Stakeholder Engagement for Development of a New Measure of Hand and Arm Function in People with Neurological Conditions

Namrata Grampurohit[1]*, Brian Dudgeon[2] and Deborah Kartin[3]

[1]Department of Occupational Therapy, Thomas Jefferson University, USA

[2]Department of Occupational Therapy, University of Alabama, USA

[3]Department of Rehabilitation Medicine, University of Washington, USA

Corresponding author:
Namrata Grampurohit,
Assistant Professor in Occupational Therapy, Thomas Jefferson University, 901 Walnut Street, Suite 637, Philadelphia, PA 19107, USA,
Email:
namrata.grampurohit@jefferson.edu

ABSTRACT

Purpose: A new generic assessment, the Hand and Arm Function Measure, with both self-reported and performance-based items was devised for people with neurological conditions using the evidence-centered design framework. The objective of this study was to gather experiences of stakeholders regarding upper extremity function in daily activities and seek opinions regarding a preliminary set of items to establish face and content validity.

Methods: This descriptive qualitative study included focus groups, cognitive interviews, and an open-ended survey. Stakeholders (n=24) were selected by purposeful sampling of content experts in rehabilitation (n=4) and people who had stroke (n=7), traumatic brain injury (n=2), Parkinson disease (n=6), and multiple sclerosis (n=5). Responses were coded and thematically analyzed by two authors independently.

Results: The construct was operationally defined and relevant items categorized based on International Classification of Functioning, Disability, and Health. The items were designed based on aspects of upper extremity function relevant to this population. A 145-item bank was generated and a preliminary set of 59 items (14 performance-based and 45self-reported) systematically identified and modified.

Conclusions: Face and content validity developed through stakeholder engagement helped generate the evidence to develop a comprehensive outcome measure in rehabilitation. Further investigation of the psychometric properties is needed.

INTRODUCTION

Neurological conditions such as stroke, Traumatic Brain Injury (TBI), Parkinson Disease (PD), and Multiple Sclerosis (MS) can cause upper extremity function impairments [1]. Clinicians, payers, patients, and all stakeholders need measures of upper extremity function that are relevant and appropriate for individuals with neurological conditions [2,3]. Reliable and valid upper extremity function measures are essential to monitor progress, set goals, determine effectiveness of intervention, and seek reimbursement for therapy services [4,5]. In order for a measure of upper extremity function to be useful for these purposes and psychometrically robust, it is critical for test items to be developed carefully with stakeholder engagement [6,7].

The existing upper extremity function measures can be categorized into performance-based and self-reported measures. Some performance-based measures, such as the Nine Hole Peg Test [8], can be used for multiple diagnostic populations [9]. Other

01

Stakeholder Engagement for Development Of a New Measure of Hand and Arm Function in People with Neurological Conditions. Physical Medicine & Rehabilitation Journal. 2019; 2(1):119.

SCIENTIFIC LITERATURE

performance-based measures are disease-specific, such as the Wolf Motor Function Test [10], which can assess upper extremity impairments in individuals post-stroke [11]. In recent years, self-reported measures for assessing upper extremity function have been developed, such as the Motor Activity Log for people with stroke [12]. Neurological conditions are unique in their wide variety of symptom presentations based on the site of lesion and no disease-specific measure can rightly capture the broad range of upper extremity deficits seen in the clinic. If there were sufficiently robust disease-specific measures available to a clinician, the feasibility of administration would still persist as a major barrier to their appropriate use. Although many outcome measures have been available for stroke, clinicians have not adopted widespread use of these measures due to limitations such as lengthy administration time, small normative sample size, poor evidence for validity of scores at varying severity, and a lack of items that assess activity and participation [13]. Further, there are no disease-specific upper extremity function measures for other neurological conditions, e.g., multiple sclerosis, and clinicians find themselves using measures developed for stroke in other neurological conditions without thepsychometric evidence to support their use. Thus, there is a need for the conceptualization and development of a measure of upper extremity function to overcome these limitations.

Upper extremity functional ability to participate in daily activities is a complex measurement construct. Currently there is no measure that assesses both upper extremity activity and participation; therefore, to thoroughly assess all aspects of this construct a clinician has to use many different types of measures. For example, an observation-based performance measure can be an indicator of the upper extremity activity and a self-reported questionnaire can provide information about participation [5]. However, in the clinic, using multiple upper extremity assessments for each patient is challenging due to the burden of administration (e.g., set up and scoring), and many essential aspects of upper extremity function, such as quality of movement, speed of task performance, and self-report still may not be adequately assessed [14]. Recent research suggests the need for two components to assess upper extremity function in the clinic: perceived use and the quality and speed of task performance [14].To address these issues; a

new measure of upper extremity function in daily activities—the Hand and Arm Function Measure (HAFM)—was conceived for use in people who have neurological conditions.

Two theoretical frameworks were considered in HAFM development. The International Classification Functioning, Disability and Health (ICF) Core Set for Hand Conditions [15]. provided structure to the core components of upper extremity function to be assessed by the measure. The Evidence-Centered Design (ECD) framework provided structure to the process of development [16,17]. The ECD framework recommends stakeholder engagement as a critical first step in the process of developing a measure. Only a few upper extremity function measures have been developed in this systematic manner [18] and none have utilized state-of-the-art measurement approaches such as the ECD framework. Thus, the purpose of this research was to conduct a qualitative descriptive study to engage content experts and people who have neurological conditions in the systematic development of the HAFM.

MATERIALS AND METHODS

The content validity of the HAFM targeted the first two stages of the study that included focus groups with content experts in the field and with individuals with neurological condition. The face validity of the HAFM targeted the next two stages including item modifications with expert panel via an open-ended survey and cognitive interviews with individuals with neurological conditions. Approval from the University of Washington Human Subjects Division (#47121) was obtained prior to commencing the study.

The study was conducted in the Rehabilitation Medicine Department and stakeholders were recruited via newsletter announcements and flyers posted at University of Washington Medical Center clinics (Seattle, WA), Evergreen Health clinics (Kirkland, WA), research websites, clinics, newsletters of registries. Participants were contacted via telephone and selected using purposive sampling. Informed consent was obtained from all individual participants included in the study. There were no refusals or dropouts in the study.

The inclusion criteria for content experts were: 21 years of age or older, English language speaker, experience of five years or more working clinically with neurological conditions; the measurement expert had to have five years or more of measurement experience. The inclusion criteria for people with

SCIENTIFIC LITERATURE

neurological conditions were: 21 years of age or older; English language speaker; in overall good health; mild, moderate, or severe upper extremity difficulty that interfered with daily activities; diagnosed by a physician at least six months prior to the study; and able to arrange travel to the University of Washington. The exclusion criteria were: concurrent neurological condition (s) that would affect upper extremity function, currently receiving Physical Therapy (PT) or Occupational Therapy (OT), limb loss or any injury to the upper extremity within the last six months, uncorrected vision or hearing problems, and memory impairment as defined by having more than two errors on the Short Portable Mental Status Questionnaire (SPMSQ) [19]. The SPMSQ scores are reported to be 86.2% sensitive and 99.0% specific [20] in detecting mild cognitive deficit in community dwelling elderly individuals.

The participants came to the University of Washington for focus groups and cognitive interviews where only researchers and participants were present during the study. A semi-structured interview guide was used for the focus groups and cognitive interviews. These sessions held from Jul 2014 to Aug 2015were audio-recorded with permission and transcribed. The open-ended survey of experts was sent by email. The two-hour focus group with experts (Jul 2014) helped shape the construct to be studied, report common practices in assessment of upper extremity function, identify professional descriptors currently used in therapy and measurement, and gather ideas about potential items through group validation. An open-ended survey was emailed to experts (Aug 2015) for their comments on each of the preliminary set of items. Disease-specific focus groups of people with neurological conditions sought to explore their prior experiences with testing of upper extremity function and to understand what aspects of upper extremity impairments affect function and participation in daily activities (Jul 2014). Cognitive interviews (Jul 2015) focused on item and task clarity, response choices, and context perceived in response to the question. At the beginning of the two-hour cognitive interviews, along with demographic data, the following standardized measures were administered: Manual Ability Measure-20 (MAM-20) [14], Patient-Reported Outcomes Measurement Information System(PROMIS) Fatigue short form 7a version 1.0, PROMIS Depression short form 8b

version 1.0, and PROMIS Pain Interference short form 6b version 1.0 [21]. In the cognitive interviews, participants completed 10-15 performance tasks, answered 30-35 self-reported questionnaire items, and discussed their perceptions about the tasks and items. The materials needed for administering the performance-based section of the HAFM cost less than $50 (USD) and included: measuring tape, paper tape, teaspoon, mug, can, pencil, pennies, lock, key, dried peas, jar, bottle, bowl, socks, beans, paper clip, nut, bolt, and paper pad.

A phenomenology approach was taken to derive themes through interpreting narrative data. Data immersion, coding, category creation, and thematic analysis were used to find patterns of meaning across data by two researchers (NG and DK) independently [22]. Dedoose version 6.2 (www.dedoose.com) was used to organize and manage the data. The researchers' notes made during and after focus groups and cognitive interviews were not coded but were reviewed as part of the analysis. The cognitive interview codes were grouped by the items. There was overall good agreement between the two coders and disagreements were resolved through discussion. There was consensus on final coding scheme as being representative of the data. The items generated by the expert panel were added to the item bank and combined with those suggested by participants with a neurological condition during focus groups. For the purpose of item development, a strategy was developed for triangulation of results from content experts and participants with neurological condition. The strategy included counting the frequency with which the items present together or co-occur among different groups. The frequency of occurrence of each item among participants with a neurological condition and among experts was combined with the frequency of co-occurrence of each item for all groups. Subsequently, the items were ranked by their total occurrence score among all groups. The top one-third of the items that had total occurrence score of six or higher were selected for further development.

RESULTS AND DISCUSSION

The expert panel included two occupational therapists, one physical therapist, and one measurement expert. Characteristics of the expert panel are provided in (Table 1). Seven key themes emerged in this study and include (in no

particular order): 1) No preference for the terms "hand function" or "hand use" to represent the construct; 2) Real-life tasks are valued in measurement and simulated tasks add sensitivity to the testing process; 3) People compensate for challenging tasks in numerous ways, which should be considered in the measure; 4) Disease-specific symptoms are important; 5) 56 items were suggested by content experts based on their experience with people with stroke, TBI, PD, and MS; 6) Fine motor skills (e.g., in-hand manipulation, coordination, and speed) and gross motor skills (e.g., reaching and lifting)are important aspects of upper extremity function; 7) Quick administration, speed and quality of movement, and inclusion of a variety of tasks are important for a measure. The high-frequency items were those that occurred repeatedly, such as using an electronic tablet (e.g., iPad), self-care, eating, using a spoon or fork, dressing, hygiene, using a pen, and handling coins.

Table 1: Characteristics of participants in the expert panel.					
Expert	Degree and Profession	Clinical Practice	Experience	Common Neurological Conditions Encountered	Other related Information
E1	MS, Occupational Therapy	Active in pediatrics, prior work with other populations	40 years clinical	Brain tumors, brachial plexus injury, cerebral palsy, epilepsy, peripheral nerve injury, polyneuropathy, stroke, and traumatic brain injury	Teaching experience
E2	PhD, Measurement	N/A	30 years research	N/A	Teaching experience; published research related to measurement in rehabilitation
E3	BS, Occupational Therapy	Active in adult care: neuro-rehabilitation, out-patient	30 years clinical	Brachial plexus injury, dementia, essential tremor, Guillian Barre syndrome, multiple sclerosis, Parkinson disease, stroke, and traumatic brain injury	N/A
E4	PhD, Physical Therapy	Active in skilled nursing facility	11 years clinical and 7 years research	Brain tumors, dementia, multiple sclerosis, Parkinson disease, stroke, and traumatic brain injury	Teaching experience;published research related to measurement in rehabilitation

Of the 20 participants with neurological conditions, six (participant number 19, 20, 22, 23, 24, 25) participated in both a focus group and a cognitive interview. Characteristics of participants with neurological condition are provided in (Table 2). More participants with severe stroke were added to get a better representation of all levels of function. Eight diagnostic-specific focus groups with four or five participants each, and subsequent one-on-one cognitive interviews were carried out.

Table 2: Characteristics of participants with neurological condition.										
Participant	Diagnosis	Age	Sex	Education	Affected hand	Current Dominance	MAM[b]	Fatigue[c]	Depression[c]	Pain[c]
1	Stroke	60	F	Bachelors	R	R	90	45.8	55.7	55
2	Stroke	70	F	Bachelors	R	R	52.4	52.2	55.7	53.8
3	Stroke	65	M	PhD	L	L	66.4	52.2	54.5	52.5
4	PD	63	F	Bachelors	R	R	67.6	47.6	62.1	52.5
5	PD	61	M	Bachelors	R	R	63.3	53.7	53.3	41
6	PD	62	M	High School	R	R	59.6	62	57.9	41
7	PD	54	M	Not reported	L	L	53.9	50.8	56.8	56.1
8	Stroke	52	F	High School	L	R[a]	55.5	50.8	62.1	41
9	Stroke	46	M	Bachelors	L	L	49.3	43.9	64.1	41
10	Stroke	60	F	Masters	Both	Both	59.6	55.1	70.3	56.1
11	MS	50	M	Bachelors	L	R	68.9	53.7	53.3	41
12	MS	52	M	Bachelors	Both	R	66.4	47.6	65.1	52.5
13	MS	66	F	Bachelors	Both	R	52	66.3	60	61.8
14	PD	65	F	Masters	Both	R	54.7	60.6	61.1	60.9
15	PD	72	M	High School	Don't know	R	100	49.2	54.5	41

SCIENTIFIC LITERATURE

16	MS	51	M	Not reported	Both	R	49.3	67.8	68.2	48.5
17	MS	50	F	Bachelors	L	R	60.5	41.9	53.3	41
18	TBI	29	M	Less than High School	Both	R	47.8	55.1	70.3	59.1
19	MS	51	M	Bachelors	R	R	68.9	53.7	53.3	41
20	MS	51	F	Bachelors	L	R	60.5	41.9	53.3	41
21	Stroke	32	F	Bachelors	R	L[a]	65.3	43.9	53.3	52.5
22	PD	63	M	High School	R	B	59.6	62	57.9	41
23	Stroke	47	M	Bachelors	L	R	49.3	43.9	64.1	41
24	PD	62	M	Bachelors	L	R	63.3	53.7	53.3	41
25	TBI	30	M	Less than High School	Both	R	47.8	55.1	70.3	59.1
26	TBI	35	M	Masters	R	L[a]	63.3	50.8	64.1	55

Note: [a]Changed dominance due to hand problems. [b]Higher scores indicate more of the trait and thus better hand function. [c]Higher scores on the Patient-Reported Outcomes Measurement Information System (PROMIS) measure indicate more of the trait, thus higher scores indicate more fatigue, more depression and more pain. MAM: Manual Ability Measure-20; Fatigue: PROMIS Fatigue short form 7a version 1.0; Depression: PROMIS Depression short form 8b version 1.0; Pain: PROMIS Pain Interference short form 6b version 1.0.

Focus group themes

Participants in the focus groups discussed the importance of hand function and reported a lack of awareness of these problems in the medical community. Five main themes informed development of the HAFM and are reported here (in no particular order): 1) Each diagnostic condition presented with a specific set of characteristics related to upper extremity function; 2) Participants valued the compensations and adaptations they used; 3) Current and prior dominance of hand was important; 4) Special considerations for upper extremity issues such as difficulty with bilateral tasks, day-to-day changes in hand function, and others were reported. 5) Participants generated items in the categories of carrying, communication, instrumental activities of daily living, movements, object handling, reach, recreation, self-care, transportation, and work. (Supplementary Tables S1 and S2) provides themes, the corresponding quotes and an example of how they impacted the measure. Items from all focus groups were pooled. The top third of the high-frequency pooled items were selected as the final item set. In keeping the ICF Core Set for Hand Conditions [15] as the theoretical framework for the measure, one item, "lifting a can," was included for theoretical consistency, even though it was not a high-frequency item. The 59 retained items were then sorted into self-reported and performance-based categories. The item development process is presented in (Figure 1).
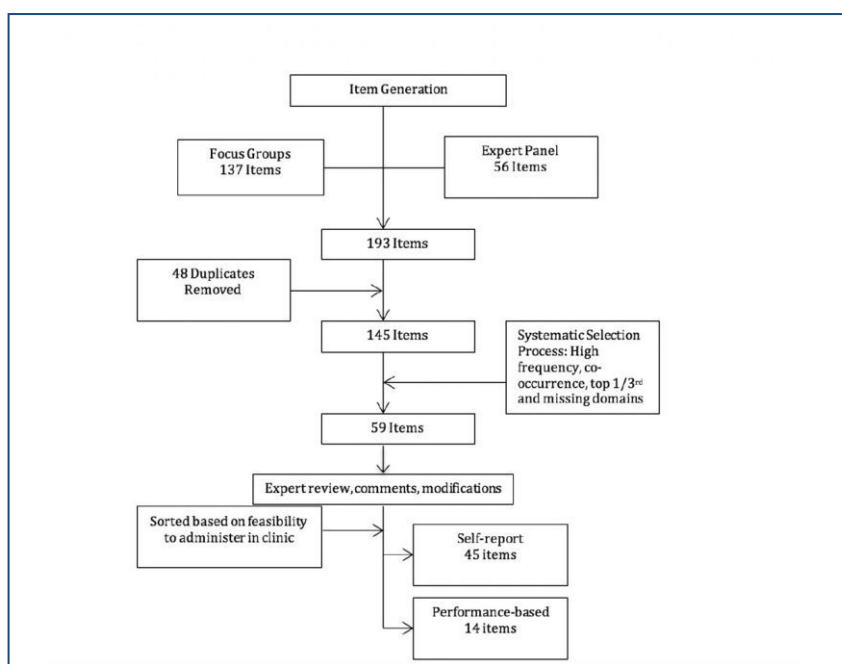

Figure 1: Item Development Process: Schematic Flow Diagram from Item Bank Development to Preliminary selection of Items.

Stakeholder Engagement for Development of a New Measure of Hand and Arm Function in People with Neurological Conditions. Physical Medicine & Rehabilitation Journal. 2019; 2(1):119.

SCIENTIFIC LITERATURE

Supplementary Table S1: Themes and Quotes of the Expert Panel Participants and their Impact on the Measure.		
Themes	**Sample quotes**	**Example of theme's impact on the measure**
Experts did not express a preference for "hand function" or "hand use" as the terms used to represent the construct.	"I pretty much use them synonymously, use and function. Just because I think I look at the framework that the ICFprovides and I just put function or classify function in their activities domain." (Participant E4)	The term "hand function" was retained for consistency.
The value of real-life tasks to be measured was noted by content experts; however, sensitivity of the simulated tasks was needed for a new measure.	"I think that it's always nice to see them do real-life things because it's more automatic. So, that's kind of how I see that as more functional and the simulated is where they may not have ever done that before or it's not as automatic to them." (Participant E1).	Real-life tasks and simulated tasks were included in the measure and terminology matched.
People compensate for challenging tasks in numerous ways and this should be considered in the measure.	"Or you feel that in the long term it may cause some other deficit like shoulder pain or something like that which you need to prevent at the right time." (Participant E1)	Compensations were acknowledged in patient information and scoring sections.
Disease-specific symptoms were important for these individuals.	"So external cues might be specific to Parkinson's, even on and off medications, it's a huge difference. It is necessary to know how long it has been since their last dose of medication." (Participant E4)	Symptoms specific to the condition were added as a symptom checklist before daily activities.
Fifty-six items were suggested by content experts from experience with patients with neurological conditions	Keys, set of keys, a zipper of some sort, buttons, or so these are irrespective of gender, irrespective of diagnoses, these could be some of the items." (Participant E3)	Items were added to the item bank.
Hand function aspects of importance were fine motor skills (e.g., in-hand manipulation, coordination, and speed), gross motor skills (e.g., reaching and lifting) and theoretical frameworks (e.g., motor control)	"Paper and pencil, puzzle, spoon and fork, something that most people have a wallet full of credit cards, money and some change, I think that would give a lot of fine-motor information and you don't necessarily have to carry it, you can use the patient's regular wallet or whatever, that seems to be a good one. Keys, set of keys, a zipper of some sort, buttons, or so these are irrespective of gender, irrespective of diagnoses, these could be some of the items." (Participant E3)	Items related to these aspects were retained in the measure.
Testing related aspects of importance were quick administration, speed of movement, quality of movement, and inclusion of a variety of tasks	"Okay, so from a measurement perspective you would want to have a variety of different tasks that we will do really tap into the different functions." (Participant E2)	Administration aspects for the measure considered these concepts.

Supplementary Table S2: Themes and Quotes from the Focus Group of Participants with Neurological Condition and their Impact on the Measure.		
Themes	**Sample quotes**	**Example of theme's impact on the measure**
Each diagnostic condition presents with specific set of characteristics	The things (about differences between people with PD previously said) that Participant 7 said so eloquently, it's different for everybody. And your progression is different for everybody. (Participant 5, PD)\nThat's why they call it a snowflake disease. (Participant 7, PD)	Disease-specific symptom checklist added to measure.
People valued the compensations and adaptations for lack of hand function in daily activities	Ya cause eating a sandwich to me was like that's difficult. So, I cut it in half and then what I usually do is I usually keep it in my hand and I don't put it down anymore. Because now it's in my hand it's like stay there until its eaten. So, it is different because you can talk about different foods because I have switched to eating foods that I know I can use one utensil. (Participant 20, MS)	Compensations were acknowledged in patient information and scoring sections.
Dominant hand function was important for daily activities,	I could use both hands but this hand is very weak so if I need to get things done fast I just use my dominant hand. Which is not helping my non-dominant hand. (Participant 10, Stroke)	Patient information section included report on prior and current dominance and the more and less affected side.
Special considerations for hand and arm function	You can't clap. (Demonstrates clapping) (Participant 2, Stroke).\nI have to really look at it (the paper cup). Keep my eye on it. But if I turned away, I wouldn't realize that I have just crushed it and spilled it. But if I really focus I could do it (keep it) right there and put it there. But I wouldn't get up and walk with it or anything. (Participant 16, MS)\nSo I mean good days and bad days, we all have them regardless of what kind. (Participant 15, PD)\nDon't test, don't just test without any offering. (Participant 11, MS)	Special considerations such as bilateral upper extremity function were added to the measure. Another consideration included manual to state explicitly that patients be informed of their scores and interpretation of the scores.

Note: PD=Parkinson Disease; MS= Multiple Sclerosis; TBI=Traumatic Brain Injury.

Cognitive interview themes

Eight cognitive interviews were conducted. The final set of 59 items used as part of cognitive interviews had 11 symptom-related questions, 34 activities of daily living questions, and 14 performance-based tasks. The detailed list of items is provided in Table 3 along with the ICF and hand function domains they represent within the construct of hand and arm function in daily activities. Each item was administered to five to seven participants, ensuring all items were trialed by at least one individual in each of the four diagnostic groups. The probing questions were presented after administering four to five items. The items were split into version A and version B with

SCIENTIFIC LITERATURE

overlapping items. Three participants were administered version-B first and five participants were administered version-A first. The reading ease was considered, e.g., Flesch-Kincaid Reading ease in Microsoft Word was 74.4% at a 5.5 grade level for the version A of self-reported section of the measure.

The feedback from the interviews resulted in substantial changes to 39 out of 59 items for the tasks, wording, and overall clarity of the item. There were no missing items as all items were discussed thoroughly with the participants.

ICF domains	Dimensions	Hand Function	Self-report items of Hand and Arm Function Measure	Performance-based items of Hand and Arm Function Measure
Activity and Participation	Gross movements in daily activities	Transport/ reach	Toilet hygiene	Hand to lower back
			Washing hair	
			Getting dressed Putting on shoes	Grabbing jar at shoulder height
		Non –prehensile skilled movements (pushing, carrying)	Clap hands	Bean can task – grasping and carrying
			Shaking hands when greeting	
			Holding an open book	
			Lifting grocery bag	
	Fine movements in daily activities	Prehension – Grip & pinch	Lifting gallon of milk	Coffee mug
			Lift cup with liquid in it	
			Open door lock with key	Open lock with key
			Open childproof bill bottle	Open spice bottle
			Open & close jar	Open jar
			Feeding yourself	Spoon use
			Cutting finger nails	Address writing
			Items in & out of wallet	Pennies in bowl – pick & place
			Clothing fasteners	Peas in bottle
			Open can	
			Brushing teeth	
			Putting on watch or jewelry	
			Cutting vegetables	
			Opening milk carton	
		In-hand manipulation	Handling credit cards or money	Pennies in bowl – pick& hold
		Non –prehensile skilled movements (pointing, turning)	Open & close Ziploc bag	Open lock with key Open spice bottle
			Turning door knob	
			Press buttons on phone or remote control	
			Using a keyboard	
			Computer mouse	
		Active haptic mode	Using a touch screen on a phone, tablet, computer, laptop	
		Passive tactile sensing	Sensation loss	
			Hypersensitivity	
Body Function/ Structure	Symptoms interfering with hand function in daily activities	Low functioning: Non-movement related aspects	Pain	
			Spasticity	
			Tremor	
			Slow movements	
			Deformity	
			Stiffness	
			Weakness	
			Coordination	
			Sleep	
			Fatigue	
Participation	Occupational roles	High functioning: High demand for productive function	Work	
			Childcare	
			School	
			Leisure	

Table 3: Hand and Arm Function Measure: Preliminary item set and the domains represented to align with the construct.

Note: ICF=International Classification of Functioning, Disability and Health.

None of the participants reported any discomfort with the number of items, time required, and administration procedures. Six themes emerged through cognitive interviews that

contributed to further refinement of the HAFM (in no particular order): 1) Relevance and clarity of items could be improved. For example, the item related to "wallet or purse" was to be

07

Stakeholder Engagement for Development of a New Measure of Hand and Arm Function in People with Neurological Conditions. Physical Medicine & Rehabilitation Journal. 2019; 2(1):119.

SCIENTIFIC LITERATURE

changed to "wallet or purse or pocket" to improve relevance. 2) Response options needed to be revised to provide more detail. For example, a comment section needed to be added. 3) Scoring needed to be clarified to improve ease of use. For example, the assessment form needed to be adapted to record scores on an initial, follow-up, and discharge.4) The set-up for the performance-based items needed to be improved. For example, one of the participants had a skin condition, which prompted the addition of explicit wording in the manual about the requirement to clean the test items after each use. 5) The demographics questions needed to be clarified. For example, the wording "hand preference" was favored over the wording "hand dominance"6) A clear explanation of symptoms needed to be added. For example, in the self-reported section of HAFM, meanings of the words spasticity, hypersensitivity, and deformity were not clear. 7) More space for certain information, such as medications, needed to be provided. (Supplementary Table S3) provides themes and quotes from the cognitive interviews.

Supplementary Table S3: Themes and Quotes from Cognitive Interviews of Participants with Neurological Condition and their Impact on the Measure.		
Themes	**Sample quotes**	**Example of theme's impact on the measure**
Relevance and clarity of items was improved with participant feedback.	Simply moving it (the can) I don't know if you are looking for my ability to accurately place it.(Participant 24, PD)	Wording of the instructions was updated for more clarity
Response options were revised to provide more detail.	I don't know what the numbers mean (Participant 30, TBI)	The patient-reported response section was revised.
Scoring was clarified to make it easy to use.	So, my left hand. So, the numbers are for you guys to add it up? (Participant 30, TBI)	The location of the scores on the document was revised.
The set-up for the performance-based items was improved.	But it's always good to inquire what adaptations have you found that help you with difficult tasks. (Participant 28, PD)	The patients were allowed to use the splints they typically use if needed.
The demographics questions were clarified.	What's your dominance right now? (Researcher NG) What do you mean? (Participant 29, TBI) Are you right-handed or left-handed? (Researcher NG)	The term "dominance" was updated to "preference" for ease of reading.
A clear explanation of the symptoms was added.	Hypersensitivity I was kind of unsure. Sometimes when it's painful, when its hypersensitivity I don't know if it's hypersensitive so I don't know. So, I looked at it as increase tactile feelings, which I don't have. (Participant 23, MS)	The terms in the symptom checklist would be defined in the next version for clarification.
More space for certain information	Did you take any medications today? (Researcher NG) Oh god yes.(Participant 27, Stroke) The reason we ask is what medications might affect functions.(Researcher NG) Epilepsy meds. It will take me a while to go through the 15 pills.(Participant 27, Stroke)	More space for writing medications on the patient information section.

Note: PD=Parkinson Disease; MS= Multiple Sclerosis; TBI=Traumatic Brain Injury.

This research contributes to the literature by demonstrating the importance of the involving stakeholders in the development of a measure of upper extremity function. The involvement of stakeholders is consistent with the guidance for researchers provided by the Patient-Centered Outcomes Research Institute [23] and by large regulatory bodies like the Federal Drug Administration [24].The contribution of stakeholders in this process also aligns with the 8-stage framework presented by Velozo and colleagues [3] who advocated the use of mixed methods in preliminary research. The use of focus groups and cognitive interviews with people who have neurological conditions strengthened the process of instrument development for the HAFM.

The scoring for HAFM continues to be developed in each iteration of this measure. The current version had self-report items scored at 3=Not at all, 2=A little, 1=Quite a lot for interference of hand function with daily tasks. The performance-based items were scored at 5=Independent, 4=Completed with compensations, 3=Completed with difficulty, 2=Partially completed, 1=Attempted, 0=Not able to do. The total score for self-report and performance-based sections was calculated separately. The final score was the percentage calculated by dividing raw score with the total points possible.

Not uncommon to qualitative studies, this study is limited by possible influences of personal researcher biases. A detailed log of researcher's notes from each focus group was kept with the intention of recording any biases, and involvement of two researchers during analysis attempted to neutralize this effect. Also, the varied backgrounds and experiences of the investigators helped reduce some of the inherent biases related to perceptions of daily activity limitations. Another limitation is

sample size; the group of 20 participants came from a limited geographical location, which may not represent all the views of people who have neurological conditions. Further, the semi-structured questions asked during cognitive interviews were developed by the researchers; input of the expert panel in designing these questions would have added relevance and strength to the methodology.

The upper extremity function level of participants in this study was 47.8 to 100 on the MAM-20, indicating mild to moderate impairment. Thus, individuals who were low functioning were not adequately represented in this group so it is possible that a complete assessment of function was not done; ongoing research will need to focus on recruiting participants with severe upper extremity impairments and at varied clinical settings to address this issue. Individuals with TBI were not well represented in this study, with a total of two participants; further efforts need to be made in order to recruit more participants with TBI in future studies. Also, the regional nature of sampling may place limits on the generalizability of the findings.

CONCLUSION

This research represents the first step in the development of a new measure of upper extremity function, the HAFM, for individuals with neurological impairments. Focus groups, cognitive interviews and the survey facilitated the grounding of the HAFM in the views of people with neurological conditions and content experts in rehabilitation and measurement. This stakeholder engagement helped establish the face and content validity for a new comprehensive outcome measure in rehabilitation. Further studies are needed to engage other types of stakeholders such as caregivers and build the evidence for reliability and validity of the HAFM.

REFERENCES

1. Kandel ER, Schwartz JH, Jessell TM, Siegelbaum SA, Hudspeth AJ. (Eds.). (2012). Principles of Neural Science, Fifth Edition (Principles of Neural Science (5th ed.). McGraw-Hill Professional, New York City, NY.

2. Kraft GH, Amtmann D, Bennett SE, Finlayson M, Sutliff MH, et al. (2014). Assessment of upper extremity function in multiple sclerosis: review and opinion. Postgraduate Medicine. 126: 102-108.

3. Velozo CA, Seel RT, Magasi S, Heinemann AW, Romero S. (2012). Improving measurement methods in rehabilitation: core concepts and recommendations for scale development. Archives of Physical Medicine and Rehabilitation. 93: S154-163.

4. Carmeli E, Patish H, Coleman R. (2003). The aging hand. The Journals of Gerontology. Series A, Biological Sciences and Medical Sciences. 58: 146-152.

5. Lemmens RJ, Timmermans AA, Janssen-Potten YJ, Smeets RJ, Seelen HA. (2012). Valid and reliable instruments for arm-hand assessment at ICF activity level in persons with hemiplegia: a systematic review. BMC Neurology. 12: 21.

6. Lehman LA, Woodbury M, Shechtman O, Wang YC, Pomeranz J, et al. (2011). Development of an item bank for a computerised adaptive test of upper-extremity function. Disability and Rehabilitation. 33: 2092-2104.

7. Esmail L, Moore E, Rein A. (2015). Evaluating patient and stakeholder engagement in research: moving from theory to practice. Journal of Comparative Effectiveness Research. 4: 133-145.

8. Oxford Grice K, Vogel KA, Le V, Mitchell A, Muniz S, et al. (2003). Adult norms for a commercially available Nine Hole Peg Test for finger dexterity. The American Journal of Occupational Therapy. 57: 570-573.

9. Asher I. (2007). Occupational Therapy Assessment Tools: An Annotated Index, 3rd Edition (3rd ed.). AOTA Press, Bethesda, MD.

10. Morris D, Uswatte G, Crago J, Cook EW, Taub E. (2001). The reliability of the wolf motor function test for assessing upper extremity function after stroke. Archives of physical medicine and rehabilitation. 82: 750-755.

Stakeholder Engagement for Development of a New Measure of Hand and Arm Function in People with Neurological Conditions. Physical Medicine & Rehabilitation Journal. 2019; 2(1):119.

SCIENTIFIC LITERATURE

11. Velstra IM, Ballert CS, Cieza A. (2011). A systematic literature review of outcome measures for upper extremity function using the international classification of functioning, disability, and health as reference. PM & R: The Journal of Injury, Function, and Rehabilitation. 3: 846-860.

12. Uswatte G, Taub E, Morris D, Vignolo M, McCulloch K. (2005). Reliability and validity of the upper-extremity Motor Activity Log-14 for measuring real-world arm use. Stroke; a Journal of Cerebral Circulation. 36: 2493-2496.

13. Ashford S, Slade M, Malaprade F, Turner-Stokes L. (2008). Evaluation of functional outcome measures for the hemiparetic upper limb: a systematic review. Journal of Rehabilitation Medicine: Official Journal of the UEMS European Board of Physical and Rehabilitation Medicine. 40: 787-795.

14. Rallon CR, Chen CC. (2008). Relationship between performance-based and self-reported assessment of hand function. The American Journal of Occupational Therapy. 62: 574-579.

15. Kus S, Oberhauser C, Cieza A. (2012). Validation of the brief International Classification of Functioning, Disability, and Health (ICF) core set for hand conditions. Journal of Hand Therapy. 25: 274-286.

16. Mislevy RJ, Almond RG, Lukas JF. (2003). A Brief Introduction to Evidence-centered Design. Educational Testing Service, Princeton, NJ.

17. Mislevy RJ. (2011). Evidence-centered design for simulation-based assessment. National Center for Research on Evaluation, Standards and Student Testing, Center for the study of evaluation, Graduate School of Education and Information Studies, UCLA, Los Angeles.

18. O'Dell MW, Kim G, Rivera L, Fieo R, Christos P, et al. (2013). A psychometric evaluation of the Arm Motor Ability Test. Journal of Rehabilitation Medicine. 45: 519-527.

19. Pfeiffer E. (1975). A short portable mental status questionnaire for the assessment of organic brain deficit in elderly patients. Journal of the American Geriatrics Society. 23: 433-441.

20. Erkinjuntti T, Sulkava R, Wikström J, Autio L. (1987). Short Portable Mental Status Questionnaire as a screening test for dementia and delirium among the elderly. Journal of the American Geriatrics Society. 35: 412-416.

21. Broderick JE, Schneider S, Junghaenel DU, Schwartz JE, Stone AA. (2013). Validity and reliability of patient-reported outcomes measurement information system instruments in osteoarthritis. Arthritis Care & Research. 65: 1625-1633.

22. Schmidt C. (2004). The analysis of semi-structured interivews. In U. Flick, E. Kardoff, I. Steinke, & B. Jenner (Eds.), A companion to qualitative research. SAGE Publications Ltd., London.

23. Patient-Centered Outcomes Research Institute. (2012). Preliminary draft methodology report.

24. U.S. Department of Health and Human Services FDA Center for Drug Evaluation and Research, U.S. Department of Health and Human Services FDA Center for Biologics Evaluation and Research, U.S. Department of Health and Human Services FDA Center for Devices and Radiological Health. (2006). Guidance for industry: patient-reported outcome measures: use in medical product development to support labeling claims: draft guidance. Health and Quality of Life Outcomes. 4:79.

Stakeholder Engagement for Development of a New Measure of Hand and Arm Function in People with Neurological Conditions. Physical Medicine & Rehabilitation Journal. 2019; 2(1):119.

SCIENTIFIC
LITERATURE

Supplementary Table S1: Themes and Quotes of the Expert Panel Participants and their Impact on the Measure.

Themes	Sample quotes	Example of theme's impact on the measure
Experts did not express a preference for "hand function" or "hand use" as the terms used to represent the construct.	"I pretty much use them synonymously, use and function. Just because I think I look at the framework that the ICFprovides and I just put function or classify function in their activities domain." (Participant E4)	The term "hand function" was retained for consistency.
The value of real-life tasks to be measured was noted by content experts; however, sensitivity of the simulated tasks was needed for a new measure.	"I think that it's always nice to see them do real-life things because it's more automatic. So, that's kind of how I see that as more functional and the simulated is where they may not have ever done that before or it's not as automatic to them." (Participant E1).	Real-life tasks and simulated tasks were included in the measure and terminology matched.
People compensate for challenging tasks in numerous ways and this should be considered in the measure.	"Or you feel that in the long term it may cause some other deficit like shoulder pain or something like that which you need to prevent at the right time." (Participant E1)	Compensations were acknowledged in patient information and scoring sections.
Disease-specific symptoms were important for these individuals.	"So external cues might be specific to Parkinson's, even on and off medications, it's a huge difference. It is necessary to know how long it has been since their last dose of medication." (Participant E4)	Symptoms specific to the condition were added as a symptom checklist before daily activities.
Fifty-six items were suggested by content experts from experience with patients with neurological conditions	Keys, set of keys, a zipper of some sort, buttons, or so these are irrespective of gender, irrespective of diagnoses, these could be some of the items." (Participant E3)	Items were added to the item bank.
Hand function aspects of importance were fine motor skills (e.g., in-hand manipulation, coordination, and speed), gross motor skills (e.g., reaching and lifting) and theoretical frameworks (e.g., motor control)	"Paper and pencil, puzzle, spoon and fork, something that most people have a wallet full of credit cards, money and some change, I think that would give a lot of fine-motor information and you don't necessarily have to carry it, you can use the patient's regular wallet or whatever, that seems to be a good one. Keys, set of keys, a zipper of some sort, buttons, or so these are irrespective of gender, irrespective of diagnoses, these could be some of the items." (Participant E3)	Items related to these aspects were retained in the measure.
Testing related aspects of importance were quick administration, speed of movement, quality of movement, and inclusion of a variety of tasks	"Okay, so from a measurement perspective you would want to have a variety of different tasks that we will do really tap into the different functions." (Participant E2)	Administration aspects for the measure considered these concepts.

Supplementary Table S2: Themes and Quotes from the Focus Group of Participants with Neurological Condition and their Impact on the Measure.

Themes	Sample quotes	Example of theme's impact on the measure
Each diagnostic condition presents with specific set of characteristics	The things (about differences between people with PD previously said) that Participant 7 said so eloquently, it's different for everybody. And your progression is different for everybody. (Participant 5, PD) That's why they call it a snowflake disease. (Participant 7, PD)	Disease-specific symptom checklist added to measure.
People valued the compensations and adaptations for lack of hand function in daily activities	Ya cause eating a sandwich to me was like that's difficult. So, I cut it in half and then what I usually do is I usually keep it in my hand and I don't put it down anymore. Because now it's in my hand its like stay there until its eaten. So, it is different because you can talk about different foods because I have switched to eating foods that I know I can use one utensil. (Participant 20, MS)	Compensations were acknowledged in patient information and scoring sections.
Dominant hand function was important for daily activities,	I could use both hands but this hand is very weak so if I need to get things done fast I just use my dominant hand. Which is not helping my non-dominant hand. (Participant 10, Stroke)	Patient information section included report on prior and current dominance and the more and less affected side.
Special considerations for hand and arm function	You can't clap. (Demonstrates clapping) (Participant 2, Stroke). I have to really look at it (the paper cup). Keep my eye on it. But if I turned away, I wouldn't realize that I have just crushed it and spilled it. But if I really focus I could do it (keep it) right there and put it there. But I wouldn't get up and walk with it or anything. (Participant 16, MS) So I mean good days and bad days, we all have them regardless of what kind. (Participant 15, PD) Don't test, don't just test without any offering. (Participant 11, MS)	Special considerations such as bilateral upper extremity function were added to the measure. Another consideration included manual to state explicitly that patients be informed of their scores and interpretation of the scores.

Note: PD: Parkinson Disease; MS: Multiple Sclerosis; TBI: Traumatic Brain Injury

Supplementary Table S3: Themes and Quotes from Cognitive Interviews of Participants with Neurological Condition and their Impact on the Measure.

Themes	Sample quotes	Example of theme's impact on the measure
Relevance and clarity of items was improved with participant feedback.	Simply moving it (the can) I don't know if you are looking for my ability to accurately place it.(Participant 24, PD)	Wording of the instructions was updated for more clarity
Response options were revised to provide more detail.	I don't know what the numbers mean (Participant 30, TBI)	The patient-reported response section was revised.
Scoring was clarified to make it easy to use.	So, my left hand. So, the numbers are for you guys to add it up? (Participant 30, TBI)	The location of the scores on the document was revised.
The set-up for the performance-based items was improved.	But it's always good to inquire what adaptations have you found that help you with difficult tasks. (Participant 28, PD)	The patients were allowed to use the splints they typically use if needed.
The demographics questions were clarified.	What's your dominance right now? (Researcher NG) What do you mean? (Participant 29, TBI) Are you right-handed or left-handed? (Researcher NG)	The term "dominance" was updated to "preference" for ease of reading.
A clear explanation of the symptoms was added.	Hypersensitivity I was kind of unsure. Sometimes when it's painful, when its hypersensitivity I don't know if it's hypersensitive so I don't know. So, I looked at it as increase tactile feelings, which I don't have. (Participant 23, MS)	The terms in the symptom checklist would be defined in the next version for clarification.
More space for certain information	Did you take any medications today? (Researcher NG) Oh god yes.(Participant 27, Stroke) The reason we ask is what medications might affect functions.(Researcher NG) Epilepsy meds. It will take me a while to go through the 15 pills.(Participant 27, Stroke)	More space for writing medications on the patient information section.

Note: PD: Parkinson Disease; MS: Multiple Sclerosis; TBI: Traumatic Brain Injury