



Characterizing Quality of Life in Caregivers of People with Parkinson's Disease and Dysphagia

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Abstract

Caring for a family member with dysphagia can negatively impact caregiver wellbeing, although little is known about how dysphagia severity or specific symptoms influence this. The purpose of this study was to examine how objective measures of dysphagia in people with Parkinson's disease influenced their caregivers' quality of life. Fifty caregivers (mainly spouses) of people with Parkinson's disease completed a caregiver quality of life survey. Results were compared to medical chart reviews, interviews, and instrumental evaluations of swallowing from the care recipients. Outcomes included caregiver quality of life score, ratings of airway invasion and pharyngeal residue, and Parkinson's disease duration. Descriptive and regression analyses were completed. All caregivers reported reduced quality of life, with 28% having severely disturbed adaptation. Every care recipient with Parkinson's disease demonstrated airway invasion and/or pharyngeal residue. Together, the combination of older care recipient age and longer disease duration was associated with poorer caregiver quality of life [adj. $R^2 = 0.10-0.12$, $p = 0.03-0.4$]. Neither airway invasion nor pharyngeal residue was related to caregiver quality of life ($p > 0.05$). Findings confirmed that caregivers of people with Parkinson's disease and dysphagia experience reduced quality of life; however, current methods of assessing caregivers' quality of life may not adequately account for dysphagia-specific burden. Results highlight the urgent need for the development of dysphagia-specific assessments of caregivers' quality of life to facilitate identification of high-risk caregivers and aid the development of support systems to improve health outcomes for caregivers and care recipients.

Keywords Parkinson's disease · Caregiver burden · Caregiver quality of life · Dysphagia · Swallowing

Introduction

Caregiver burden has become a topic of great interest given the aging population and consequent changes to the healthcare landscape. With longer life expectancy comes a rise in chronic diseases and increasing demands on healthcare resources. In recent years, finding ways for patients with chronic diseases to live at home safely, while maintaining

quality of life (QOL), has become a healthcare priority. Ultimately, the responsibility of ensuring patient safety and wellbeing at home often falls to formal (i.e., paid and trained) or informal (i.e., unpaid family members or close friends) caregivers. In 2015, it was estimated that 17% of adults in the U.S.A. provide informal caregiving to another adult, with spouses/partners comprising 11% of informal caregivers [1]. Unfortunately, while it is well documented that patients benefit from receiving care in the home [2–5], the burden placed on informal caregivers is often high [6].

Quality of life, as defined by the World Health Organization, encompasses “individuals' perceptions of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards and concerns” ([7], p. 11). Definitions of caregiver QOL are scarce in the literature, although a relationship seems to exist between caregiver QOL and caregiver burden, which is directly affected by factors such as perceived social support, availability of

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rest, and relationship to the care recipient [8]. However, caregiver QOL can be maintained even in the presence of substantial burden [9]; thus, it may be more helpful to consider caregiver QOL as the *outcome* of the caregiving process [8, 9] rather than an appraisal of the caregiving situation.

The majority of theoretical models developed to explain the stress of caregiving have been based on Transactional Stress Theory [10]. More specifically, the model which has had the most influence on the theoretical understanding of QOL in caregivers has been the Pearlin Stress Process Model [11–14]. Pearlin et al. describe the dynamic changes to relationships between the caregiver and care-receiver over time that can translate into burden, physical, and psychological reactions in some caregivers. Briefly, background and contextual factors (e.g., socioeconomic characteristics, the relationship dynamic, access to resources) may directly influence the caregiver's adaptation to the stress involved in caregiving. The authors describe various types of primary stressors (i.e., specific conditions/experiences/activities that are problematic and fatiguing) which can proliferate in other areas of the caregiver's life (e.g., finances, work, family dynamics), leading to secondary stress. For example, caregivers who work outside of the home may experience pressure both from the demands of their job as well as the demands of providing care within the home. The caregiver's background, contextual factors (e.g., socioeconomic characteristics, the relationship dynamic, access to resources), and self-assessment of their response to the situation (e.g., diminished feelings of self-esteem or competency) also directly influence their adaptation to the stress involved in caregiving. For example, reductions in household income, increased expenditures, family grievances, or differences in opinion regarding care may all have causal associations to increased caregiver stress. According to this model, caregiver QOL is not static, rather, it is a dynamic mix of circumstances, experiences, reactions, and resources that vary not only between caregivers, but also in their impact on caregivers' health. However, the resulting stress can be mitigated by various mediating factors, such as coping strategies and social support.

Most people with neurodegenerative disorders, such as Parkinson's disease, are cared for at home, with spouses as the main informal caregiver [15]. Caring for a spouse or family member with neurologic disease has been recognized to negatively impact the caregiver's physical, mental, financial, marital, and psychosocial wellbeing, with an increased risk of persistent distress, ill-health, and depression [16, 17]. Family members of people with Parkinson's disease (PWPD) report poorer QOL that is associated with factors such as the care recipient's cognitive impairment, health status, ability to participate in activities of daily life, and disease progression, among others [17].

In the initial stages of Parkinson's disease, dysphagia (impaired swallowing) may be present, but is usually not severe [18]. Signs include hypokinetic (slow) and/or variable lingual and palatal movements, leading to symptoms of impaired bolus preparation and propulsion, reflux, and esophageal hypomobility [18–21]. With disease advancement comes worsening swallowing deficits including prolonged mastication, tongue 'pumping' (repetitive back-and-forth movements of the tongue), difficulties manipulating and clearing the bolus, and difficulties initiating swallowing, leading to increased mealtime duration [20, 22, 23]. Diet modification is also often necessary. In addition, reduced swallowing frequency (leading to sialorrhea/excess saliva [24]), and coughing or choking due to the aspiration of oropharyngeal contents [25, 26] are common. The presence of tremor can also make it difficult for PWPD to use utensils and self-feed [27]. The consequences of dysphagia symptoms for caregivers of PWPD are substantial and include extra or separate preparation of food to make swallowing easier for the person with Parkinson's disease; longer meal-times; avoiding eating out in public or dining with visitors; substantial cleaning following meals; feelings of guilt; and anxiety around choking or the inability to sustain adequate nutrition and hydration [27]. Despite the high number of stressors which can potentially result in burden for these caregivers, the specific impact of dysphagia on caregiver QOL in PWPD has not been studied.

While not specific to caregivers of PWPD, there is an emerging body of literature examining caregiver burden and QOL in people with dysphagia. Caregivers of people with motor neuron disease [28], dementia [29], stroke [30], head and neck cancer [31, 32], and community-dwelling older adults [6, 33], consistently identify the presence of dysphagia and concerns about their loved ones' swallowing as a source of major stress and burden. Caregivers report fear and stress related to making significant adaptations to accommodate meal requirements, finding the 'right' food or menu item, and the possibility of their loved one choking or becoming malnourished [31]. These fears and stressors are compounded by disruptions to family life when the family cannot eat together, go out to eat, or eat their favorite foods [31, 32]. It is unsurprising then, that dysphagia has been associated with social withdrawal, depression, and reduced QOL for caregivers of people with dysphagia [6, 27–32]. Limitations of previous work in this area have included: potential sampling bias (i.e., by relying on patient self-report to identify dysphagia) and treating dysphagia as a binary (i.e., present/absent) variable, limiting our understanding of the potential effects of dysphagia severity, specific dysphagia symptoms, or progression of swallowing decline on caregiver QOL.

Supporting caregivers and maximizing their QOL has become an important issue, since poor caregivers' QOL

may negatively impact care recipients' health and QOL [34, 35]. Therefore, better understanding the impact of dysphagia severity and disease-specific factors on caregiver QOL in Parkinson's disease is of utmost importance. This information will be vital for clinicians to use in order to facilitate the earlier identification of caregivers at risk of reduced QOL and ultimately design and implement appropriate support systems to improve caregiver QOL. Therefore, the purpose of this study was to examine how objective (i.e., instrumental) measures of dysphagia in PWPDP influenced their caregivers' QOL. We hypothesized, based on the Pearlin Stress Process Model [11–14] and previous work in heterogeneous populations [6, 28–33] that the severity of swallowing outcomes (i.e., airway invasion, residue) would significantly influence caregiver QOL, when controlling for disease severity and age.

Methods

In this retrospective, cross-sectional study, data were extracted from a clinical research database of people with neurogenic disease from a metropolitan area dating from 2016 to 2020. All procedures were carried out in accordance with the Declaration of Helsinki, and ethical approval was granted by the Local Institutional Review Board.

Subjects

PWPDP and their primary informal (i.e., unpaid) caregivers were selected for this study. Eligibility criteria were (1) a diagnosis of idiopathic Parkinson's disease by a movement disorders neurologist; (2) no evidence of other neurologic comorbidities (e.g., stroke, traumatic brain injury); (3) no history of head or neck cancer; and (4) having a caregiver who completed the caregiver burden survey within 2 months of the care recipient's instrumental swallowing evaluation. Participants who did not undergo instrumental evaluation of swallowing were excluded from the analysis.

Equipment and Materials

Flexible endoscopic evaluations of swallowing (FEES) were used to quantify swallowing outcomes. FEES equipment consisted of a 3.0-mm diameter flexible distal chip laryngoscope (ENT-5000, Cogentix Medical, U.S.A.) and a video system with integrated LED light source LCD display (DPU-7000A, Cogentix Medical). Equipment was routinely tested, and all recordings were archived in duplicate.

Participants with Parkinson's disease were presented with a variety of foods and liquids, dyed to maximize visualization of residue during the FEES [36]. Liquids included three 90 cc trials of filtered tap water (International Dysphagia

Diet Standardisation Initiative, IDDSI [37] level 0) in a 6 oz cup. To achieve blue, green, and white coloring, six drops (~0.2 cc) of blue dye (Chef-O-Van Food Coloring, Rockford, Ohio, U.S.A.), green dye (Chef-O-Van Food Coloring), or three teaspoons (~24 g) of barium powder (E-Z-PAQUE barium sulfate for suspension, 96% w/w; E-Z-EM Canada, Inc., Anjou, Canada) were added to each cup. Three 1 oz medicine cups containing 10 cc thin liquids, as well as one 5 cc and one 20 cc thin liquid were also presented. For solid food trials, three drops (~0.1 cc) of blue or green dye were added to pre-prepared pineapple pieces with juice (Dole Food Company, California, U.S.A.) to achieve a dyed mixed consistency (IDDSI level 7) food. Two pieces of dyed pineapple and ~3 cc juice were extracted onto teaspoons. Two teaspoons of vanilla pudding (IDDSI level 4; Hunt's Food Company, California, U.S.A.) were extracted and left undyed, as well as a Sunshine Crispy Original Saltine cracker (IDDSI level 7; Sunshine Biscuits, Illinois, U.S.A.). All participants with Parkinson's disease were in the 'on' phase of their medication cycle at the time of testing.

Procedure

FEES were completed by a speech-language pathologist with expertise in FEES, without the use of anesthetic or vasoconstrictors. The endoscope was passed transnasally and positioned within the oropharynx such that the pharynx, larynx, and subglottis were in view. Following each swallow, the endoscope was advanced into the laryngeal vestibule to visualize residue patterns within the larynx and subglottis.

The following standard protocol of liquid and solid bolus presentations was followed for all participants: 5 cc liquid (cued swallow), 20 cc liquid (cued swallow), three 10 cc liquids, three 90 cc thin liquid sequential swallowing challenges, two pudding swallows, two mixed-consistency swallows, and a Saltine cracker challenge. If necessary, modifications to the protocol were made for participant safety—for example, if gross aspiration was observed on a large volume bolus that the participant was not able to eject sufficiently, the same large volume was not administered further. If compensatory strategies were used (e.g., chin tuck), these swallows were omitted from data analysis. The order of blue, green, and barium liquids was random, and boluses were self-administered.

Caregiver QOL was measured using an online version of the Scale of Quality of Life of Caregivers, SQLC [38]. This 16-item scale evaluates three important domains of caregiver burden, namely, the ability to undertake professional activities, social and leisure activities, and responsibilities. Items are scored to provide a measure of psychosocial adaptation of the caregiver, ranging from full adaptation (141–145 points), mildly disturbed adaptation (100–140 points), moderately disturbed adaptation (86–99

points), and severely disturbed adaptation (< 85 points). Caregivers were provided with a secure link using the Research Electronic Data Capture (REDCap) [39, 40], from which they could complete the survey at their leisure. If forms were not completed prior to the patient's FEES appointment, the caregiver was provided with a paper copy to complete upon arrival, and scores were manually entered into REDCap. Items were automatically scored using REDCap.

Outcome Measurement

Demographic information was collected and recorded from participant interviews and medical chart review. Data collected from medical charts included age, disease duration (defined as the number of years since diagnosis), and disease severity [measured by the Movement Disorders Society Unified Parkinson's Disease Rating Scale (MDS-UPDRS) criteria]. Data regarding diet [measured by the International Dysphagia Diet Standardization Initiative (IDDSI) criteria] were collected during participant interviews and involved selecting the score that was most reflective of participants' typical diet.

FEES recordings were de-identified and stored digitally for off-line analysis. All recordings were initially rated by two blinded, independent raters who were trained in FEES analysis. Each video segment was examined for the presence of bolus residue in the valleculae and pyriform sinuses after the final clearing swallow. A visual-analog scale (VAS) was used to estimate the percentage of the structure/space on which residue was present, ranging from 0 to 100% [41]. The average percent residue across the two residue locations (i.e., valleculae and pyriform sinuses) was then calculated for a total pharyngeal residue score and carried forward for statistical analyses.

Raters also measured airway invasion using the Penetration-Aspiration Scale, PAS [42]. The worst PAS score across the FEES protocol was selected for statistical analyses. Following FEES rating, scores from the two raters were compared. If the raters' VAS ratings were in disagreement by > 10%, or if the difference in ratings was between the presence/absence of residue (i.e., VAS rating ≥ 1 versus VAS rating = 0), a senior, blinded rater was used to determine the final residue rating. Similarly, if raters' PAS scores were in disagreement, the third rater determined the final PAS score. Initial agreement between the two raters was measured by dividing the number of discrepant trials by the total number of trials rated for each anatomic landmark.

Statistical Analysis

IBM SPSS Statistics Version 26 (IBM Corporation, Armonk, New York, USA) was used to analyze the data. Disease severity ratings, often measured by the MDS-UPDRS criteria, were only available for 20/50 participants, due to differences in reporting practices across local neurologists. For this reason, disease duration was used as a proxy measure of disease severity for all participants, given that the two are closely related [43].

In addition to the amount of pharyngeal residue, we hypothesized that bolus properties—namely, whether the bolus was a liquid or a solid—may have influenced dysphagia presentation and thus, caregiver QOL. For this reason, the relationship between dysphagia outcomes and caregiver QOL was analyzed separately for liquids and solids.

Descriptive statistics were used to characterize patterns of residue and airway invasion in participants with PD. To characterize caregiver QOL, raw scores were grouped by severity level using the cut-offs provided in the SQLC [38]. No caregivers scored within the range of normal QOL and few caregivers scored within the range of moderately impaired QOL. Thus, the scores of moderate and severe QOL impairment were combined, leaving two groups: mildly impaired, and moderate-severely impaired QOL.

Initially, hierarchical multiple regression was used to determine if the addition of swallowing-specific outcomes (i.e., pharyngeal residue score, airway invasion) improved the prediction of caregiver QOL over and above age and disease duration. In these models, SQLC scores were treated as a continuous variable, and PAS scores were grouped by depth of airway invasion (i.e., none, PAS 1; penetration, PAS 2–5; aspiration, PAS 6–8) and treated as a categorical variable. Separate models were run for solids and liquids. Age and disease duration were entered into the first block, and pharyngeal residue and PAS scores were entered into the second block. Statistical significance was set at $p \leq 0.05$ (two tailed).

Next, binomial logistic regression was used to determine if there were differences in pharyngeal residue and PAS scores between caregivers with mildly impaired versus moderate-severely impaired QOL, controlling for age and disease duration. In these models, SQLC scores were treated as categorical (i.e., mildly impaired, and moderate-severely impaired QOL), as were PAS scores (described above). Separate models were run for solids and liquids. Age and disease duration were entered into the first block, and pharyngeal residue and PAS scores were entered into the second block. Statistical significance was set at $p \leq 0.05$ (two-tailed).

Results

The total sample was composed of 50 patient-caregiver dyads. Demographic information for the participants with Parkinson's disease is listed in Table 1. The majority of caregivers (47/50, 94%) were spouses/partners; the remaining primary caregivers were adult children 4% (2/50) or a close friend (1/50, 2%).

The majority of participants received all of the liquid and solid trials described in the FEES procedures above. However, 8 participants did not participate in any 90 cc thin liquid trials due to increased risk for gross aspiration, 4 participants refused the pineapple and/or cracker, 1 participant brought in their own food, and 13 participants did not participate in any solid food trials due to their involvement in another research protocol. Initial inter-rater agreement (prior to expert rater review) was 77.62% for pharyngeal residue ratings and 72% for PAS ratings.

Characteristics of Caregivers Experiencing Reduced Quality of Life

All caregivers reported experiencing reduced QOL. The majority of caregivers were characterized as having severely disturbed adaptation (28%) or mildly disturbed adaptation (66%) (Fig. 1). Nearly one quarter (12/50, 24%) of caregivers indicated that caring for the PWPd made them feel depressed. The main activities of daily life that caregivers reported as having less or no time for included: leisure activities (15/50, 30%), helping out other relatives (13/50,

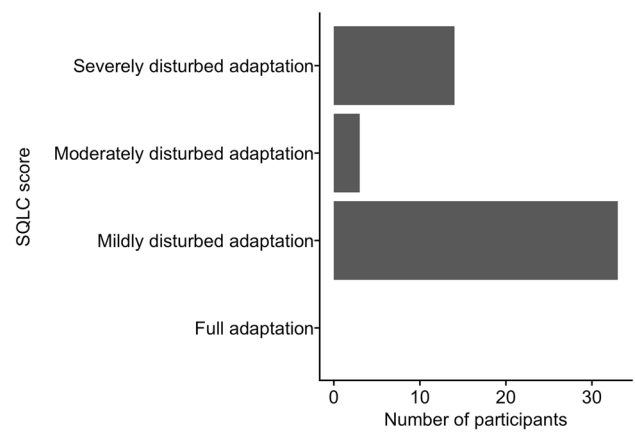


Fig. 1 Distribution of Scale of Quality of Life of Caregivers (SQLC) scores

26%), and household responsibilities (e.g., cooking; 10/50, 20%). The most concern expressed by caregivers was related to transportation, with 21/50 (42%) caregivers reporting that assistance was required for the care recipient to drive or use public transport, and 19/50 (38%) reporting that assistance was required for the care recipient to go for a walk/go shopping. Relating to meals, 9/50 (18%) caregivers expressed concern about the care recipient being able to warm up food that was left for them, and nearly one quarter (11/50, 22%) expressed concern about the care recipient being able to take food that was left out for them.

Patterns of Airway Invasion and Residue in Care Recipients with Parkinson's Disease

Every PWPd had objective symptoms of dysphagia, including airway invasion and/or pharyngeal residue. The frequency of PAS scores is listed in Table 2. For solids, 24/40 participants with PD (60%) had no airway invasion (PAS 1), 11/40 (28%) had penetration (PAS 2–5), and 5/40 (13%)

Table 1 Demographic information from participants with Parkinson's disease

Age (years), mean (SD), range	71 (7), 49–87
Sex	11 F, 38 M
Disease duration (years), mean (SD), range	9 (7), 1–31
MDS-UPDRS score, mean (SD), range	27 (11), 8–54
IDDSI food: frequency (percent)	
Regular/easy-to-chew diet	44 (88%)
Soft and bite-sized diet	3 (6%)
Minced and moist diet	2 (4%)
Pureed diet	1 (2%)
Liquidized diet	0 (0%)
IDDSI drink: frequency (percent)	
Thin liquids	47 (94%)
Mildly thick liquids	0 (0%)
Moderately thick liquids	2 (4%)
Extremely thick liquids	1 (2%)

SD standard deviation, F female, M male, MDS-UPDRS Movement Disorders Society Unified Parkinson's Disease Rating Scale, IDDSI International Dysphagia Diet Standardization Initiative

Table 2 Frequency of highest Penetration-Aspiration Scale scores for liquids and solids among participants with Parkinson's disease during instrumental evaluation of swallowing

Highest PAS score	Frequency	
	Liquids	Solids
1	11	24
2	0	3
3	11	3
4	2	1
5	13	4
6	1	0
7	4	4
8	8	1

PAS Penetration-Aspiration Scale

had aspiration (PAS 6–8). For liquids, 11/50 (22%) had no airway invasion, 26/50 (52%) had penetration, and 13/50 (26%) had aspiration.

All participants with PD demonstrated pharyngeal residue. The mean pharyngeal residue score for solids was 14.74% (1.50–48.50%), with a standard deviation of 12.06%. The mean pharyngeal residue score for liquids was 16.06% (6.00–43.00%), with a standard deviation of 8.61%. Additional information regarding the specific bolus texture/amount that resulted in the highest PAS, vallecular residue, and pyriform sinus residue scores can be found in the Supplementary Material.

Relationship Between Dysphagia Symptoms and Caregiver Quality of Life

Solids

Results from hierarchical regression analyses revealed that the initial model containing age and disease duration significantly influenced caregiver QOL [$F(2, 36) = 3.55$, adj. $R^2 = 0.12$, $p = 0.04$], with the combination of older care recipient age and longer disease duration associated with increased SQLC scores (i.e., poorer QOL). However, neither age nor disease duration alone was independently predictive of caregiver QOL ($p > 0.05$). Neither the addition of PAS scores (airway invasion) nor pharyngeal residue scores to the model significantly improved the ability to predict caregiver QOL ($p > 0.05$, Figs. 2, 3).

The initial logistic regression model predicting the severity of caregiver QOL impairment (i.e., mild versus moderate severe) using age and disease duration was significant [$\chi^2(2) = 8.95$, $p = 0.01$]. The model explained

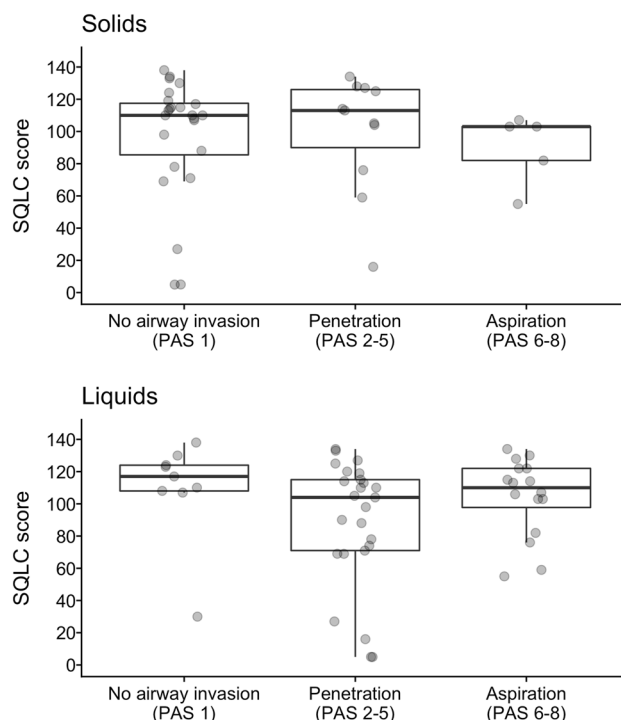
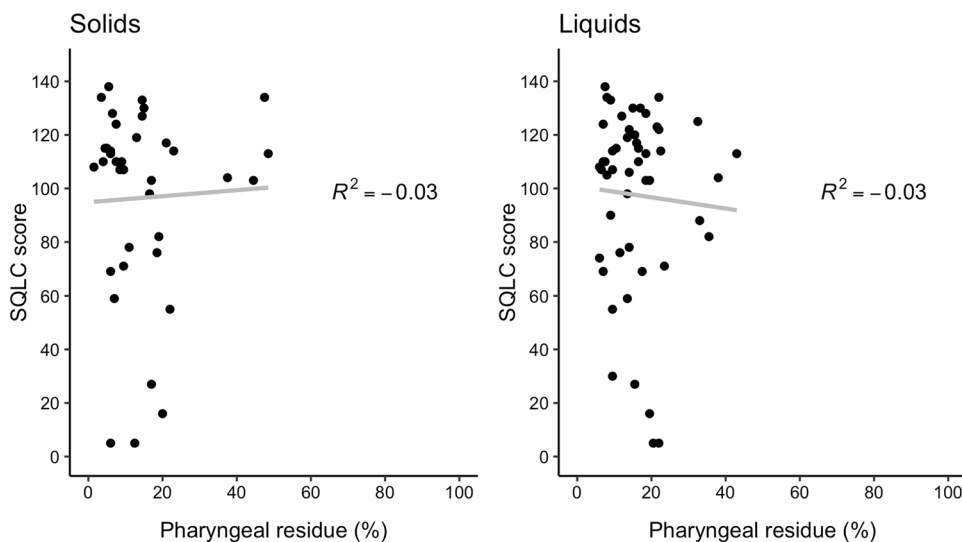


Fig. 3 Comparison of Scale of Quality of Life of Caregivers (SQLC) scores with patterns of airway invasion (measured by the Penetration Aspiration Scale score) observed during instrumental evaluations of swallowing for solids and liquids. PAS scores of 1 indicate no airway invasion, PAS scores of 2–5 indicate penetration and scores of 6–8 indicate aspiration. SQLC scores of 141–145 represent full adaptation, scores of 100–140 represent mildly disturbed adaptation, scores of 86–99 represent moderately disturbed adaptation, and scores < 85 represent severely disturbed adaptation

30.0% (Nagelkerke R^2) of the variance in QOL impairment and correctly classified 75.7% of cases. However, only disease duration emerged as a significant predictor of severity

Fig. 2 Comparison of Scale of Quality of Life of Caregivers (SQLC) scores with percentage pharyngeal residue observed during instrumental evaluations of swallowing solids and liquids. SQLC scores of 141–145 represent full adaptation, scores of 100–140 represent mildly disturbed adaptation, scores of 86–99 represent moderately disturbed adaptation, and scores < 85 represent severely disturbed adaptation. Adjusted R^2 is provided



of QOL impairment [$B(SE) = 0.17 (0.08)$, $p = 0.03$] with the odds of moderate-severely impaired QOL increasing by 1.19 [95% CI = 1.01–1.39] for every additional year of PD. Neither the addition of PAS scores (airway invasion) nor pharyngeal residue scores to the model significantly improved the ability to predict QOL impairment ($p > 0.05$).

Liquids

Results from hierarchical regression analyses revealed that the initial model containing age and disease duration significantly influenced caregiver QOL [$F(2, 49) = 3.73$, adj. $R^2 = 0.10$, $p = 0.03$], with the combination of older care recipient age and longer disease duration associated with increased SQLC scores (i.e., poorer QOL). However, neither age nor disease duration alone was independently predictive of caregiver QOL ($p > 0.05$). Neither the addition of PAS scores (airway invasion) nor pharyngeal residue scores to the model significantly improved the ability to predict caregiver QOL ($p > 0.05$, Figs. 2, 3). However, with the exception of one participant, all of the ratings of moderately or severely disturbed adaptation were from caregivers whose care recipient demonstrated airway invasion (Fig. 3).

The initial logistic regression model predicting the severity of caregiver QOL impairment using age and disease duration was significant [$\chi^2(2) = 6.77$, $p = 0.03$]. The model explained 17.5% (Nagelkerke R^2) of the variance in QOL impairment and correctly classified 74.0% of cases. Again, only disease duration emerged as a significant predictor of severity of QOL impairment [$B(SE) = 0.13 (0.06)$, $p = 0.04$] with the odds of moderate-severely impaired QOL increasing by 1.14 [95% CI = 1.01–1.28] for every additional year of PD. Neither the addition of PAS scores (airway invasion) nor pharyngeal residue scores to the model significantly improved the ability to predict QOL impairment ($p > 0.05$).

Discussion

Informal caregivers can experience reduced QOL, leading to negative health consequences for both the caregiver and care recipient [34, 35]. The presence of dysphagia in care recipients is suggested to contribute to reduced caregiver QOL [6, 28–32]. In the present study, we sought to objectively quantify swallowing outcomes—including airway invasion and post-swallow pharyngeal residue—and measure the association with caregiver QOL in Parkinson's disease. Although findings confirmed that caregivers of PWPDP are likely to report reduced QOL, results did not support a relationship between caregiver QOL and severity of swallowing outcomes in care recipients.

The main finding of reduced caregiver QOL is consistent with the literature in Parkinson's disease [15–17, 44].

Increased caregiver burden is generally expected in the later stages of the disease, when both motor and non-motor symptoms are greater and there is increased dependence on caregivers. This finding is also consistent with the literature in dysphagia [6, 27–32], where the presence of swallowing difficulties has been associated with increased burden in caregivers of patients with a wide range of peripheral and/or neurological disease. This can be interpreted in the context of the Pearlin Stress Process Model [11–14], where the management of dysphagia is a primary stressor with the potential to influence the caregiver's life in terms of finances (e.g., costs of thickened liquids, associated medical appointments), work (e.g., flexibility in hours and location to allow the caregiver to be at home), and/or family dynamics (e.g., providing assistance during mealtimes), leading to secondary stress, such as strain in maintaining roles at work or at home. The caregiver's reaction to the consequences of dysphagia may contribute to their overall QOL.

Contrary to our original hypothesis, dysphagia severity (defined in terms of extent/response to airway invasion and amount/location of pharyngeal residue) did not influence ratings of caregiver QOL. There are several possible explanations for this finding. First, the tool used to measure QOL in caregivers—the SQLC—does not contain specific questions relating to swallowing and may not have been sensitive to detect dysphagia-related reductions in caregiver QOL. The SQLC is a validated tool with satisfactory internal consistency, as well as convergent validity with disease duration and stage, mental status, and motor functioning [45]. However, a dysphagia-specific tool that takes into account factors such as social functioning, time, and assistance required for mealtimes, financial and emotional costs of dysphagia, fear of watching your loved one cough or choke with meals, and other related factors, may be more sensitive at measuring dysphagia-related changes to caregiver QOL. Currently, there is no validated tool available that measures dysphagia-related QOL in caregivers of people with dysphagia, although work is underway to address this [46].

Second, our sample was skewed towards participants with a relatively shorter disease duration (52% had Parkinson's disease for 6 years or less). It is possible that including more participants with a longer disease duration may have revealed new relationships between caregiver QOL and dysphagia severity; this is a suggested direction for future research. Nevertheless, it is noteworthy that 100% of caregivers in the present study reported at least a mild level of reduced adaptation/QOL, with more than one quarter identifying themselves as having severely reduced adaptation/QOL. This finding suggests that factors other than dysphagia may have a greater influence on caregiver QOL in the early stages of Parkinson's disease. Previous work has identified that symptoms of Parkinson's disease such as falls, depression, confusion, and hallucinations

are all independent predictors of caregiver QOL in the early stages of Parkinson's disease [17]. Comparatively, symptoms of dysphagia can be subtle or even silent, and the consequences of dysphagia—such as weight loss and pneumonia—can take weeks to manifest. It is, therefore, understandable that gross motor and/or psychiatric symptoms may represent a greater threat to caregiver QOL than dysphagia in the initial stages of the disease. Further research is required to understand whether this dynamic changes with disease progression. It is also possible that the presence of multiple factors influencing caregiver QOL may mask the specific effect of dysphagia.

A third possible explanation for our findings is that the caregivers in our study were unaware or unaffected by their care recipient's experience of dysphagia symptoms. PWPDP may underestimate symptoms of dysphagia due to cognitive deficits and/or sensory changes affecting the upper airway [47–49], making them less likely to report changes to swallowing. Indeed, there is a high prevalence of silent aspiration in PWPDP, and self-report has been shown to be an unreliable indicator of swallowing ability [27, 50]. As a result, a person with a PAS score of 8 (i.e., profound pharyngeal dysphagia and silent aspiration) could report their symptoms in a similar way as a person with a PAS score of 3 (i.e., mild dysphagia and no aspiration). If this is the case, dysphagia-related caregiver burden might be more closely related to patient symptom report than dysphagia severity. Alternatively, dysphagia may be a largely personal and private experience that, even when reported, has little effect on a caregiver's QOL. This explanation seems less likely when taken in the context of previous work that has identified increased caregiver burden and stress when the care recipient has dysphagia [6, 27–33], although it should be noted that previous work in this area relied on self-report to diagnose dysphagia, thus, potentially excluding patients with low symptom-reporting accuracy. The role of under- or over-reporting of dysphagia symptoms on caregiver QOL is a suggested direction for future research.

Despite the fact that caregiver QOL was not specifically related to dysphagia severity in this study, it was the case that all caregivers reported reduced QOL and all care recipients had dysphagia. Although the ability to measure dysphagia-specific caregiver QOL is limited by the tools that are currently available, there are steps that clinicians can take to begin to address caregiver QOL when working with PWPDP and their families. Clinicians are encouraged to measure caregiver QOL informally, by probing for potential areas where QOL may be reduced. From there, caregivers can be supported with appropriate resources. When coping strategies and social supports are in place, the burden associated with caring for a person with dysphagia may be somewhat alleviated [15]. Additionally, although the presence of dysphagia may decrease QOL for some caregivers, there

can also be benefits to caregiving, such as satisfaction from feeling needed, or from acquiring new knowledge or skills [51], and clinicians can play a role in facilitating this development. Indeed, as part of a multidimensional approach to treatment, Namasivayam-MacDonald and Shune [6] suggest that, during care planning, caregivers should be considered as patients in their own right.

This study was not without limitations. First, as discussed above, the SQLC may not be sensitive to dysphagia-specific caregiver QOL, limiting the extent to which relationships between features of dysphagia in care recipients and caregiver QOL could be drawn. However, at the time of data collection, there was no validated tool that measured dysphagia-related caregiver QOL. In this context, the descriptive findings from the present study may offer the most valid insights into the influence of dysphagia on caregiver QOL. Second, our sample was relatively homogenous, i.e., majority male, married, living independently, relatively short disease duration, and on regular diets. This is not an uncommon demographic in Parkinson's disease, and, given that reduced caregiver QOL was identified in 100% of caregivers in the sample, we are confident that our findings remain valid. However, specifically analyzing the effects of variables such as diet restrictiveness, caregiver sex, and caregiver-care recipient relationship may reveal new relationships to caregiver QOL and is a suggested line for future research. Third, airway invasion was quantified using the worst PAS score, which carries the risk of over-characterizing an individual's airway protective function as impaired. However, extracting the worst PAS score is the most common approach that the authors have identified in the literature, and, in the case of the present study, has the benefit of capturing impairment across a range of assessment protocols. Finally, it is worth pointing out that the majority of evidence regarding caregiver QOL in PWPDP, including the present study, comes from cross-sectional research, which limits the ability to determine causal relationships between predictor variables and caregiver QOL. Future research would benefit from exploring the relationships between dysphagia severity and caregiver QOL in a longitudinal fashion, with emphasis on objective measurement of swallowing outcomes and a validated tool for swallowing-related QOL in caregivers.

Results of this study confirm that caregivers of PWPDP and dysphagia experience reduced QOL and highlight the need for the development of a measurement tool specific to dysphagia-related QOL. Awareness of the factors contributing to QOL in caregivers of PWPDP and dysphagia is important for the development of family-centric interventions to relieve the effects of caregiving and support caregivers in their role. Protecting the health and wellbeing of caregivers promotes home-based care environments that are safe and sustainable. A less burdened caregiver

ultimately benefits PWPDP by improving health outcomes, maximizing QOL, and maintaining a high level of functioning at home.

Supplementary Information The online version contains supplementary material available at <https://doi.org/10.1007/s00455-021-10299-z>.

Declarations

Conflict of interest The authors confirm that they have no relevant conflicts of interest to disclose.

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