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Research Report

Validation Study of the Parkinson's Disease Stigma Questionnaire (PDStigmaQuest)

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Abstract.
Background: Stigma is a relevant aspect of Parkinson's disease (PD). Specific stigma tools are needed to address the complex construct of stigma in PD comprehensively.
Objective: To test the dimensionality and psychometric properties of the newly developed Parkinson's Disease Stigma Questionnaire (PDStigmaQuest).
Methods: In this multi-center, cross-sectional study including PD patients and healthy controls, the dimensionality of the PDStigmaQuest was examined through exploratory factor analysis. Acceptability and psychometric properties were investigated. PDStigmaQuest scores of patients and healthy controls were compared.
Results: In total, 201 PD patients and 101 healthy controls were included in the final analysis. Results suggested high data quality of the PDStigmaQuest (0.0001% missing data for patients). The exploratory factor analysis produced four factors: felt stigma, hiding, enacted stigma: rejection, and enacted stigma: patronization, explaining 47.9% of variance. An optional work domain for employed patients was included. Moderate floor effects and skewness, but no ceiling effects were found. Cronbach's alpha of 0.85 indicated high internal consistency. Calculated item-total correlations met standard criteria. Test-retest reliability was high ($r_s = 0.83$). PDStigmaQuest scores correlated significantly with other stigma measures ($r_s = 0.56$–0.69) and were significantly higher in patients than in healthy controls and higher in patients with depressive symptoms than in those without.
Conclusions: The patient-reported 18-item PDStigmaQuest showed strong psychometric properties of validity and reliability. Our results suggest that the PDStigmaQuest can be used to assess and evaluate stigma comprehensively in PD, which will improve our understanding of the construct of PD stigma.

Keywords: Stigma, quality of life, validation study, questionnaire

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INTRODUCTION

Stigma is a determinant factor for Parkinson's disease (PD) patients' quality of life (QoL).[1–3] For the term "stigma", numerous definitions have been proposed in the last decades.[4–7] In the field of chronic illnesses, stigma is often studied as health-related stigma, a "social process, experienced or anticipated, characterized by exclusion, rejection, blame, or devaluation that results from experience or reasonable anticipation of an adverse social judgment".[8] Scambler and Hopkins initially introduced the distinction between felt stigma, including fear of being stigmatized and feelings of shame associated with the disease, and enacted stigma, referring to actual experiences of discrimination.[9,10] Fox et al. established a more complex stigma framework, including anticipated stigma (expectations of stigmatization), experienced stigma (actual stigmatization), and internalized stigma (adopting others' negative beliefs).[11]

Stigma plays a significant role in PD: For example, patients report experiences of being mislabeled as drunk, being stared at, feeling like being a burden to others, and feeling ashamed.[12–15] Especially in the early disease stages, patients try to hide PD-related symptoms from others.[13,16] Importantly, stigma can cause social isolation, obstruct seeking medical care, and is associated with non-motor symptoms (NMS) like depression and anxiety.[17–22]

However, our current knowledge of stigma in PD is sparse and mainly based on qualitative studies. There is currently no specific tool available for addressing the highly complex construct of stigma in PD comprehensively. To our knowledge, to date, mainly generic stigma measures for chronic illnesses or the PD Questionnaire 39 (PDQ-39) stigma subscale consisting of four items have been applied in PD.[23–27] Therefore, our objective was to develop and validate a stigma questionnaire specific to PD patients to help to address and evaluate PD stigma. The development process and data of the pilot study presenting the preliminary version of the patient-reported Parkinson's Disease Stigma Questionnaire (PDStigmaQuest) have been reported previously.[28] Here, we report validation data from the new PDStigmaQuest.

METHODS

Study design and participants

This multi-center (Cologne University Hospital, Germany; Movement Disorders Hospital in Beelitz, Germany) and cross-sectional validation study included patients with a diagnosis of PD according to the MDS criteria[29] and non-spousal and non-caregiver healthy controls. In- and outpatients were approached for participation in the study. For recruitment of both patients and controls, posters and flyers were used. Exclusion criteria were: age <18 or >90 years, moderate to severe medical conditions other than PD that could have interfered with the ability to complete the study, impaired hearing or sight interfering with study participation, significant cognitive impairment or insufficient knowledge of the German language based on the judgment of the examining health professional, and inability to consent. Additional exclusion criteria for patients were: PD of non-idiopathic form or other clinically relevant neurological diseases besides PD. Additional exclusion criteria for healthy controls were: PD diagnosis or other neurological or psychiatric disorders.

All participants were included between August 2022 and January 2024.

Ethical aspects

All participants provided written informed consent. The study was performed under the principles of the Declaration of Helsinki. The local ethics committee approved the study protocols (Cologne vote: 21-1385; Beelitz vote: 2023-85-BO). The study was registered at the German Clinical Trials Register: DRKS00025513.

Procedures and materials

Patients were tested under regular medication (MedON). Firstly, participants were asked about sociodemographic data, PD patients additionally about their disease history and current treatment. After that, different tools were assessed in German:

The German-language PDStigmaQuest is a patient-reported questionnaire developed based on literature, clinical experience, focus groups, and PD patients' and caregivers' feedback.[28] The version resulting from the pilot study consisted of 25 items based on the stigma concept by Fox et al. adapted for PD including two optional items for employed PD patients.[11,28] To test for uncomfortableness related to PD symptoms, the first item included sub-items to evaluate specific motor symptoms and NMS separately. Five items were reverse-scored items to control for response bias and avoid negative wording.[30] Each item was rated on a five-point Likert scale from

"never" (0) to "always" (4) regarding the past four weeks. Only item 1 (uncomfortableness related to PD symptoms) included the option "symptom not applicable" for sub-items. The PDStigmaQuest total score was calculated as the sum of all item scores. Since some items directly referred to the disease (e.g., "I try to hide my Parkinson's symptoms from others"), these could not be answered by healthy controls without PD. The controls were asked to fill in the following generally formulated items: item 1 (uncomfortableness with symptoms), 4 (feeling worth as much as others), 5 (feeling like a burden to others), 7 (feeling useless), 8 (self-respect), 11 (being seen as mentally impaired), 15 (decisions taken by others), 19 (being interrupted), 21 (being taken seriously), and 23 (others acting as feeling uncomfortable in the presence of the patient/control).

For retest evaluation, all patients were asked to complete only the PDStigmaQuest a second time 7–14 days after initial completion.

Beyond the PDStigmaQuest, the following self-rated scales and questionnaires were administered:

- The Stigma Scale for Chronic Illness (SSCI) contains 24 items measuring the stigma of chronic illnesses such as PD, Alzheimer's dementia, or epilepsy.[26] It consists of two subscales: self-stigma and enacted stigma. These are rated on a 5-point scale from "never" (1) to "always" (5), resulting in a maximum total score of 120. Higher values indicate higher stigma levels.
- The PDQ-39 is the most frequently used questionnaire for QoL in PD.[27,31] It contains 39 items in eight different domains. The items are rated on a 5-point scale from "never" (0) to "always" (4). As the domains contain different numbers of items, the domain scores are standardized on a summary index (SI) score from 0 (no impairment) to 100 (maximum impairment). In this study, only the stigma subdomain was used. Due to the domains' high internal consistency (stigma: Cronbach's $\alpha = 0.80$), they are often used independently of other domains.[32,33]
- The Beck Depression Inventory II (BDI-II) is an instrument measuring depression severity.[34] It consists of 21 items assessed on a 4-point scale (0–3), resulting in a maximum total score of 63. Higher scores indicate higher depression levels.
- The Hospital Anxiety Depression Rating Scale (HADS) is a scale for anxiety and depressive states.[35] It consists of 14 items divided into two subscales for anxiety and depression, each including 7 items. These are rated on a 4-point scale (0–3), resulting in a maximum score of 21 points for each subscale.

The following clinician-rated tools were administered:

- The Montreal Cognitive Assessment (MoCA) is a short screening test for mild cognitive impairment with various tasks testing the following cognitive domains: Short-term memory, visual-spatial abilities, executive functions, language, attention, concentration, working memory, and orientation.[36] A maximum total score of 30 (maximum performance) can be achieved.
- The Movement Disorders Society Unified Parkinson's Disease Rating Scale III (MDS-UPDRS III) is a clinician-based rating scale for motor function in PD.[37] It includes 18 items rated on a scale from "normal" (0) to "severe" (4) for different motor aspects (e.g., rigidity, tremor), most of them rated separately for the left and right side of the body, resulting in a maximum total score of 132 (maximum impairment). The MDS-UPDRS III additionally contains a Hoehn and Yahr (HY) classification for motor staging of PD, ranging from stage 0 (no signs of disease) to 5 (wheelchair bound or bedridden unless aided).

The SSCI, PDQ-39 stigma domain, and MDS-UPDRS III were only applied in patients.

Sample size

For conducting exploratory factor analyses (EFA), a minimum of 200 participants is proposed for questionnaires with up to 40 items.[38] A ratio of patients to controls of 2:1 was used based on other validation studies.[39,40]

Data analysis

Descriptive statistics for demographic and clinical characteristics were calculated. The Shapiro-Wilk test was applied to test for normal distribution of data. Levodopa equivalent daily dose (LEDD) was calculated according to the formula of Tomlinson et al.[41] The score of PDStigmaQuest item 1 (uncomfortableness with symptoms) was calculated by summing up the scores of all applicable symptoms and dividing the sum by the number of applicable symptoms. Data

quality was explored by the proportion of missing data points.

Only for patients, the following analyses were conducted:

(1) *Dimensionality:* For this analysis, the optional stigma domain for employed patients was left out as many patients were retired ($n = 137$). Further, based on participant feedback, patients had problems answering the reverse-scored items. We found that 15.4% of patients initially answered at least one reverse-scored item in the direction of the other non-reversed items, which was subsequently crossed out and changed. In 11.9% of patients, at least one answer to the reversed items did not match the other answers. Since we aimed to develop an easy-to-use and reliable tool, we decided to remove these five items prior to analysis, avoiding potentially biased item characteristics. Inter-item correlations between all other stigma items were calculated, and items mostly showing correlations <0.3 or >0.9 with other items were removed due to potential lack of fit with other items or collinearity.[42] A principal axis EFA with oblique rotation (promax, kappa = 4) was applied as many PDStigmaQuest items were right-skewed, and principal axis EFA does not make distributional assumptions. Oblique rotation was chosen as we assumed that the questionnaire's stigma factors would correlate and oblique rotation permits correlation between factors.[43] The Kaiser-Meyer-Olkin (KMO) measure was applied for testing sampling adequacy. Values >0.7 are considered middling, values >0.8 meritorious, and values >0.9 marvelous.[44] KMO values for individual variables should be >0.5. Bartlett's test of sphericity was used for testing the adequacy of the correlation matrix.

(2) *Acceptability* was tested through floor and ceiling effects (percentage of extreme values $\leq 15\%$) and skewness.[45] For the latter, limits were –1 and +1.[46]

(3) *Internal consistency*: Cronbach's alpha was calculated for PDStigmaQuest as a whole as some domains only consisted of 2 items (standard value ≥ 0.7).[30,40,47] The work domain not applying to many patients resulted in systematic data loss for internal consistency analysis. Therefore, only for calculating Cronbach's alpha, items referring to work left out by unemployed/retired patients were coded as zero.[48] This approach was considered acceptable because the work domain not applying to a patient also means that the patient cannot be confronted with stigma at work. Further, for every item, corrected item-total correlation (standard value ≥ 0.3) and inter-item correlations (standard value > 0.20 and < 0.75) in every domain were calculated.[30,49]

(4) *Test-retest reliability* was investigated through Spearman correlation between the initial assessment and the retest (7–14 days later) PDStigmaQuest total score.

(5) *Convergent validity* was tested through correlations with other stigma measures (SSCI, the stigma domain of the PDQ-39; $r_s > 0.50$).[50]

(6) *Known-groups validity:* Based on the well-established relationship between stigma and depression, known-groups validity was tested by comparing PDStigmaQuest scores in patients with and without depressive symptoms using a Mann-Whitney *U* test.[21] We hypothesized that stigma scores should be higher in PD patients with than without depressive symptoms. To identify patients with depressive symptoms, the cut-off BDI-II ≥ 14 was used as originally suggested by Beck et al. for detecting mild depression.[34,51]

The following analyses were conducted with data from patients and controls:

(1) *Group confirmation:* To ensure that relevant PD symptoms were not similarly present in healthy controls, Mann-Whitney *U* tests were conducted between PD patients and healthy controls for the following scales: MoCA total, BDI-II total, HADS-A, and HADS-D. Tests were corrected for multiple comparisons according to the Benjamini-Hochberg procedure.

(2) *Comparison of PDStigmaQuest scores*: To compare PD patients' and healthy controls' stigma scores, a Mann-Whitney *U* test was conducted with new stigma scores summing up only items answered by both groups. We hypothesized that stigma scores should be higher in PD patients than in healthy controls.

All analyses were conducted using Statistical Package for Social Sciences (SPSS; version 28.0). *P*-values < 0.05 were considered statistically significant.

RESULTS

Demographic characteristics

In total, 201 PD patients and 101 healthy controls matched by age and sex were included in the final analysis. Demographics and clinical characteristics are presented in Table 1.

Data quality and dimensionality

In patients, there was one missing data point in the PDStigmaQuest (0.0001% missing). In healthy controls, no data were missing.

Correlations of item 6 (feeling responsible for PD) were ≥ 0.3 with only two other items, thus item 6 was removed. KMO was 0.84 for the remaining items, showing that our sampling was adequate. Further, KMO values for individual variables were all > 0.5. Bartlett's test of sphericity was statistically significant ($p < 0.001$), indicating that our correlation matrix was appropriate for conducting a factor analysis. Kaiser-Guttman criterion extracting factors with eigenvalues > 1 suggested a four-factor solution explaining 46.0 % of the variance. In the pattern matrix, loadings of item 16 on the factors were all < 0.30, so this item was dropped.[42] Subsequently, the EFA was completed again showing a four-factor solution explaining 47.9% of the variance (Table 2): 8 items loaded onto a factor interpreted as "felt stigma", 3 items loaded onto a factor measuring "hiding", 3 items loaded onto a factor measuring "enacted stigma: rejection", and 2 items loaded onto a factor measuring "enacted stigma: patronization". The factor felt stigma was correlated with the other factors hiding ($r = 0.50$), enacted stigma: rejection ($r = 0.39$), and enacted stigma: patronization ($r = 0.36$). Factors enacted stigma: rejection and enacted stigma: patronization were also moderately correlated ($r = 0.42$).

PDStigmaQuest scores and acceptability

Descriptive statistics of PD patients' PDStigmaQuest scores and acceptability parameters are shown in Table 3. The maximum total score for (self-)employed patients summing up the final 18 items is 72, while unemployed/retired patients can achieve a total score of 64 (without the two items referring to work). In PD patients, floor effects were found for the domains hiding, enacted stigma: rejection, enacted stigma: patronization, and optional domain of work, but not for the domain felt stigma and total score. No ceiling effects were found. A moderate skewness was found for domains hiding and enacted stigma: rejection.

Internal consistency

Cronbach's alpha was 0.85 for the whole scale. Inter-item correlations and corrected item-total correlations are presented in Table 4.

Table 1
Demographics and clinical characteristics of patients with Parkinson's disease and healthy controls

	Patients	Healthy controls	*p*
Sex (female)	34.3%	45.5%	0.058
Age (y)[a]	64.4 ± 9.7: 32–86	62.5 ± 10.1: 42–87	0.110
Education (y)[a]	15.3 ± 3.1: 7.5–23	16.2 ± 3.3: 9–27	**0.036**
Family status			0.908
Married	74.1%	70.3%	
Single	9.0%	10.9%	
Divorced	10.9%	11.9%	
Widowed	6.0%	6.9%	
Occupation			**<0.001**
(Self-)employed	30.3%	58.4%	
Retired	67.2%	37.6%	
Other	2.5%	4.0%	
Disease duration (y)[a]	7.9 ± 5.0: 0.4–26.8	N/A	N/A
LEDD (mg)[b]	667.6 ± 464.9	N/A	N/A
MDS-UPDRS III (score)[b]	26.0 ± 12.3	N/A	N/A
HY (stage)[c]	2.0 (2.0–3.0)	N/A	N/A
Treated with DBS	22.9%	N/A	N/A

DBS, Deep brain stimulation; HY, Hoehn and Yahr; LEDD, Levodopa equivalent daily dose; MDS-UPDRS III, Movement Disorders Society Unified Parkinson's Disease Rating Scale III; N/A, not applicable. Note: Significant differences are highlighted in bold. [a]Mean ± *SD*: range [b]Mean ± *SD* [c]Median (Interquartilerange).

Table 2
Pattern matrix of the final exploratory factor analysis

	Item	Factor			
		1	2	3	4
	Felt stigma				
1	In the presence of others, I feel uncomfortable … [list of symptoms]	**0.629**			
3	I am unhappy about how my Parkinson's symptoms affect my appearance.	**0.563**			
5	I see myself as a burden to others.	**0.647**			
7	I feel useless.	**0.613**			
9	I worry about how others may react to my Parkinson's disease.	**0.539**	0.364		
10	I worry about how others will perceive me when my Parkinson's disease progresses.	**0.648**			
11	I am afraid that others could consider me mentally impaired.	**0.467**			
20	Because of my Parkinson's symptoms, others have looked at me.	**0.403**			
	Hiding				
2	I feel uncomfortable when others address me regarding the treatment of my Parkinson's disease (e.g., pills, patches, pump, or deep brain stimulation).		**0.413**		
12	I try to hide my Parkinson's symptoms from others.		**0.758**		
13	I have kept my Parkinson's disease secret from someone.		**0.900**		
	Enacted stigma: rejection				
17	Friends or family members have turned away from me because of my Parkinson's disease.			**0.632**	
22	I have got invited by others less often than prior to my Parkinson's disease.			**0.701**	
23	Others have behaved as if my presence made them feel uncomfortable.			**0.676**	
	Enacted stigma: patronization				
15	I have experienced others making decisions for me before I can make them for myself.				**0.770**
19	I have experienced others not letting me talk.	0.320			**0.422**

PD, Parkinson's disease. Note: Loadings < 0.3 omitted.[42] Items assigned to the respective factor in bold.

Table 3
Distribution and acceptability of PDStigmaQuest domain scores for patients with Parkinson's disease

	Mean	*SD*	Minimum	Maximum	Maximum achievable	Floor effect (%)	Ceiling effect (%)	Skewness
Felt stigma	9.2	5.4	0	27.9	32	3.0	0	0.6
Hiding	2.4	2.7	0	12.0	12	35.8	0.5	1.2
Enacted stigma: rejection	0.8	1.5	0	7.0	12	67.2	0	2.2
Enacted stigma: patronization	1.8	1.7	0	6.0	8	31.3	0	0.7
Optional: work domain ($n = 64$)	1.7	1.5	0	5.0	8	31.3	0	0.4
Total Score	14.7	8.9	0	43.91	72	2.0	0	0.6

PDStigmaQuest, Parkinson's Disease Stigma Questionnaire.

Test-retest reliability

Spearman correlation between initial and retest PDStigmaQuest total score was 0.83 ($n = 147$, $p < 0.001$).

Convergent validity

The final PDStigmaQuest correlated moderately with the PDQ-39 stigma domain ($r_s = 0.56$, $p < 0.001$) and strongly with the SSCI total score ($r_s = 0.69$, $p < 0.001$).

Known groups validity

Final PDStigmaQuest scores were higher in patients with depressive symptoms (mean = 20.8, $SD = 9.6$) than in patients without depressive symptoms (mean = 13.1, $SD = 8.1$, $p < 0.001$).

Comparison of patients and controls

Descriptive characteristics of MoCA total, BDI-II total, HADS-A, and HADS-D score for PD patients and controls are presented in Table 5. All examined

Table 4
Internal consistency analysis for patients with Parkinson's disease

	Item	*n*	Inter-item correlation	Item-total correlation
	Domain 1: Felt stigma			
1	Uncomfortableness related to PD symptoms	200	**0.34–0.52**	**0.63**
2	Uncomfortableness related to PD appearance	200	**0.29–0.52**	**0.51**
3	Feeling like a burden to others	200	**0.27–0.51**	**0.51**
4	Feeling useless	200	**0.27–0.51**	**0.53**
5	Worries about reactions to PD	200	**0.28–0.70**	**0.62**
6	Worries about reactions to disease progression	200	**0.27–0.70**	**0.64**
7	Fear of being seen as mentally impaired	200	**0.29–0.48**	**0.56**
8	Being observed	200	**0.27–0.36**	**0.45**
	Domain 2: Hiding			
9	Feeling uncomfortable being asked about PD treatment	201	**0.40–0.43**	**0.46**
10	Hiding of PD symptoms	201	**0.43–0.66**	**0.67**
11	Concealing PD	201	**0.40–0.66**	**0.65**
	Domain 3: Enacted stigma: rejection			
12	Rejection by friends and family members	201	**0.42–0.45**	**0.51**
13	Being invited less often	201	**0.45–0.49**	**0.56**
14	Others acting as feeling uncomfortable in the presence of the patient	201	**0.42–0.49**	**0.54**
	Domain 4: Enacted stigma: patronization			
15	Decisions taken on behalf of patient by others	201	**0.44**	**–**
16	Being interrupted	201	**0.44**	**–**
	Optional domain: Work			
17	Fear of devaluation at work	64	0.18	**–**
18	Unfair treatment at work	64	0.18	**–**

PD, Parkinson's disease. Note: In bold are inter-item correlations > 0.2 and item-total correlations ≥ 0.3, representing preferable item characteristics.[30,49] New numeration of items refers to the final questionnaire.

clinical characteristics were significantly higher in PD patients than in controls.

New stigma scores summing up only items in the final PDStigmaQuest answered by PD patients and controls were higher in PD patients (mean = 5.5, $SD = 3.9$) than in healthy controls (mean = 3.3, $SD = 2.5$, $p < 0.001$).

Table 5
Comparison of patients with Parkinson's disease and healthy controls regarding relevant clinical characteristics

	Patients			Healthy controls			*p*
	n	Mean	*SD*	*n*	Mean	*SD*	
MoCA total	197	26.1	2.6	98	27.7	1.9	**<0.001**
BDI-II total	197	8.8	6.2	100	5.0	5.0	**<0.001**
HADS-A	194	4.5	3.2	101	3.6	2.9	**0.016**
HADS-D	194	4.1	3.2	101	2.3	2.3	**<0.001**

BDI-II, Beck Depression Inventory II; HADS, Hospital Anxiety and Depression Scale; MoCA, Montreal Cognitive Assessment. *Note:* Mann-Whitney *U* tests between patients with Parkinson's disease and healthy controls to analyze differences in relevant clinical characteristics. Bold font highlights significant results, $p < 0.05$; All *p*-values are corrected for multiple comparisons using Benjamini-Hochberg procedure.

DISCUSSION

In this study, we report validation data from the PDStigmaQuest, the first questionnaire specifically and comprehensively addressing stigma in PD. Our results illustrate that the new PDStigmaQuest, consisting of 18 items, is a valid and reliable self-reported questionnaire to comprehensively assess and evaluate stigma in a real-life PD population. Face validity can be assumed as experts, PD patients, and caregivers developed and reviewed the scale.[28]

Data quality and dimensionality

The assessment of the PDStigmaQuest revealed high data quality with only 0.0001 % missing data in patients and no missing data in controls. Results from EFA indicated sufficient construct validity. EFA identified 4 factors: felt stigma, hiding, enacted stigma: rejection, and enacted stigma: patronization. We additionally included an optional work domain for employed patients. The identified factors only partially overlap with our initially assumed domains based on one of the latest stigma conceptualizations: uncomfortableness, internalized stigma, anticipated stigma, hiding, and experienced stigma.[11] Instead, the factors identified in the EFA align with the earlier

stigma model by Scambler and Hopkins, consisting of felt and enacted stigma, extended by the domain of hiding.[9] Our proposed domains of uncomfortableness, internalized stigma, and anticipated stigma were grouped as only one factor: felt stigma. Our initially assumed domain experienced stigma was divided into two aspects: rejection and patronizing by others. These also represent two components of the enacted stigma concept by Scambler and Hopkins.[9] Our additional hiding domain specific to PD patients due to their partly concealable condition was preserved, with one additional item previously assigned to uncomfortableness (feeling uncomfortable being asked about PD treatment), which could have resulted from the high correlation between the factors felt stigma and hiding ($r = 0.50$). Fox et al. based their stigma concept on stigma insights concerning mental illness, which may explain the differences observed to the findings of our study in PD.[11] In contrast, Scambler and Hopkins investigated stigma for persons with another neurological disease, epilepsy, in which stigma is conceptually closer to PD stigma.[9]

Acceptability

Moderate floor effects were found for domains of hiding, enacted stigma: rejection, enacted stigma: patronization, and work. In the context of other health conditions, it is often reported that felt stigma, including fear of being stigmatized, is significantly more prevalent than experiences of enacted stigma, e.g., rejection and patronization.[52,53] Especially enacted stigma: rejection items refer to extreme forms of stigma, including breaking off contact to the PD patient.[54] Therefore, this domain was expected to show floor effects. We nevertheless included these items since they are essential to portraying stigma in PD. Furthermore, it was shown that hiding efforts are more prevalent in the early stages of the disease, potentially leading to moderate floor effects of the domain hiding in our PD cohort representing the general PD population.[13] Floor effects of the domain work could be explained by the fact that some PD patients stated to be self-employed and, therefore, the stigma items might not be fully applicable. Future studies should differentiate between employed and self-employed patients when investigating work-related stigma. To date, little is known about PD patients' work-related stigma, which has to be investigated more intensively in the future, representing an important stigma aspect in PD.[55,56] Notably, there were no floor effects for the total score and ceiling effects were absent. In summary, the results indicated an appropriate acceptability of the final PDStigmaQuest.

Internal consistency and test-retest reliability

For the final PDStigmaQuest, Cronbach's alpha was 0.85, indicating high internal consistency. Inter-item correlations in the different stigma domains were all satisfactory except for work items. In this domain, one item represents felt stigma and the other enacted stigma experiences according to Scambler and Hopkins.[9] The differential prevalence of the two stigma aspects could have led to low inter-item correlations with higher values on the felt stigma than on the enacted stigma item. However, given the importance of employment for PD patients, we consider including both stigma types within an optional domain for employed patients necessary. All calculated item-total correlations for domains met standard criteria. Test-retest correlation of the final PDStigmaQuest was 0.83, indicating high test-retest reliability.

Convergent and known-groups validity

The final PDStigmaQuest showed satisfactory correlations with other stigma measures, suggesting high convergent validity. Furthermore, data provided evidence for adequate known-groups validity due to the difference in PDStigmaQuest scores between patients with and without depressive symptoms. This finding is consistent with previous stigma literature, showing higher stigma levels in patients with higher depression levels.[19,21,22,57,58]

Comparison of PD patients and healthy controls

Comparing PD patients and healthy controls regarding relevant PD symptoms, we observed higher scores in patients than in healthy controls, providing evidence for a representative control group. New stigma scores summing up only items in the final PDStigmaQuest answered by PD patients and controls were significantly higher in PD patients, suggesting that the stigma experiences in the PDStigmaQuest are not equally made by elderly people without PD and rather represent PD-specific experiences.

Limitations

This validation study also has some limitations. Firstly, the two items referring to work-related stigma ($n = 64$) could not be explored with regard to dimensionality as for EFA, a minimum of 200 cases is required.[38] However, we decided to retain these items due to their high importance for employed PD patients. Furthermore, since these items represent a dimension of stigma that can only be experienced by a subgroup of patients, treating this domain separately and not as a part of other stigma aspects affecting the general PD population was considered appropriate. Secondly, only seven items of the final PDStigmaQuest were applicable also to healthy controls and therefore, could be compared to PD patients. All other items were not completed by healthy controls as they included PD-related wording like "because of my Parkinson's disease" and already implied that healthy controls cannot have these experiences at all. Thirdly, there was a difference between PD patients and healthy controls in years of education as well as employment status. Although to our knowledge, education has not been associated with stigma in PD, it has been identified as an influencing factor in other conditions such as epilepsy so that it would be reasonable controlling years of education in further studies.[59] The difference in employment status is somehow expected as PD patients retire 4–7 years earlier than the general population.[60] Lastly, the field's current understanding of stigma in PD remains limited, highlighting the need for cross-validation of our findings in different PD cohorts, with a special need for investigating stigma in different countries and socio-cultural backgrounds as well as providing longitudinal data.

Conclusions

In conclusion, our results indicate that the patient-reported PDStigmaQuest has strong psychometric properties of validity and reliability and is helpful in assessing and evaluating PD-specific stigma. In future, the PDStigmaQuest can be applied to understand the different aspects of PD stigma and their potential influencing factors, e.g., demographics, and its relationship to clinical characteristics in more detail. This might contribute to improve the management of stigma in clinical practice and, as a consequence, patients' QoL.

Future studies validating the PDStigmaQuest in different languages and independent multi-cultural PD cohorts are warranted.

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CONFLICT OF INTEREST

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DATA AVAILABILITY

The data used to support the findings of this study are available from the corresponding author upon reasonable request.

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