

METHODOLOGY, MECHANISMS & TRANSLATIONAL RESEARCH SECTION

Original Research Articles

Reliability and Validity of the Range of Motion Scale (ROMS) in Patients with Abnormal Postures

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Disclosure: This study is part of TREND (Trauma Related Neuronal Dysfunction), a Dutch consortium that integrates research on epidemiology, assessment technology, pharmacotherapeutics, biomarkers, and genetics on complex regional pain syndrome type 1. TREND is supported by a government grant (BSIK03016). D.E. van Rooijen, S. Lalli, C. Maihöfner, J. Marinus, A.G. Munts, A.A. van der Plas, and J.J. van Hilten declare no conflicts of interest. C.S. McCabe is funded by the NIHR, UK. M.A.J. Tijssen

received an unrestricted research grant from Ipsen Pharmaceutical and Allergan, Inc. for studies and teaching workshops on dystonia and from Ipsen to finance a specialized dystonia nurse. B.P. van de Warrenburg has received research support from the Prinses Beatrix Fonds, The Netherlands Brain Foundation, the Gossweiler Foundation, the Royal Dutch Society for Physical Therapy, Radboud University Nijmegen Medical Centre, and the Biobanking and Biomolecular Research Infrastructure (BBMRI-NL). A. Albanese received honoraria from TEVA, Ipsen, Merz, and Medtronic and research support from Allergan.

Abstract

Objective. Sustained abnormal postures (i.e., fixed dystonia) are the most frequently reported motor abnormalities in complex regional pain syndrome (CRPS), but these symptoms may also develop after peripheral trauma without CRPS. Currently, there is no valid and reliable measurement instrument available to measure the severity and distribution of these postures. The range of motion scale (ROMS) was therefore developed to assess the severity based on the possible active range of motion of all joints (arms, legs, trunk, and neck), and the present study evaluates its reliability and validity.

Methods. Inter- and intra-rater reliability of the ROMS was determined in 16 patients with abnormal sustained postures, who were videotaped following a standard video protocol in a university hospital. The recordings were rated by a panel of international experts. In addition, 30 patients were clinically tested with both the Burke-Fahn-Marsden (BFM) scale as well as the ROMS to assess construct validity.

Results. Inter-rater reliability for total ROMS scores showed an intra-class correlation coefficient (ICC) of 0.85. The majority of the scores for the separate

joints (13 out of 18) demonstrated an almost perfect agreement with ICCs ranging from 0.81 to 0.94; of the other items, one showed fair, one moderate, and three substantial agreement. The ICCs for the intra-rater reliability ranged from moderate to almost perfect (0.68–0.98). Spearman's correlation coefficients between corresponding body areas as measured with the ROMS or BFM were all above 0.82.

Conclusion. The ROMS is a reliable and valid instrument to evaluate the severity and distribution of sustained abnormal postures.

Key Words. Rating Scale; Abnormal Postures; Active Range of Motion; Reliability; Validity; Complex Regional Pain Syndrome

Introduction

Sustained abnormal postures of body parts may occur in a number of conditions, either as an initial feature or as a phenomenon in later stages, such as in dystonia. While isolated dystonia (previously named primary dystonia [1]) is characterized by sustained or intermittent muscle contractions that cause abnormal, often twisting, repetitive movements and/or postures [1,2], the term fixed dystonia has been used to describe cases who exhibit abnormal sustained postures without any mobile component [3,4]. Given that the phenotype of fixed dystonia differs significantly from its mobile counterpart, the term “fixed dystonia” is in fact inappropriate, for which reason we decided to use the name “sustained abnormal postures” instead of fixed dystonia throughout this article. The pathophysiology of sustained abnormal postures is still unclear and under debate [5,6], but the phenomenon is frequently observed after a peripheral trauma, as the most common movement disorder in complex regional pain syndrome (CRPS), or in the context of a functional syndrome [5–8]. Regardless of the etiology of these sustained abnormal postures, there is currently no appropriate measurement instrument available to rate their severity, to monitor the progression of the disorder, or to evaluate the effects of treatment over time. To date, the sustained abnormal postures are usually scored with the Burke-Fahn-Marsden (BFM) scale [9] or with the psychogenic movement disorders scale [10]. However, neither of these scales was designed to evaluate sustained abnormal postures. The BFM is primarily intended for rating abnormal movements in isolated dystonia, whereas the psychogenic movement disorders scale was developed to rate all types of movement disorders without distinguishing between the various features of dystonia.

Here, we introduce a new rating scale in which the severity of sustained abnormal postures is based on the impairment to voluntary move the affected body parts. Although the term sustained abnormal postures may evoke an image of complete absence of joint mobility, some active movement may still be possible [11–13]. Sustained abnormal

postures are typically most pronounced at the fingers and hands, and/or ankles and feet, but other joints of the limbs may be affected as well [14], together with the neck or trunk [6,15,16]. Taking these considerations into account, the range of motion scale (ROMS) was developed to evaluate the distribution and the severity of sustained abnormal postures by rating the active range of motion of all joints of the arms and legs, as well as of the neck and trunk. In the present study, we investigate the reliability and validity of the ROMS in a population of patients with sustained abnormal postures.

Methods

Participants

Sixteen patients (14 females) with sustained abnormal postures were invited to participate in the evaluation of the reliability of the ROMS, while 30 patients (26 females) agreed to participate in the validity study of the ROMS (Table 1; 11 patients participated in both studies). In most patients, the abnormal postures were developed after a (often peripheral) trauma. Patients with contractures or other causes of joint stiffness that were so severe that it prohibited any motion of the joint were not recruited. CRPS was diagnosed according to the 1994 consensus criteria of the International Association for the Study of Pain (“Orlando criteria”) [17]. Consecutive patients visiting the outpatient clinic or participating in other studies who fulfilled the inclusion criteria were invited to participate. All patients were 18 years or older, procedures were carried out in accordance with the Declaration of Helsinki, and informed consent was obtained from all participants. The ethical committee of the Leiden University Medical Center approved of the study's protocol.

Development and Structure of the ROMS

Ten raters with expertise in movement disorders (eight neurologists, one physiotherapist and one health professional) from seven different centers participated in this study. A prototype of the scale was developed among two centers (Leiden and Milano) using a consensus strategy. All raters were involved in a pilot study to test the feasibility of the scale and the video protocol. The final study version was defined after addressing suggestions for improvement after the pilot phase.

The ROMS (Supporting Information Appendix S1) contains 18 items—reflecting the involved body areas—which include four joints of each arm (fingers, wrist, elbow, shoulder), four joints of each leg (toes, ankle, knee, hip), the trunk, and neck. Response options range from 0 to 3 and are based on the magnitude of the active range of motion, with option 1 including an additional evaluation of movement fluency and velocity. To enhance feasibility and efficiency, and reduce administration burden, the examined movements in the ROMS are restricted to one plane, i.e., flexion and extension movements in the sagittal plane. Moreover, scores are judged by eye, i.e., goniometers are not used.

Table 1 Patient characteristics

	Reliability Study	Validity Study
Number of patients*	16	30
Number of patients with CRPS	10	23
Age at onset	32.8 (SD $\pm$ 12.3)	29.5 (SD $\pm$ 19.9)
Disease duration (of abnormal postures)	10.2 (SD $\pm$ 6.1)	9.7 (IQR 5.8–16.2)
Precipitant event		
Peripheral trauma	11	22
Period of extreme fatigue and fever	—	1
Syncope	—	1
Postanoxic encephalopathy	1	—
Intense pain (spontaneous)	1	1
While doing sports	1	1
Spontaneous	2	4

*Eleven patients participated in both studies.

CRPS = complex regional pain syndrome; IQR interquartile range; SD = standard deviation.

Total scores, as well as the number of affected joints can be calculated.

Reliability

To evaluate the inter-rater and intra-rater reliability of the ROMS, 16 patients were videotaped using a standard video protocol. These patients were selected because they differed in the degree of joint mobility impairment as well as in the distribution of symptoms. Throughout the patient recruitment, the type of affected joints and a global severity score (minor or severely affected) were registered to ensure that, across the sample, per body region at least four affected joints that exhibited different degrees of impairment were collected. One patient with affected trunk was specifically invited after a database search. No extra clinical assessments prior to enrollment of the study were performed. This sample thus allowed for an adequate coverage of the items and range of the scale. Patients were verbally and visually instructed by the experiment leader to move each joint in the sagittal plane smoothly from maximum flexion to maximum extension. The movements were filmed from the lateral side, except for inversion and eversion of the feet, which were recorded from the frontal side. Seven raters from five centers participated in the intra-rater reliability study. Each rater received a DVD at baseline and was asked to rate all patients according to the instructions of the scale. Each rater evaluated the videotape independently and was therefore blind to the scores of other raters. This procedure was repeated 2–4 weeks after the first session, using a videotape with the same content, but with the patients presented in a different random order.

The inter-rater reliability was assessed with the intra-class correlation coefficient (ICC; two-way random effects model for agreement). These values were calculated for the overall agreement between the different raters (total

scores), as well as for each individual component per body region (left and right sides combined) for both the baseline evaluation and the retest. A classification of strength of agreement was interpreted according to Landis and Koch's criteria [18]: slight (0.00–0.20), fair (0.20–0.40), moderate (0.41–0.60), substantial (0.61–0.80), and almost perfect (0.81–1.00).

The ICC (one-way random effects model) was also used to calculate the intra-rater reliability, i.e., the agreement between the baseline measurement and the retest for both the total score as well as each individual body area.

Validity

Thirty patients were clinically assessed with both the ROMS and BFM scale by one rater (DEvR) to evaluate construct validity. The correlation between scores of the same body region as evaluated by the BFM and ROMS was calculated using Spearman's rank order correlation coefficient for non-normally distributed data. For the ROMS, the sum score of four joints of the arms and legs was used. A correlation of >0.60 was considered supportive of construct validity.

Results

Inter-Rater Reliability

The ICC of the total score was 0.85, both when baseline and retest measurements were calculated. According to Landis and Koch's criteria, ICC for each separate joint at baseline demonstrated almost perfect agreement in 13 out of the 18 assessed joints (Table 2).

For all other joints, a substantial agreement was found, except for the left knee (moderate agreement) and the left shoulder (fair agreement). The reliability of the individual

Table 2 Inter-rater reliability for each joint using intra-class correlation coefficients

Joint(s)	Baseline	Retest
Left fingers	0.64	0.56
Left wrist	0.81	0.68
Left elbow	0.75	0.67
Left shoulder	0.30	0.41
Right fingers	0.94	0.94
Right wrist	0.90	0.92
Right elbow	0.88	0.89
Right shoulder	0.86	0.88
Neck	0.82	0.80
Trunk	0.77	0.57
Left hip	0.86	0.86
Left knee	0.59	0.82
Left ankle	0.95	0.96
Left toes	0.83	0.91
Right hip	0.88	0.90
Right knee	0.94	0.95
Right ankle	0.93	0.92
Right toes	0.83	0.88

A two-way random effects model for agreement was used. All results were $P < 0.001$, except for the left shoulder at baseline, which was $P = 0.001$.

joints observed during the retest was fairly similar to that of the baseline results, except for a lower agreement of the trunk. When left and right sides were combined for the limb joints, all ICC values demonstrated almost perfect reliability (Table 3).

Intra-Rater Reliability

High reliability scores were shown between the first and second measurement for the seven raters. The lowest score found for the individual body parts was 0.68 for the left elbow (Table 4). The ICC of the total scores was 0.93.

Table 3 Inter-rater reliability for limbs with right and left side combined, using intra-class correlation coefficients

Joint(s)	Baseline	Retest
Fingers	0.93	0.93
Wrist	0.91	0.91
Elbow	0.89	0.89
Shoulder	0.85	0.87
Hip	0.87	0.89
Knee	0.84	0.91
Ankle	0.94	0.94
Toes	0.83	0.89

A two-way random effects model for agreement was used; $P < 0.001$ for all tests.

Table 4 Intra-rater reliability (test-retest)

Joint(s)	ICC
Left fingers	0.77
Left wrist	0.81
Left elbow	0.68
Left shoulder	0.78
Right fingers	0.97
Right wrist	0.94
Right elbow	0.91
Right shoulder	0.90
Neck	0.91
Trunk	0.76
Left hip	0.89
Left knee	0.81
Left ankle	0.97
Left toes	0.93
Right hip	0.95
Right knee	0.98
Right ankle	0.93
Right toes	0.88

ICC = intra-class correlation coefficient (using a one-way random effects model); $P < 0.001$ for all tests.

Validity

Correlations between the composite scores of the corresponding body areas as measured with the BFM or ROMS were all significant ($P < 0.001$) and ranged from 0.83 (right leg) to 0.98 (neck). Correlation coefficients for the other body areas were: right arm: $r = 0.93$; left arm: $r = 0.91$; trunk: $r = 0.86$; and left leg: $r = 0.94$.

Discussion

The ROMS demonstrated good reliability and validity, and hence meets the criteria for a sound clinimetric instrument to measure the severity of abnormal sustained postures in fixed dystonia. Total scores displayed a high ICC between raters, which also held true for the ICC of the individual body areas. An exception was the fair-to-moderate reliability of the left shoulder, most likely caused by the low variation in scores in this joint as shown by visual inspection of the data for this joint (nearly all patients had a score of 0 or 1) [19]. The almost perfect reliability of the shoulder when the left and right sides were combined supports this assumption.

As to date no appropriate measurement instrument was available to evaluate the severity and distribution of abnormal postures; researchers used the BFM scale [20,21]—which was originally developed for isolated (or primary) dystonia [9]—or used no measurement instrument at all [3,22]. The ROMS has several advantages over the BFM scale. Firstly, the score of the BFM contains a provoking factor, which depends on the presence in time. Although this factor does include the response option “dystonia present at rest,” this distinction is not useful in sustained

abnormal postures as the abnormal postures are (nearly) always present and is therefore not made in the ROMS. Secondly, the provoking factor is combined with a severity factor, which is based on an impairment level instead of a symptom level in the limbs, for example, "grasping an object" in the evaluation of the arms. Moreover, the BFM does not differentiate between involvements of several joints in the same limb, hence the maximum score for an arm can be achieved even when only the fingers are affected. In contrast, the ROMS takes all the joints into account and anchors the severity on the limitation of a simple voluntary movement in a similar way for all body areas. Extensive experience with patients with abnormal posturing learned that the condition is nearly always associated with restrictions in voluntary movement and therefore active range of motion serves as a good indicator of the severity of the abnormal posture. Thirdly, the total score of the BFM also includes scores of the face, mouth, speech, and swallowing, body areas that may be involved in isolated dystonia. However, sustained abnormal postures are typically present in the extremities [13,14], therefore the body areas above the neck were not incorporated in the ROMS. Lastly, and in line with the above-mentioned limiting factors, the BFM is insensitive to small changes [9]. For instance, a case with sustained abnormal postures of all joints of the arm that fully recovers after treatment will only improve 7% on the BFM (8 out of 120 points), while the same improvement would show a change of 22% (12 points) on the ROMS. Despite the differences in content between the two scales, the correlations between the corresponding body areas were high. This result was not unexpected, as the presence of abnormal postures in dystonia should give a score on either scale, and therefore correlations between both were supposed to be present.

A potential source of error could have been the addition of "slow speed or clumsiness" as an extra dimension to score 1. Although this is a subjective element, the pilot phase showed that some patients do reach a full range of motion, but with considerably slower speed or clumsiness, which could not be related to incorrect instructions of the patients. As the reliability scores were high, this extra dimension did not seem to have influenced the results. A limitation of the current approach is that restricted range of motion can be the consequence of other disorders, and is not specific to abnormal postures in the context of dystonia. However, this instrument is not intended as a diagnostic tool, but as a simple instrument to quickly assess the severity of the condition. Whether the impairment is due to a coexisting contracture or another cause of the sustained abnormal posture should be kept in mind, but is considered of less importance in the rating of the ROMS, as long as the main diagnosis for the examined patients is known. The ROMS can potentially be used in a variety of other neurological disorders where limitations in range of motion or abnormal sustained postures may occur, such as camptocormia and sequels of spasticity. In contrast to other rating scales in this area—such as the Ashworth scale where assessment is based on increase in muscle tone in response to passive movement and the Tardieu scale where the quality of the

muscle reaction to passive stretch is scored—the ROMS focuses on active range of motion and can therefore be of added value. However, reliability should be separately tested before it can be applied to other conditions. Moreover, before the ROMS is used to monitor longitudinal changes or measure treatment responses, responsiveness should be determined in each disorder. Given that patients may perform differently during voluntary or involuntary movements, another potential limitation of the ROMS is that only active range of motion (i.e., voluntary execution) is addressed. This is an aspect of sustained abnormal posturing that is encountered in some patients and should therefore be kept in mind during examination. On the other hand, disability in daily life is based on the impairments encountered during voluntary movement. The ROMS therefore evaluates an important source of disability.

In conclusion, the ROMS is a reliable and valid instrument to easily investigate the distribution and severity of sustained abnormal postures. Further clinimetric testing of the scale is required before it can be used in other disorders presenting with abnormal postures.

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Supporting Information

Additional Supporting Information may be found in the online version of this article at the publisher’s web-site:

Appendix S1. Instructions and score form of the ROMS.

Range Of Motion Scale (ROMS) - Instructions

This scale evaluates the active range of motion (AROM) of joints in disorders that are characterized by abnormal postures, which are presumed to have a neurological origin (e.g. fixed/tonic dystonia, spasticity).

The AROM is scored only during active flexion and extension (i.e. sagittal plane, transversal axis) of each joint of the arms, legs, neck and trunk. Patients are instructed to perform flexion and extension movements as far as possible, with two repetitions. During the execution of movements, the severity score is based on the maximum active range of motion of the whole movement range of that particular body part. For the hip and trunk, lying flat on the back on a bench and standing upright are assumed to reflect full extension; upright sitting is also assumed to reflect full extension of the trunk.

If the back of the thighs or knees do not touch the bench if the patient is in supine position, this may be due to limited extension of the hip or knee and additional movements must be performed:

- Full extension of the knee while the hip maintains the same angular position; if this movement is possible, the problem is located at the hip;
- If extension of the knee is not possible, the patient needs to (be) move(d) to the side of the bench to examine whether the hip extends further if the (upper and lower) leg is moved in the direction of the floor beside the bench.

The best performance of the two repetitions is scored. The severity scores of fingers and toes are based on the range of motion of the most affected digit.

0 is scored when a full AROM (i.e. from maximal flexion to maximal extension) is achieved without any difficulties;

1 is scored when more than 50% of AROM is reached, or when a full AROM is reached with slower speed or clumsiness;

2 is scored when less than 50% of AROM is reached;

3 is scored when no or almost no movement ($\leq 5\%$) can be performed.

NB: if there are movements that cannot be assessed (e.g. because of amputation or presence of a plaster cast), please score Not Applicable (NA).

Patients are instructed to perform a fluent movement; a score of 1 should be recorded if movement speed or fluency clearly deviate from what would be expected from a fluently executed movement by that patient (provided that the patient has an AROM >50%).

Each joint is scored, even when there is no observed abnormal posture at rest.

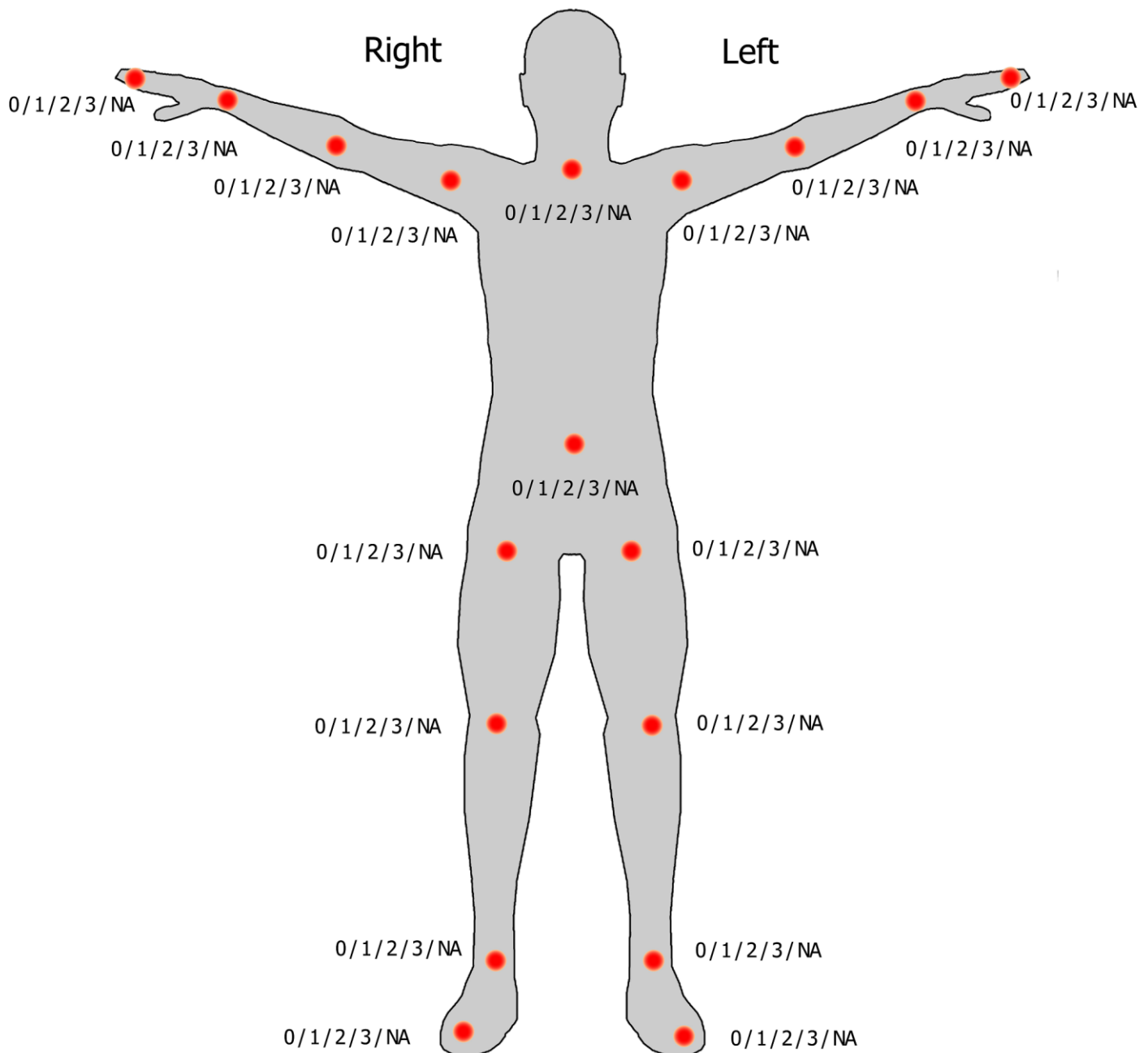
The number of affected joints is calculated by the summation of all joints with a score ≥ 1 . The total severity score is calculated by the summation of all separate severity scores. The sum score is adjusted if body parts cannot be examined, in which case the maximum obtainable score of the involved body parts is subtracted from the total.

ROMS

Patient number:
Observer:

Date:

0: full AROM
1: AROM 50% - 99% (or slow speed or clumsiness)
2: AROM < 50%
3: no AROM (or almost no AROM [$\leq 5\%$])
NA: not applicable



Total severity score: / (maximum is 54)

Total number of affected joints

Average severity: / (total severity score/total number of affected joints)