Title: **Stigma as a key determinant of health-related quality of life in Parkinson’s disease**

Abstract

*Introduction:* People with Parkinson’s disease (PD) may experience stigma due to their visible features of movement and communication difficulties. This paper aimed to examine the role of stigma in health-related quality of life (QOL), after controlling for personal and clinical characteristics.

*Methods:* This is a preliminary analysis of a subset of baseline data from the Social Self-Management of Parkinson’s Disease Study (SocM-PD), an ongoing 3-year prospective cohort study. 73 people with PD ($M_{age} = 65.72$, 29 women) participated in this study.

Hierarchical multiple regression analyses were used to determine the role of stigma in QOL, after controlling for gender, disease severity, depression, and motor experiences of daily living.

*Results:* Significant correlations were found between QOL with gender ($r = .26$), disease severity ($r = .38$), depression ($r = .65$), motor experiences of daily living ($r = .71$), and stigma ($r = .83$). After controlling for the significant covariates, stigma made a significant and unique contribution to the explanation of QOL by 13.7 % ($p < .001$). A final hierarchical multiple regression with stigma and the 4 covariates revealed an overall model that explained 77.8% of
the total variance of QOL ($F[5, 63] = 48.79, p < .001$).

**Conclusion:** Stigma appears to be a key determinant of QOL in people with PD. The results suggest the importance of further understanding stigma in PD to develop possible intervention strategies. Future work is also needed to verify the results with a larger and longitudinal dataset of the SocM-PD.

Keywords: Health-related quality of life; Parkinson’s disease; Stigma
1. Introduction

As first documented by Dr. James Parkinson in 1817 and later depicted in the classic picture by Gowers in 1896 [1], a person’s Parkinson’s disease (PD) can be observed from the masked face, stooped posture, clenched and trembling arms, and shuffling gait. The image connotes deformities and disability, which are likely to lead to a situation in which the individual is disqualified from full social acceptance, defined by Goffman as stigma [2].

Experience of stigma in people with PD has been described vividly and analyzed in-depth in qualitative research [3-7]. Stigma emerges from the interaction between the individual with PD and the “outside world”. People with PD may feel stigmatized (or shamed, embarrassed, disgraced) when encountering responses of others, such as staring, questioning, and avoiding, to their visible features of movement and communication difficulties. Being aware of their symptoms transmitting a message of not being socially competent and of being labeled as deviate, the individual with PD may try to disguise the symptoms and later withdraw from a public into a private world when they can no longer hide their symptoms.

Quantitative research also has validated the source of stigma by surveying public attitudes toward PD [8] and by examining others’ impressions of PD symptoms [9-11]. Moore and Knowles [8] reported that nearly half of the respondents considered stigma was attached
to this disease. And those who believed there was stigma had more negative attitudes to PD.

In addition, Tickle-Degnen [9] showed that facial masking may bias health care practitioners’ views of people with PD as more depressed, less sociable, and less cognitively competent than their actual attributes. Moreover, Hemmesch [10, 11] reported that higher facial masking or higher abnormal bodily movement (including tremor and related movement disorders) of individuals with PD elicited more negative first impressions in older adult observers. Along with these negative attitudes and biased impressions embedded in our society, people with PD are likely to feel stigmatized and thus suffer from psychological distress.

Health-related quality of life (QOL) is a broad construct pertaining to subjective well-being, taking into account body function, activity and participation, and environment.

Therefore, it is likely that psychological distress and social withdrawal resulting from stigma would lead to decreased QOL. To the best of our knowledge, however, research in PD has only reported that stigma, measured in a subscale of a PD-specific QOL measure, is a determinant of depression [12]. No research has examined the degree to which stigma plays a role in overall feelings of quality of health in PD. In contrast, research about other diseases (e.g., epilepsy [13], HIV [14], lung cancer [15]) has documented that stigma predicted a significant amount of variance of QOL.
Identifying determinants of QOL is important to guide the development of interventions. In a systematic review of determinants of QOL in PD [16], the factors that had been examined included demographic variables, disease characteristics, non-motor symptoms, and motor symptoms. Among the reviewed studies that used various factors and analysis models, depression was found to be the most constant predictor of QOL in the final model. In a later study with path analysis [17], in which depression was not included, limitations in performing self-care activities as measured by the Unified Parkinson’s Disease Rating Scale (UPDRS) Part II was reported to contribute the most to QOL in the model. Given the above findings, it is of interest to know whether stigma would be a determinant of QOL in PD after controlling for depression and limitations in self-care activities.

A series of research by Tickle-Degnen on daily living with PD has demonstrated social concerns as a primary focus for individuals with PD [18]. The disease may not only cause impairments in body function, but also likely to interact with the socio-cultural context to influence QOL, as illustrated in our aforementioned review of stigma in PD. Therefore, the purpose of this project was to examine (1) the relationship of experienced stigma with depression and QOL, (2) the role of stigma in predicting QOL after controlling for personal and clinical characteristics, and (3) the type of stigma that would contribute to QOL.
2. Methods

2.1. Subjects

This paper analyzed a subset of baseline data from the Emergence and Evolution of Social Self-Management of Parkinson’s Disease Study (SocM-PD), an ongoing 3-year prospective cohort study [18]. Inclusion criteria were (a) diagnosis of idiopathic PD utilizing the United Kingdom Parkinson’s Disease Society Brain Bank clinical diagnostic criteria, (b) modified Hoehn and Yahr stage 1 through 4, (c) score $\geq 26$ on the Mini-Mental Status Exam, (d) home setting within travel distance to study locations, (e) able to communicate clearly and in English with research staff, (f) interested in participating and willing and able to provide informed consent. Participants were on antiparkinsonian medications. Protocols were approved by the institutional review boards of Boston University and Tufts University. All participants provided written informed consent before the testing and interview began.

2.2. Measurements

Measures reported in this paper were part of a larger interview and testing protocol of SocM-PD [18]. Study data were collected and managed using REDCap electronic data capture tools [19] hosted at Tufts University. REDCap (Research Electronic Data Capture) is a secure, web-based application designed to support data capture for research studies.
2.2.1. Stigma

The 24-item Stigma Scale for Chronic Illness (SSCI) was developed to measure stigma experienced by people with chronic neurological disorders including PD [20]. It contains two subscales: felt stigma and enacted stigma. The felt stigma scale asks questions about the respondent’s feelings, for example, embarrassment, worry, and self-blame. The enacted stigma subscale asks questions about the behavior of others toward the respondent, for example, avoiding contact, staring, and being unkind. Each item is rated as 1 = never, 2 = rarely, 3 = sometimes, 4 = often, and 5 = always. A higher score indicates a higher frequency of experiencing stigma. A systematic review [21] suggests that the SSCI has good content validity and fair internal consistency.

2.2.2. Depression

The 15-item Geriatric Depression Scale (GDS) is a self-report measure to detect depressive symptoms in older adults [22] and is appropriate for use in PD as it excludes many symptoms overlapping between depression and PD [23]. The GDS is reported to have adequate discriminant validity for a diagnosis of depressive disorder at a cutoff of 5, with a higher score meaning more depressive. In addition, the GDS is reported to have good internal consistency and test-retest reliability.
2.2.3. Motor experiences of daily living

The UPDRS-Part II is the second subscale of the UPDRS, which concerns self-perceived motor experiences of daily living [24]. A higher score indicates a more severe impact of motor symptoms on their ability to complete activities of daily living. Clinimetric testing suggests satisfactory internal consistency and concurrent validity.

2.2.4. QOL

The 39-Item Parkinson’s Disease Questionnaire (PDQ-39) assesses life concerns of people with PD [25]. It is composed of a summary index (PDQ-SI) and eight domain scores – mobility, activities of daily living (ADL), emotional well-being, stigma, social support, cognition, communications, and bodily discomfort. A higher score indicates a more frequent self-perceived difficulty in QOL. Psychometric testing suggests that the PDQ-39 has adequate reliability and validity [26].

2.3. Statistical Analyses

Descriptive statistics were used to summarize demographic and clinical characteristics, as well as scores in study measures. Student’s t test was used to compare the felt and enacted stigma scores of the SSCI. Pearson’s correlation coefficients were computed between the study variables. Finally, hierarchical multiple regression was used to estimate the contribution
of stigma in the explanation of the variance of QOL, after controlling for significant covariates. All analyses were performed by IBM SPSS statistics 22 for Windows.

3. Results

Table 1 shows the demographic and clinical characteristics of the study participants (N = 73). A majority of our participants (63%) had household income above $50,000 USD, which was at the average-to-high income level in New England area. Due to missing data on the PDQ-39 and SSCI, four participants were excluded pair-wise during statistical analyses. For SSCI, one participant never had any felt stigma, while 14 participants never had any enacted stigma (20.3%). About half of the participants experienced more-than-rare (>= 2) felt stigma and 15.9% of the participants experienced more-than-rare enacted stigma. In addition, the participants reported significantly higher felt stigma than enacted stigma (t = 9.99, p < .001).

Table 2 shows the Pearson’s correlation coefficients among the study variables. Overall stigma had significant correlations with Hoehn & Yahr (H&Y) stage, GDS and UPDRS Part II. PDQ-SI was significantly correlated with gender and H&Y stage, and strongly correlated (r > .50) with GDS, UPDRS Part II, and SSCI (overall, felt, and enacted stigma). The overall stigma of SSCI had correlation coefficients r above .40 with all domains of the PDQ-39.
Those factors that were significantly correlated with PDQ-SI were entered into the regression model as covariates (i.e., gender, H&Y stage, GDS, UPDRS Part II). A simultaneous multiple regression with the four covariates and stigma revealed an overall model that explained 77.8% of the total variance of PDQ-SI ($F[5, 63] = 48.79$, $p < .001$, Table 3). Overall stigma provided a unique and significant explanation of the variance of QOL over and above that of gender, H&Y stage, GDS, and UPDRS Part II, by 13.7% ($p < .001$). When the domain scores of the PDQ-39 were entered as the outcome variable, overall stigma provided significant explanation of the variance of PDQ-39 in domains of ADL, emotional well-being, stigma, social support, and communications, in addition to that of the covariates.

To explore the relative importance of felt vs. enacted stigma in the explanation of the variance of QOL, these two stigma scores were entered stepwise as independent variables into the hierarchical multiple regression model in addition to the covariates. Table 4 shows that felt stigma, rather than enacted stigma, explained a significant amount of variance of PDQ-SI and domains of ADL, emotional well-being, stigma, social support, and communication.
4. Discussion

The important findings of this study are that stigma appears to be a key determinant of QOL in people with PD. Experienced stigma was strongly related to depressive symptoms and motor experiences of daily living. Those who reported higher stigma tend to have more depressive symptoms and more motor difficulties in daily living. Moreover, after controlling for gender, disease severity, depression, and motor experiences of daily living, we found that stigma, especially felt stigma, made a significant and unique contribution to the explanation of the variance in QOL, including motor, emotional, and social domains. Our study is the first one to demonstrate the important role of stigma in QOL of PD.

Stigma has been a popular issue in diseases such as epilepsy, HIV, and lung cancer. However, research on stigma in PD is just in the beginning stage; only one study reported stigma as a determinant of depression [12]. Our findings of stigma as a key determinant of QOL are consistent with research in epilepsy [13], HIV [14], and lung cancer [15]. In PD, although many studies have examined determinants of QOL, those studies only examined features related to personal and clinical characteristics [16], and failed to include predictive factors emerging from interaction with the socio-culture context. Feeling of stigmatization manifests itself when the individual with PD has to interact with others or has to show up in
public. The significant contribution of stigma to QOL suggests that one’s subjective well-being is inseparable from the socio-cultural context.

By breaking down overall stigma into felt and enacted stigma, we further found that felt stigma, compared to enacted stigma, was experienced by more participants and to a stronger degree. Moreover, felt stigma appears to be the main type of stigma contributing to QOL. Our results are in parallel with research findings in epilepsy [27, 28] that felt stigma was more prevalent than enacted stigma and that felt stigma may be in its own right a profound source of psychological distress and QOL problems.

Given the critical role of felt stigma in QOL, it is important to track where felt stigma may come from. In Corrigan’s model [29], public attitudes lead to enacted stigma, which in turn results in felt stigma. Our post hoc meditation analysis seems to support the model by finding a significant indirect effect of enacted stigma on QOL through felt stigma (Lower Limit Confidence Interval=4.22, Upper Limit Confidence Interval=12.02, $p < .05$, after controlling for depression and motor experiences of daily living. Although the scores of enacted stigma suggest that most of our participants seldom experienced that others discriminated against them, it seems that even a slight encounter of discrimination aggravated their negative feelings about themselves from having PD (i.e., felt stigma).
However, some participants had felt stigma without having any enacted stigma (n=13, 18.84%). This may suggest that the perception of PD as stigmatizing may be anchored in what people with PD accept to be the commonly held view of PD and its visible symptoms among the lay community. In a survey of public attitudes toward PD [8], more than 70% of the respondents reported somewhat or greatly worried about potential consequences of PD, such as physical disability, being dependent on others, being a burden, and not being able to socialize. As public attitudes shape the context in which people with PD live in, those negative attitudes toward PD are likely to become one source of felt stigma once people get diagnosed. Similarly, negative impressions of visible features of movement and communication difficulties, such as considering abnormal bodily movement and facial masking as physical disability and compromised mental health [10], may also constitute one source of felt stigma.

In this study, participants with PD who experienced higher stigma were likely to also have more depressive symptoms and poorer QOL across motor, emotional, and social domains. The findings should raise the awareness of stigma of PD in health care professions and general public. As stigma is inseparable from the socio-cultural environment, it is important to recognize the social meaning of PD and PD-related symptoms. Future work with
longitudinal data is needed to validate the temporal sequence of felt and enacted stigma for possible causal inference. In addition, examining moderating and mediating factors in the relationship between stigma and QOL may help develop intervention strategies. For example, would social resources, such as musical or exercise group classes mixed of people with and without PD, decrease experienced stigma of individuals with PD and negative attitudes towards PD in general public, and thus lead to better QOL?

This paper presents preliminary results from a subset of baseline data from the ongoing 3-year SocM-PD project [18]. Some limitations of this study should be noted. First, negative wordings in SSCI, UPDRS Part II, and PDQ-39 may have exaggerated the correlation coefficients among these measures [30]. Second, the results that felt stigma, rather than enacted stigma, explained a significant amount of variance of QOL may be partly attributable to the score distribution that felt stigma had a larger variance than enacted stigma. Third, sampling bias may exist because most of our participants had at least bachelor’s degree and were in relatively high socioeconomic status. Therefore, the results may not be generalizable to people with PD who have lower educational level or socioeconomic status. Our findings provide beginning evidence about one aspect of social life with PD, and should be verified with a larger and longitudinal dataset of the SocM-PD in the future.
5. Conclusions

This study provides preliminary evidence showing stigma, especially felt stigma, as a key determinant of QOL in PD. The results suggest the importance for health care practitioners and general public to be aware of stigma issues related to PD and its symptoms. The results of this study should be verified with a larger and longitudinal dataset of the SocM-PD with on-going data collection. Future work is needed to identify moderating and mediating factors in the relationship between stigma and QOL to develop intervention strategies.
Conflict of interest

The authors declare no conflict of interest.

Acknowledgments

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