved upon standardized core outcome measurement set/battery of instruments for CRPS Type 1 research studies [43]. Griev et al (2017, p. 1083) goes on to state that "the development of a core measurement set would facilitate the pooling and comparison of data to answer specific research questions agreed upon as internationally important and relevant for the advancement of CRPS treatment" [43]. Therefore, when looking explicitly at the movement system with reference to CRPS Type 1 of the upper limb, the author of this paper also highlights that if the tools that have been briefly described in this paper are to be used effectively, it may potentially be more efficacious if they are included in a single-combined instrument or set battery of instruments that fully assesses and addresses the movement system of the affected upper limb. A future task necessitates the compilation of current assessment tools into one standardized instrument or the innovation of a set of instruments that may be validated and tested for reliability. Thus, through the implementation of well-constructed translational research, when examining the movement system of individuals diagnosed with CRPS Type 1 in an area of the upper limb, a new more-inclusive instrument or conglomeration of tools may be used in clinical practice with this population group.

Therefore, it is also suggested that these tools also need to be further modified through evidence-based research in terms of being body-region specific. In summary, although for example the BFM, UDRS and GDS do look at different body-parts in terms of dystonic dysfunction, instruments should be produced, tested for validity and reliability, specifically looking at an individual body part that is displaying dystonic features. In the case where there may be more than 1 body part presenting with symptoms of dystonia, separate instruments should evaluate each body part independently, to create a more in-depth evaluation of the movement system with reference to the body region/s being evaluated. With reference to the development of more body location directive instruments, building scoring systems that are both simple to utilize and time efficient, should be further addressed in the future, incorporating good quality and quantity outcome measures.

Finally, to emphasize, this type of examination should be completed using a thorough biokinesio-psychosocial approach. Thus, items in each instrument should facilitate the various components of the biokinesio-psychosocial model to gain an in-depth view of the movement system in a patient with CRPS Type 1 through an all-inclusive patient-centered approach.

Examples of Central Physiological, Anatomical and Psychosocial Mechanisms at Play in Various Movement System Disorders in CRPS Type 1

Central Nervous System Pathophysiological and Anatomical Mechanisms

Galer et al. (1995) suggest that 'neglect-like' symptoms in patients with CRPS Type 1 are mainly descriptive of abnormalities that produce difficulties with movement [44]. 'Neglect-like' symptoms may be understood as a combination of dysregulation between the 'biokinesio-psycho' aspect of the biokinesio-psychosocial model. Of the 11 cases that Galer et al. (1995) describe, a common theme was that the patients experienced a sense of disconnection from their affected limb and needed to pay specific attention to the limb in order to be able to move it [44]. When the patients were able to move the affected limb, initiation of movement was generally slow (hypokinetic) and execution of the movement also revealed reduced speed (bradykinetic) [44]. However, when patients were motivated by the clinicians to move, there was a significant improvement around the above movement parameters [44]. This result displays the strong effect that the therapeutic alliance can have on patients' outcomes [45,46]. Previous data seems to suggest that 'neglect-like' symptoms in patients with CRPS Type 1 may affect sensory perception in these individuals [47]. However, the majority of the data points in the direction that 'neglect-like' symptoms in patients with CRPS Type 1 mainly affects their movement [47]. Furthermore, it is proposed the mechanisms behind 'motor neglect' and 'learned nonuse' are different, although the behaviors of the patients share common features [47]. Punt et al's (2013) argument proceeds to suggest that reduced use of an affected CRPS Type 1 limb may appear to correspond more with 'learned nonuse' than actual 'motor neglect' [47]. Ultimately, 'learned nonuse' or 'neglect-like' symptoms are generally symptomatic of a primary centrally based dysfunction that may have a secondary impact on peripheral, structural, and mechanical systems that will present with a movement dysfunction/s.

Based on the Neurobehavioral Questionnaire [48], 2 measures are used to examine patients with either CRPS Type 1 or other CP conditions in an affected limb [48]. These 2 measures include motor and cognitive 'neglect-like' symptoms and involuntary movements (dystonic like movements) [48,49]. In research by Brink et al (2021), it was shown that both the above components measured through the Neurobehavioral questionnaire were reported more often by patients with CRPS Type 1 than patients with other CP conditions in an affected limb [49]. In addition, it was found that patients with CRPS Type 1 who reported these symptoms more frequently than those in the other group of patients, also described higher pain intensity and extra somatic symptoms [49]. Thus, there seems to be an association between 'neglect-like' symptoms and involuntary movements and increased degree of pain and somatic symptomology. Of interest, the motor and cognitive 'neglect-like' symptoms had a larger relationship to lower versus upper limb pain, whereas dystonic-like movements were, although still motor by definition, had a greater association with depression [49]. Therefore, it is once again clear to assess psychological variables

when treating patients with CRPS Type 1, in addition to motor and other signs and symptoms that occur in patients with CRPS Type 1. Once again, integrating a full biokinesio-psychosocial model into assessment and treatment of this population group is vital.

Proprioception, in relatively simple terms, is the ability of a body part to know where it is in space. Proprioception utilizes both central and peripheral processing of information to arrive at the conclusion of whether individuals can account for where their limb is in space. Bank et al (2013), in their investigation, found that impaired central processing of proprioceptive information in patients with CRPS Type 1, contributed significantly to motor dysfunction in this group of patients [50]. Thus, a clinician being able to identify proprioceptive impairments through a thorough biokinesio-psychosocial assessment of their patients with CRPS Type 1, should manage via correct therapeutic management, to aid in the recovery of motor dysfunction in patients with CRPS Type 1.

Finally, adding on to the idea of central mechanisms at play within CRPS Type 1, Schilder et al (2012) investigated motor control, via a kinematic analysis, in patients with CRPS Type 1 in the hands [51]. This research found that patients with CRPS Type 1 demonstrated noticeable bradykinesia and akinesia [51]. Of most interest, these symptoms presented in both the affected and unaffected sides which suggested altered central processing in this pathology [51]. In addition, patients with dystonia performed less well overall in comparison to patients without dystonia [51]. Intriguingly, motor impairments were unrelated to quantity and quality of pain [51]. Schilder et al (2012), however, does not specify the amount of time that the study's participants had been experiencing CRPS Type 1. In contrast, De Jong et al (2011) found that pain intensity in patients with CRPS Type 1, significantly limited movement and function [52]. The current review, however, argues that motor impairments that may be independent from pain would most likely fall in the category of post-acute CRPS Type 1 i.e., rather when the condition is in the sub-acute to chronic stages. The above refers to a critical segment of the argument proceeding in this paper; more than one MSDx may be necessary for this condition, and this may be related to the stage of the pathology when the patient presents to the clinician.

Psychosocial Factors

Bruehl et al (2006, p.430) states that "psychological and behavioral factors can exacerbate the pain and dysfunction associated with complex regional pain syndrome (CRPS) and could help maintain the condition in some patients"[53-56]. Pain-related fear, including fear avoidant behavior, is a unique forecaster of functional limitations in patients with CRPS Type 1 [50,53-55]. Thus, if there is a clear correlation between pain-related fear and functional limitations or movement system limitations in patients with CRPS Type 1, then this should be an essential treatment target for this population [52]. Therefore, there are interventions such as Cognitive Behavioral Therapy (CBT), other psychological approaches that fall under the umbrella of CBT, Therapeutic Pain Neuroscience Education (TPNE), graded activity, graded

exposure, pacing and other psychoeducational modalities, that target the fear-avoidant consequence of pain that do not directly set out to reduce pain, but rather aim to decrease pain-related fear [16,21,23,52,58,66]. As such, these effective modalities of treatment assist in eliminating the common and often debilitating assumption in patients with CP that certain movements may lead to further injury and damage, and thus cause further pain [52].

CBT, which has evidence to be extremely effective in the general CP world [64,67-74], is likely to be beneficial in the treatment of CRPS Type 1 [53,75-77]. Bruehl et al (2006) adds that successful usage of CBT modalities requires acknowledgement of the unique problems in patients with CRPS Type 1 [53]. As recognized earlier in the paper, patients with CRPS Type 1 are frequently witnessed to present with persistent learned disuse of their affected and unaffected body part/s [47]. CBT used with other non-invasive interventions, some of which were highlighted previously, are commonly used in these cases and have proven to be an effective treatment modalities for this component of the condition [53]. In particular, the CBT approach of graded exposure in vivo (systematic desensitization/progressive gradual exposure) in patients with CRPS Type 1 with fear avoidant behavior patterns, has been shown to be fruitful in decreasing levels of self-reported pain-related fear, pain intensity, movement and functional disability and self-reported peripheral abnormalities [78]. Hence, effective management of CRPS Type 1 necessitates that these psychosocial and behavioral characteristics be addressed as part of a cohesive interdisciplinary treatment approach [53]. Therefore, it is clear, that incorporating an evaluation and identification of possible fear avoidant behaviors and other psychosocial factors in a movement system evaluation in patients with CRPS Type 1 is necessary to provide suitable treatment for necessary patients and hence navigate towards the best possible outcomes. Once again, using the newly termed biokinesio-psychosocial assessment and treatment approach should importantly further enhance assessing and treating patients with CRPS Type 1 through the framework of a movement system-based approach.

Novel Movement System Diagnoses in CRPS Type 1 of The Upper Extremity: A Biokinesio-Psychosocial Approach

This review will now go on to describe three newly formulated movement system differential diagnoses by the author of this review, for patients with CRPS Type 1 in the upper limb. Movement system classification schemes will fall under the three separate MSDxs which in turn will further explore the condition under the wings of the biokinesio-psychosocial model. The three MSDxs, as will be explored further, are based on the stage of the condition when the patient may present themselves to the clinician as well as the degree to which central mechanisms and peripheral mechanisms may be at play. Importantly, the author also wishes to highlight independent of the stage of the condition, that these MSDxs are fluid and thus may bounce from one to the other in both directions during the required ongoing assessment and management of patients with CRPS Type 1 in the upper limb. In relation to the 3 MSDxs, 3 separate 'classification tools' have been briefly formulated that may mirror the specific classification

scheme being described. These instruments, for the purpose of this paper, may be titled as a 'questionnaire' or 'classification tool'. However, it is key to note that these tools have not been measured for validity and reliability and thus they are merely being preliminary described in this paper. Future research should measure the psychometric properties of these tools.

Acute Pseudomotor Fear Avoidant Movement Disorder (APFAM)

APFAM is a movement system diagnosis that the author of this review has derived with the primary aim to highlight how the movement system may present in the initial/acute stages of a CRPS Type 1 diagnosis. Although the focus of this paper is CRPS Type 1 of the upper limb, this diagnosis may be applied to CRPS Type 1 of the lower limb, however the classification system may need to be adjusted. This paper, with reference to APFAM, will focus on an approximate timeframe of the first 1-2 months after the onset of symptoms. Within the initial stages of CRPS Type 1, patients generally present with severe pain, swelling, trophic changes and more often than not, display 'motor' and/or movement deficits such as reduced range of motion, akinesia, bradykinesia, dystonia and others. The reason the author has placed 'motor' in 'inverted commas', is that these motor impairments/movement disorders, may initially, in the acute stages, present as definitive primary movement disorders/motor impairments. However, they may not actually be impairments or disorders at this stage in the pathological process of CRPS Type 1, but rather a secondary result of diminished movement resulting from fear avoidant behavior as a consequence of the primary complaint being pain. Thus, a MSDx label of APFAM; the 'pseudo' component focuses on the idea that it may present as a motor problem but in fact may not be an absolute centrally based motor/movement dysfunction at this stage of the condition. An application of the diagnosis of APFAM to a movement system classification scheme will now be presented.

The APFAM movement classification system was developed based on a previous paper's appendix (appendix 2 from Norton, 2007) which was presented in The Diagnosis Dialogue which took place at Regis University, Denver, Colorado USA [79]. To add, other movement disorders noted in CRPS Type 1, some of which have been outlined earlier in this paper, have also been included in this classification system. Under the umbrella of APFAM, the author has identified numerous movement disorders and other relevant symptoms that should navigate a practitioner's clinical reasoning towards a diagnosis of APFAM, and additionally assist in directing evidence-based treatment. The following is a list of items that the author of this review has compiled that should be included in APFAM. The categorization format is divided into 3 categories: Pain, Movement (including biokinesiological as part of the biokinesiological-psychosocial model), and Psychosocial. According to this classification system, an individual should have one or more symptoms/disorders present in each category, to be given the APFAM MSDx. The 'other' subcategory under the 'Movement' category should contain other movement and/or motor disturbances the clinician may find including either centrally and/or peripherally based movement/motor dysfunction/s.

A) **Pain:** Generally irretraceable in nature, hyperalgesia, allodynia, hyperpathia, and a multitude of further pain signs and symptoms.

B) **Movement:** Loss of joint mobility, mobility deficit, reduced muscle power, coordination problems, fractional movement deficit, perceptual complications (including 'neglect-like' symptoms), proprioceptive deficit, musculoskeletal deconditioning and cardiopulmonary deconditioning. As briefly asserted earlier in this review, although movement disorders such as for example myoclonus akinesia, bradykinesia, dystonia, and other movement disorders, have been shown to generally present after the acute/subacute stages of the disease, they certainly can still appear in the earlier stages [4], [7]. Therefore, the author of this paper names them here under this category and provides a space in the APFAM Classification Tool/Questionnaire for these disorders under the heading 'other' falling under the 'movement' subcategory.

C) **Psychosocial:** Fear Avoidant behavior, anti-social patterns of behavior, overall decreased QOL as well as many other psychosocial factors identified through 1.) Observation, 2) patient interview and 3) questionnaires/instruments such as Fear-Avoidance Beliefs Questionnaire (FABQ), Tampa Scale for Kinesiophobia (TSK), Pain Catastrophizing Scale (PCS), the 36 Item Short Form Survey (SF-36) which usefully evaluates essential parts of an individual's psychosocial wellbeing, as well as numerous other instruments.

The above categories have been clearly set out in a preliminary assessment format questionnaire/ classification tool (Figure 1). The categories within the questionnaire/classification tool have also been established in a manner which should allow clinicians to formulate clear clinical reasoning with regards to the diagnosis and potential management of the presenting patient with CRPS Type 1.

Acute Pseudomotor Fear Avoidant Movement Disorder (APFAM) Questionnaire/Classification Tool

A. Pain			
Type of Pain and pain descriptions (A)	Present or Absent	Measures: (interview, questionnaires observation, other)	Possible Pain Mechanism: (mechanical/noiceptive, central-spinal/cortical)
Allodynia			
Hyperalgesia			
Other			
A. Movement			
Movement Disorder (B)	Present or Absent	Measures:(Interview, Observation, Tools used, other)	Description
Loss of Joint Mobility			
Mobility Deficit			
Muscle Power Deficit			
Coordination Deficit			
Fractional Movement Deficit			
Perceptual Deficit (Including neglect)			
Proprioceptive Deficit			
Musculoskeletal Deconditioning			
Cardiopulmonary Deconditioning			
Other			
B. Psychosocial			
Psychosocial Factors (C)		Identification Through:	Description
		<ul style="list-style-type: none"> • Observation • Patient interview • Questionnaires/instruments 	
Fear Avoidant Behavior/s			
Other Psychosocial			

Figure 1: Acute Pseudomotor Fear Avoidant Movement Disorder (APFAM) Classification Tool (Questionnaire).

As mentioned earlier, if there is more than one MSDx, an individual may move back and forth between the MSDXs depending on various factors. Thus, when entertaining a potential MSDx such as APFAM or in that case any other MSDx, clinicians, as such, should further consider the following potential variables: stage of condition, specific type of day the individual is having when visiting the treating clinician (i.e., greater stress, more anxiety etc., work-related issues, family problems, other comorbid health conditions), along with many other biokinesio-psychosocial factors and variables. Therefore, ultimately, there very well could be many grey areas that overlap amongst the MSDx of APFAM

and other newly derived MSDXs by the author which are about to be explored.

Complex Fear Avoidant Movement Disorder-Type 1(CFAM-1)

CFAM-1 is the second MSDx related to CRPS Type 1 developed by the author of this paper (Figure 2). This MSDx may be used primarily in the subacute to chronic stages of the condition. However, as mentioned previously, it is not to say that it cannot be used earlier on if the patient's presentation allows for such. The major distinguishing factor between a CFAM-1 diagnosis and an APFAM diagnosis is the presence of motor changes particularly

founded on CNS disruption. CNS changes, outlined previously in this paper, are evidenced not only through pain presentations such as allodynia and hyperalgesia (noted in APFAM as well), but in movement disorders as well such as proprioception, dystonia, bradykinesia, akinesia, myoclonus and tremor, as examples [51]. Notably again, these CNS movement disorder conditions may still occur earlier on in the diagnosis of CRPS Type 1 and thus may still be included in an APFAM diagnosis. To highlight, an APFAM diagnosis does not necessitate the inclusion of one or more of the above CNS motor dysfunctions, whereas the CFAM-1 does indeed require one or more of these CNS movement ailments. An individual presenting with CFAM-1 must have one or more of the aforementioned CNS movement disorders or any other centrally based movement disorders. Ultimately, to be given the CFAM-1 MSDx, an individual should have one or more symptoms/ disorders as demonstrated earlier, present in each category (pain, central movement disorders, other movement disorders and biopsychosocial signs and symptoms).

If the individual still presents in the subacute to chronic stages of the condition above but, does not present with any of the these or other CNS movement disorders as discussed above, but presents with other movement disorders under the APFAM classification

scheme, the individual may receive a diagnosis called Complex Fear Avoidant Movement Disorder-Type 2 (CFAM-2). Please refer to Figure 3 for an outline of the MSDx classification tool / questionnaire for CFAM-2. Again, what's critical in a CFAM-2 diagnosis as noted with APFAM and CFAM-1, is the presence of centralized pain. In addition to all or some of the movement disorders that fall under the APFAM category, a patient must at least have allodynia and/or hyperalgesia and potentially other symptoms of pain, as well as fear avoidant patterns of behavior and other psychosocial elements to be diagnosed under CFAM-2. Again, there is no restriction regarding the amount of movement/ motor pathology items and psychosocial manifestations to be classified under the CFAM-2. The difference between CFAM-2 and APFAM, primarily, is the stage of the condition and the fact that the motor problems are, at this stage, most likely not 'pseudomotor' but actual motor dysfunction such as, for example, reduced range of motion and strength due to the length of having had the condition at time of examination. Again, patients may present with central movement disorders as with the APFAM and CFAM-1, and these movement deficits should be demarcated under the 'other' subcategory falling under the 'Movement' category belonging to the CFAM-2 classification tool/questionnaire.

Complex Fear Avoidant Movement Disorder-Type 1 (CFAM-1) Questionnaire/Classification Tool

A. Pain

<i>A. Type of Pain and pain descriptions</i>	<i>Present or Absent</i>	<i>Measures:(interview, questionnaires observation, other)</i>	<i>Possible Pain Mechanism: (mechanical/nociceptive, central-spinal/cortical)</i>
<i>Allodynia</i>			
<i>Hyperalgesia</i>			
<i>Other</i>			

B1. Central Movement Disorders

<i>Central Movement Disorder (B1)</i>	<i>Present or Absent</i>	<i>Measures:(Interview, Observation, Tools used, other)</i>	<i>Description</i>
<i>Dystonia</i>			
<i>Bradykinesia</i>			
<i>Akinesia Muscle Power Deficit</i>			
<i>Myoclonus</i>			
<i>Tremor</i>			
<i>Proprioception Deficit/s</i>			
<i>Other Central Movement Disorders</i>			

If the individual at this point does not present with any of the above 'central movement disorders', proceed to the movement classification scheme under **Complex Fear Avoidant Movement Disorder-Type 2 (CFAM-2)**. If the patient does present with any of the 'central movement disorders', proceed to the next section named 'other movement disorders'.

B2. Other Movement Disorders:

<i>Other Movement Disorder (B2)</i>	<i>Present or Absent</i>	<i>Measures:(Interview, Observation, Tools used, other)</i>	<i>Description</i>
<i>Loss of Joint Mobility</i>			
<i>Mobility Deficit</i>			
<i>Muscle Power Deficit</i>			
<i>Coordination Deficit</i>			
<i>Perceptual Deficit (Including neglect)</i>			
<i>Proprioceptive Deficit</i>			
<i>Musculoskeletal Deconditioning</i>			
<i>Cardiopulmonary Deconditioning</i>			
<i>Other</i>			

C. Psychosocial:

<i>Psychosocial Factors (C)</i>	<i>Identification Through:</i>	<i>Description</i>
	<ul style="list-style-type: none"> • <i>Observation</i> • <i>Patient interview</i> • <i>Questionnaires/instruments</i> 	
<i>Fear Avoidant Behavior/s</i>		
<i>Other Psychosocial</i>		

Figure 2: Complex Fear Avoidant Movement Disorder-Type 1 (CFAM-1) Classification Tool (Questionnaire).

Complex Fear Avoidant Movement Disorder - Type 2 (CFAM-2) Questionnaire/Classification Tool

A. Pain

<i>Type of Pain and pain descriptions (A)</i>	<i>Present or Absent</i>	<i>Measures:(interview, questionnaires observation, other)</i>	<i>Possible Pain Mechanism: (mechanical/nociceptive, central-spinal/cortical)</i>
<i>Allodynia</i>			
<i>Hyperalgesia</i>			
<i>Other</i>			

B. Movement

<i>Movement Disorder (B)</i>	<i>Present or Absent</i>	<i>Measures:(Interview, Observation, Tools used, other)</i>	<i>Description</i>
<i>Loss of Joint Mobility</i>			
<i>Mobility Deficit</i>			
<i>Muscle Power Deficit</i>			
<i>Coordination Deficit</i>			
<i>Fractional Movement Deficit</i>			
<i>Perceptual Deficit (Including neglect)</i>			

<i>Musculoskeletal Deconditioning</i>		
<i>Cardiopulmonary Deconditioning</i>		
<i>Other</i>		
C. Psychosocial		
<i>Psychosocial Factors (C)</i>	Identification Through: <ul style="list-style-type: none"> • <i>Observation</i> • <i>Patient interview</i> • <i>Questionnaires/instruments</i> 	Description
<i>Fear Avoidant Behavior/s</i>		
<i>Other Psychosocial</i>		

Figure 3: Complex Fear Avoidant Movement Disorder-Type 2 (CFAM-2) Classification Tool (Questionnaire).

Referring to Figures, 1, 2 and 3 respectively, as with APFAM, the classification schemes for CFAM-1 and CFAM-2 are divided into 3 categories (pain, movement and psychosocial) and have been clearly set out in an assessment format table. The only difference between CFAM-1 and APFAM is the 'movement' category which is subdivided into B1 and B2; 'Central movement disorders' and 'Other movement disorders' respectively. Notably, although symptoms such as perceptual deficit/neglect like-features and proprioceptive problems fall under category B2 in CFAM-1, they are still certainly considered as central dysfunctional ailments. Therefore, one might decide to categorize them under the category of 'other central movement disorders' within B1. The categories within the table have also been created in a way which should permit the clinician to construct clear clinical reasoning with regards to the diagnosis and potential management. Again, as with APFAM, to diagnose either CFAM-1 or CFAM-2, an individual should have one or more symptoms/disorders present in each category.

It is necessary to point out that the majority, if not all, of the items listed in APFAM, CFAM-1 and CFAM-2 may be addressed through direct evidence-based PT and other movement based clinical professions' interventions. Evidence-based treatment, for example GMI, can play a critical role in addressing pain, many of the outlined movement disorders as well as fear avoidant behavior patterns identified in APFAM, CFAM-1 and CFAM-2.

In a situation where a clinician may not be comfortable treating the specific MSDx, it would be the clinician's duty to direct an appropriate referral to a potentially specialized health-care provider in the field. Finally, if any of the disorders or impairments fall outside the treating clinician's scope of practice, it would be necessary to identify this and therefore refer to the appropriate clinical professional i.e., psychiatrist, psychologist, social worker, occupational therapist, medical doctors and furthermore specialists in the field of pain. Incorporating a thorough interdisciplinary model into the care of patients with CRPS Type 1, should navigate a strong and all-encompassing movement system assessment and treatment approach. Along with paying attention to the content of this paper to date, it is suggested that with regards to the CFAM-1 and CFAM-2, the biokinesio-psychosocial model can once

again be further dissected and understood in further detail moving forward so to potentially translate its meaning into clinical practice and future translational research.

Conclusion

In conclusion, as Van Sant (2017, p. S10) suggests, "for the profession to advance, we need to identify aspects of clinical presentation that are relevant to our practice, establish MSDxs, and begin the hard work of researching the effectiveness of our interventions to enable prediction of likely outcomes" [13]. Although 'the profession', as stated by Van Sant (2017) is referring to PT, this quote can certainly include all healthcare providers that deal with movement and motor-based pathologies. The current review, whilst emphasizing a newly labelled biokinesio-psychosocial model, has endeavored to supply 3 novel movement system based differential diagnoses to patients with CRPS Type 1 in the hand, and has in turn strived to produce a movement system classification system that correlates to each one of these diagnoses. Moreover, creating such MSDxs and classification systems should hopefully steer the ship for more detailed analyses surrounding interventions within movement-based clinical occupations. Ultimately, the aim is to allow for the best possible outcomes for patients with CRPS Type 1. CRPS Type 1, as a purely medical diagnosis, has traditionally and potentially been viewed merely within the boundaries of a biopsychosocial framework. This review has attempted to take this approach one step further by incorporating an all-important kinesiology component to this model i.e., a biokinesio-psychosocial framework. Therefore, the author has provided an introductory means to which PTs and other rehabilitative experts may start to think about assessing and treating patients with CRPS Type 1 through a movement system paradigm. The current paper focused on the biokinesio-psychosocial movement system paradigm with reference to CRPS Type 1 in the upper extremity. Therefore, future articles should present the evaluation and diagnosis of patients with CRPS Type 1 in the lower limb, whilst again taking a position around the movement system through a biokinesio-psychosocial lens. Bruehl et al. (2006) suggests that an integrated interdisciplinary or multidisciplinary team allows for many patients with CRPS Type 1 to accomplish substantial progress in function and ability

to manage pain [53]. Moving forward, it is recommended that classification schemes and MSDxs are well validated and tested for reliability to apply them in clinical practice. Finally, this paper has highlighted CRPS Type 1 in the adult population with minimal reference to the pediatric population. Further reviews should focus specific attention on the assessment and diagnosis of children with CRPS Type 1, according to the movement system approach whilst again embracing a biokinesio-psychosocial model.

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Appendix: Radboud skills questionnaire (RASQ), translated version.

(For additional information concerning the scores, see table 2)

How to answer:

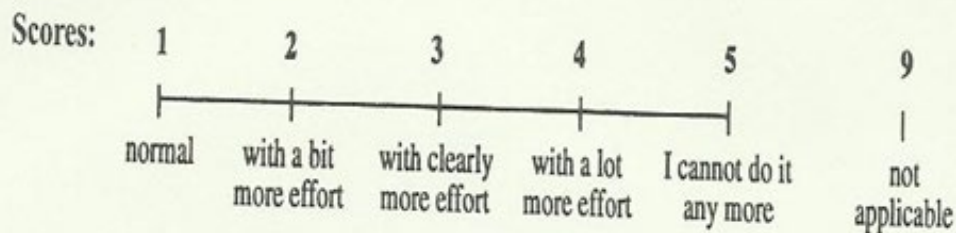
This list concerns some activities and skills of daily living.

Please answer the questions in accordance with the way you have performed the activities recently (over the past few days). Please compare the way you used to do the activities before the disease of your hand(s) to how you do them now.

For example: you cannot use your hand well while dressing. Regarding the items concerned with dressing, you should indicate whether you do this as you have always been used to, with more effort, or whether you need the assistance of an other person (for example your partner).

If necessary you can give an average score if there are different answers to the same question. For example: if—regarding sports—you normally go swimming and cycling, and you can cycle with a bit more effort, but swim with a lot more effort, you answer with '3': 'with clearly more effort'.

You can choose only one answer. If you have not performed an activity lately, for example going on holiday, please imagine the situation and try to give a score of between 1 and 5. Only if you never do a specific activity, choose the score '9'.



RASQ
(translated from Dutch)

	normaal	with a bit more effort	with clearly more effort	with a lot more effort	I cannot do it any more	not applicable
	1	2	3	4	5	9
I Personal care						
1. Personal hygiene						
1 wash upper and lower parts of the body	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
2 dry upper and lower parts of the body	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
3 look after face	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(put on make-up, shave, depilate)						
4 look after finger and toenails	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(to cut, file, laquer)						
5 look after hair	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(to wash, comb, dry, style)						
6 to brush teeth (electrically or manually, including	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
putting toothpaste on brush, taking care of false teeth)						
2. Toilet hygiene						
7 toilet hygiene in general	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(for example use of toilet paper, sanitary towels)						
3. Dressing						
<i>Upper body</i>						
8 to put on / take off underwear	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(for example bra, undershirt, corselette)						
9 to put on / take off upper clothing	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(for example blouse, sweater, T-shirt, necktie)						
<i>Lower body</i>						
10 to put on / take off underwear	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(for example underpants)						
11 to put on / take off upper clothing	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(for example trousers, skirt)						
12 to put on / take off socks	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(including elastic stockings, tights)						
13 to put on / take off shoes (including tying shoelaces)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
14 to handle fasteners (for example buttons, cufflinks, belts,	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
buckles, zippers)						
4. Eating and drinking						
15 handle fork, spoon, knife and cup in the usual way	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(putting food into your mouth)						

RASQ
(translated from Dutch)

	normal	with a bit more effort	with clearly more effort	with a lot more effort	I cannot do it any more	not applicable
	1	2	3	4	5	9
II Domestic activities						
5. Housekeeping (including handling necessary appliances)						
16 to clean and scrub	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
17 to wring out dish / floor clothes.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
18 to Hoover.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
19 to change a dustbin bag	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
20 to make beds.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
21 do the washing up (wash and dry).....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
6. Meal preparation						
22 to go shopping	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(for example to handle supermarket trolley or basket, to transport and put away shopping)						
23 to take money out of your purse	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
24 to prepare a meal.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(to chop or cut meat or vegetables, to peel potatoes, to prepare vegetables)						
25 to pour, for example out of a pan.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
26 to prepare sandwiches (including slicing cheese).....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
27 to cut meat on a plate.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
28 to carry a tray with some full cups of coffee / tea	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
29 to carry a crate of beer (or something similarly heavy).....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
7. Taking care of clothes						
30 doing a handwash	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
31 hanging out the washing.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
32 to fold and iron the washing	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
III Other activities						
8. Recreational activities						
33 participating in sports (individually or in a team)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
34 doing needlework, knitting, woodwork	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(odd jobs, building models, etcetera)						

RASQ
(translated from Dutch)

	normal	with a bit more effort	with clearly more effort	with a lot more effort	I cannot do it any more	not applicable
	1	2	3	4	5	9
9. Social activities						
35 going on outings, excursions	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
36 going on holiday (also staying overnight)	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
37 playing with animals or children	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
10. Other Items						
38 using a typewriter or PC	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
39 writing.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
40 using scissors	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
41 to open the front door with a key	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(also when necessary pushing or pulling the door)						
42 transportation to shops, work etcetera on a bicycle.....	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
43 transportation to shops, work etcetera in a car.	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
44 transportation to shops, work etcetera using public transport	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
11. Work						
45 performing your occupation	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>	<input type="radio"/>
(paid or unpaid; excluding household activities)						

Appendix 2: The Vividness of Movement Imagery Questionnaire-2 (VMIQ-2)

Name: _____ **Age:** _____
Gender: _____ **Sport:** _____
Level at which sport is played at (e.g., Recreational, Club, University, National, International, Professional)
Years spent participating in this sport competitively:.....

Movement imagery refers to the ability to imagine a movement. The aim of this questionnaire is to determine the vividness of your movement imagery. The items of the questionnaire are designed to bring certain images to your mind. You are asked to rate the vividness of each item by reference to the 5-point scale. After each item, circle the appropriate number in the boxes provided. The first column is for an image obtained watching yourself performing the movement from an external point of view (External Visual Imagery), and the second column is for an image obtained from an internal point of view, as if you were looking out through your own eyes whilst performing the movement (Internal Visual Imagery). The third column is for an image obtained by feeling yourself do the movement (Kinaesthetic imagery). Try to do each item separately, independently of how you may have done other items. Complete all items from an external visual perspective and then return to the beginning of the questionnaire and complete all of the items from an internal visual perspective, and finally return to the beginning of the questionnaire and complete the items while feeling the movement. The three ratings for a given item may not in all cases be the same. For all items please have your eyes CLOSED.

Think of each of the following acts that appear on the next page, and classify the images according to the degree of clearness and vividness as shown on the RATING SCALE.

RATING SCALE. The image aroused by each item might be:

- Perfectly clear and as vivid (as normal vision or feel of movement) RATING 1
 Clear and reasonably vivid RATING 2
 Moderately clear and vivid RATING 3
 Vague and dim RATING 4
 No image at all, you only “know” that you RATING 5
 are thinking of the sk

VMIO-2 continued

Item	Watching yourself performing the movement (External Visual Imagery)					Looking through your own eyes whilst performing the movement (Internal Visual Imagery)					Feeling yourself do the movement (Kinaesthetic Imagery)				
	Perfectly clear and vivid as normal vision	Clear and reasonably vivid	Moderately clear and vivid	Vague and dim	No image at all, you only know that you are thinking of the skill	Perfectly clear and vivid as normal vision	Clear and reasonably vivid	Moderately clear and vivid	Vague and dim	No image at all, you only know that you are thinking of the skill	Perfectly clear and vivid as normal feel of movement	Clear and reasonably vivid	Moderately clear and vivid	Vague and dim	No image at all, you only know that you are thinking of the skill
1.Walking	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
2.Running	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
3.Kicking a stone	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
4.Bending to pick up a coin	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
5.Running up stairs	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
6.Jumping sideways	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
7.Throwing a stone into water	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
8.Kicking a ball in the air	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
9.Running downhill	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
10.Riding a bike	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
11.Swinging on a rope	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5
12.Jumping off a high wall	1	2	3	4	5	1	2	3	4	5	1	2	3	4	5

Appendix 3: Burke-Fahn-Marsden Scale (BFM)

The Fahn-Marsden (BFM) Scale: Movement Scale

Region	Provoking factor	Severity	Weight factor	Product	
Eyes	0-4	X	0-4	0.5	0-8
Mouth	0-4	X	0-4	0.5	0-8
Speech					
Swallow	0-4	X	0-4	1.0	0-16
Neck	0-4	X	0-4	0.5	0-8
R arm	0-4	X	0-4	1.0	0-16
L arm	0-4	X	0-4	1.0	0-16
Trunk	0-4	X	0-4	1.0	0-16
R leg	0-4	X	0-4	1.0	0-16
L leg	0-4	X	0-4	1.0	0-16
			Sum:		
			Maximum=120		

<p>I. Provoking Factor</p> <p>A. General</p> <ol style="list-style-type: none"> 0. No dystonia at rest or with action 1. Dystonia only with particular action 2. Dystonia with many actions 3. Dystonia on action of distant part of body or intermittently at rest 4. Dystonia present at rest <p>B. Speech and swallowing</p> <ol style="list-style-type: none"> 0. Occasional, either or both 1. Frequent either 2. Frequent one and occasional other 3. Frequent both <p>II. Severity Factors</p> <p>Eyes</p> <ol style="list-style-type: none"> 0. No dystonia 1. Slight. Occasional blinking 2. Mild. Frequent blinking without prolonged spasms of eye closure 3. Moderate. Prolonged spasms of eyelid closure, but eyes open most of the time 4. Severe. Prolonged spasms of eyelid closure, with eyes closed at least 30% of the time <p>Mouth</p> <ol style="list-style-type: none"> 0. no dystonia present 1. Slight. Occasional grimacing or other mouth movements (e.g., jaw opened or clenched; tongue movement) 2. Mild. Movement present less than 50% of the time 3. Moderate dystonic movements or contractions present most of the time 4. Severe dystonic movements or contractions present most of the time 	<p>Speech and swallowing</p> <ol style="list-style-type: none"> 0. Normal 1. Slightly involved; speech easily understood or occasional choking 2. Some difficulty in understanding speech or frequent choking 3. Marked difficulty in understanding speech or inability to swallow firm foods 4. Complete or almost complete anarthria, or marked difficulty swallowing soft foods and liquids <p>Neck</p> <ol style="list-style-type: none"> 0. No dystonia present 1. Slight. Occasional pulling 2. Obvious torticollis, but mild 3. Moderate pulling 4. Extreme pulling <p>Arm</p> <ol style="list-style-type: none"> 0. No dystonia present 1. Slight dystonia. Clinically insignificant 2. Mild. Obvious dystonia, but not disabling 3. Moderate. Able to grasp, with some manual function 4. Severe. No useful grasp <p>Trunk</p> <ol style="list-style-type: none"> 0. No dystonia present 1. Slight bending; clinically insignificant 2. Definite bending, but not interfering with standing or walking 3. Moderate bending; interfering with standing or walking 4. Extreme bending of trunk preventing standing or walking <p>Leg</p> <ol style="list-style-type: none"> 0. No dystonia present 1. Slight dystonia, but not causing impairment; clinically insignificant 2. Mild dystonia. Walks briskly and unaided 3. Moderate dystonia. Severely impairs walking or requires assistance 4. Severe. Unable to stand or walk on involved leg
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Appendix 4: Unified Dystonia Rating Scale (UDRS)

Factor/area	Criteria
Duration	
0	None
0.5	Occasional (<25% of the time); predominantly submaximal
1.0	Occasional (<25% of the time); predominantly maximal
1.5	Intermittent (25–50% of the time); predominantly submaximal
2.0	Intermittent (25–50% of the time); predominantly maximal
2.5	Frequent (50–75% of the time); predominantly submaximal
3.0	Frequent (50–75% of the time); predominantly maximal
3.5	Constant (>75% of the time); predominantly submaximal
4.0	Constant (>75% of the time); predominantly maximal
Motor severity	
Eyes and upper face	
0	None
1	Mild: increased blinking or slight forehead wrinkling ($\leq 25\%$ maximal intensity)
2	Moderate: eye closure without squeezing or pronounced forehead wrinkling (>25% but $\leq 50\%$ maximal intensity)
3	Severe: eye closure with squeezing, able to open eyes within 10 seconds or marked forehead wrinkling (>50% but $\leq 75\%$ maximal intensity)
4	Extreme: eye closure with squeezing, unable to open eyes within 10 seconds or intense forehead wrinkling (>75% maximal intensity)
Lower face	
0	None
1	Mild: grimacing of lower face with minimal distortion of mouth ($\leq 25\%$ maximal)
2	Moderate: grimacing of lower face with moderate distortion of mouth (>25% but $\leq 50\%$ maximal)
3	Severe: marked grimacing with severe distortion of mouth (>50% but $\leq 75\%$ maximal)
4	Extreme: intense grimacing with extreme distortion of mouth (>75% maximal)
Jaw and tongue	
0	None
1	Mild: jaw opening or tongue protrusion $\leq 25\%$ of possible range or forced jaw clenching without bruxism
2	Moderate: jaw opening or tongue protrusion >25% but $\leq 50\%$ of possible range or forced jaw clenching with mild bruxism secondary to dystonia
3	Severe: jaw opening and/or tongue protrusion >50% but $\leq 75\%$ of possible range or forced jaw clenching with pronounced bruxism secondary to dystonia
4	Extreme: jaw opening or tongue protrusion >75% of possible range or forced jaw clenching with inability to open mouth
Larynx	
0	None
1	Mild: barely detectable hoarseness or choked voice or occasional voice breaks
2	Moderate: obvious hoarseness or choked voice or frequent voice breaks
3	Severe: marked hoarseness or choked voice or continuous voice breaks
4	Extreme: unable to vocalize
Neck	
0	None
1	Mild: movement of head from neutral position $\leq 25\%$ of possible normal range
2	Moderate: movement of head from neutral position >25% but $\leq 50\%$ of possible normal range
3	Severe: movement of head from neutral position >50% but $\leq 75\%$ of possible normal range
4	Extreme: movement of head from neutral position >75% of possible normal range

Factor/area	Criteria
Shoulder and proximal arm (right and left)	
0	None
1	Mild: movement of shoulder or upper arm $\leq 25\%$ of possible normal range
2	Moderate: movement of shoulder or upper arm 25% but $\leq 50\%$ of possible normal range
3	Severe: movement of shoulder or upper arm 50% but $\leq 75\%$ of possible normal range
4	Extreme: movement of shoulder or upper arm 75% of possible normal range
Distal arm and hand including elbow (right and left)	
0	None
1	Mild: movement of distal arm or hand $\leq 25\%$ of possible normal range
2	Moderate: movement of distal arm or hand 25% but $\leq 50\%$ of possible normal range
3	Severe: movement of distal arm or hand 50% but $\leq 75\%$ of possible normal range
4	Extreme: movement of distal arm or hand 75% of possible normal range
Pelvis and proximal leg (right and left)	
0	None
1	Mild: tilting of pelvis or movement of proximal leg or hip $\leq 25\%$ of possible normal range
2	Moderate: tilting of pelvis or movement of proximal leg or hip 25% but $\leq 50\%$ of possible normal range
3	Severe: tilting of pelvis or movement of proximal leg or hip 50% but $\leq 75\%$ of possible normal range
4	Extreme: tilting of pelvis or movement of proximal leg or hip 75% of possible normal range
Distal leg and foot including knee (right and left)	
0	None
1	Mild: movements of distal leg or foot $\leq 25\%$ of possible normal range
2	Moderate: movements of distal leg or foot 25% but $\leq 50\%$ of possible normal range
3	Severe: movements of distal leg or foot 50% but $\leq 75\%$ of possible normal range
4	Extreme: movements of distal leg or foot 75% of possible normal range
Trunk	
0	None
1	Mild: bending of trunk $\leq 25\%$ of possible normal range
2	Moderate: bending of trunk 25% but $\leq 50\%$ of possible normal range
3	Severe: bending of trunk $>50\%$ but $\leq 75\%$ of possible normal range
4	Extreme: bending of trunk $>75\%$ of possible normal range

Appendix 5: Global Dystonia Rating Scale (GDS)

The global score is an overall score for the body area. The investigator rates the patient in relationship to all patients. If the dystonia changes during the examination, the rating for the maximal dystonia is recorded.

Each body area is rated from 0 to 10:

- 0: No dystonia present in that body area
- 1: Minimal dystonia
- 5: Moderate dystonia
- 10: Most severe dystonia

Ten body areas are tested: 1) Eyes and upper face, 2) lower face, 3) jaw and tongue, 4) larynx, 5) neck, 6) shoulder and proximal arm, 7) distal arm and hand including elbow, 8) pelvis and upper leg, 9) distal leg and foot, and 10) trunk.

Body Area	No Dystonia Present (0)	Minimal Dystonia (1)	Moderate Dystonia (5)	Most Severe Dystonia (10)
Eyes and Upper Face				
Lower Face				
Jaw and tongue				
Larynx				
Neck				
Shoulder and Proximal Arm				
Distal arm and hand including elbow				
Pelvis and Upper Leg				
Distal Leg and Foot				
Trunk				

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