

# Dynamometry of intrinsic hand muscles in patients with Charcot–Marie–Tooth disease

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**Abstract—Background:** Several problems are associated with manual muscle testing and dynamometry in the hands of patients with Charcot–Marie–Tooth (CMT) disease. **Objective:** To evaluate the efficacy of the Rotterdam Intrinsic Hand Myometer (RIHM) to directly measure intrinsic hand muscle strength in CMT disease. **Methods:** We measured hand muscle strength and hand function in 41 patients with CMT disease. **Results:** RIHM measurement of intrinsic strength had excellent reliability. We found overlapping RIHM strength values in Medical Research Council grades 3 to 5. High grip and pinch strength could be found in patients with severe intrinsic muscle weakness. RIHM measurements were more strongly correlated with fine motor skills of the hand than grip and pinch strength. **Conclusions:** The Rotterdam Intrinsic Hand Myometer is a reliable instrument to measure intrinsic hand muscles strength in patients with Charcot–Marie–Tooth disease, providing more detailed information than manual muscle testing and a more direct assessment of intrinsic muscle loss than grip and pinch dynamometers.

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The Medical Research Council (MRC) 0 to 5 scale is often used to manually quantify hand muscle weakness in patients with Charcot–Marie–Tooth (CMT) disease<sup>1,2</sup> because it is easy to administer and differentiates between muscles that cannot contract (grade 0) and muscles with small contractile properties (grades 1 and 2). However, manual muscle testing is not very sensitive in detecting change within grades 3 to 5.<sup>3–8</sup> Alternatively, pinch and grip strength dynamometers are used to measure hand strength.<sup>9,10</sup> However, these instruments evaluate intrinsic and extrinsic hand muscles in combined action.<sup>11</sup> As a result, they may underestimate intrinsic muscle weakness in CMT disease as the extrinsic hand muscles are less severely affected.<sup>12,13</sup>

The Rotterdam Intrinsic Hand Myometer (RIHM) is a hand-held dynamometer<sup>13–15</sup> that directly measures intrinsic hand muscle strength. In this study, we evaluated the use of the RIHM to measure intrinsic hand muscles in isolation in patients with CMT disease. To do so, we evaluated the reliability and compared the RIHM outcomes of the intrinsic hand muscles with manual muscle testing measurements and generally accepted dynamometers for hand and

wrist muscle strength. To assess whether the application of the RIHM measurements in patients with CMT disease provides additional information to pinch and grip strength dynamometers, we compared the relation between the strength measures and two activity measures of the upper extremity function.

**Methods. Participants.** We recruited patients from the rehabilitation outpatient clinic of the Erasmus Medical Center (the Netherlands) to participate in this study. All patients were diagnosed with CMT disease using electrophysiologic, clinical, or DNA analysis and were between ages 18 and 80. Patients were excluded if they had comorbidity that could interfere with muscle strength or hand function or if they had been operated on both hands. If patients were operated on one hand, only data from the nonoperated hand were included in the analysis. Written informed consent was obtained from all subjects.

**Measurements.** All measurements were performed by the same researcher. All subjects were asked for the duration (in years) since they first had noticed hand problems. In addition, we asked for hand dominance and for the most important impairments in hand function as experienced by the patient, such as fatigue, pain, or clumsiness. Visual analogue scales (VASs; scored between 0 and 100) were used to indicate pain and fatigue. Sensory loss was assessed using the Weinstein Enhanced Sensory Test at six locations at the palmar side of the hand.<sup>16</sup> The nylon monofilaments are labeled from 2.83 to 6.65, corresponding to the force required to bend the filament. The average of the six locations was calculated.

**Strength measurements.** Manual muscle testing and RIHM measurements (figure 1) were performed for three intrinsic muscle groups, that is, abduction of the thumb, abduction of the index finger, and abduction of the little finger. Manual muscle testing was scored based on the MRC scale, adapted for assess-

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Figure 1. Illustration of the strength measurement of the abduction of the index finger using the Rotterdam Intrinsic Hand Myometer.

ment of hand muscles.<sup>17,18</sup> The details of the RIHM and the measurement protocol have been described elsewhere.<sup>13,15</sup> In short, during the RIHM measurements, patients were asked to hold the finger or thumb in the instructed position while the force was slowly increased until the subject would not be able to maintain the same position (break test). The places at which the force was recorded in the RIHM measurements were similar to the anatomic reference points used during manual muscle testing.<sup>18</sup> When the manual muscle testing grade was less than 3, RIHM dynamometry was not possible because no resistance could be given and a “0” score was recorded. The reliability of the RIHM measurements has not been established in CMT disease. Therefore, in a separate session, we evaluated 15 patients with MRC grades of >2 for the three intrinsic hand muscle groups (abduction of the thumb, abduction of the index finger, and abduction of the little finger). To establish intraobserver reliability, two measurements were performed by the same researcher for the three intrinsic muscle groups. In addition, to establish interobserver reliability, the same patients were also evaluated by a second researcher.

Maximal isometric contraction of grip, tip pinch, and key pinch were measured using a Lode handgrip and pinch-grip dynamometer. Grip force measurements were performed with the Lode handgrip dynamometer (handle position 2), similar to the Jamar hand dynamometer, and with 4.6-cm distance between the handles.<sup>19</sup> Tip pinch and key pinch were measured with the Lode pinch-grip dynamometer, which is similar to the Preston pinch dynamometer.<sup>20</sup> Tip pinch was measured with the index finger on the top and the thumb below and with the other fingers flexed. Key pinch was measured with the thumb on the top and the radial side of the index finger below.

Muscle strength of the wrist flexor and extensor strength was measured using a microFET-2 hand-held dynamometer.<sup>21</sup> The patient was asked to hold the wrist in dorsal or palmar flexion, while the examiner increased the resistance on the wrist until the hand moved (break test). For wrist extension, force was measured at the mid location of the third metacarpal level. For wrist flexion, force was recorded at the center of the palm of the hand.

During all strength measurements, patients were seated and were verbally encouraged to produce their maximal voluntary contraction. To minimize the influence of fatigue, the measurements for the right and the left hand were alternated. For all measurements, the mean of three maximum voluntary contractions was recorded.

**Hand function.** Hand function was measured using three items selected from the Sollerman test assessing fine object manipulation.<sup>22</sup> The standardized Sollerman Hand Function Test<sup>23</sup> includes 20 tasks based on the most common hand grips performed during daily living. For patients recovering from peripheral nerve injuries at the wrist or distal forearm level,

the selected items were found to maximally correlate with the outcome of the full Sollerman test in these patients.<sup>22</sup> These selected items are 1) picking up four coins from surface and putting into purses mounted on a board, 2) picking up four nuts and screwing to bolts, and 3) doing up four buttons. The tests were done with the dominant hand and item scores ranged from 0 to 4 (0 indicates that “the task cannot be performed at all” and 4 indicates that “the task is completed without any difficulty within 20 seconds and with the prescribed hand grip of normal quality”). Outcomes were presented as the sum of the three items (range 0 to 12).

The Disabilities of the Arm, Shoulder, and Hand (DASH) questionnaire was used as a standardized outcome measure of upper-extremity disability from the perspective of the patient. The DASH is a 30-item self-report questionnaire that measures upper-extremity disability with a five-response option for each item (1 = “no difficulty” to 5 = “unable”).<sup>24,25</sup> The sum of the responses is transformed to a DASH score between 0 and 100, with higher DASH scores indicating increased disability. The DASH has good validity, test-retest reliability, and responsiveness to study musculoskeletal disorders and its usefulness as a measure to monitor upper-extremity conditions has been confirmed.<sup>26</sup> In the current study, we used the standard part of the DASH, omitting the two optional sections on sports/music (four items) and work activities (four items).

**Statistical analysis.** Statistical analysis was performed using SPSS 12.0 for Windows. Results are presented as means  $\pm$  SD or numbers. Intraclass correlation coefficients (ICCs) were calculated to determine intra- and interobserver reliability. In addition, the SEM and the smallest detectable difference (SDD) were calculated.<sup>13,27</sup> The SEM reflects the variability of measurements due to repetition and random error. The SDD reflects the smallest change that can be detected in a subject.

A paired sample *t* test was used to test for differences between the dominant and nondominant hand. Relations between the manual muscle strength parameters and hand function were investigated using the nonparametric Spearman correlation coefficient. Relations between the dynamometry measurements of the arm and hand muscles were investigated using the parametric Pearson correlation coefficient. A probability value of  $\alpha \leq 0.05$  determined significance.

**Results.** Forty-nine of the patients of the Department of Rehabilitation Medicine of the Erasmus Medical Center were eligible for the study. From these patients, 43 patients (87%) agreed to participate. The patients were relatively diverse in terms of age and duration since the onset of the disease. Most of the patients had either type I (32%) or type II (37%) CMT disease or had not been diagnosed with a specific type of CMT disease (27%). The most frequently mentioned hand problem concerned the manipulation of small objects (63%).

We found no significant differences in the muscle strength, hand function, and sensibility between the dominant and the nondominant hand. Therefore, we present only the results of the dominant hand. As two subjects underwent operations on their dominant hand, these two subjects were excluded from the analysis ( $n = 41$ ) (table 1).

**Reliability of RIHM measurements.** The ICCs for the intraobserver reliability (table 2) ranged between 0.92 and 0.98 (average 0.94). For the interobserver reliability, ICCs ranged between 0.86 and 0.97 (average 0.93).

**Strength measurements.** Table 3 provides the group descriptive data of the strength measurements. Correlations were found between manual muscle testing and RIHM measurements of the intrinsic hand muscles (table E-1 on the *Neurology* Web site at [www.neurology.org](http://www.neurology.org); figure 2; for all correlations,  $p < 0.01$ ). However, despite these correlations, we found overlapping strength values of the intrinsic muscle strength in MRC grades 3 to 5.

**Table 1** Subject characteristics (n = 41)

Variable	Group descriptive data
Age, y	44.1 ± 15.2 (18–80)
Height, m	1.75 ± 0.10 (1.58–1.98)
Gender, M/F	22/19
Body wt, kg	76.6 ± 16.3 (42–140)
Time since onset hand problems, y	12.4 ± 12.6 (1–45)
Type CMT disease, no. (M/F)	
I	13 (5/8)
II	15 (8/7)
III	2 (1/1)
Unknown	11 (8/3)
Reported hand problems, %	
Manipulation of small objects	63
Fatigue	29
Loss of hand strength	24
Clumsiness	20
Weinstein Enhanced Sensory Test	3.77 ± 0.89 (2.83–6.65)
Pain (VAS)	30.7 ± 18.6 (0–90)
Fatigue (VAS)	51.3 ± 29.1 (0–100)
Sollerman	8.1 ± 2.7 (2–11)
DASH	28.7 ± 17.8 (0–70.8)

Values indicate means ± SD (range), numbers, or percentages. Time since onset of hand problems is based on 35 patients, because 6 patients never had experienced any hand problems or could not recall the onset of their hand problems. Weinstein Enhanced Sensory Test filament data indicate the mean of the six locations measured in each subject.

VAS = visual analogue scale; DASH = Disabilities of the Arm, Shoulder, and Hand questionnaire.

The correlations within each of the three groups of dynamometer measurements (RIHM, pinch and grip, and wrist dynamometry) were consistently higher than the correlations between these groups. The correlation coefficients between maximal isometric contraction of grip, tip pinch, and key pinch ranged between 0.79 and 0.89 (table E-1). Similarly relative high correlations coefficients were found between wrist flexion and extension (0.89) and between the intrinsic muscles (between 0.79 and 0.86). In

contrast, the correlations between the strength measurements that predominantly assess the extrinsic hand muscles (grip and wrist strength) and the intrinsic muscles strength (abduction of thumb, index finger, and little finger) were consistently lower than within each group. Comparing the correlations between muscle groups, the correlations between the wrist strength measurements and the RIHM measurements were the lowest (between 0.31 and 0.54), whereas the correlations between grip strength and intrinsic muscles strength were between 0.55 and 0.65. Comparing grip, pinch, and key strength, we found that pinch grip was most strongly correlated with the strength of the intrinsic hand muscles.

Figure 3 visualizes the relations between grip strength, wrist flexion strength, and intrinsic muscle strength in more detail, showing a pattern in which a relatively high grip strength or wrist flexion strength can correspond with both a high or a low intrinsic muscle strength. In other words, whereas patients with a relatively low grip strength always have a relatively low intrinsic muscle strength, a good grip strength does not necessarily correspond with a good strength of the intrinsic muscles.

**Dynamometry and hand function.** Comparing the correlations between the different strength measurements and activities of daily living and disability measured with the modified Sollerman and the DASH (table E-2), we found that the Sollerman test, which specifically measures fine motor tasks, was more strongly correlated with the intrinsic muscle strength. In contrast, the DASH, which is a more global measure of upper extremity function, was more strongly correlated with the extrinsic strength measurements. These differences in the correlations between RIHM and pinch and grip outcome measures indicate that the different measurements evaluate different aspects of muscle loss of the upper extremity.

**Discussion.** In the current study, we investigated if the RIHM is able to measure the intrinsic hand muscles in isolation, as in manual muscle strength testing, while having the advantage of dynamometry to detect smaller changes in the MRC grades 3 to 5 in patients with CMT disease. We compared a number of instruments to assess muscle strength of the hand in patients with CMT disease and specifically studied the efficacy of direct dynamometer assessment of the intrinsic hand muscles using the RIHM

**Table 2** Intra- and interobserver repeatability of Rotterdam Intrinsic Hand Myometer measurements

	Intraobserver			Interobserver		
	ICC	SEM, N	SDD, N	ICC	SEM, N	SDD, N
Abduction thumb dominant	0.93 (0.80–0.98)	4.5	12.6	0.93 (0.80–0.98)	4.1	11.4
Abduction thumb nondominant	0.95 (0.83–0.98)	3.9	10.7	0.96 (0.87–0.99)	3.3	9.2
Abduction index finger dominant	0.98 (0.94–0.99)	1.9	5.3	0.97 (0.91–0.99)	2.1	5.8
Abduction index finger nondominant	0.93 (0.80–0.98)	3.3	9.2	0.95 (0.86–0.98)	2.4	6.8
Abduction little finger dominant	0.93 (0.81–0.98)	2.1	5.9	0.90 (0.73–0.99)	2.1	5.7
Abduction little finger nondominant	0.92 (0.79–0.97)	2.2	6.2	0.86 (0.70–0.96)	2.6	7.2

Shown are the intraclass correlation coefficient (ICC) with the 95% CI between brackets, the SEM, and the smallest detectable difference (SDD). SEM and SDD in Newtons.

**Table 3** Group values for strength measures of patients with Charcot-Marie-Tooth disease

Variable	Male, mean $\pm$ SD						Female, mean $\pm$ SD					
Grip strength, N	293.3 $\pm$ 146.7						175.9 $\pm$ 98.5					
Tip pinch strength, N	35.6 $\pm$ 25.2						27.6 $\pm$ 13.8					
Key pinch strength, N	57.0 $\pm$ 30.9			39.7 $\pm$ 24.4								
Wrist flexion, N	158.6 $\pm$ 91.8						85.4 $\pm$ 45.9					
Wrist extension, N	131.3 $\pm$ 75.9						69.9 $\pm$ 45.9					
Abduction thumb, N	32.8 $\pm$ 28.3						24.4 $\pm$ 17.5					
Abduction index finger, N	19.6 $\pm$ 19.4						17.2 $\pm$ 12.8					
Abduction little finger, N	14.6 $\pm$ 10.6						9.6 $\pm$ 7.6					

	MRC grade, % of patients											
	Male						Female					
	0	1	2	3	4	5	0	1	2	3	4	5
Abduction thumb	18	0	5	5	46	27	11	5	5	5	32	42
Abduction index finger	14	9	0	14	50	14	5	11	0	11	42	32
Abduction little finger	5	5	5	18	59	9	5	5	0	16	58	16

Values indicate group mean and SD (dynamometry in Newtons) or the percentage of patients scored with Medical Research Council (MRC) grades 0 to 5 during the manual muscle strength testing.

as compared with pinch and grip strength dynamometry and manual muscle testing.

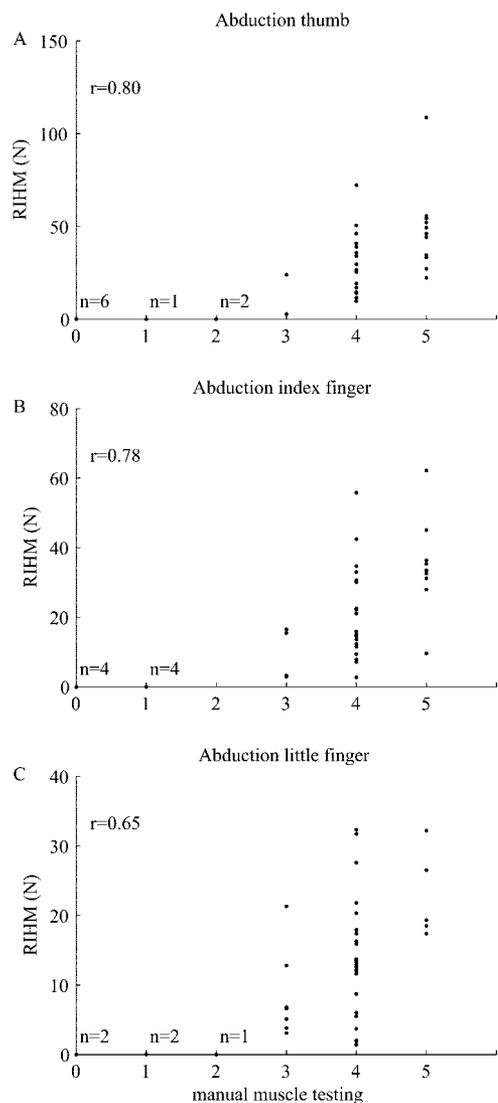
On average, the ICC of the intraobserver reliability of the three muscles groups for the dominant and nondominant hand was 0.94; for the interobserver reliability, the average ICC was 0.93. Although the reliability data are based on a relatively small number of subjects, the outcomes are comparable with findings in patients with intrinsic muscle loss due to ulnar and median nerve injuries, where ICCs of the interrater repeatability for the RIHM measurements were 0.94 or higher.<sup>13</sup> The ICCs are comparable with pinch and grip strength data in other patient groups<sup>19</sup> and are appropriate to study the recovery and function of the intrinsic muscles of the hand in individual subjects. The SEM and SDD values found in the current study were comparable with those found in patients with median or ulnar nerve injuries.<sup>13</sup> The SEM and SDD provide insight into the errors that can be expected in RIHM measurements of the intrinsic hand muscles in patients with CMT disease and provide a reference when evaluating clinical change.

The strength values found in the current study are generally in line with findings reported in the literature. For example, the grip strength measured in 86 patients with CMT disease<sup>2</sup> was approximately 170 and 280 N (women and men) compared with 176 and 293 N in the current study. In another study on 20 patients with CMT disease,<sup>10</sup> a mean grip force was found of 227 N in women and men combined. In the same study, mean key-pinch forces of 48 N and mean tip-pinch forces of 69 N were found, which are higher than the values found in the current study

(28 for women and 36 for men for the tip pinch and 40 and 57 for key pinch).

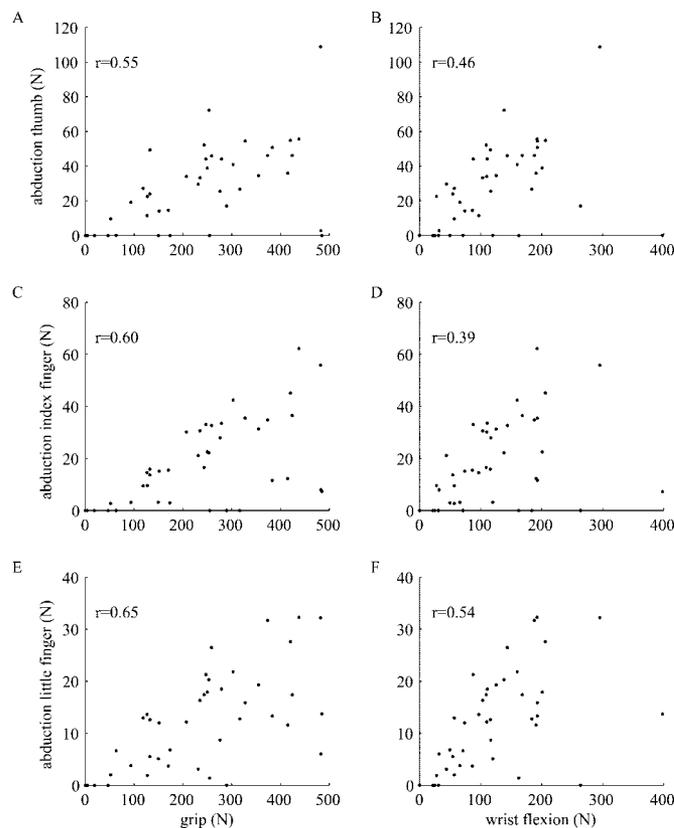
Although the number of values in some of the MRC grades are relatively small, the data indicate that there are overlapping RIHM strength values for the intrinsic hand muscles within the manual muscle testing grades 3 to 5. For example, the strength measured with the RIHM in patients with MRC grade 4 for the abduction of the index finger ranged between 2 and 56 N, whereas the strength within the MRC grade 5 ranged between 9 and 62 N (figure 2). These overlapping values are in line with other studies on knee muscle strength<sup>28</sup> and on shoulder and elbow muscle strength<sup>29</sup> reporting similar overlapping values in MRC grades 3 to 5. In addition to the overlapping strength values, we found only modest correlations between manual muscle testing and dynamometry of the intrinsic hand muscles, ranging between 0.65 and 0.80. To our knowledge, the relation between manual muscle testing and dynamometry has not been studied before for the intrinsic hand muscles. However, these findings are also in line with literature on other muscle groups, reporting correlations between 0.48 and 0.90.<sup>7</sup> Overall, the current data therefore support other studies concluding that although manual muscle testing can be a useful and easy to administer clinical tool, dynamometers may be more appropriate to detect relevant changes in MRC grades of 3 and higher, to detect differences between body sides, or to detect deficits in patients relative to normal.<sup>3,7</sup>

Upon comparison of the correlations between the RIHM measurements and the other dynamometry measurements (pinch, grip, and wrist strength), the



**Figure 2.** Scatter plots visualizing the relation between the manual muscle testing and Rotterdam Intrinsic Hand Myometer (RIHM) measurements in the individual subjects. Patients with a manual muscle testing grade of less than 3 were not assessed with the RIHM but scored as 0. The number of patients scoring grades 0 to 2 are indicated in the figure. Significant correlations were found between the manual muscle testing and RIHM measurements. However, much overlap can be seen between the force levels recorded in patients with different manual muscle testing grades. Spearman correlation coefficients ( $r$ ) of the visualized relations are shown in each plot.

highest correlations were found between RIHM measurements and tip and key pinch (between 0.58 and 0.83), whereas the correlations between the RIHM and the grip strength were lower (between 0.55 and 0.65). The relatively high correlation between RIHM and pinch measurements may reflect the important contribution of the intrinsic hand muscle to the total pinch strength. In a study on healthy subjects,<sup>11</sup> it was shown that temporary motor block of the intrinsic hand muscles decreased grip strength by 49% and key pinch by 85%, indicating that key pinch is more strongly related to intrinsic muscle strength than



**Figure 3.** Scatter plots visualizing the relation between maximal isometric contraction of the grip and the intrinsic hand muscles using the Rotterdam Intrinsic Hand Myometer (A, C, E) and the relation between the maximal isometric contraction of wrist flexion and the same intrinsic hand muscles (B, D, F). Pearson correlation coefficients ( $r$ ) of the visualized relations are shown in each plot.

grip strength. As expected, we found that wrist strength, which is not directly influenced by intrinsic muscle loss, had the smallest correlation with intrinsic muscle strength (correlation coefficients between 0.31 and 0.54).

The comparison of the strength measurements with two different hand function tests further indicates that RIHM, pinch, grip, and wrist muscle strength measurements evaluate different aspects of hand function in CMT disease. It was found that the RIHM measurements were most strongly related to the modified Sollerman task, which measures fine motor task of the hand and fingers. In contrast, the more global assessment of the upper extremity with the DASH was most strongly related to the muscle strength of the wrist flexors and extensors.

The hand function in the current study was measured using the DASH as well as a selection of the Sollerman test. The selection of the Sollerman items was based on research on patients with peripheral nerve injuries that showed similar patterns of muscle loss as patients with CMT disease, that is, intrinsic muscle loss with less or no extrinsic muscle loss, and was shown to be highly correlated with the total Sollerman test.<sup>22</sup> Although the selection consists of

only three items, the relatively high correlation between the Sollerman test and the intrinsic hand strength measurements (between 0.62 and 0.67) indicates that these items can be used to assess function of the intrinsic hand muscles.

From the scatter plots between grip, wrist flexion, and RIHM measurements, it can be seen that a reduced grip and wrist strength is always accompanied by a reduced intrinsic muscle strength. However, we found that the opposite was not always the case: A high grip or wrist strength can also be found in patients with little or even no (MRC < 3) intrinsic muscle strength (figure 3). Although we did not investigate how patients with severe intrinsic muscle loss were able to create high grip strengths, this might be due to training or compensation of the extrinsic hand muscles. As CMT is a chronic and slowly progressive disease, patients may have adapted by training their extrinsic hand muscles over the years to maintain sufficient grip strength. A similar compensatory mechanism to create grip strength has been reported in patients with ulnar nerve injuries.<sup>12,15</sup> An instrument like the RIHM that directly measures intrinsic muscle strength may be more appropriate than grip and pinch strength dynamometers to draw conclusions about the strength of the intrinsic hand muscles.

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