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Validity of the 12-Item World Health Organization Disability Assessment Schedule 2.0 (WHODAS 2.0) in individuals with Huntington disease (HD)

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Abstract

Purpose—The reliability and construct validity of the 12-item World Health Organization Disability Assessment Schedule 2.0 (WHODAS 2.0) was examined in individuals with Huntington disease (HD).

Methods—We examined factor structure (confirmatory factor analysis), internal consistency reliability (Cronbach's alpha), floor and ceiling effects, convergent validity (Pearson correlations), and known-groups validity (multivariate analysis).

Results—Results of a confirmatory factor analysis replicated the six factor latent model that reflects the six separate scales within the WHODAS 2.0 (understanding and communicating; getting around; self-care; getting along with others, life activities; participation). Cronbach's alpha for the scale was 0.94, suggesting good internal consistency reliability. The WHODAS demonstrated a ceiling effect for 19.5% of participants; there were no floor effects. There was evidence for convergent validity; the WHODAS demonstrated moderate significant correlations with other general measures of health-related quality of life (HRQOL; i.e., RAND-12, EQ5D). Multivariate analyses indicated that late-stage HD participants indicated poorer HRQOL than both early-stage HD and prodromal HD participants for all HRQOL measures.

Conclusions—Findings provide support for both the reliability and validity of the WHODAS 2.0 in individuals with HD.

Keywords

Neuropsychology; Neuropsychological Assessment; World Health Organization Disability Assessment Schedule 2.0 (WHODAS 2.0); Confirmatory Factor Analysis; Psychometrics

The World Health Organization Disability Assessment Schedule 2.0 (WHODAS 2.0) is a generic measure designed to capture functioning and disability, important aspects of health-related quality of life (HRQOL), according to the Internal Classification of Functioning, Disability, and Health (ICF) framework [1; 2]. This measure has been recommended for determination of functional decline secondary to psychiatric illness by the Diagnostic and Statistical Manual of Mental Disorders, Fifth Edition (DSM-V) [1]. The WHODAS, which comes in 12-, 32- and 36-item versions [1; 2], examines activity limitations and restrictions for six different tasks: 1) understanding and communication; 2) self-care; 3) mobility (getting around); 4) interpersonal relationships (getting along with others); 5) work and household roles (life activities); and 6) community and civic roles (participation); it is available in 12 different languages (English, Spanish, French, German, Chinese, Danish, Dutch, Hungarian, Bangala, Hindi, Serbian and Tamil). While the 36-item version has been examined in several different populations [1; 3–22], the 12-item version has only been examined in a handful of studies [2; 23–25]. The purpose of this paper is to examine the psychometric properties of the 12-item WHODAS 2.0 in individuals with Huntington disease (HD), a devastating autosomal dominant neurodegenerative disease affecting approximately 1 out of 17,000 individuals across North America, Europe, and Australia [26]. HD is a triad disorder characterized by motor, cognitive and behavioral symptoms. Individuals with the CAG Huntingtin gene expansion for the disease typically develop symptoms leading to a clinically definite motor diagnosis around age 40, ~20 years prior to death [27].

To date, two studies have been conducted examining the WHODAS 2.0 in individuals with prodromal HD (i.e., positive CAG gene test but absence of unequivocal motor diagnosis) [20; 23] using the Predict-HD cohort [28; 29]. The first study [20] examined the 36-item version of the WHODAS 2.0 over a 3-year period. Findings supported internal consistency for both the self-report for prodromal participants, as well as the proxy-report by their companions (Cronbach's alphas were 0.92 for both) [20]. Validity of the WHODAS was suggested by self- and proxy-reports of worse functioning for individuals with high disease burden relative to controls and significant self- and proxy-reports of declines in functioning over time for medium- and high-burden prodromal participants [20]. There was also concordance between self-report and proxy report for participants with low and medium disease burden, but not high disease burden (participants with high disease burden self-reported less functional decline than did their companions' proxy reports) [20]. Finally, the WHODAS indicated larger declines than the gold standard HD measure of functional capacity (the Total Functional Capacity scale [TFC] [30]) in individuals with medium disease burden relative to controls [20] suggesting that the WHODAS may be particularly sensitive to change in earlier stages of prodromal HD, and may be useful in clinical trials aimed at these stages.

The second study compared the 12-item version to the 36-item version of the WHODAS in HD [23]. For the 12-item WHODAS 2.0, individuals with prodromal HD showed baseline group differences (those with medium and high burden reported significantly worse functioning than controls), whereas proxy ratings by companions showed both baseline (those caring for individuals with high disease burden reported worse functioning than controls) and longitudinal group differences (those caring for individuals with medium and high disease burden reported worse functioning than controls) [23]. Companions also proxy reported worse functional decline over time for the high burden participants on the 12-item WHODAS 2.0 [23]. Finally, the 12-item WHODAS detected longitudinal change better than the 36-item WHODAS in the medium progression group [23]. These results support the utility of the 12-item WHODAS 2.0 in prodromal HD, especially in those with medium and/or high disease burden.

Neither of these studies focused on the psychometric properties of the 12-item WHODAS, and neither included individuals with manifest HD [20; 23]. Thus, information on the psychometric properties of this measure across the full disease spectrum in HD is needed. While existing psychometric data for the 12-item WHODAS in the general population support its reliability (i.e., Cronbach's $\alpha = 0.86$), its utility across samples, cultures and points in time has been questioned due to poor item characteristic curves [2]. Furthermore, while one study found evidence to support the 6-factor structure in the 12-item WHODAS [25], another study conducted in older adults in seven different developing countries was only able to support a single factor structure [24]. Given the equivocal psychometrics findings of the 12-item WHODAS 2.0, more work is needed to examine its reliability and validity.

Thus, the purpose of this manuscript is to examine the reliability and construct validity of the 12-item World Health Organization Disability Assessment Schedule 2.0 (WHODAS 2.0) in individuals with both prodromal and manifest HD. Specifically, we attempted to replicate the six-factor structure of the WHODAS 2.0. We also examined internal consistency reliability, floor and ceiling effects, as well as convergent and known-groups validity.

Method

Individuals with prodromal and/or manifest HD were invited to participate in this study. Participants needed to be at least 18 years old, able to read and understand English, and have either a positive test for the CAG expansion for HD and/or a clinical diagnosis of HD. Participants were recruited from specialized treatment centers at the University of Michigan, the University of Iowa, the University of California-Los Angeles, Indiana University, Johns Hopkins University, Rutgers University, Struthers Parkinson's Center, and Washington University; in addition, the majority of the prodromal HD participants were recruited through the Predict-HD study, which includes a well characterized, prodromal HD sample [31].

Measures

WHODAS 2.0.[1]—The WHODAS 2.0 [1] assesses generic function-related HRQOL across six subdomains: understanding and communication, self-care, mobility, interpersonal

relations, work and household roles, and community and civic roles. The WHODAS 2.0 is comprised of 12 self-report items that are rated on a scale of 0 to 4, with summed total scores ranging from 0 (highest level of health) to 48 (low health). Administration time is less than 5 minutes. The WHODAS was completed on-line through assessmentcenter.net; participants had the option of completing this measure independently during an in-person study visit, independently at home, or with assistance from a study team member. Assistance could include physical assistance (i.e., for those with physical difficulties using a mouse) or administration using an interview style (i.e., for individuals who had difficulty reading due to oculomotor impairments).

RAND-12 Health Status Inventory (HSI)[32]—The RAND-12 HSI [32] is a 12-item self-report measure designed to assess general health; it has been developed to provide a shorter, yet valid, alternative to the RAND-36 [32]. Physical and mental health composite scores (PHC and MHC) are computed using scores of the 12 questions, ranging from 0 (low health) to 100 (highest level of health). Administration time is less than 5 minutes. Reliability and validity of the RAND-12 has been established in previous studies [33–36]. The Rand-12 was completed on-line through assessmentcenter.net; participants had the option of completing this measure independently during an in-person study visit, independently at home, or with assistance from a study team member (types of assistance described above).

EQ5D [37]—The EQ5D[37] is a standardized measure of health status developed by the EuroQol Group in order to provide a simple, generic, self-report measure of HRQOL. The EQ5D Health Scale scores range from 0 (low health) to 100 (highest level of health) while the EQ5D Index Value scores range from 0 (low health) to 1 (highest level of health). Administration time is less than 5 minutes. The EQ5D has established reliability and validity in both the general population [38–41] and several clinical populations [42–45]. The EQ5D was completed in-person using a paper/pencil format; participants were given the option of completing this independently, or with assistance from a study team member (types of assistance described above).

TFC[30]—The TFC [30] is a 5-item clinician-rated measure that provides an index of day-to-day functioning across the domains of occupation, finances, domestic chores, activities of daily living and care level. Scores range from 0 (low functioning) to 13 (highest level of functioning) with higher scores indicating better functioning. The TFC is one of the most frequently used measures in HD research [46]. In this study TFC scores were used to classify participants with an HD diagnosis as either early stage (sum scores of 7–13) or later-stage (sum scores of 0–6). The TFC was administered in person, by a study clinician.

Data Analysis

Confirmatory Factor Analysis—To determine whether the standard 6-factor structure of the WHODAS 2.0 fit this sample, a confirmatory factor analysis (CFA) was implemented using Mplus (Version 7.11). The estimated correlation among factors and model fit indices were checked to assess model fit. In order to have acceptable fit, a model should have a

comparative fit index (CFI) greater than 0.90, Tucker-Lewis index (TLI) greater than 0.90, and Root Mean Square Error of Approximation (RMSEA) less than 0.10 [47–50].

Internal Consistency Reliability—Cronbach's alphas were calculated and a critical cut-off of 0.70 was considered minimal acceptable reliability.

Floor and Ceiling Effects—Floor and ceiling effects were calculated by identifying the proportion of participants that either had the lowest or the highest possible scores for the WHODAS and EQ5D; for the RAND-12, persons with composite scaled scores between 0–10 were considered to have floor effects, and persons with scores from 90–100 were considered to be at the ceiling.

Convergent Validity—Pearson correlations were examined to determine the relationships between the WHODAS 2.0 and other general measures of HRQOL (i.e., EQ5D and RAND-12); correlations less than 0.3 were considered poor, 0.3 – 0.6 adequate, and 0.6 or greater were good to very good evidence of convergent validity [51].

Known-Groups Analyses—A multivariate analyses was conducted to identify group differences (i.e., prodromal, early- or late-HD) on the HRQOL measures (WHODAS 2.0, EQ5D Health Scale, EQ5D Index Value, RAND-12 Physical Health, and RAND-12 Mental Health). We hypothesized that prodromal participants would report better functioning than both manifest HD groups, and that early-HD participants were report better functioning than late-HD participants.

Results

Four hundred seventy-seven (477) individuals with prodromal and/or manifest HD participated in this study. Participants were sampled to represent the entire continuum of HD symptomatology; 190 individuals had prodromal HD (CAG > 35, but did not yet have an HD clinical diagnosis) and 285 had manifest HD ($n = 196$ participants were earlier-stage HD and $n = 89$ were later-stage HD). Participants ranged in age from 18 to 81 years ($M = 48.86$, $SD = 13.22$). The majority of participants were Caucasian (96.6%); 1.7% were African American, 1.5% were classified as “other,” and 0.2% were unknown. Participants' education ranged from 4 to 26 years ($M = 15.13$, $SD = 2.90$). Groups did not differ on gender, $\chi^2(2, N = 475) = 2.392$, $p = .30$. While there were group differences in education, $F(2, 470) = 9.824$, $p < .0001$, these differences were small; early- ($M = 14.85$, $SD = 2.93$) and late-HD ($M = 14.31$, $SD = 2.62$) had 1 to 1.5 years less education relative to the prodromal HD group ($M = 15.80$ years, $SD = 2.88$). In addition, significant differences were seen for age (as symptoms are progressive with age), $F(2, 472) = 41.601$, $p < .0001$, with individuals who were prodromal ($M = 42.87$, $SD = 12.67$) being significantly younger than the early HD group ($M = 51.67$ $SD = 12.12$) and the late HD group ($M = 55.63$, $SD = 11.50$). Additionally, the early HD group was younger than the late HD group, $F(2, 472) = 41.601$, $p < .0001$. Table 1 includes the descriptive statistics for the three HRQOL measures.

Factor Structure

Results indicated that a 6-factor structure (understanding and communication; self-care; mobility; interpersonal relationships; work and household roles; community and civic roles) of the WHODAS fit the data well; CFI = 0.99, TLI = 0.99, and RMSEA = 0.02; see Figure 1.

Internal Consistency Reliability

Cronbach's alpha for the WHODAS 12 item scale was 0.94 (CI: .926–.944). For the six subdomains, Cronbach's alpha was .90 for Self-Care (CI: .879–.915), .89 for Mobility (CI: .868–.908), .83 for Life Activities (CI: .801–.861), .82 for Cognition (CI: .782–.848), .74 for Getting Along (CI: .690–.783), and .74 for Participation (CI: .685–.780).

Floor and Ceiling Effects

None of the HRQOL measures demonstrated floor effects (0% for the WHODAS, Rand-12 Physical Health Composite score, and Rand-12 Mental Health Composite Score; 0.2% for the EQ5D Health Scale; and 0.6% for the EQ5D Index Scale). Both WHODAS and EQ5D demonstrated ceiling effects (19.5% for the WHODAS; 23.9% for the EQ5D Index Scale; and 8.9% for the EQ5D Health Scale); ceiling effects were not present for the Rand-12 Physical Health Composite score, and Rand-12 Mental Health Composite Score (0% for both).

Convergent Validity

The WHODAS demonstrated moderate significant correlations with other general measures of HRQOL (i.e., EQ5D and RAND-12); correlations ranged from -0.41 to -0.76 (see Table 2).

Known-Groups Analyses

Multivariate analyses indicated that late-stage HD participants indicated poorer HRQOL than both early-stage HD and prodromal HD participants for all measures of HRQOL except the RAND-12 Mental Health, Pillai's Trace = .521, $F(10,922) = 32.520$, $p < .0001$, partial $\eta^2 = .261$; see Table 3.

Discussion

The purpose of this study was to examine the reliability and validity of the 12-item version of the WHODAS 2.0 in individuals with HD across the disease spectrum. We were able to replicate the 6-factor structure of the instrument that reflects: 1) understanding and communication; 2) self-care; 3) mobility (getting around); 4) interpersonal relationships (getting along with others); 5) work and household roles (life activities); and 6) community and civic roles (participation) in this population. In addition, convergent validity was supported by significant moderate correlations with other generic measures of HRQOL. While there was a ceiling effect for the WHODAS 2.0, this was comparable to the ceiling effect found for the comparative measures. Regardless, the WHODAS 2.0 was able to differentiate between individuals within prodromal, early-and late-stage HD.

Support for the 6-factor structure of the 12-item version of the WHODAS 2.0 is consistent with other studies examining the 36-item version [1; 3; 4; 6; 21; 52]. For the two studies that examined the factor-structure of the 12-item version of the WHODAS 2.0, only one was able to confirm the factor structure [25]; the other found support for a one-factor solution (for 6 of 7 developing countries) [24]. One possibility for the discrepancy between this study and the Sousa [24] study is that different populations were examined (older adults versus HD), and that impairments on all WHODAS domains in older adults may account for the single factor solution. It is also possible that the discrepancy between this study and Sousa [24] is due to differences in low-income countries relative to higher income countries. More work is needed to further examine these discrepancies. Similarly, support for reliability is also consistent with other previously published studies that demonstrate that Cronbach's alphas were good to excellent for the 36-item version [1; 4–6; 9–12; 14; 15; 17; 18; 21; 52] and good for the 12-item version [2]. Our examination of the reliability of the individual subscales indicated that, despite only 2 items per subscale, there was excellent internal consistency reliability for self-reported ADLs (i.e., Self-Care), good reliability for cognition and activities (i.e., Cognition and Life Activities), and adequate reliability for social engagement (i.e., Getting Along and Participation). This would provide support for using the individual subscales in studies that target a specific dimension of disability (e.g., an intervention designed to improve ADLs might focus on the ADL subscale score).

Our findings complement the findings of Downing and colleagues [20] who compared the 36-item WHODAS to the Total Functional Capacity (TFC) scale, the gold standard functional measure for HD. The 36-item WHODAS was able to detect worse functioning in prodromal HD participants with high disease burden at baseline, and longitudinal functional decline in medium and high groups as reported by companions. In addition, our findings also complement findings of Kim and colleagues [23] who found baseline group differences for medium and high burden prodromal groups, and baseline and longitudinal group differences for companion-rated assessments of individuals with medium and high disease burden reported worse functioning than controls) [23]. Specifically, the current analysis provides support for the utility of the 12-item version of the WHODAS 2.0 in individuals with both prodromal and manifest HD by demonstrating reliability, validity, and the ability to differentiate level of function by disease progression groups across the HD spectrum.

While this study provides support for the reliability and validity of the WHODAS 2.0 in individuals with HD, it is important for us to acknowledge several limitations. First, while our study examined individuals across the stages of the disease process, the reliability of self-report data in individuals in the later stages of the disease is questionable. Thus, future work should examine the utility of proxy reports and/or data from other sources when examining those in the later stages of the disease process; such information will provide a better understanding of the clinical utility of this measure. For example, Downing and colleagues [20] found that companions of prodromal HD participants in the medium and high disease burden groups reported functional decline over time on the WHODAS while their partners with the HD gene mutation did not, indicating possible reduced personal awareness of functional decline in prodromal HD. Furthermore, while the WHODAS 2.0 provides a reliable assessment of generic HRQOL, and it appears to be more sensitive to change over time than an HD-specific functional measure (i.e., the TFC [30]) [20], there is

still a need for measures that capture the aspects of HRQOL that are specific to HD and encompasses more than functional domains [46; 53]. In order to enroll in this study, prodromal participants were required to know their gene status; this also resulted in education differences between the prodromal and symptomatic participants. Our participants may also not be representative of people with the HD gene mutation who do not participate in research. Thus it is unclear if findings would be generalizable to those individuals that are at-risk for HD and are not aware of their gene status. Persons positive for the HD gene mutation who know their gene status might also do more self-monitoring than those who have not undergone HD gene testing [54]. In addition, this study only examined English speaking participants in the United States. Given previous work in developing countries that did not support a 6-factor model [24], future work might also focus on examining the psychometric properties of the WHODAS across different countries and in different languages. Finally, this study focused on cross-sectional data; this precluded our ability to examine specific measurement properties such as responsiveness to change and minimally important change score. Future work is needed to examine these measurement properties; such information will be important in informing the clinical utility and sensitivity of this measure in individuals with HD.

Regardless of these limitations, this study provides support for the WHODAS 2.0 as a valid tool for evaluating HRQOL in individuals with HD. Our findings help provide further support for administration of the 12-item version, as the instrument's 6-factor structure is consistent with that of the 36-item version. Additionally, the instrument has good correlation with the other generic HRQOL items in this study (EQ5D, RAND-12). Although future work is needed to examine this measure's sensitivity to change over time in individuals with manifest HD, this study suggests that this measure can effectively differentiate between individuals within different stages of the disease process. Findings also suggest that the WHODAS 2.0 may provide clinically useful information to providers and clinicians managing HD symptoms.

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Figure 1.
6-factor structure for the World Health Organization Disability Assessment Schedule 2.0 (WHODAS 2.0)

Table 1

Descriptive Statistics for the HRQOL measures

HRQOL Measure	Prodromal HD			Early HD			Late HD			All		
	N	Mean	SD	N	Mean	SD	N	Mean	SD	N	Mean	SD
WHODAS 2.0	190	3.45	4.86	196	9.37	8.52	89	20.65	10.91	477	9.14	9.95
EQ5D												
Health Scale	189	85.50	10.06	195	77.94	14.79	89	74.47	21.22	474	80.33	15.28
Index Value	190	0.89	0.12	196	0.80	0.14	89	0.71	0.16	477	0.82	0.16
RAND-12												
Physical Health	190	53.87	5.58	192	46.88	9.18	87	38.17	9.09	471	48.06	9.78
Mental Health	190	49.87	9.32	192	47.60	10.99	87	49.60	11.42	471	48.85	10.53

Note. HRQOL = Health-related quality of life; WHODAS 2.0 = World Health Organization Disability Assessment Schedule 2.0; EQ5D=EuroQOL5D. HD staging data was missing for n = 2 participants; thus the number of participants for the total Sample (N=475) exceeds the sum of the three subgroups (N=473).

Table 2
Pearson correlations between the WHODAS 2.0 and other measures of HRQOL

Measures	WHODAS 2.0	
	N	r
EQ5D		
Health Scale	474	−0.49
Index Scale	477	−0.65
Rand-12		
Physical Health	471	−0.76
Mental Health	471	−0.41

Note. HRQOL = Health-related quality of life; WHODAS 2.0 = World Health Organization Disability Assessment Schedule 2.0; EQ5D=EuroQol5D; all $p < .0001$

Table 3

HRQOL for prodromal, early-, and late-stage HD

	Prodromal HD (N=189) Mean (SD)	Early-HD (N=191) Mean (SD)	Late-HD (N=87) Mean (SD)	F	Partial η^2
WHODAS 2.0 ^{a,b,c}	3.47 (4.87)	9.27 (8.49)	20.89 (10.81)	148.75*	0.391
EQ5D Health Scale ^{a,b}	85.50 (10.06)	78.11 (14.80)	74.34 (21.39)	21.13*	0.083
EQ5D Index Scale ^{a,b,c}	.89 (.11)	.81 (.13)	.71 (.16)	65.71*	0.221
Rand-12					
Physical Health ^{a,b,c}	53.84 (5.58)	46.90 (9.20)	38.17 (9.09)	120.49*	0.342
Mental Health	49.85 (9.34)	47.76 (10.80)	49.60 (11.42)	2.15	0.009

Note. HRQOL = Health-related quality of life; WHODAS 2.0 = World Health Organization Disability Assessment Schedule 2.0; EQ5D=EuroQol5D; all $p < .0001$

^a indicates significant univariate differences between prodromal and early stage HD

^b indicates significant univariate differences between prodromal and late stage HD

^c indicates significant univariate differences between early and late stage HD