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ORIGINAL REPORT

MANUAL DEXTERITY IN HEREDITARY MOTOR AND SENSORY NEUROPATHY TYPE 1A: SEVERITY OF LIMITATIONS AND FEASIBILITY AND RELIABILITY OF TWO ASSESSMENT INSTRUMENTS

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Objective: **To assess the prevalence and significance of impaired manual dexterity in hereditary motor and sensory neuropathy type 1a (HMSN 1a), with the Sollerman hand function and the Functional Dexterity test, and compare the reliability and agreement of the tests.**
Design: **Descriptive cross-sectional study.**
Subjects: **Forty-nine subjects with HMSN 1a.**
Results: **Forty-six (94%) subjects had an abnormal Sollerman sum score (< 80) for the dominant hand. The most difficult subtests required finger grips such as pulp, tripod and lateral pinches. Dexterity scores of both hands were categorized as "moderately functional". Test-retest reliability was excellent for the Sollerman test, with intraclass correlation coefficients between 0.98 and 0.99 (95% confidence interval (CI) 0.97–0.99), and good for Functional Dexterity test scores with correlation coefficients between 0.83 and 0.95 (95% CI. 71–0.97). The 95% limits of agreement between Sollerman tests showed that differences greater than 3 points can be interpreted as a change in dexterity. The Functional Dexterity test limits were wide.**
Conclusion: **Impaired manual dexterity is common among subjects with HMSN 1a, stressing that the evaluation of dexterity is an essential element of the functional assessment. Both tests are able to detect impaired manual performance in HMSN 1a. For monitoring of disease progression and the effects of treatment programmes the Sollerman test is most suitable.**

Key words: hereditary motor and sensory neuropathies, CMT, hand, manual dexterity, Sollerman hand function test, Functional Dexterity test.

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INTRODUCTION

Hereditary motor and sensory neuropathy (HMSN), also known as Charcot-Marie-Tooth disease (CMT), is a group of neuropathies, characterized by slowly progressive, distal muscle weakness and wasting, and sensory loss predominantly in the feet and hands. Impairments in hand function are frequently reported in HMSN (1–6), but studies that focus on the implications of impaired hand function for the activities of daily life are sparse. Nowadays, both in clinical trials and in clinical practice, functional assessments are carried out in addition to widely used symptom-oriented measures (7).

Manual dexterity is of utmost importance in performing activities of daily living and is described as the ability to move the hands easily and skilfully, to work with the hands in turning and placing motions (8). Many patients with HMSN with affected hand function complain of reduced manual dexterity, and the evaluation of limitations in this field has recently received more attention (5, 9–12).

Although numerous manual dexterity tests exist, there is no test available specifically for HMSN. Well-known tests, such as the Nine-hole-peg test, the Box and Block test, the Purdue pegboard test, the Jebsen test and the Sollerman hand function test (SHT), have all been used for the evaluation of dexterity in HMSN (1, 5, 10, 12). However, with the exception of the SHT, these tests provide data only on the speed of hand and finger use (13). Based on a previous explorative study (12), we believe that a manual dexterity test for HMSN should include additional aspects of dexterity, such as grasp patterns, precision and accuracy, co-ordination and bilateral tasks. The SHT seems to be appropriate, but empirical data to support the use of this test in HMSN is lacking. The drawback of the SHT is that it is time-consuming and may not be suitable for use in daily clinical practice. We therefore added the rapidly administered Functional Dexterity test (FDT), a time-scored test that also incorporates qualitative aspects of movement during the manipulation of pegs.

This paper aims to evaluate manual dexterity and the suitability of the SHT and FDT in subjects with HMSN. The research questions addressed in this study are: to what extent is manual dexterity, as measured with the SHT and the FDT, impaired; and how do these manual dexterity tests perform in terms of feasibility, reliability (homogeneity, test-retest) and agreement in subjects with HMSN?

METHODS

Participants

All patients with HMSN 1a, known at the Department of Rehabilitation and the Department of Neurology of the Academic Medical Center

in Amsterdam, were invited to participate (n = 63). We included only subjects with HMSN 1a in order to achieve a genetically homogeneous group of the most prevalent subtype of HMSN. Inclusion criteria were: (*i*) diagnosis of HMSN type 1a, confirmed by DNA study showing duplication on chromosome17p11.2–p12; and (*ii*) age between 18 and 70 years. Subjects were excluded if any other disabling disorder in their medical history might influence hand function and if they had difficulty understanding Dutch.

The study was approved by the medical ethics committee of our hospital and all subjects gave their consent to participate.

Measurement instruments

Manual dexterity was assessed with the SHT (14) and the FDT (15).

The SHT assesses unilateral and bilateral handgrip function and reflects the 7 most common grip types used in daily life: pulp pinch, lateral pinch, tripod pinch, 5-finger pinch, diagonal volar grip, transverse volar grip, and spherical volar grip. This test has been used to evaluate manual dexterity in various conditions affecting hand function (16–20), after hand surgery (14, 21) and repair of peripheral nerve injury (22). Intra- and inter-rater reliability is good (14, 19). Twenty subtests are scored on a scale from 0 to 4 points. Subjects with normal manual dexterity should achieve a total of 80 points with the dominant hand and 77–79 points with the non-dominant hand. Both hands can be tested within 45 min (14).

The FDT measures the ability to perform a tripod pinch through the timed manipulation of pegs (administration time about 5 min). A tripod pinch pattern is frequently used during daily activities such as eating, writing and tying (14, 15). This grip pattern in particular may become problematical when the intrinsic muscles of the hands are affected. The examiner records the time, in sec, it takes the subject to turn over 16 pegs, as quickly as possible, with one hand. A 5-sec penalty is added each time the subject supinates the arm or touches the board for assistance. If a peg is dropped, time is stopped, and a 10-sec penalty is added. Two scores are obtained: (*i*) the initial time score to complete the test, and (*ii*) the combined time score with penalty scores added to the initial time score. According to the classification of Aaron & Jansen. (15) FDT scores can be classified into categories, ranging from "functional" to "non-functional". Intra- and inter-rater reliability of the FDT test appeared to be good, construct validity has been confirmed and preliminary normative data are available (15).

Protocol

Information concerning subject characteristics was collected as part of a larger, descriptive cross-sectional study on the determinants of manual dexterity in HMSN 1a. Manual dexterity was measured twice with a minimal interval of 5 days. To reduce variability, the same investigator (AV) took all measurements at the same location.

Data analysis

Scores on SHT and FDT were analysed using descriptive statistics. The ability to perform the tests was employed as an empirical indicator of feasibility. Homogeneity (internal consistency) of the SHT was expressed in Cronbach's α. Test-retest reliability of the SHT sum scores and the FDT raw scores was assessed by calculating intraclass correlation coefficient's (ICC) and the 95% confidence interval (CI) of the ICC, from a random effects one-way analysis of variance. A lower limit of the CI of at least 0.75 was considered as good test-retest reliability (23, 24). Systematic differences between visits were tested with Student's *t*-tests. Agreement of measurements was analysed according to the Bland-Altman method (25). For all analyses, an alpha level of $p < 0.05$ was used. All data was analysed using the SPSS 12.0.1 statistical program.

RESULTS

From the group of 63 subjects with HMSN 1a, 53 were willing to participate in this study. Four subjects were excluded; 3 due to co-morbidity and one due to alcohol abuse. Characteristics of the final study sample (n = 49, 78% response rate) are shown in Table I. Forty-two subjects were willing to return for a second evaluation of manual dexterity (mean interval of 27 days).

Table I. *Characteristics of the study sample (n = 49)*

Characteristics	
Sex, *n* (%)	
male	21 (43)
female	28 (57)
Age (years)	
mean (SD)	46.8 (11.7)
range	21–69
Age per stratum, *n* (%)	
18–39 years	13 (26.5)
40–59 years	29 (59)
≥ 60 years	7 (14.3)
Hand dominance, *n* (%)	
right	48 (98)
left	1 (2)
Disease duration* (years)	
mean	31.5
median	30.7
range	2.7–60.1
Hand involvement, *n* (%)	
yes	37 (75.5)
no	11 (22.5)
missing	1 (2)
Duration hand involvement† (years)	
mean	11.6
median	6.7
range	0–54.8
Working status, *n* (%)	
yes	32 (65.3)
no	16 (32.7)
not applicable	1 (2)

*Time since first symptoms of hereditary motor and sensory neuropathy.
†Time since first symptoms of the hand.

Ninety-four percent of the subjects had an abnormal SHT sum-score for the dominant hand (< 80) and 59% of the subjects for the non-dominant hand (< 77). The distributions of the SHT scores appeared to be skewed, showing marked clustering between 70 and 79 points. Although the sum-scores ranged from 33 to 80 for the dominant hand and from 41 to 80 for the non-dominant hand,

Table II. *Score distributions of the Sollerman hand function test (SHT) and the Functional Dexterity test (FDT) (n=49)*

Test	Hand	Median (P25;75)	Range	Mean (SD)
SHT	D	76 (67.5; 78)	33–80	
	ND	76 (69; 77.5)	41–80	
FDT-I (sec)	D	25.1 (21.9; 36.3)	16.2–93.0	31.0 (14.7)
	ND	28.5 (24.5; 37.8)	18.5–73.0	33.2 (13.2)
FDT-C (sec)	D	29.0 (22.9; 49.2)	16.2–149.4	
	ND	33.4 (26.4; 49.7)	20.8–144.8	

The sum-scores of the SHT are given in points; SHT norm scores for the dominant hand: 80 points and non-dominant hand: 77–79 points. FDT-I: initial FDT time score; FDT-C: combined FDT score (with penalties). D: dominant hand; ND: non-dominant hand; SD standard deviation.

the P25 indicates that the majority (75%) of the subjects had a sum-score of 67.5 or more for the dominant hand (Table II).

No difference was found between the SHT sum-scores for the dominant and non-dominant hands (mean difference –0.5, standard deviation (SD) 4.9, $p = 0.45$). SHT scores did not correlate significantly with age ($r_s = –0.27$, $p = 0.06$), but a negative correlation with disease duration was found ($r_s = –0.34$, $p = 0.02$, Fig. 1).

None of the 20 SHT subtests were performed within reference limits by all subjects (Table III). Subtest 8 (pick up nuts and put on bolts) stands out, with 90% of the subjects scoring below normal with the dominant hand and 96% with the non-dominant hand, respectively.

Median and percentile scores of the FDT are presented in Table II. Fine hand use of subjects with HMSN 1a ranged from functional to non-functional, with an average FDT score indicating a "moderately functional" level. Compared with the initial FDT time scores, the median of the combined scores were 3.9 sec longer for the dominant hand and 4.9 sec longer for the non-dominant hand, respectively. Large differences between initial time scores and combined scores were seen for the P75 scores in particular, with differences of 9.9 and 11.9 sec, respectively.

Feasibility of the SHT and FDT was good. Although we observed various compensatory movement patterns, all subjects were able to complete both tests within the reported administration time (14, 15) and test scores were obtained in all subjects.

The homogeneity of the SHT subtests scores was high for both the dominant ($\alpha = 0.96$) and the non-dominant hand ($\alpha = 0.95$), indicating good internal consistency. The test-retest reliability was excellent for the measurements of the SHT. ICCs ranged from moderate to good for initial FDT scores and from good to excellent for FDT combined scores (Table IV). There were no systematic differences in SHT and FDT scores between visits.

For the dominant hand, the 95% limits of agreement between the 2 SHT measurements were –3.1 to 2.9 points, and for the non-dominant hand –4.1 to 3.1 points (Table IV). Since the Bland-Altman plots of the FDT showed that the difference between 2 measurement values was proportional to their mean value, limits of agreement were calculated on log-transformed data. The limits of agreement for the FDT were wide (Table IV).

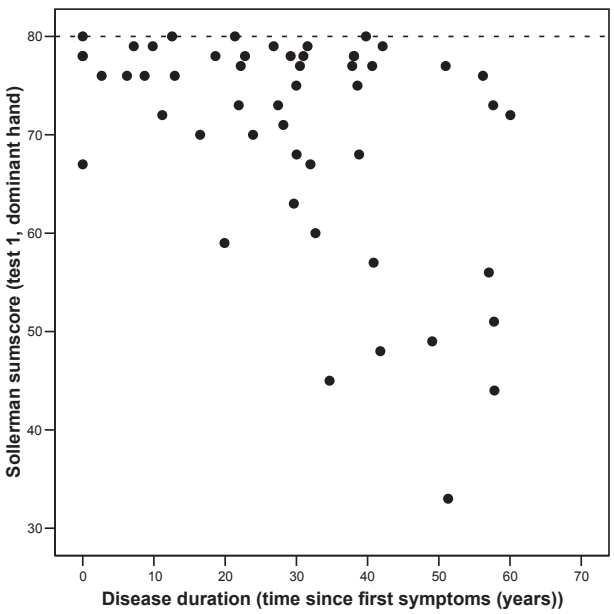

Fig. 1. Relationship between manual dexterity and disease duration.

Table III. *Results of the Sollerman hand function test (SHT) subtests*

Subtest		Expected grip	Hand	% below norm
1	Put key into Yale lock, turn 90˚	Pulp pinch, lateral pinch	D	29
			ND	22
2	Pick coins up from flat surface, put into purse mounted on wall	Pulp pinch	D	37
			ND	25
3	Open/close purse	Pulp pinch, lateral pinch	D	41
			ND	47
4	Pick up coins from purses	Pulp pinch	D	33
			ND	45
5	Lift wooden cubes over edge 5 cm in height	5-finger pinch	D	33
			ND	33
6	Lift iron over edge 5 cm in height	Transverse volar grip	D	16
			ND	12
7	Turn screw with screwdriver	Diagonal volar grip	D	18
			ND	12
8	Pick up nuts and put on bolts	Pulp pinch, lateral pinch, tripod pinch	D	90
			ND	96
9	Unscrew lid of jars	Spherical volar grip	D	29
			ND	25
10	Do up buttons	Pulp pinch, lateral pinch	D	41
			ND	33
11	Cut modelling clay with knife and fork	Tripod pinch, diagonal volar grip	D	33
			ND	20
12	Put tubigrip stocking on the other hand	Lateral pinch, 5-finger pinch	D	20
			ND	20
13	Write with a pen	Tripod pinch	D	31
			ND	41
14	Fold paper, put into envelope	5-finger pinch, lateral pinch	D	33
			ND	33
15	Put paper clip on envelope	Pulp pinch, lateral pinch	D	35
			ND	33
16	Pick up telephone-receiver and put it to the ear	Diagonal volar grip	D	14
			ND	12
17	Turn door-handle 30˚	Transverse volar grip	D	10
			ND	10
18	Pour water from 1 litre paper milk package	5-finger pinch	D	20
			ND	18
19	Pour water from jug	Transverse volar grip	D	10
			ND	12
20	Pour water from cup	Pulp pinch, lateral pinch	D	37
			ND	33

The % scores represent, with regard to the subtest, the percentage of subjects with a manual dexterity score below normal performance. Scores are obtained from the first administration of the SHT.
D: dominant hand; ND: non-dominant hand.

DISCUSSION

The results of this study show that impaired manual dexterity is a common finding among subjects with HMSN 1a, and

Table IV. *Reproducibility: intraclass correlation coefficient (ICC) and Bland and Altman tests*

		ICC (one-way random)		Bland and Altman				
Test	Hand	ICC coefficient	95% CI	$\bar{d}$	SE of $\bar{d}$	95% CI for $\bar{d}$	SDdiff	95% limits of agreement
SHT	D	0.99	0.98 → 0.99	–0.07	0.23	–0.54 → 0.39	1.5	–3.1 → 2.9
	ND	0.98	0.97 → 0.99	–0.50	0.28	–1.06 → 0.06	1.8	–4.1 → 3.1
FDT-I (sec)	D	0.87	0.77 → 0.93	–1.76	1.00	–3.77 → 0.25	6.45	–29.0 → 28.0
	ND	0.83	0.71 → 0.91	0.14	1.21	–2.31 → 2.59	7.86	–31.0 → 49.0
FDT-C (sec)	D	0.93	0.88 → 0.96	–0.67	1.96	–4.63 → 3.29	12.71	–44.0 → 65.0
	ND	0.95	0.91 → 0.97	–1.04	1.51	–4.10 → 2.02	9.81	–39.0 → 52.0

ICC values are presented with their 95% confidence interval; the 95% limits of agreement of the FDT-I and FDT-C scores are expressed as ratios (derived by taking the anti-logs of the limits of agreement calculated on log-transformed data).
$\bar{d}$: mean difference; SE of $\bar{d}$: standard error of the mean difference; 95% CI for $\bar{d}$: 95% confidence interval for the mean difference; SDdiff: standard deviation of the differences; SHT: Sollerman hand function test; FDT-I: initial score of Functional Dexterity test; FDT-C: combined FDT score. D: dominant hand; ND: non-dominant hand.

on average, fine hand use can be categorized as "moderately functional". Activities that require finger grips such as the pulp pinch, the tripod pinch and the lateral pinch, are most limited.

Reduced manual dexterity, described in the International Classification of Functioning, Disability and Health (ICF) as "fine hand use", may hamper the execution of many daily activities of subjects with HMSN 1a and lead to restrictions in participation (7). For the rehabilitation management of upper extremity related disabilities the evaluation of manual dexterity with a dexterity test, qualified in an ICF perspective as capacity, is of crucial importance. It provides information about the patient's ability to execute a task or an action. Because activities are the primary focus of rehabilitation, manual dexterity needs to be objectified, not merely as an outcome measure, but also for a better understanding of the causes of activity limitations and of the consequences of the impairments in body functions for hand use. These relationships need to be known to design and evaluate rehabilitation interventions.

In this study we have used the SHT and the FDT. Previously we used the Jebsen test of hand function to evaluate manual dexterity in subjects with HMSN type I and II (12). Although the Jebsen test also includes functional tasks, it only measures the time needed to perform a task. As a consequence, with this test some HMSN subjects were classified as having normal manual dexterity, although they showed various compensatory movements. Since the objective of evaluating manual dexterity is to provide data about the speed, accuracy and manner of hand and finger use, manual dexterity should be evaluated with a test that also incorporates qualitative aspects of movement (8). The SHT and FDT both provided, besides speed, additional information on the quality of movement. The need of subjects to use compensatory movement patterns is incorporated into both scoring systems, and with the SHT an evaluation of the various grip patterns commonly used in daily life was obtained and a broad spectrum of functional tasks is evaluated. Compared with the FDT, the tasks of the SHT are more representative for activities of daily living and also include bilateral tasks. The SHT therefore better reflects the ability of HMSN 1a subjects to use their hands in daily life. The FDT, on the other hand, is less time-consuming.

The relatively high sum scores on the SHT suggest that the severity of the limitations in dexterity is mild. However, it should be realized that the SHT measures *overall* hand function (14). Low scores on the SHT are obtained only if *all* grip patterns are severely affected. HMSN 1a subjects performed relatively well on those subtests that required a volar grip, while activities that require finger grips, such as like the pulp pinch and the tripod pinch, were difficult to perform. Picking up nuts and putting them on bolts using a tripod pinch was the most difficult task. Similar SHT scores were found in other studies of patients with affected hand function due to Dupuytren's contractures (16), and after long-term haemodialysis (20).

Only 2 studies on manual dexterity in HMSN were found (1, 10). Miller et al. (1) reported poor performance on a Purdue pegboard and, in keeping with our results, the greatest impairments were found in areas requiring the most precise function. However, data on reliability and agreement of the Purdue pegboard in HMSN subjects were not reported. More recently, Svensson et al. (10) evaluated the manual dexterity and reliability of the Nine-hole-peg test and the Box and Block test in 20 subjects with various types of CMT. In line with the results of our study, subjects with CMT were much slower than age- and sex-matched norms. The test-retest reliability of both tests was found to be good, but it must be realized that the various compensatory hand grip techniques of HMSN subjects are not taken into account with these tests. Furthermore, as opposed to the SHT, the Nine-hole-peg test and Box and Block test provides solely information on the use of a pinch grip.

With respect to representativeness of our findings, selection bias might have occurred. Subjects with more upper limb impairment could be more willing to participate than those with less impairment. However, a high percentage (77.8%) of the known HMSN 1a subjects at our departments participated in this study, selection was made only on the basis of diagnosis and 22.5% of our study sample did not experience any upper limb involvement (Table I).

Opinions differ about the natural course of HMSN 1a in adulthood. A recent study suggests that disease duration is associated with the severity of signs and symptoms (3). Our study findings support this hypothesis (Fig. 1), but this should further be evaluated in prospective follow-up studies.

An essential requirement of all outcome measures is that they are feasible, valid and reproducible. Feasibility of the SHT and FDT was good. All subjects were able to complete both dexterity tests, varying from subjects with no limitations to subjects with severe involvement of the upper limb.

The test-retest reliability of the SHT measurements was excellent and moderate to excellent for FDT scores. The Bland and Altman 95% limits of agreement tests showed that, for example, for the dominant hand only a decline between 2 SHT measurements of more than 3 points can be interpreted as a real decline in manual dexterity. The limits of agreement for the initial and combined scores of the FDT showed rather wide ranges. This has to be taken into account when tests are used for longitudinal evaluation of manual dexterity of individual subjects with HMSN. Together with the fact that, compared with the FDT, the SHT tasks better represent activities of daily living, we propose the SHT be used in HMSN 1a for monitoring disease progression and evaluating hand therapy programmes. The FDT can serve as a quick test to assess the severity of impaired dexterity.

This study did not aim to provide full validity and reliability of the SHT and FDT in subjects with HMSN. For that purpose, studies that focus on inter-observer reliability and concurrent validity are warranted.

In conclusion, impaired manual dexterity is common in HMSN 1a, and evaluation of fine hand use should therefore be included as an essential part of the functional assessment. Both the SHT and the FDT dexterity tests render detailed information on various aspects of dexterity in subjects with HMSN 1a. Based on the representation of daily life activities and reliability, the SHT should be used in clinical practice, to monitor disease progression and the effects of hand treatment programmes, and in research to evaluate the relationships between dexterity on the one hand and impairments in body functions and activity limitations on the other. Better insight into these relationships is needed in order to develop adequate, evidence-based interventions.

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