



Reachable workspace analysis is a potential measurement for impairment of the upper extremity in neuralgic amyotrophy

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Abstract

Introduction/Aims: Neuralgic amyotrophy (NA) is a multifocal neuropathy involving the nerves of the upper extremity, limiting functional capability and reducing range of motion. The reachable workspace (RWS) is a computerized three-dimensional analysis system that evaluates the relative surface area (RSA) of an individual's arm reachability and has shown utility in several neuromuscular disorders. The aims of this study were to examine the ability of the RWS to quantitatively detect limitations in upper extremity active range of motion in patients with NA, and correlate these with other upper extremity functional outcome measures.

Methods: Forty-seven patients with NA and 25 healthy age- and sex-matched controls were measured with the RWS. Study participants' RSAs were correlated with scores on the Shoulder Rating Questionnaire (SRQ), the Disabilities of Arm Shoulder and Hand (DASH) questionnaire, and upper extremity strength measurements using hand-held dynamometry.

Results: Patients with NA showed significantly lower values in the affected arm for all quadrants (except for the ipsilateral lower quadrant) and total RSA compared with controls ($P < 0.001$). We found moderate correlations between the reachable workspace, the DASH questionnaire result ($r = -0.415$), and serratus anterior muscle strength ($r = 0.414$).

Discussion: RWS is able to detect limitations in active range of motion of the affected arm in patients with NA, and is moderately correlated with upper extremity functional measures. RWS can demonstrate impairment of the affected upper extremity in NA and it has potential as a clinical outcome measure.

Abbreviations: AROM, active range of motion; BMI, body mass index; DASH, disabilities of arm shoulder and hand; EMG, electromyography; HHD, hand-held dynamometry; NA, neuralgic amyotrophy; RSA, relative surface area; RWS, reachable workspace; SQR-DLV, Shoulder Rating Questionnaire---Dutch language version; SRQ, Shoulder Rating Questionnaire.

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KEYWORDS

activity limitations, neuralgic amyotrophy, physical examination, range of motion, upper extremity

1 | INTRODUCTION

Neuralgic amyotrophy (NA) is a relatively common multifocal mononeuropathy, with a yearly incidence of 1:1000, that often involves brachial plexus nerves.^{1,2} The large majority of patients with NA will have residual symptoms and functional limitations even long after nerve recovery.^{3,4} These patients usually have limitations in upper extremity active range of motion (AROM) that impact reaching and overhead activities.⁴ Physical and occupational therapy, targeting motor control of the scapula and arm, can improve scapular motor control during daily activities using the upper extremities.⁵ An appropriate therapy program can thus result in an increase in functional capacity, due to more efficient movements that enable patients to use their upper extremities more frequently and with larger AROMs.^{2,5} These AROMs are typically assessed with goniometry, which is time-consuming and has suboptimal reproducibility due to considerable influence of training and experience.⁶ Moreover, goniometry provides degrees of movement for a specific joint in a single plane, whereas decreased functional capacity is usually related to multiple movement restrictions in multiple joints and planes.

As an alternative to goniometry, a sensor-based reachable workspace (RWS) measure to evaluate the proximal upper extremity AROM and arm reachability has been developed.⁸⁻¹¹ The RWS allows simultaneous tracking of multiple joints of the upper extremity in a series of protocolized movements, which correlates with activities of daily living.⁷ The system shows sufficient accuracy and robustness in tracking upper extremity movement when compared with more complex and expensive motion capture systems.^{8,9} As described in previous work, the RWS outcomes are shown as total and quadrant envelope relative surface areas (RSAs) between 0 and 1, with increasing values indicating more arm mobility. The RWS produces a visual representation of the RSAs (approximately as hemispherical shape) that provides patients and clinicians with objective information about the functional AROM. The RWS has shown to be a valid and reliable outcome measure in other neuromuscular diseases and musculoskeletal conditions of the upper extremity.¹⁰⁻¹³ The aims of this study were to examine the ability of the RWS to quantitatively detect limitations in upper extremity AROM in patients with NA and correlate RSAs with patient-reported outcomes for functional capacity and hand-held dynamometry of the upper extremity.

2 | METHODS

All data were collected as part of the NA-CONTROL study,¹⁴ a randomized, controlled trial investigating the effects of an outpatient rehabilitation program for patients with NA in The Netherlands.

For this study, we performed a cross-sectional analysis of baseline RWS data collected from participants in the NA-CONTROL study.

Patients with diagnosed NA who were referred to the Neuromuscular Center of the Radboud University Medical Center were included for the period from March 1, 2018 to March 16, 2020. Inclusion criteria were: right hand dominance, right-sided symptoms that included scapular dyskinesia, age 18 years or older, and not being in the acute phase of NA (>2 months after onset). Age- and sex-matched healthy controls were recruited through the university's healthy participants database, with right hand dominance and without current or previous shoulder complaints or other comorbidities (eg, muscular or neurological disorders). Hand dominance was confirmed based on a score of at least 40 on the Edinburgh Handedness Inventory.¹⁵

The sample size was based on a power analysis for the primary outcome measure of the NA-CONTROL study (ie, functional capacity of the upper extremity as measured with the Shoulder Rating Questionnaire---Dutch language version [SRQ-DLV]).⁶ Previous RWS studies in patients with neuromuscular disorders had similar or smaller sample sizes.^{7,10,12,16} A detailed description of recruitment procedures and a full list of inclusion and exclusion criteria can be found in the design paper of the NA-CONTROL study.⁶

Written informed consent was obtained from all study participants before participation, and the study was approved by the accredited regional medical ethics committee of Arnhem-Nijmegen (2017-3740, v3.0, NL63327.091.17). This study was registered at ClinicalTrials.gov under NCT03441347.

The RWS system utilizes three-dimensional motion tracking by a sensor to provide arm movement trajectory data that can be used to reconstruct an individual's reachable workspace. We used a relatively inexpensive, portable, commercially available sensor system (Kinect 2sensor; Microsoft Corp), which has demonstrated excellent reliability and clinical utility.⁸⁻¹¹ RWS measurements were performed according to a previously published standardized protocol.⁹ Briefly, subjects followed a video movement protocol, which covered shoulder movements within cardinal planes, while subjects were seated in front of the three-dimensional sensor, under the supervision of one of the investigators (R.L.). Subjects were instructed to reach as far as they could with their arm in several vertical and horizontal sweeps, while keeping the elbow extended and without twisting the body or leaning forward. RSAs were reported as a total of all quadrant data summated or as individual quadrant data, split into four frontal quadrants: the (Q1) upper medial, (Q2) lower medial, (Q3) upper lateral, and (Q4) lower lateral.¹¹ We also calculated an above-shoulder RSA, consisting of the sum score of Q1 and Q3. Figure S1 provides a schematic representation of the measurement protocol, the RSA quadrants, and a visual hemispherical representation of frontal reachable workspace RSA.

For patients with NA, patient-reported limitations in functional capacity of the upper extremity were evaluated using the SRQ-DLV and Disabilities of Arm Shoulder and Hand (DASH) questionnaire.^{17,18} Both questionnaires measure functional capacity of the upper extremity and have shown adequate reproducibility in various populations with shoulder pathology.¹⁹ Patients with NA also underwent muscle strength measurements. Handgrip strength was measured with a hand-held dynamometer (Jamar; Sammons Preston Rolyan). Maximal forces exerted during internal and external humerus rotation and for the serratus anterior muscle, while resisting scapular movement in the

scapular plane in 90 degrees of shoulder flexion, were measured with a digital manual muscle dynamometer while resisting axial scapular pressure in 90 degrees of scaption following a procedure described elsewhere (MicroFET2; Hoggan Scientific).^{20–22}

Individual participants underwent all physical measurements during a single day, in a fixed order. All measurements were performed by the same investigator (R.L.).

Data were analyzed using SPSS version 25 (IBM Corp). Descriptive statistics were used to evaluate baseline data. RSA data, strength measures, and patient-reported outcomes were evaluated for normality,

Characteristic	HC	NA	Mean difference	p value
Male, n (%)	15 (52%)	29 (62%)		
Female, n (%)	14 (48%)	18 (38%)		
Age, years	43 ± 1.7	43.1 ± 1.6	0.1 ± 2.5	0.971
BMI, kg/m ²	24.8 ± 0.6	24.7 ± 0.6	−0.1 ± 2.3	0.917
Strength				
Handgrip right, kg	41.4 ± 9.1	37.1 ± 2.3	−4.4 ± 3.4	0.206
Handgrip left, kg	39.3 ± 8.9	40.3 ± 1.6	1.0 ± 2.6	0.711
Pinchgrip right, kg	6.0 ± 1.8	5.4 ± 0.3	0.6 ± 0.5	0.771
Pinchgrip left, kg	5.3 ± 1.3	5.4 ± 0.2	0.9 ± 0.7	0.247
Keygrip right, kg	10.7 ± 1.9	10.0 ± 0.5	0.7 ± 0.8	0.352
Keygrip left, kg	9.9 ± 2.6	10.8 ± 0.4	0.9 ± 0.7	0.200
Exorotation right, N	122.2 ± 27.1	88.1 ± 6.3	−34.0 ± 9.5	0.001
Exorotation left, N	120.8 ± 26.4	127.4 ± 7.0	1.47 ± 16	0.524
Endorotation right, N	188.0 ± 10.4	141.2 ± 10.7	−46.9 ± 16.4	0.006
Endorotation left, N	187.2 ± 10.7	188.6 ± 10.7	1.47 ± 16.2	0.927
Serratus anterior right, N	256.3 ± 7.3	188.5 ± 8.8	−67.8 ± 13.0	<0.001
Serratus anterior left, N	252.2 ± 7.1	231.7 ± 8.4	−20.5 ± 11.0	0.107

Note: Values expressed as mean ± standard error of measurement or mean ± standard error of difference. Abbreviations: BMI, body mass index; HC, healthy controls; kg, kilograms (force); N, Newtons (force); NA, neuralgic amyotrophy subjects.

Quadrants	HC RSA	NA RSA	Difference	p value
Left Q1	0.226 ± 0.008	0.205 ± 0.007	−0.021 ± 0.011	0.048
Left Q2	0.181 ± 0.006	0.166 ± 0.004	−0.015 ± 0.010	0.045
Left Q3	0.225 ± 0.003	0.222 ± 0.002	−0.003 ± 0.006	0.264
Left Q4	0.221 ± 0.001	0.223 ± 0.001	0.001 ± 0.007	0.369
Left Total	0.853 ± 0.011	0.816 ± 0.010	−0.037 ± 0.003	0.015
Right Q1	0.234 ± 0.009	0.114 ± 0.067^a	−0.120 ± 0.002	<0.001
Right Q2	0.172 ± 0.022	0.131 ± 0.033^a	−0.041 ± 0.008	<0.001
Right Q3	0.239 ± 0.011	0.175 ± 0.072^a	−0.064 ± 0.015	<0.001
Right Q4	0.237 ± 0.001	0.228 ± 0.029 ^a	−0.009 ± 0.006	0.006
Right total	0.881 ± 0.012	0.647 ± 0.024^a	−0.234 ± 0.034	<0.001

Note: Data expressed as mean ± standard error of the mean.

Abbreviations: HC, healthy controls; L, left upper extremity; NA, patients with neuralgic amyotrophy; Q, quadrant; R, right upper extremity; RSA, relative surface area; Sig, Significance based on Kruskal-Wallis test.

^aSignificant differences for left and right arm within NA group. Bold indicates statistically significant difference (Kruskal-Wallis test).

TABLE 1 Characteristics of study participants

TABLE 2 Group differences in reachable workspace RSA

visually through histograms, and with the Shapiro-Wilk test. Body mass index (BMI) and strength differences between patients with NA and controls were evaluated with an independent-samples *t* test. To correct for multiple comparisons, a Bonferroni correction for multiple testing was applied, and alpha was set to 0.003 (0.05/14 dependent variables). Due to non-normality, differences in total and quadrant RSA values of the affected and dominant right side between patients with NA and healthy controls were evaluated using the Kruskal-Wallis test. Differences in total and quadrant RSA between the left (unaffected) and right (affected) side within the patients with NA were evaluated with a Wilcoxon signed rank test.

Spearman correlations were calculated for the total RSA and the combined above-shoulder RSA (Q1 + Q3) of the affected and dominant right arm with functional capacity of the upper extremity (SRQ-DLV and DASH scores) and strength measurements of ipsilateral handgrip, serratus anterior, and internal and external rotation strength. Interpretation was as follows: 0.00 to 0.09 = negligible, 0.10 to 0.39 = weak, 0.40 to 0.69 = moderate, 0.70 to 0.89 = strong, and 0.90 to 1.00 = very strong.²³ To correct for multiple comparisons, a Bonferroni correction for multiple testing was applied, and alpha was set to 0.005 (0.05/10 dependent variables)

for the RSA comparisons and to 0.008 (0.05/6 dependent variables) for the correlations.

3 | RESULTS

A total of 47 NA patients were measured (29 men [62%]; mean age, 44 ± 12 years). Mean and median time since onset were 16 (± 32) and 8 months, respectively. A total of 25 healthy age- and sex-matched control subjects were included. One healthy control was excluded due to a pre-existing shoulder problem that was missed at initial screening, but became apparent during the assessment period. This left 24 control individuals for the final analysis. One patient was excluded from the RWS analyses due to potential protocol violations. Mean BMI did not show significant differences between patients with NA and controls. Serratus anterior strength, exorotation strength, and grip strength were significantly lower for the patients with NA. For five participants, BMI could not be calculated, as their weight and height was not recorded because they did not complete the full NA-CONTROL study protocol. Table 1 provides a full overview of subjects' characteristics.

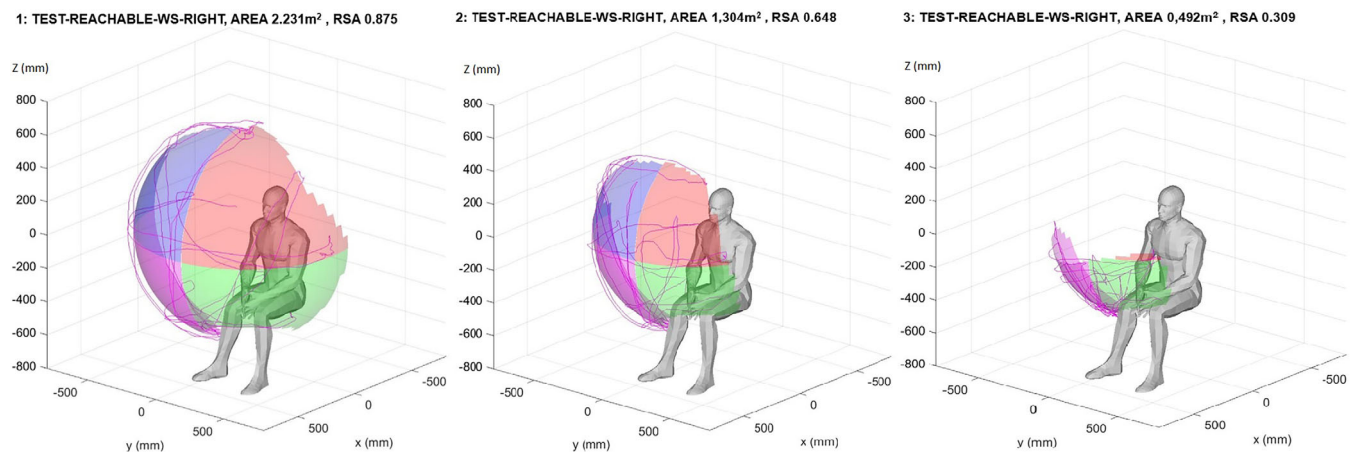


FIGURE 1 Visual representation of relative surface area in 1—healthy control; 2—moderately limited patient with neurologic amyopathy (NA); and 3—severely limited patient with NA.

TABLE 3 Spearman correlation coefficients for reachable workspace with functional capacity of upper extremity questionnaires and strength measurements in patients with NA

	Total RSA right (affected) arm	<i>p</i> value	Above-shoulder RSA right (affected) arm	<i>p</i> value
SRQ-DLV	0.278	0.071	0.256	0.097
DASH	-0.415 ^a	0.006 ^a	-0.394	0.009
SA strength	0.414 ^a	0.006 ^a	0.445 ^a	0.003 ^a
Grip strength	0.274	0.072	0.391	0.010
Exorotation strength	0.391	0.010	0.353	0.019
Endorotation strength	0.299	0.052	0.290	0.56

Abbreviations: DASH, Disabilities of Arm Shoulder and Hand questionnaire; RSA, relative surface area; SA, serratus anterior; SRQ_DLV, Shoulder Rating Questionnaire—Dutch language version.

^aStatistically significant.

3.1 | RWS measurements

Patients with NA had a significantly lower total RSA score compared with healthy controls. Quadrants 1, 2, and 3, were each significantly lower in patients with NA compared with healthy controls, with the largest difference found for Q1 (upper medial quadrant). Within the patients with NA, Q1-3 also showed significantly lower RSA scores when comparing the affected with the nonaffected side. Mean differences for each quadrant are shown in Table 2. Figure 1 provides a visual representation of the differences.

Larger total RSA and combined above-shoulder RSA correlated with greater serratus anterior muscle strength. Total RSA correlated negatively with DASH scores. No other correlations were found between RSA and functional capacity of the upper extremity questionnaires or strength measurements. All correlations are presented in Table 3.

4 | DISCUSSION

The total reachable workspace of the affected arm in patients with NA, as measured by RSA, was significantly reduced compared with the reachable workspace of healthy controls. Most of the reduction in the NA group's reachable workspace was due to limited reachability in the upper quadrants (reaching above the shoulder level). RSAs of the affected right arm were significantly lower than those of the nonaffected left arm within the patients with NA in Q1-3 as well. The RSA measure correlated moderately with the DASH score as well as with the affected limb's serratus anterior strength. These results indicate that the RWS can be useful for quantification of functional upper extremity limitations in patients with NA.

The lower RSA values for patients with NA compared with healthy controls in the upper shoulder quadrants (Q1 and Q3) were expected, as functional limitations during above-shoulder activities of the affected upper extremity are frequently reported.^{2,4,24} However, interestingly, we also found that the lower contralateral quadrant RSA of the affected arm (ie, Q2) was significantly lower in patients with NA compared with healthy controls. Previous studies showed that limitations in above-shoulder-level AROM are typical in NA.^{3,4,25} Our finding indicates that impaired AROM in patients with NA and scapular dyskinesia is not limited to above-shoulder-level movements, but may extend to movements below shoulder level as well, further impacting an individual's function. Although our patients with NA had RSAs similar to those of patients with facioscapulohumeral dystrophy with "severe scapular involvement" (as indicated by Ricci clinical severity scores ≥ 5 ^{7,26}), similar reductions in below-shoulder level RSAs were not seen in individuals with facioscapulohumeral dystrophy.⁷ The fact that patients with NA had reduced below-shoulder-level AROM may be explained by strain and muscle rigidity. Scapular dyskinesia in NA causes overloading of periscapular muscles such as the rhomboids and levator scapulae, and subsequent strain and rigidity in complex shoulder movements.^{2,5} This rigidity limits the lateral translation and internal rotation of the scapula, which are needed to

position the humerus across the body, possibly resulting in a decrease in AROM for the affected arm to the contralateral side.²⁷ This may indicate that patients with facioscapulohumeral dystrophy have less rigidity and primarily demonstrate weakness of (peri)scapular muscles.^{7,16} The patients in this study did show lower strength compared with healthy controls, but strength values were well over Medical Research Council score 3 for the arm- and scapular-pivoting musculature, and should not have affected the capability to move the arm for the duration of the protocol. However, we cannot be certain that capsular thickening or intra-articular glenohumeral problems did not play a role in the patients with NA.

Higher RSAs correlated with better functional capacity as measured by the DASH questionnaire. The moderate correlation observed may be explained by the fact that some items of the DASH questionnaire consist of activities that represent distal rather than proximal upper extremity tasks, such as cutting food, writing, or turning a key. This may also be the reason why we did not find a significant correlation for the above-shoulder RSA (Q1 + Q3) and the DASH questionnaire. Unexpectedly, the SRQ-DLV did not correlate significantly with the RWS measurements. In previous studies, the SRQ did show responsiveness to change in functional capacity of the upper extremity in patients with NA.⁵ However, the SRQ-DLV items strongly focus on symptoms and activity limitations and is a measure of limitations. Only 3 of 30 SRQ-DLV items specifically relate to a larger AROM of the affected upper extremity, which may explain the lack of correlation with the RWS.

Muscle strength of patients with NA was significantly lower for the serratus anterior, exorotation, and handgrip; however, the mean strength produced cannot explain the limitations in Q1-3. The correlation of RWS with serratus anterior muscle strength is in line with our expectations, as this muscle is the main scapular stabilizer. However, the correlation was also moderate. There are three possible reasons for this moderate correlation. First, the procedure used for the assessment of serratus anterior muscle strength has limited reproducibility. The measurement error is quite substantial, with a reported standard error of the mean of 15.8 N, due to considerable influence of the assessor handling the dynamometer.²⁰ Second, 39 of the 47 patients with NA reported pain during the day of the assessments, possibly limiting the strength they were able to produce.²⁰ Third, patients with NA also show coordinative deficits in scapular movement and positioning, possibly related to maladaptive cortical adaptation after peripheral nerve damage,²⁸ which may have influenced serratus anterior muscle strength recruitment during testing.

We did not perform electrodiagnostic (EDx) testing on most of the patients in this study. At our center, EDx testing is performed only when there is a genuine differential diagnosis to explore, because it is not needed to establish the NA diagnosis.²⁹ Moreover, nerve conduction studies are often normal in NA, and paraspinial electromyographic (EMG) abnormalities can be found in NA, which makes it difficult to differentiate from cervical radiculopathy.³ Those who had their serratus anterior muscles studied with needle EMG usually showed either mild neurogenic changes or no abnormalities when the clinical evaluation of SA strength testing was normal. The strength measurements

did not show any noticeable differences within the patient group, which is supported by the relatively small standard deviations of the mean scores; this leads us to conclude that damage to a specific nerve is unlikely to have caused the limitations found in these patients.

We believe the RWS may become a promising tool to quantify and evaluate AROM of the upper extremity in patients with NA, both in clinical practice and in research. However, more research is needed to establish reproducibility, validity, and responsiveness to change in RSA for functional upper extremity limitations in people with NA and other peripheral nervous system disorders affecting the upper extremity.

In conclusion, the RWS is suitable for objective assessment of functional shoulder limitations in patients with NA. The moderate correlation of the RWS with other functional outcome measures suggests that the RWS measure reveals additional information about functional (dis)ability of the upper extremity compared with capacity questionnaires and strength measurements. Therefore, the RWS is a potentially valuable clinical outcome measure for the assessment of functional upper extremity limitations caused by NA.

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CONFLICT OF INTEREST

The authors declare no potential conflicts of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICAL PUBLICATION STATEMENT

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

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SUPPORTING INFORMATION

Additional supporting information can be found online in the Supporting Information section at the end of this article.

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