Measuring Quality of Life in Huntington's Disease Using a Proxy Approach: Validation of the HD-mQoL-prx Measure

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Background

Challenges of Quality of Life Assessment:
Cognitive decline in Huntington's Disease (HD) patients limits effective self-reporting of quality of life (QoL), complicating monitoring efforts.

Role of Proxy Reporting:
Proxy-reported measures enable caregivers to assess patients' QoL, ensuring continued evaluation despite communication and/or cognitive symptoms.

HD-mQoL-prx Development:
The HD-mQoL-prx provides a structured and validated tool for proxy-based QoL assessment based on the needs-based model.

A holistic representation of QoL:
Incorporating insights from those close to patients can help reflect a wider understanding of their well-being and inform care decisions throughout disease progression.

Aims

- **Develop** a proxy-reported instrument to assess the QoL of HD patients unable to self-report due to cognitive decline.
- **Ensure** measurement is consistent across disease stages by capturing caregiver perspectives alongside the patient experience.
- **To validate** the scale using Rasch Measurement Theory and Classical Test Theory, confirming reliability, unidimensionality, and clinical utility.
- **Create** a crosswalk table to be able to estimate a patient's self-reported score based on their proxy score.

Methods

Participants: Data were collected from HD patients and their caregivers.

Instrument Development: A 49-item questionnaire was created based on in-depth qualitative interviews across the UK, Germany, Czechia, and Italy. It was available in both self-reported and proxy-reported formats.

Psychometric Validation: Rasch Measurement Theory was primarily applied to develop and refine the HD-mQoL and HD-mQoL-prx scales.

Score Comparability: Test equating methodology using common items was employed to assess measurement equivalence and generate a crosswalk table for converting proxy scores to patient-reported equivalents.

Participants

Sample: 150 caregivers of individuals with HD were recruited across the UK (n=61), Germany (n=65), and Italy (n=24).
Demographics: The majority were female (58.1%) and most were spouses or partners of the HD patients (70.3%).
Age: The median age of participants was 59 years (IQR: 51–67), with country-specific variation.

Table 1: Study participants characteristics

PARTICIPANT CHARACTERISTICS	UK (N=61)	GERMANY (N=65)	ITALY (N=24)	TOTAL (N=150)
Female	38 (63.3%)	34 (53.1%)	14 (58.3%)	86 (58.1%)
Male	22 (36.7%)	30 (46.9%)	10 (41.7%)	62 (41.9%)
Median Age (IQR)	61.0 (54-67)	60.0 (54-68)	50.5 (41-59)	59.0 (51-67)
Spouse/Partner	47 (78.3%)	47 (73.4%)	10 (41.7%)	104 (70.3%)
Son/Daughter	4 (6.7%)	6 (9.4%)	3 (12.5%)	13 (8.8%)
Sibling	2 (3.3%)	0 (0.0%)	6 (25.0%)	8 (5.4%)
Parent	5 (8.3%)	10 (15.6%)	3 (12.5%)	18 (12.2%)
Friend	0 (0.0%)	0 (0.0%)	1 (4.2%)	1 (0.7%)
Other	2 (3.3%)	1 (1.6%)	1 (4.2%)	4 (2.7%)

Results

Data Analysis Process:
Stage 1: Self- and proxy-reported items were analysed separately using Rasch Measurement Theory. Misfitting items were identified and removed through tests of model fit, item residuals, local dependency, DIF, unidimensionality, PSI, and targeting.

Key outcome: The proxy measure was refined from 49 items to 23 items, ensuring strong fit to the Rasch model. The final scale demonstrated unidimensionality, excellent reliability (PSI > 0.85), good targeting, and coverage of all key QoL themes (Figure 1).

Stage 2: Patient and proxy datasets were combined and analysed jointly to assess their measurement equivalence.

Key outcome: Rasch diagnostics confirmed excellent model fit (p=0.498), no DIF between patient and proxy responses, and unidimensionality across both measures (Figure 2). These findings support placement on a shared continuum and enable score conversion via a crosswalk table.

Figure1: Person-Item map of HD-mQoL-prx: demonstrating scale targeting and coverage

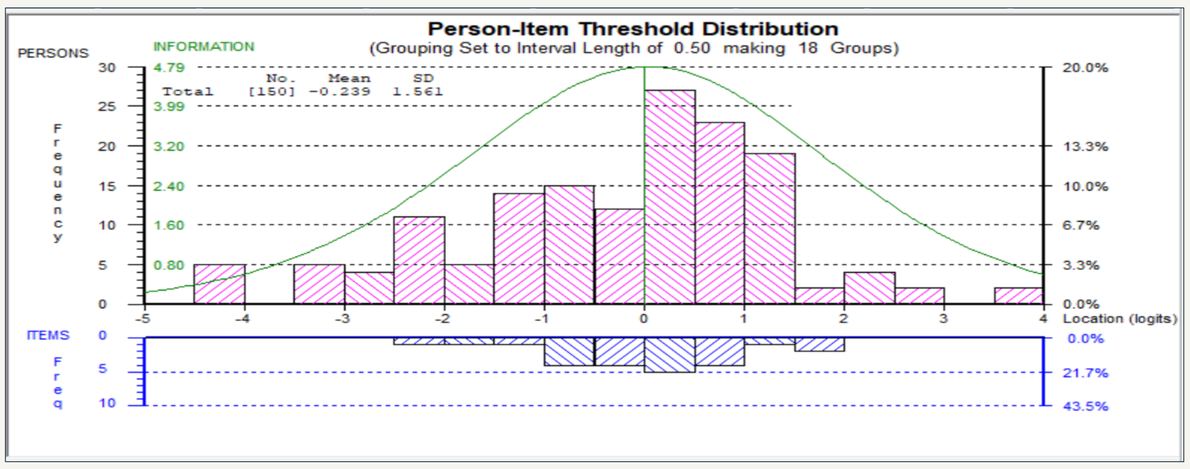
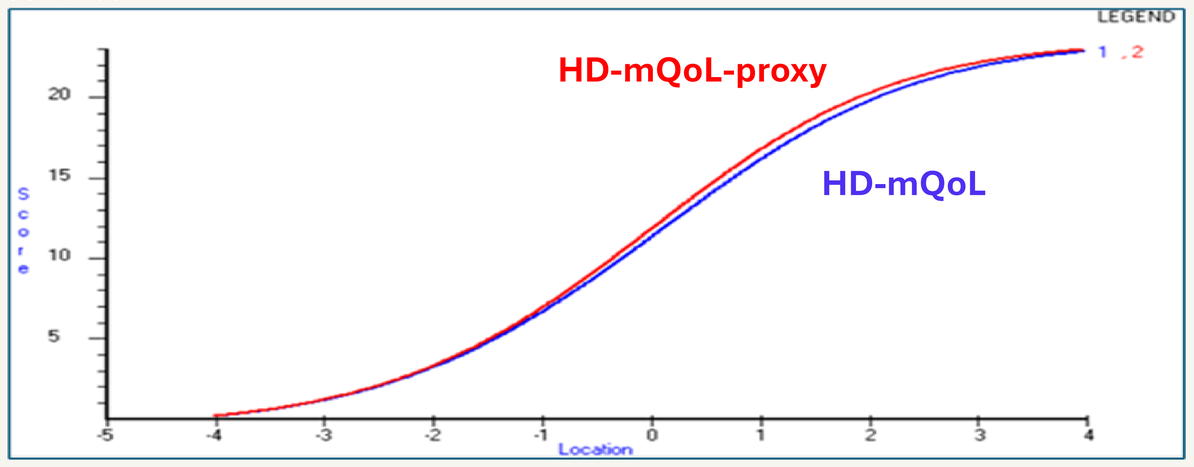


Figure 2: Patient & Proxy measures: demonstrating equivalence on a shared Rasch continuum



Conclusions

Validation: The HD-mQoL-prx was rigorously validated using Rasch Measurement Theory and Classical Test Theory, confirming strong psychometric properties and unidimensionality.

Measurement Equivalence: Anchoring proxy scores to the HD-mQoL ensured comparability between self- and proxy-reported data, bridging the gap across disease stages.

Clinical Utility: The measure enables continuous, holistic QoL monitoring in Huntington's Disease, even when cognitive decline limits self-reporting, supporting patient-centred care and informed decision-making.

Future Directions: Further research should explore longitudinal responsiveness and cross-cultural validation to strengthen clinical applicability.

Impact: HD-mQoL-prx fills a critical gap in HD-specific QoL assessment, offering a unique needs-based approach that supports both clinical practice and research.

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