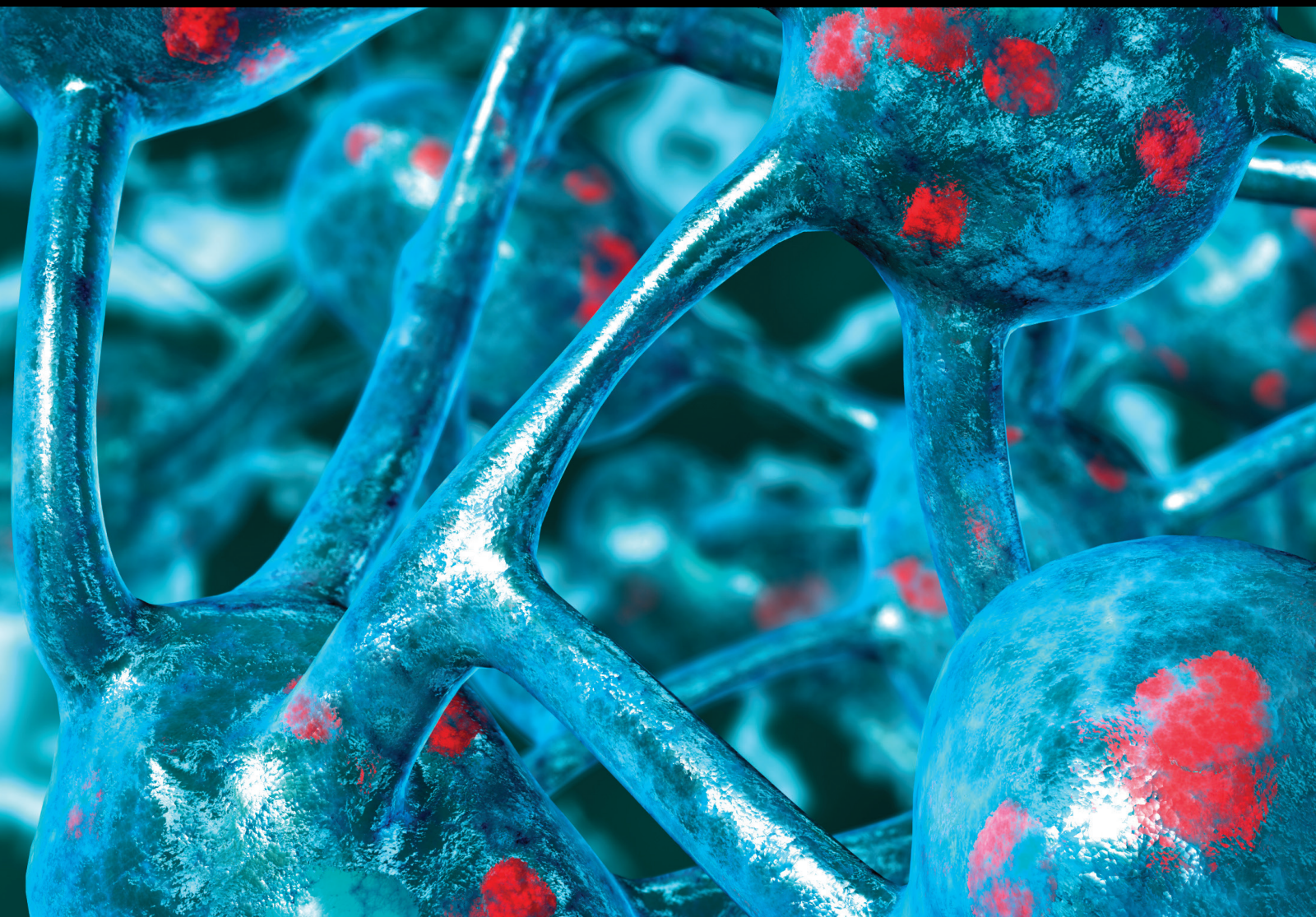# Integrated and Personalized Care Approaches, Levels of Support, and Social Inclusion in Parkinson's Disease 2021

Lead Guest Editor: Mari Carmen Portillo
Guest Editors: Line Kildal Bragstad, Anita Haahr, and María Victoria Navarta Sánchez

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Contents

Research Article

Exploring the Caregiver Role after Deep Brain Stimulation Surgery for Parkinson's Disease: A Qualitative Analysis

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This pilot study aimed to explore how caregiver spouses make sense of themselves one and five years after their partner's deep brain stimulation (DBS) surgery for Parkinson's disease. 16 spouse (8 husbands and 8 wives) caregivers were recruited for the interview. Eight struggled to reflect on their own lived experience and primarily focused on the impact of PD on their partners, such that their transcripts were no longer viable for interpretative phenomenological analysis (IPA). A content analysis showed (1) how these 8 caregivers shared less than half as many self-reflections than the other caregivers, (2) that there was a bias to reflect on their partner's experience answering the opening question, (3) the bias continued when answering subsequent questions, and (4) there was a lack of awareness of this bias. No other patterns of behaviour or themes were able to be extracted. The remaining 8 interviews were transcribed and analysed using IPA. This analysis discovered 3 inter-related themes: (1) DBS allows carers to question and shift the caregiver role, (2) Parkinson's unites and DBS divides, and (3) seeing myself and my needs, DBS enhances visibility. How these caregivers interacted with these themes depended on when their partners were operated. The results suggested that spouses maintained the role of caregiver one year post DBS because they struggle to identify themselves in any other way but were more comfortable reassociating into the role of spouse 5 years post surgery. Further inquiry into caregiver and patient identity roles post DBS is recommended as a means of supporting their psychosocial adjustment after surgery.

1. Introduction

Deep brain stimulation (DBS) is effective in controlling the motor symptoms of Parkinson's disease (PD) for 5 or more years [1, 2]. DBS has been described as creating a "biographical disruption" for the patient as it can change the way they interact in the world quite suddenly once PD symptoms are improved [3]. This biographical disruption can be associated with poor psychosocial adjustment post DBS [4] due to the sense of loss related to the care once received in comparison to the independence possible post DBS [5] and challenges around identity regarding the merging with technology [3, 6, 7].

The DBS journey is thought to be comprised of 4 stages [8]. The presurgery stage focuses on the decision-making process patients and their caregivers use to decide to opt for DBS surgery. The second stage focuses on the surgery itself and the physical support needed along with the clinical goal-setting. The third stage focuses on the changes that DBS creates in symptoms, behaviours, and roles. The fourth stage refers to the patient's reengagement with life as well as their perceptions of the future. These third and fourth stages are the most relevant with regards to how life is experienced 1 and 5 years post DBS. The impact of DBS surgery has yet to be explored with regards to shifts in caregiver role and identity over time after the surgery.

According to the "social identity theory," we make sense of ourselves in the context of the social groups we ascribe to, and how we engage in the behaviours we consider appropriate to those groups [9]. Over time, spouses, partners, and other family members who care for an ill relative, become

depended upon for care, practical, and emotional support, potentially emphasizing the social role of the caregiver [10, 11]. As PD progresses, it can become harder for caregivers to remain employed, socialise independently, and feel connected to others [12, 13]. This can reinforce the caregiver role as it becomes more challenging to engage in any other social role(s) [14].

The caregiver identity theory suggests that there are 5 phases which caregivers can transition through during their caregiving "career" [15]. The first two phases are comprised of the caregivers becoming more involved in care which extends beyond a family, or a friendship role, which starts the process of identifying with the role of caregiver. In phase three, carers tend to be more involved in such activities as personal grooming of the ill person. This can put both parties ill at ease, and the individual is most likely now to identify with the role of caregiver around fifty percent of the time. Caregivers enter phase four when their lives are heavily dominated by their caregiving role. Phase five is characterised by the reverting back to an earlier phase of caregiver identity, thanks to a change in circumstances. DBS may be one such circumstance.

Caring for a spouse can be physically, emotionally, socially, existentially, and financially demanding and can leave caregivers feeling isolated, invisible, and in-need [16]. The concept of caregiver burden is not new and has been evaluated in many studies of PD [17]. There are also countless studies that have examined the effect of caregiving on identity [18]. Many studies have focused on the identity change perceived by caregivers of their caree [19]. Other studies have looked at how these changes can be managed and facilitated (Haahr et al. [20]). Most studies tend to focus heavily on the caregiver's experience of the person with PD with whom they live and the way that has impacted their own daily life. Themes have emerged such as "A sense of freedom embracing life" (Haahr et al. [21]). This theme focuses on the shifts in the patient's physical state which have given caregivers more freedom. Restoration of the "old self" [22] gives insight into the return of the person with PD's functional abilities and mobility and how that affects their personality. Themes such as, "being different after DBS" [23], "clinical management of personality change" [22], and "the challenge of changes and constraint" (Haahr et al. [21]) all focus on the adjustments needed to be made by caregivers to manage the changes DBS has made to the person with PD. All of these studies have given us important insights into how caregivers experience the changes in their caree's physical, psychological, and emotional state. However, there has been little exploration of how caregivers perceive their own evolution as individuals and their caregiver role. Caregivers can feel a sense of loyalty towards the person they care [24], which can overshadow their own needs [16]. By maintaining so much focus on their caree, there is a lack of information regarding the caregiver's personal journey. This pilot study aimed to fill this gap in the literature by investigating how caregiver spouses make sense of themselves, as well as their caregiving role one and five years post DBS surgery.

2. Methods

2.1. Design. This study used semistructured interviews employing an interpretative phenomenological analysis (IPA) (Smith et al. [25]) approach to understand the lived experience of caregivers 1 year or 5 years after their spouse's DBS surgery to treat PD. IPA was the analysis of choice due to the double hermeneutic that is used in its approach. With each question posed, participants are invited to reflect and make sense of their lived experience. They are given the space to reflect about how they think and respond to the circumstances of their life. As they share these reflections, the researcher is then invited to do the same (Smith and Shinebourne [26]). A summative content analysis [27] was used for those interviews which did not meet the threshold of caregiver personal reflections needed for IPA, as it displayed the measurable differences between those caregivers who shared self-reflections and those who were more restrained.

2.2. Participants. IPA requires a relatively small and homogenous sample. An equal number of male and female spouses were recruited. Participants were heterosexual, British, or Irish, over the age of 50, married for at least 10 years, and living with their spouses who were diagnosed with PD at least 10 years prior to interview. Spouses with PD had bilateral DBS of the subthalamic nucleus (STN) and were either 1 or 5 years post DBS at the time of interview. All caregivers who met the recruitment criteria were contacted from the patient list of the Functional Neurosurgery Unit at the National Hospital of Neurology and Neurosurgery in London. However, to ensure a sample size of 16, the threshold of time post surgery was changed to 12–18 months and 4-5 years.

Caregivers who met the recruitment criteria were contacted by phone or e-mail and were invited for interview. 16 participants were interviewed in total; 8 participants were interviewed 12–18 months post their spouse's surgery, and 8 participants were 4-5 years post their spouse's surgery.

2.3. Rapport Building. In a study employing interview methodology, an essential element of data collection is rapport building. It has been noted that people who share similar experiences of illness can create a unique connection because they are able to give instrumental advice and emotional support as their empathy comes from a place of shared knowledge [28]. This has been researched in the realms of peer support, but the same principles can be true for qualitative research, and hence can help facilitate interviews.

For this reason, the interviewer disclosed to each participant that she had experience of caring for a spouse with PD who had undergone DBS surgery. This information was welcomed by the participants, and many reflected that it was comforting to speak to someone with similar lived experience. While this disclosure by the interviewer seemed to lift some barriers with regards to self-disclosure by the caregivers, it may have created other challenges as some participants seemed to expect the interviewer to understand

their experiences without them having to share details. This meant that in such cases, the interviewer had to make a greater effort to elicit the detailed responses required for IPA.

2.4. Procedure. Ethics approval was granted by the HRA and Health and Care Research Wales (REC Ref 18/LO/1368). Informed consent was obtained from all participants.

A semistructured interview schedule of 10 questions with prompts was devised, aimed at understanding the lived experience of caregiver spouses approximately 1 and 5 years post their spouse's DBS surgery. A pilot interview was held, and the interview schedule adjusted to make sure that the questions were clear enough for the participants to understand. A set of prompts were used to help guide the participants reflect on the questions.

An example of this is with the question, "How does DBS affect how you think about your partner who has PD?" The following prompts were used: "Has the surgery affected the way you view your partner? Has DBS affected how you care and your involvement in their care? How has that affected the way you feel about them? Has that been a difficult change?"

Interviews lasted around 60 minutes. Interviews were conducted by SS. 3 interviews took place in the participant's homes. The participants arranged to be alone at home to allow for maximum comfort with regards to disclosure. The remaining 13 interviews took place in the privacy of the hospital clinic. All interviews were recorded and transcribed verbatim. All names were changed to codes of which the first was the letter W or H depending on if they were a husband or wife, a number, and then Y1 or Y5 depending on when their spouse was operated. Any identifying information was removed to protect confidentiality.

2.5. Data Analysis

2.5.1. Interpretive Phenomenological Analysis. The first stage of analysis was conducted following the guidelines set out by Smith, Flowers, and Larkin (Smith et al. [25]). Each interview was analysed in isolation of the others to maintain objectivity regarding each participant's lived experience. Interviews were subjected to an inductive process which started by annotating transcripts with initial observations in the margin. These notes were then converted into "personal experiential statements," (PES's) in the other margin of the transcript.

Furthermore, analysis made it clear that the caregivers could be distinguished into two groups. The interviews of the first group had in excess of 50 PES's (group 1, $n = 8$) and were analysed using IPA. Those with less than 30 PES's (group 2, $n = 8$) did not provide enough data for an IPA analysis. Changing the style of analysis to fit the data was not an option as this lack of self-reflection shown by group 2 was worthy of note, and hence their interviews were analysed using a content analysis [29].

All of the PES's for group 1 were collated, and connections between the various statements were considered and clustered into groups. "Personal experiential theme's"

(PET's) emerged for each cluster. All of the PET's were then collated and clustered according to the connections that could be made between them, and 2-3 superordinate themes per transcript emerged. At each stage of analysis, checks were conducted by other members of the research team to make sure the first author was not projecting any of her own personal experiences on the data.

A table for each transcript's superordinate themes, with the relevant PET's, PES's, and supporting statements from interviews was created. Table 1 presents an abbreviated example taken from H3Y1's interview. The first superordinate theme that emerged from the interview was, "If she's well, I'm well." This theme emerged from the two PET's, "Living a combined life" and "The caring caree irrespective of DBS." 2 supporting PES's and keywords have been shown to exemplify the process.

These tables were used to compare across cases. All of the superordinate themes from the year 1 interviews were clustered together from which 3 themes emerged. The first focused on identity, the second on individuality, and the third on personal needs. When the year 5 superordinate themes were clustered, a similar pattern emerged allowing for the 3 main themes presented in Table 2, to emerge.

Tables were created, for each of these themes, with the supporting superordinate theme, PET and PES from each individual interview in one column, and supporting quotes in the adjacent column. These tables were split into two sections: 12–18 months post DBS (Group A) and 5 years post DBS (Group B).

2.5.2. Content Analysis. Considering that the focus of this study is on identity, the content analysis took place in two stages. The first stage of analysis was comprised of quantifying how much these caregivers shared their own life experience by counting PES's compared to the caregivers in group 1.

The second stage of the content analysis was aimed at looking for any other patterns in the data which could help us to make sense of these caregiver's experiences. Unfortunately, very few meaningful patterns emerged beyond the ways in which these participants answered the questions posed, due to a lack of data.

3. Results

Data analysis revealed that two groups of caregivers could be distinguished as follows:

> Group 1: those caregivers who had above 50 personal experiential statements during the interview
>
> Group 2: those caregivers who had below 30 personal experiential statements during the interview

3.1. Group 1: Caregivers Who Expressed Their Own Personal Experiences of Caregiving. Table 3 shows the three themes that emerged across post-DBS year 1 and year 5 interviews.

The focus of this paper is on how spousal caregivers redefine their self within their caregiver role post DBS. PD

TABLE 1: Breakdown of how superordinate theme 1 from husband 3 year 1's interview evolved.

Themes IPA interview table husband 3 year 1	Keywords taken from transcript
Superordinate theme 1: if she is well, I am well	
PET1: living a combined life	
	I do not think of myself as (a carer)
	It is just our life. . .
Prior to and post DBS, he feels the effects of PD so much he feels as if he has it too	We actually say, you know, we have got Parkinson's, if you like so it is more inclusive
His life is very entwined with hers, DBS makes their conjoined lives better now but he cannot stop their combined life from declining	It is like a combined life, is how I see it really
	We thought well this is not working very well so we will do it ourselves
PET 2: the caring carer irrespective of DBS	
He likes how well he cares for her and is proud that he maintained his relationship as a husband rather than carer prior to DBS	I have to help with all that. . .
	I do not always wait to be asked either, it is quite a sort of delicate. . .
He desperately wants to ease as much of her suffering as possible and fears that he is not able to post DBS	I worry sometimes, I am not being thoughtful enough, have missed something. . . anxious
	I: is that even now after DBS?
	H3Y1: generally, yeah

TABLE 2: Number of PES's per participant.

Group 1 participant	Number of PES's	Group 2 participant	Number of PES's
W2Y1	51	W1Y1	23
W4Y1	66	W3Y1	26
H1Y1	76	H2Y1	29
H3Y1	53	H4Y1	13
W1Y5	54	W2Y5	29
W4Y5	52	W3Y5	25
H3Y5	66	H1Y5	24
H4Y5	50	H2Y5	18

TABLE 3: Themes and subthemes by group.

Theme 1: identity	DBS allows carers to question and shift the caregiver role
Group A (12–18 months)	Am I more than just a caregiver?
Group B (years 5)	The carer identity has shifted
Theme 2: independence	Parkinson's unite and DBS helps divide in time
Group A	The combined self
Group B	DBS brings balance in relationships through acceptance
Theme 3: personal needs	Seeing myself and my needs, DBS enhances visibility
Group A	I am still invisible, DBS has not helped enough
Group B	Acceptance means it is easier to live for me

shifts the way caregiver spouses see themselves, their partners, and their roles within their marriages. DBS causes new shifts, forcing them once more to review all these aspects.

It has often been reported that people with chronic illness adopt a sick role and caregivers adopt a caregiver role. However, DBS has the potential to improve the physical symptoms of PD almost overnight which can alter how these two roles proceed to manifest following surgery. These interviews highlight elements of the journey the spouses interviewed had taken in their caregiving role post DBS surgery. Each of the themes that emerged will be presented in two sections: 12–18 months and 5 years post-DBS. Each group was comprised of 2 wives and 2 husbands. Comparisons between these groups will be focused on in the discussion.

3.1.1. 12–18 Months Post DBS. Theme 1: DBS allows carers to question and shift the caregiver role.

Am I more than just a caregiver?

All 4 of the spouses who were interviewed 12–18 months post DBS stopped working prior to surgery so that they could care for their partners, reinforcing the caregiver role. W2Y1 describes a newfound sense of freedom since her husband's surgery because,

"It gave me a bit more freedom, probably, so I didn't feel so bad about going out and doing things and I joined the gym and yeah, I joined the gym, and I, that's when I went back to teaching"

We note here the guilt W2Y1 alludes to having experienced prior to surgery, when she went out without her husband. It is not clear if there is an effect of gender, but in this study, there was a difference in how the carer husbands

reacted to their caregiving carees compared to the wives interviewed.

"I have worked since I was 16, in fact before that really, so suddenly I didn't have that role and then there was a possibility that I didn't have another role so that was a bit tricky, it soon became apparent that I at least had one of those roles." H1Y1

H1Y1 describes how he completely surrendered to his caregiving role. Everything he did was for his wife, freeing him from any guilt. For as much as it was a smooth transition from his working role to his caring role, it is the potential disruption to his caregiving role that he struggles with. We see from the previous quote, H1Y1 is so entrenched in his caregiver role, he struggles to imagine life without it now that he no longer works. The potential success of DBS would have left him feeling redundant and in search of a new way of seeing himself.

W4Y1 showed more resentment towards her situation than H1Y1 when she states:

"I don't think he does understand, he'll say to me I've got Parkinson's, yes he has but he, I don't think he realizes that actually it's not just him that's got Parkinson's and I could, I could have not stopped working, I could have carried on working."

W4Y1 is not only angry with the effects PD is having on her life, she is struggling because her husband seems "blind" to her suffering and the loss she has experienced by giving up work. When she says, *"it's not just him that's got Parkinson's."* She is identifying with her husband's illness because PD is affecting her life as much as it has affected his.

Theme 2: Parkinson's unites and DBS helps divide.

The combined self as follows:

W4Y1 has just shown us how PD seems to create a sense of merged self, whereby the caregiver starts identifying with their spouse's illness. The caregiver's life can be so governed by their partner's condition that all four caregivers identified with their partner's illness and sick role as well as their caregiver role. This identification with the PD role is well displayed in both husbands' narratives.

H3Y1 describes his lived state as a "combined self," suggesting that he too fails to see himself as an autonomous individual. He states, *"We actually say, you know, we've got Parkinson's, if you like, so it's more inclusive."*

Unlike W4Y1 who is identifying with the sick role because of her own inner pain, H3Y1 is identifying with his wife's PD out of solidarity. He wants us to know that he is not just supporting his wife on her journey, he is very much a part of her PD journey. PD tends to make the patient feel isolated, H3Y1 does not want his wife to feel alone in her suffering hence he states, "we have Parkinson's."

H1Y1 shares how he experiences this phenomenon, when describing his fears around DBS as follows:

"I do tend to see myself through (my wife's) lens really in the sense that I'm kind of basically about her, really, and I have been for a long time. I mean this is one of the things that people go, "oh you're a carer" and I am a carer and I do feel like I'm a carer."

To an extent, this explains H1Y1's relief that DBS did not give his wife full independence, since his caregiver role forms a major component of his current identity. He may not be as explicit as to state that he has PD, but caring for his wife seems to give him a sense of purpose. "I'm kind of basically about her" suggests that caring for her doesn't just make his life meaningful, it encompasses the full experience of his life, hence his reticence to relinquish this identity.

Caregiver spouses described how their lives were filled with many restrictions prior to DBS. They all described a reality where they were overcome by tending to the needs of their spouses, and hence their own needs became less visible. They were only able to focus on what PD allowed. DBS starts to shift this reality, in the first year, as it lessened the needs and demands of the PD patients on most of the spouses. We will see how this becomes more pronounced after 5 years.

Theme 3: Seeing myself and my needs, DBS enhances visibility.

I am still invisible, DBS has not helped enough.

When H1Y1 stated, *"I do tend to see myself through (my wife's) lens really in the sense that I'm kind of basically about her,"* we note that irrespective of DBS, all of his attention remains on his wife's needs.

DBS grants the PD patient improvement of their motor symptoms, and hence some independence has the potential to create a break in that combined self and allow caregivers the opportunity to tend to themselves but that means caregiver spouses need to become more self-aware.

Caregivers would need to acknowledge the separation that has occurred between them and their partners post surgery, allowing them to see themselves and behave as separate entities. One could expect the success of surgery to determine the extent of this separation, as the carers and their needs become more visible. However, this had not happened for any of the caregivers who were interviewed 1-year post surgery irrespective of the success of surgery. Whether it was seeing friends, having time alone or just recognising their own self, most of the spouses struggled to recognise or address their needs or individual state 1-year post DBS.

H3Y1's wife has had a good result with DBS. She is much more mobile and independent which should allow him the freedom to see himself as an autonomous partner, allowing him to recognise and tend more to his own needs and desires. When asked to reflect on the best part of DBS and how this had impacted on his life he responded:

"the best bit really is, is seeing her life get better, that's, and I know that's impacted, that's obviously good for me too but what I mean is, that's been the greatest pleasure to see that she can now do more, and the windows are not shutting quite as much as they were, on us, you know I wouldn't push off for a holiday or something and do something like that for myself if you like."

H3Y1 is so used to his "combined life," he still only ever reflects on his wife's experience. When he says *"that's obviously good for me too,"* it is evident that any improvements that DBS has made to his life seem secondary. Throughout the interview, he expresses how he has never felt the need to address his needs, he places hers above his, and even now he shirks away from any mention of him doing so when he states, *"I wouldn't push off for a holiday or something and do something like that for myself."* H3Y1 still focuses his attention on his wife and relies on her state to determine how he feels and functions in life.

Like H3Y1, W2Y1's husband has also had a good result from DBS, but she is more able to reflect on how it has impacted on how she experiences life.

"After the DBS we started on a different sort of level, right, and where I felt more, I felt more freedom. And I didn't feel that I had to be constantly checking on him."

We see here that unlike H3Y1, W2Y1 is aware of the improvement DBS has made to her husband and the impact it has made on her own life. She is no longer needed to constantly keep a watchful eye on him. On the contrary, she can set her sights on herself. However, she also stated:

"I was tired, and I just couldn't be bothered doing things and he was tired anyway with Parkinson's and he couldn't be bothered. We started to make excuses not to go out with our friends and we've always had a really, really busy social life. And, we've loads of friends and friends that we've had since school, and I just started to think, "I just don't have the energy anymore." And, that's sort of stayed with me I haven't really come back from that."

We see how different life was for W2Y1 prior to PD and DBS. She and her husband enjoyed socializing with friends until PD stopped them. The fact that they have maintained their childhood friends shows to what extent these connections are meaningful. However, even though DBS had given her the freedom to contemplate bringing socializing back into her life, she was struggling. For W2Y1, reengaging with the world was reliant on the energy she has not recovered and consequently she is unable to socialise. DBS has given her clarity on what is important, but it has not given her the strength to go after it.

All year 1 spouses failed to change the way in which they engaged with the world even though DBS changed their circumstances. The positive shifts that had occurred within their physical lived experience had not trickled down to their emotional life. Some still struggled to see themselves as anything other than care partners, while others did not have the strength to change their way of being.

3.1.2. 5 Years Post DBS. Theme 1: DBS allows carers to question and shift the caregiver role.

The carer identity has shifted.

All of the spouses in Group 1 who were interviewed 5 years post DBS, shared this ability to let go of the sense of responsibility to care for all of their partner's needs, allowing them to act like carers less and act like spouses more again. H4Y5 sought some support through a counsellor who helped him to recognise,

By allowing his wife to support him, he has brought balance back into their relationship. They no longer interact as caregiver and recipient, there is more of an equal exchange of care and support.

The 2 wives interviewed in this group did not reflect so much on how their husbands could now support them in the way H4Y5 did. They both discussed how they made more of an assertive effort to make this shift out of their caregiver roles by addressing the ways in which they interacted with their husbands.

W1Y5 remembers pre-DBS,

"It just put a strain because I became more of a carer, more than an equal and I obviously changed."

The strain of managing her husband changed how she saw herself. Now, 5 years since she says,

"I make a conscious effort not to help him sometimes and to sit back and let him get on with things rather than, you know, before his DBS I used to have to help him out of the chair and now if he's struggling a little bit I will just let him get on with it."

DBS has relieved that sense of responsibility she once felt. She now allows herself to allow him to be more independent and gives herself the freedom to sit back, while he tends to his own needs.

W4Y5 has also handed responsibility back to her husband; however, this has more of an emotional than a physical responsibility. She stated as follows:

"sometimes I feel like his carer, don't get me wrong, sometimes I feel like his carer and I do tell him, "I feel like your carer today rather than you wife," um and he sort of says, ok I'll do this and tell me what I need to do to make you feel like my wife. Cos sometimes it just feels like that sometimes that I'm doing everything constantly, reminding him to take his tablets, reminding him to do this, reminding and I say to him, hold on this week has been like a carer, you need to sort yourself out, and then we go back to him being the husband that I first met."

Through her verbal communication about being treated as a carer or a wife, W4Y5 is making her husband feel responsible for the way he makes her feel through his actions. She points out to him the behaviours which cause those role shifts within their relationship so that he can make the changes that help them to maintain their spousal roles. DBS has given W4Y5 the space to make sense of how PD impacts her relationship with her husband, and now she works hard to make sure she is not robbed of her spousal role again.

Theme 2: Parkinson's unites and DBS divides.

DBS brings balance in relationships through acceptance.

The 4 spouses, in this group, showed no signs of identifying with their spouses' sick role in the way the year 1 spouses did. They seemed more confident about their partners being more responsible for themselves. As we saw when W4Y5 stated,

"I say to him, hold on this week has been like a carer, you need to sort yourself out, and then we go back to him being the husband that I first met."

These spouses had more confidence asserting their autonomy.

The husbands described how these shifts in autonomy occurred in terms of practicality. H4Y5's wife started working from home post DBS, but surgery meant she was now able to share more in the daily chores, allowing there to be more of a sense of equality within the home at a practical level.

"DBS prolonged the time that she could do that, so that was a good thing but that's, that's been a bigger shift, you know, in the last few years, in terms of the kind of practicalities and so on, if she was still trying to do the job, you know, I'd be doing more still but because she's gone and got more flexibility now, about, you know she's working at home a lot and so on, so umm, you know, in terms of, you know, cooking."

The wives described more of an emotional journey towards finding their own autonomy. W1Y5 shared:

"it was after the DBS because I think what happened was I suddenly realised I had been carrying a lot of baggage and that was enough that (my husband) was better, and I had to do something and reclaim my life a little bit."

DBS alleviated the emotional "baggage" that weighed her down. She stated:

"I needed to reclaim my life a little bit, so I decided to take a year out and spend more time at home, do more of the things that I enjoyed because I think that I had lost myself in the caring, mothering, and working role"

"Reclaiming her life" appears to be about redefining who she is by her actions. Prior to DBS, she needed to fulfil many roles imposed on her: carer, worker, and mother. However, DBS helped facilitate a shift in W1Y5's behaviour, allowing her more time to do what she liked, allowing her to see herself as more than just a carer or mother, and distancing her enough from the PD life and her husband's sick role, so that these no longer dominated her life.

Theme 3: seeing myself and my needs, DBS enhances visibility.

Acceptance means it is easier to live for me.

This shift in responsibility did not just allow spouses to redefine their sense of self, it also allowed them to start identifying and start addressing their needs, something which was evident in the previous section when W1Y5 started "reclaiming" her life.

H3Y5's wife did not feel a huge benefit from DBS. She still struggled with pain, mobility issues, and low mood. However, the small shift that did occur gave some relief to her husband, enough for him to start seeing his own needs.

Knowing she is more independent at home has given him the freedom to go to work and socialise with friends guilt free. He even expressed:

"it got to the stage last year where I said look I need to get away I'm gonna go away for a week on my own,"

Since his wife's DBS surgery, H3Y5 recognised his need for independence and his need for a holiday. DBS gave him the opportunity to address those needs.

H4Y5's wife was also struggling with mood and mobility issues. Unlike H3Y5, who has taken to socializing without his wife and going on holiday, H4Y5 had recognised a need for more emotional support.

"as the effects of it have become more difficult in that period before DBS and again in the last couple of years I suppose that those, I think I've got better at talking to other people."

Prior to DBS, he described what it was like hiding his emotions thinking that was the best way of supporting his wife. However, DBS gave them a brief interlude, one in which he was able to become more self-aware and recognise his own struggles with the effects of the illness. As his wife's PD has started to progress, he now recognised his own needs for emotional support and has learnt to rely on his friends.

3.2. Group 2: Caregivers Who Did Not Express Their Own Personal Experiences of Caregiving. A content analysis was used to look for any other themes or patterns that could emerge from the interviews from the participants in group 2. The data collected was not rich enough to add anything to our understanding of caregiver identity post DBS surgery, even using a content analysis. Gender did not seem to have any effects on identity and, unlike group 1, neither did time since surgery. The location of where the interview took place did not seem to influence self-disclosure either, as one of the three participants who were interviewed at home was very comfortable sharing their experience and was hence in Group 1. The only overarching pattern worth noting was the strategies which these caregivers used in answering the questions posed, and Table 4 shows these three strategies. There was a constant desire by these participants to discuss their partners' lives rather than their own.

3.2.1. Strategy 1: A Bias When Answering the Opening Question. The first interesting strategy of note was regarding how the participants answered the opening question. The interest and focus of the study on their own experiences as carers, was emphasized to the participants at the time of recruitment and before the interviews began. All the interviews were started by asking the participants,

"Can you describe what life was like for you before your husband had DBS surgery, how did your spouse's Parkinson's disease impact on you?"

H2Y5 answered,

"The main problem was she had good and bad days, and the main problem was that a lot of the tablets wouldn't work because one of the symptoms of Parkinson's, she sweats a lot, so she'd have a very good day and a very bad day. She gave up work years ago."

W1Y1 answered,

"It, I think it figured quite a lot in his decision to retire because he, as he says, knowing what he knows now, felt he had the symptoms 3 years before."

These quotations illustrate how both participants launched into descriptions of their partners' symptoms and reactions to those symptoms, when asked about their own experience as a caregiver. This initial response suggests that these caregiver spouses may have a natural bias to consider their spouse with PD before themselves. When their spouses had been operated, was of no consequence, none of the 8 of the spouses described the impact PD had on their lives, a phenomenon that did not occur in the other caregiver group.

When it was reflected back to each of these participants that they had shared their spouse's experiences, they were once again asked to consider and describe how PD had affected their own lives. Similar responses were given.

Table 4: Strategies used by participants in group 2 when answering questions.

Strategy 1	A bias when answering the opening question
Strategy 2	Continuation of bias when answering subsequent questions
Strategy 3	Awareness of bias when answering questions

As we see from the Table 5, only one wife fully engaged with the question and reflected on how her life had been changed by PD. One husband and one wife started to consider the effects of PD on their lives but then reverted to describing their spouses' symptoms. One wife deflected the question by describing the impact PD had on her children, and the other four participants in this group described once again, their partner's symptoms, with very little portrayal of how this had affected how they felt or operated in the world.

3.2.2. Strategy 2: Continuation in Bias When Answering Subsequent Questions. As the interviews continued, this same pattern of behaviour persisted with all the participants. They either avoided, deflected, or reflected predominantly on their partners' lives. A good example of this is displayed by W1Y1.

"W1Y1: *he did say he thought he would be dead by last Christmas if he hadn't had the surgery and I absolutely believe he would have driven the car into a tree or a wall. I absolutely believe he would have done that.*

Interviewer: How did that impact on you? Was there any impact on your relationship or you, knowing that you are out all day, you have got this stress, you have got this fear and then after a few years, once everything started to get worse, you have now got this added stress that he is telling you, I might not be here by next Christmas? How did that make you feel?

W1Y1: Well no he didn't, it was just the way he was, the not eating, he lost a lot of weight. I mean he could stand to lose some weight to be fair and he is a much better weight now than he was, so, but it seemed to be quite quick, it was quite sudden, now again with all the movement that there is with Parkinson's you are going to lose weight also the fact that he wasn't eating as he used to eat, which wasn't enormous plates full of food but a normal diet and that was worrying."

After describing her husband's symptoms, she shares the disturbing information that her husband had struggled with suicidal thoughts prior to DBS. When asked to reflect on how this impacted on her, her answer barely relates to the question asked. She seems to find solace in describing her husband's symptoms rather than spending too much time reflecting on her own personal thoughts and fears.

As the participants described their spouses' difficulties, a consistent effort was made by the interviewer to remind them that although their accounts were valid, they were being asked to consider how these difficulties impacted on

their own emotional and physical wellbeing. 5 of the 8 participants spent minimal time self-disclosing and focused purely on their spouse as illustrated with W3Y1.

"*Interviewer: And how did that impact on your relationship with your husband? How was that for you?*

W3Y1: Well I suppose he at times feels guilty that he has this need, and, but he has become, I suppose his personality is different than it would have been years ago, much more anxious about things, everything, and life is lived around Parkinson's really."

Two of the eight participants asked that one of their children be present during the interview. H4Y1's daughter interjected often during the interview when her father avoided questions. She made statements such as,

"*when it first happened, you were a bit, everyone was a bit sad at the start and then as it progressed sometimes it wasn't nice.*"

And

"*you do worry, don't you, you do worry because mum is, one thing you don't have to worry about drinking or like no health things.*"

Yet, even these prompts from his daughter did not inspire H4Y1 to share any details about his own experience.

3.2.3. Strategy 3: Awareness of Bias When Answering Questions. Seven participants in this group seemed unaware of this bias towards describing their spouses' experience rather than their own. The one remaining participant in this group, outrightly stated that he preferred to speak about his spouse rather than himself. As we see from the following quote, his feelings seemed to be fully dependent on his wife's state of physical and emotional health.

"*Interviewer: You described how DBS has made her more positive as she is able to do more, can you now describe what it has been like for you?*

H1Y5: Well again I know I keep talking about her I suppose but I was pleased for her I was pleased that she felt this way and I still walk too fast, so I haven't really got much out of it but I was very pleased for her that she was getting this treatment."

4. Discussion

4.1. The Issues with Self-Disclosure. Conducting this study was not without its challenges and those challenges have raised some questions which are worthy of discussion. The first challenge of note is the issue that arose during data collection and analysis. Half of the 16 participants did not share enough reflections regarding their own lived experience for an IPA analysis. They disregarded, deflected,

TABLE 5: Strategies used by participants answering first question.

	Ability to reflect on first question	Ability to reflect and answer once question was repeated	Reflected once question was repeated but reverted to avoidance again	Deflected	Avoided answering once question was repeated
Number of participants group 1 1 year post DBS	4	N/A	N/A	N/A	N/A
Number of participants group 1 1 year post DBS	4	N/A	N/A	N/A	N/A
Number of participants group 2 1 year post DBS	0	0	2	0	2
Number of participants group 2 5 years post DBS	0	1	0	1	2

or chose not to answer many questions about their experience, and chose to speak predominantly about their ill partners. This could be indicative of something personal that was playing out for these carers. Carers can feel invisible [16], and these carers may have been uncomfortable with the level of visibility an interview afforded them, but it would be wrong to make any assumptions as to why they shared so few reflections, with such a small amount of data.

The method of data collection may have also impacted on the participants' ability to share. Other studies have used multiple interviews as a means of building rapport and engendering psychological safety [30, 31] to allow for deeper levels of self-disclosure. By the time it became clear that this was an issue within the data, the UK had been locked down due to the COVID-19 pandemic. The decision was made to continue analysing the data as collected, as interviewing caregivers virtually via Zoom or Microsoft Teams, with their spouses under the same roof, could potentially create new barriers to self-disclosure. Considering the current events, it would have also been difficult to compare the lived experience of caregivers in lockdown, to those who had been interviewed 4–6 months prior. If this study were to be replicated, consideration should be given to using a series of multiple interviews.

For as much as one's identity is created through one's own narrative, it also has a social context [32]. Individuals make sense of themselves through their interactions with others. By placing all attention on their partners, the thoughts and feelings about their own experiences were lost to the interviewer. This behaviour could suggest elements of "role engulfment and loss of self" which occurs "when the role of caregiver and responsibilities of caring begin to consume a person, leaving little time for other activities and behaviours that may have defined the person previously" [33]. This was prevalent in all the caregivers in group 2. In line with the caregiver identity theory [15], the introduction of an intervention such as DBS should have helped these caregivers to shift their attention away from their partners and more on to themselves, and yet this shift was not evident at interview, a phenomenon which is worthy of note and further exploration. This shift was however, present in the remaining 8 caregivers in group 1.

4.2. Shifts in Caregiver Identity: DBS Creates Possibilities.
In line with the proposed stages operational post DBS [8], all of the caregivers in group 1 were open to the shifts in their roles thanks to the changes in their spouses' symptoms and behaviours. We see from the first theme "DBS allows carers to question and shift the caregiver role" an evolution occurring between the two time points. The subtheme for the 4 participants left in the 12–18 months postsurgery group is, *Am I more than just a caregiver?* It has been noted that in the first year post DBS surgery, a positive result can give patients and spouses a sense of liberation and a less favourable result can bring on the need to reconcile with disappointment [31]; Haahr et al. [21]. These emotions were in part reflected in the testimonies of the participants.

W4Y1's anger when she stated, "*it's not just him that's got Parkinson's and I could, I could have not stopped working, I could have carried on working,*" shows her awareness of the choice she made to step into her caring role as she describes her disappointment that DBS has not given her any respite. It has been noted before that women struggle more with lack of freedom and excessive demands being placed on them by their partner's disease progression [34]. Whereas, all the caregivers in this group seemed to show some level of role engulfment, W4Y1 is not so engulfed that she is unaware of the lack of freedom her caregiving role affords her. We sense that this may be why she is associating with the role of the patient, as it offers her more than her caregiver role.

H1Y1 seemed much more comfortable and welcoming of the engulfment of his caregiver role. This could fit with Hughes' theory of master identity [35]. The master identity is formed when other identities are lost and the prevailing identity, in this case the role of caregiver, overshadows all others. H1Y1 explained that he can only see himself as a caregiver. Once faced with the potential loss of that identity we can postulate that his fear was triggered by the "burden of normality" [6] that has been described by patients post-DBS. Adjusting back to a more "normal" way of living can be daunting, particularly when it affects one's identity. H1Y1 is very comfortable in his role and feels united with his wife in their management of PD, DBS could shift that dynamic.

4.3. Shifts in Caregiver Identity: The Separation of Self.
The second theme "Parkinson's unites and DBS divides" explores this shift further. H3Y1 refers to his "combined

self," a concept which has been seen in other studies (Haahr et al. [21]) and was alluded to by all of the other caregivers in the earlier group. In the first year post DBS, the master identity of caregiver, if indeed one has arisen, has the potential to be challenged if the person with PD receives benefit from DBS surgery. The 4 of the 8 caregivers interviewed 4-5 years post DBS did not allude to this sense of unity. They felt more comfortable asserting new identity roles. W1Y5 referred to this as "reclaiming her life" which could be translated to, she reclaimed her other identities.

H4Y5 displays how his wife's reengaging in activities in the home has facilitated his ability to have more control over his own life. He is at ease with reclaiming his life. It has been reported that some people with PD struggle to involve themselves in activities of daily living post-DBS surgery which can lead to marital dissatisfaction [36]. H4Y5 reported feeling closer to his wife because she is engaging more in her role of partner, DBS has helped this couple to divide themselves off from the roles of caregiver and patient.

Feeling a sense of control while living with illness has been shown to improve life satisfaction and depression in caregivers [37]. People with an internal locus of control are motivated to engage in efforts which allow them that sense of control. They are also more likely to profit from psychological interventions [38]. This can lead us to the question of whether they could also benefit from surgical interventions such as DBS. These 8 participants, whose interviews were analysed, suggest that DBS has restored a sense of control back to them, improving their sense of wellbeing and their sense of self.

4.4. Shifts in Caregiver Identity: Remembering One's Self.

The third theme, "seeing myself and my needs, DBS enhances visibility" encompasses the two subthemes, "*I'm still invisible, DBS hasn't helped enough*" and "*Acceptance means it's easier to live for me*". Unlike previous studies that have shown a direct correlation between the spouse's treatment success and the caregiver's change in life experience (Haahr et al. [21]), the results of this study highlight the harsh reality that sense of self is not only dependent on external factors such as the caree's state of health. The results of DBS surgery vary from individual to individual, a reality that was very much captured by the experiences of all 16 participants. For the four caregivers who contributed to the subtheme of invisibility, elements of role engulfment still seemed visible, hence H3Y1's comment, "*the best bit (about DBS) really is, is seeing her life get better.*" That shift from phase four of caregiver identity, where the caregiver's lives are heavily dominated by their caregiving role, to phase five, where the individual reverts back to an earlier phase of identity [15], is starting to occur but it seems to be still in process.

The caregivers in the second group all seemed more in line with phases 2 or 3 of the caregiver identity theory. They were still involved in some of their partner's care but had a greater sense of self even though their spouses were starting to experience more PD symptoms. This may be described as redefining PD, something which occurs after the first year after surgery, once when both spouses find new ways to interact with PD due to the biographical shift caused by DBS [31]. These

caregivers displayed good coping mechanisms such as acceptance of their partners' state of health and seeking out social support. Improving coping strategies through a short course of cognitive behavioural therapy one year post surgery has been shown to benefit caregivers post DBS for PD [39]. The caregivers who were operated 4-5 years prior have a better sense of self than those interviewed at the earlier time point.

4.5. Creating Future Resources.

Many research teams have mentioned the need for pre and postoperative support for couples who embark on the DBS journey [36, 40, 41]. Attention is often given to the impact of expectations on satisfaction and burden [42, 43] with suggestions for interventions prior to DBS aimed at managing expectations and nurturing coping strategies [44]. Once more, this shows a bias towards the management of physical symptoms post DBS.

Managing PD is a complex process. The progression of the disease can cause one's sense of self and identity to shift during its life course (Haahr et al. [45]). DBS surgery is one more shift which must prompt more changes the within the self. Using the triadic model of multiple conversations with a nurse is worthy of consideration. Individualised meetings among nurse, patient, and spouse have been shown to shift the focusing on the physical adjustment process post DBS to the emotional adjustment process (Haahr et al. [20]). Repeated meetings have shown to create a safe space, where both members of the couple are invited to share their perceptions around everyday life, coping strategies, and expectations, allowing not only for them to be understood and guided by the DBS nurse but also giving the couple time to appreciate and have deeper understanding of each other. When we consider the testimonies of W4Y1 and H3Y1, we sense an imbalance in these caregiver's relationships with their spouses, as so much attention is focused on their spouse. This manner of intervention can bring balance to how the couple manage the adjustment process post surgery by giving each of them equal importance, time, and space to share. Individual therapy may also be considered as it can allow caregivers time to reflect and express their feelings, separate from their caree. In this case, it could help these spouses to process and separate out their own life journey from their partners. The results of this study suggest further explorations of the self-identity of the patient and the caregiver following future interventions, may be of value.

Health professionals could also consider approaches from positive psychology for innovative ways of helping caregivers to redefine their sense of self. Character strength interventions, whereby participants are asked to engage in those aspects of themselves, such as love of learning or creativity, can enhance pleasure and meaning independent of the caring role [46]. Facilitating access to psychological therapy, discussing the mobilization of social support networks and preoperative discussions around "readjustment" should all be considered [4].

5. Conclusions

DBS has the potential to restore a sense of self and agency, as displayed by half of the caregivers interviewed. However,

post DBS, itcan take time for spouses to make the shift from identifying as caregiver back to seeing themselves as partner.

After years of various patterns of behaviour becoming the norm within the relationship between people with PD and their spouses, DBS may offer caregivers the opportunity to create new behavioural patterns and those behavioural shifts can help give rise to new ways of identifying. As described previously, the handing of control and responsibility back to their spouses, was a process which many caregivers faced, which can cause distress for both partners at the beginning of their DBS journey. The more we understand these dynamics, the more clinical teams can support couples in relation to the emotional and psychological shifts that can occur post DBS, alongside the physical changes in the PD patient.

The differences in identity between the two subgroups of caregivers in group 1, supports the notion that identity is something that evolves over time. Healthcare providers should consider having conversations around identity, at various moments in time, to help support the shifts that occur for both the person with PD and the caregiver. Whether it is at the beginning of the PD journey, or at seminal moments such as pre- or post-DBS surgery, awareness is key. Making both parties aware of the potential effects PD can have on roles and responsibilities within the home and within one's partnership can help couples to behave more mindfully when possible, allowing for more control over any identity shifts.

This was a small study, and the ability to recruit more caregivers was cut short by the COVID-19 pandemic. Nevertheless, the results suggest a need for further exploration of the impact of DBS on caregiver identity and the effects of time on their psychosocial adjustment post surgery. That half the spouses found it hard to discuss their own reality is concerning and worthy of note. It suggests that some caregiving partners need substantial encouragement and support in understanding their own lived experiences prior to and post DBS. If caregiving spouses understand their identity and recognise their needs, outside of their caregiving roles, this may help facilitate psychosocial adjustment post surgery, not only for themselves but also for their spouse who has PD.

5.1. A Reflexive Statement from the First Author. I have been intimately connected to PD for over 20 years, and still I am learning about the effects of this illness. I am a therapist who has worked with people with PD and their caregivers for over 10 years. I have also been a caregiver, one who cared for a husband with PD for over 20 years and supported him through the DBS experience. One of the challenges I feared facing, in this research project, is keeping the balance between using my knowledge to inform my work, without projecting my life experience onto it.

In IPA, we speak about convergence and divergence. We look for the similarities that unite personal experience, while also looking for the differences that keep those experiences unique. This project had me reflecting consistently on those convergences and divergences. So, many of my own life experiences were mirrored in the testimonies shared, yet the way in which each individual described those life experiences and the impact they had; they were so different to my own journey.

When you live PD, and yes I mean live PD, one does not live with PD as if it is an addition to one's life; one eats, sleeps, and breathes PD. When you live PD, you start to recognise that no matter how much you have in common with others who live PD, your experience will always be unique and that can be isolating. So, we turn to others and look for those convergences, those strands of commonality that help us to feel like we are not alone in our suffering. We take comfort that someone recognises some of our challenges, and we give gratitude that there are resources that address those issues that so many of us share.

Interviewing this amazing cohort of caregivers was a privilege and a humbling experience, and I am grateful to all the participants who gave up their time to share their experiences with me so that I can help others understand them enough to help them through their challenges. I am also incredibly grateful to the other authors and my colleagues, who supported me through this study as they helped me to disentangle my own emotions and focus on what's important.

Data Availability

Access to patient interview transcripts is restricted due to patient confidentiality.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

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Research Article

The Lived Experience of Caregiving and Perception of Service Provision among Family-Caregivers of People with Late-Stage Parkinson's: A Qualitative Study

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Background. The complex nature of late-stage Parkinson's requires multiagency support and leads to an increased burden on family members who assume a multiplicity of responsibilities. The aim of this study is to further understand the lived experiences of family-caregivers and their perception of, and satisfaction with, service provision. *Methods.* This qualitative substudy was a part of the European multicentre Care of Late-Stage Parkinsonism (CLaSP) project. Purposive sampling resulted in a sample of eleven family-caregivers of people with late-stage Parkinson's, who were interviewed using semistructured open-ended questions. Thematic analysis followed. *Findings.* Three overarching themes were developed from the data: ensuring continuous support is vital to providing care at home, perceiving unmet service provision needs, and advocating and co-ordinating all aspects of care take their toll. These themes include not only experience of services that caregivers find supportive in order to deliver care but also of disjointed care between multiple agencies, a perceived lack of Parkinson's expertise, and there was a lack of anticipatory future planning. The constancy and scope of the family-caregiver role is described, including the need to project manage multiple aspects of care with multiple agencies, to be an advocate, and to assume new roles such as managing finances. Multiple losses were reported, which in part was mitigated by gaining expertise through information and support from professionals and organised and informal support. *Conclusion.* The intricacies and consequences of the family-caregivers' role and their experience of service provision indicate the need to acknowledge and consider their role and needs, fully involve them in consultations and provide information and joined-up support to improve their well-being, and ensure their continuous significant contribution to the ongoing care of the person with Parkinson's.

1. Background

The complexity and multifaceted nature of advancing Parkinson's requires ongoing primary care, specialist services, and increasing long-term support from multiagency health and social services. In addition, increased use of secondary care services with increased risk of hospitalization follows falls or infections [1], often precipitating care-home placement [2]. Whilst at home, there is an increased reliance on family members, frequently a spouse, adult offspring, or sibling [3], who become caregivers providing physical, social, domestic, and emotional care in the home, for up to

16 hours per day [4]. The caring demands related to reduced mobility and multiple fluctuating, incapacitating, and upsetting nonmotor symptoms (NMSs) of Parkinson's have been shown to negatively affect the physical, social, financial, psychological, and quality of life (QoL) of family members [5–9], with a higher burden compared to caregivers of older adults in general [10].

Parkinson's has no clear trajectory or duration, and its resulting dependency can therefore vary in years from diagnosis to death [11], with a range of between 1 and 21 years of caregiving reported [4, 10]. In addition to duration, caregiving strain increases and QoL reduces by increased age

and health needs in caregiving-spouses [12], and working-age offspring may be navigating life stage demands on top of caring demands. It has been found that, across conditions, such informal (unpaid) care had an estimated value of £132 billion annually in 2015, almost double its value in 2001 [13]. Despite caregiver strain being well documented, especially in relation to the burden of specific Parkinson's symptoms including significant impairment and falls; behavioural and cognitive changes; and neuropsychiatric symptoms of depression, apathy, and impulse disorders [5–9], and the roles and key activities assumed [4, 10], there is limited knowledge about the lived experience of family-caregivers in relation to their perception of service needs and provision for those with late-stage Parkinson's who have high degrees of disability. It is important to understand in depth the caregivers' experience and perception of service provision to comprehend the gaps they perceive and therefore often fill. Addressing unmet needs can potentially equip and support carers in multifaceted ways to continue offering support [14–16]. This is of broad significance as evidence suggests that caregiver strain is a strong predictor for the selection of institutionalised care for those with later-stage Parkinson's [17, 18], and the largest direct cost of Parkinson's is typically nursing home costs and inpatient care [19].

These are important considerations given the extensive provision of care by family-caregivers. Also given the predicted increase in prevalence and global burden due to Parkinson's [20]; and that the population is aging and Parkinson's occurs in people over the age of 60 [21]. Gaining insights into the experiences of family-caregivers and their perceptions of service use is therefore warrented. The findings are of value to help support those caring for people with Parkinson's and also applicable to family-caregivers of other progressive neurodegenerative conditioons. The aim of this study was therefore to facilitate an in-depth exploration and further comprehend the lived experience of caregiving for late-stage Parkinson's and the perception of service needs and provision from the family-caregivers' perspective in England.

2. Methods

2.1. Design.
This study had an explorative qualitative approach, using semistructured interviews [22] with family-caregivers. Interview data were analysed using thematic analysis [23], and reported guidelines by the Standards for Reporting Qualitative Research framework (COREQ) [24].

2.2. Ethics.
The study was granted ethical approval from Camden and Kings Cross Research Ethics Committee, London (IRAS 143636.14/LO/0367). Written informed consent was obtained from all participants.

2.3. Sampling and Participants.
Participants were purposively sampled [25] to obtain a breadth of ages, genders, living situations, and disability of the person with Parkinson's (PwP); they were providing care for from the English

cohort ($n = 123$) of the European "Care of Late-Stage Parkinsonism" (CLaSP) study [26].

Participants in the present study ($n = 11$) were family-caregivers of those with late-stage Parkinson's (findings reported elsewhere [27]). The PwP were caring for had been diagnosed according to UK Parkinson's Disease Society Brain Bank clinical diagnostic criteria [28], for at least seven years, and with disease severity stage 4 or 5 during the "On" state on the modified Hoehn and Yahr Scale (H&Y) [29, 30], or significant disability indicated by a score of 50% or below on the Schwab and England scale [31]. As analysis took place alongside the interview process, recruitment ceased once there was confidence that saturation was reached [32], that is, saturation was identified as attained when additional interviews did not reveal any new, extra information related to the study aim.

The recruitment process was closed when the sample consisted of 11 family-caregivers, the majority of whom were female spouses, living in their own homes with the PwP in urban or suburban areas in and within a 50-mile radius of London, England. In one case, their partner with Parkinson's was residing in a nursing home. The sample included three daughters and one sibling (Table 1).

2.4. Procedure.
Participants were recruited through general practitioners' (GPs) surgeries, NHS hospital outpatient clinics, Parkinson's charities, and specialist neurologists in and within Greater London, England. Initial recruitment approaches were made by clinicians, where invitation letters and information sheets were given to potential participants who were asked to return reply-slips to the research team should they wish to take part in the study. Following confirmation of eligibility, and the chance to ask questions, written informed consent was obtained.

Interviews used a study topic guide specifically for family-caregivers (Table 2) which was based on study objectives and developed by members of the CLaSP consortium involved in the qualitative arm of the project (see also [27]) and was further refined during application. Open-ended questions explored the perceived impact on life situations, needs, opinions about care and services, personal challenges and the positives of being a family-caregiver, deficits and barriers to care provision, and future care decisions. Prompts and probes were used, and responses were summarised to ensure that the information revealed during the interviews was correctly understood.

The interviewers had healthcare and/or psychology backgrounds. They were further trained for the study by qualitative methods experts, and therefore had the skills to build rapport and encourage information sharing during the interviews. All but one interview took place at the participant's place of residence, with one interview taking place in a private room in a nursing home. In all cases, interviews took place between the interviewer and the participant alone so that open discussion was facilitated. This included the eight instances where both the family-caregiver and PwP participated in the CLaSP qualitative study (PwP findings using a PwP-specific topic guide reported elsewhere: [27]). Interviews took an average of 60 to 90 minutes, were

TABLE 1: Participant characteristics (family-caregivers), $n = 11$, and characteristics of the persons with Parkinson's they cared for.

Demographic details of participants (family-caregivers)	
Gender	
Women (n)	10
Men (n)	1
Relationship with person with Parkinson's	
Spouse (n)	7
Daughter (n)	3
Sister (n)	1
Living arrangements	
Spouse living with a person with Parkinson's (n)	6
Spouse living alone. Person with Parkinson's residing in a nursing home (n)	1
Family member living separately but visiting regularly (n)	3
Family member living with a person with Parkinson's (n)	1
Characteristics of person with Parkinson's	
Duration since PD diagnosis	
Range (years)	8–27
Mean (years)	17
H&Y stage	
Stage 4 (n)	4
Stage 5 (n)	7
Age	
Range (years)	70–88
Average (years)	78
Education	
Range (years)	8–16
Average (years)	12

recorded using a digital recorder, transcribed verbatim by the interviewers, deidentified and data stored securely. The interviews presented below use a unique study ID. Interviews took place in a process separate from the quantitative data collection for the main CLaSP study [26], and the qualitative interviews and analysis took place over a period of twelve months during 2016.

2.5. Analysis. Content thematic analysis [23] was applied to identify, analyse, and describe themes as suggested by Clarke and Braun [23], and data were managed using NVivo 11 Pro [33]. An inductive approach was taken, and transcripts were read repeatedly by the first and second authors (JR and SC) to build an overview of all content and develop initial coding lists, concentrating on the study aims. The separate code lists were compared (JR and SC) and combined to create a coding frame, which was reviewed (CL, SI, and AS) and applied line-by-line to all data of interest (JR and SC). Codes were discussed throughout the process with all authors, and previously coded data were revisited (JR and SC) whenever new codes were identified. Themes and the definition of categories and subcategories were developed, and interpretations were discussed regularly with all authors throughout the analysis process to ensure validity. Trustworthiness was ensured by constantly reviewing the raw data, and supportive and reflective quotes were identified and selected.

3. Findings

In late-stage Parkinson's, the family-caregivers' lives become increasingly focused on the needs of the PwP and interactions with service providers, which changed the shape and content of the family-caregivers' lives, as reflected in three overarching themes, each with two to three subthemes shown in Table 3, and supported by additional quotes in the Supplementary Table (available here) entitled "Exemplar quotes."

3.1. Ensuring Continuous Support Is Vital to Continue Providing Care at Home. When family-caregivers spoke about "having needs met" this was rarely about their own specific needs, but instead related to those of the PwP which if met had benefits or positive consequences for the family-caregivers. The supports that enabled family-caregivers to continue providing home-based care without excessive impact on themselves were through *making use of multiple sources of support to manage life with Parkinson's* and *continuously finding the right information is vital to becoming an expert.*

3.1.1. Making Use of Multiple Sources of Support to Manage Life with Parkinson's. Family-caregivers felt responsible for managing often deteriorating situations, and support with this was seen as coming through the availability, efficiency, and good relationships with known local service providers,

TABLE 2: Family-caregiver interview topic guide.

Interview with family-caregiver living at home with a person with Parkinson's	Interview with family-caregiver of a person with Parkinson's living in a nursing home
(i) Personal needs and meeting of needs	(i) Needs and meeting of needs for both parties
(ii) Opinion about professional care received	(ii) Opinion about the residential care facility (including staff competence and medication)
(iii) Impact on personal health and life situation	(iii) Influence on care provided at the facility
(iv) Availability and opinion of respite services	(iv) Decision-making process to relocate to institutional setting (when, by whom, reasons, and information available)
(v) Opinion of residential care facility/nursing home, and if considered as a future option	(v) Feelings related to the move, at the time and in the present
(vi) Reasons current care situation	(vi) Management of care prior to the relocation and what would have been necessary to continue living there
(vii) Personal challenges in caring	(vii) What is missing?
(viii) Positive aspects of caring	(viii) Personal challenges
	(ix) Positive aspects

TABLE 3: Family-caregiver themes.

3.1. Ensuring continuous support is vital to continue providing care at home:	3.2. Perceiving unmet service provision needs:	3.3. Advocating and co-ordinating all aspects of care takes its toll:
3.1.1. Making use of multiple sources of support to manage life with Parkinson's	3.2.1. Experiencing fragmented and insufficient care for a complex condition	3.3.1. Assuming a project manager role
3.1.2. Continuously finding the right information is vital to becoming an expert	3.2.2. Lack of anticipatory planning for the future	3.3.2. Managing a constancy of demands
		3.3.3. Perceiving personal loss

specifically Parkinson's Disease Nurse Specialist (PDNS), social workers, and occupational therapists (OTs). Their professional input of specialist knowledge including about medications and symptom control, about financial support, and the provision of specialist equipment such as hoists or bathing aids, helped ensure the needs of the PwP were appropriately met. This subsequently supported family-caregivers, as the provision of equipment such as a wheelchair meant the couple could go out of the home and maintain some social contact. Similarly, the interview data revealed that changes in the PwP behaviour benefitted from specialist input for appropriate management which facilitated the family-caregiver continuing to providing care at home, and appropriate advice helped relieve the sense of responsibility and isolation:

"*She is on the other end of the phone and I can you know say*, (*name of specialist nurse*)" *I am desperate.*" And she will say "*Right I will be over tomorrow morning*" if I can sort of survive (...) if I have any problems over medication, I just give her a ring and ask her to come over or we go and see her in the clinic or something such as this and talk, talk the medication through (1094).

In addition, to support from health professionals, organisations in the voluntary sector facilitated couples remaining in their homes through the provision of assistive equipment, and importantly their service identified appropriate workers to assist with home maintenance; a task that for many elderly couples would otherwise be difficult or prohibitively expensive. Moreover, practical and emotional support for family-caregivers also came from other family members, longstanding friends, and the wider community, including faith communities and neighbours:

"We have lovely neighbours as well who have been over in the night to help me once when (PwP) fell. It was one in

the morning, I went and knocked on their door and, immediately (neighbour) came over, you know. Picked (PwP) up. Took him up to bed and tucked him up" (1094).

Support was also found through Parkinson's and generic caregivers support groups, which provided emotional support and a "safe space" to share the experience of being a family-caregiver. In a couple of cases family-caregivers took on organisational roles within both Parkinson's and community support groups, providing them with purpose, responsibility akin to previous employment, and respite away from personal caregiving demands: "*and I go to that on my own and that helps me*" (1064). A break from caregiving responsibilities was also found when the PwP spent time elsewhere for respite care, which was sometimes organised with the help of social workers or on the advice of PDNS, although often accompanied by a sense of guilt in the family-caregiver, was restorative and sometimes seen as essential for family-caregivers.

3.1.2. Continuously Finding the Right Information Is Vital to Becoming an Expert. Family-caregivers felt they carried the ultimate responsibility of providing or managing care and the interview data revealed that they either recognised the importance of becoming experts on the needs of the PwP and about Parkinson's, or became experts out of necessity, and that ongoing information was specifically identified as important in equipping them. Ongoing information was required as the condition advanced, particularly as information provided early on had often been forgotten or needs unexpectedly changed. Relevant information was obtained through healthcare professionals, through research and charity websites for example Parkinson's UK, and some support groups offered structured information and facilitated experiential information from peers, which was reported as being of value:

"Well, it is all Parkinson's patients with a Parkinson's nurse and they usually have a subject and it could be diet,

constipation, and exercise. And they have an hour or so with coffee and biscuits and things so you can talk" (1094).

3.2. Perceiving Unmet Service Provision Needs.
Family-caregivers spoke about the current service provision they received as being *experiencing fragmented and insufficient care for a complex condition* and that there was a *lack of anticipatory planning for the future*.

3.2.1. Experiencing Fragmented and Insufficient Care for a Complex Condition.
The family-caregivers described the care system as being complex, fragmented, inefficient, inflexible, overstretched, and understaffed, resulting in negative outcomes for those with Parkinson's and stressful ramifications for themselves. Family-caregivers described that care delivery was predominantly community-based, varied geographically, and service structure meant that there was a range of professionals and agencies that they had to liaise with. These included care agencies and formal carers, a variety of nurses including district, practice, elderly care, and PDNS, also social workers, OT's, physiotherapists, falls teams, and secondary care including neurology outpatients, and hospices. The result of which often resulted in fragmented care and additional burden to family-caregivers:

"He was getting all this sort of fragmented bits of care that were not the kind of coming together (...) a meeting to bring it all together. (...) I think I was trying to get some more care. Or was I trying to organise some respite? I was very tired" (1103).

Lack of specialist care was highlighted as problematic, with some not wishing to contact certain professionals feeling that they would not understand or be able to provide the required symptom management. In addition, it was felt that some formal carers did not have an adequate understanding of Parkinson's, thus creating discomfort about the care delivered:

"Some of the girls (formal carers) are lovely and some of them are absolutely diabolical. The problem is with the carers is that they do not really understand about the Parkinson's. They clearly had no training on it. You know some of them have come in and they even, they do not mean to do it nastily, but where my dad goes "b, b, b, ba, and b" (carer demonstrating the difficulty the PwP experienced when speaking) they take the mickey out of him and go "b, b, b come on (name), b, b, and b" "(carer demonstrating how formal carers mimicked the speaking pattern of the PwP), and repeat it back. They do not even realise how cruel that is" (1071).

3.2.2. Lack of Anticipatory Planning for the Future.
Managing daily life often consumed family-caregiver time and energy meaning the focus was primarily on the present rather than the future, and the unpredictable symptoms and uncertain disease trajectory also meant that managing and adapting to the immediate were more practicable. Some who would prefer to look ahead to the future were, however, inhibited by the PwP: "*Mum does not like to think about the future*" (1106), whilst others were in environments, usually a hospice, where future considerations were encouraged:

"And they do things like living wills and end of life care (...). So, that is very good (...) it makes you think (...) rather than just thinking about them in your head, actually verbalising them a bit which is a very good idea actually! So, and I think some people, well you all shy away from it a bit do not you because it is the last thing you really want to be thinking about" (1094).

The preference and intention of many family-caregivers were to continue looking after the PwP at home, nevertheless, future institutional care was often viewed as inevitable, despite previous poor experiences: "*The poor Parkinson's sufferers were trying to eat, cut their food up, and could not, and they were just whipping the plates away*" (1064); or concerns about the financial complexities and a lack of clarity or support in how to manage that: "*So social services were saying that it was a nursing need and nursing were saying that it is a social need*" (1071); or promises made to the PwP: "*You will never put me into a care home will you?*" and so, of course, I say "*no I will not,*" I mean I can't say "*yes one day I will,*" because that would be horrible really" (1064); or the fear that the person would deteriorate once admitted: "*I think he will go in there and he will nose dive*"(1071).

Despite any current difficulties, participants still deferred the decision for care to take place other than at home as a future option rather than something to consider in the present. Instead, decisions became superfluous when admissions to a nursing home became unavoidable, for example, in response to unmanageable deterioration, or when increasing demands were in conflict with commitments such as needing to work in paid employment or care for other family members: "*And I have spoken to me dad about it and I have said to him, "I have got to put my kids first, same as you would have put me first, you have got to go*"(1071). In these two examples, social services became involved in finding suitable alternative accommodation, however, participants did not speak about other health professionals or services advising or becoming involved in any preparation for the future.

3.3. Advocating and Co-Ordinating All Aspects of Care Take Its Toll.
The consequences of fragmented care from multiple care agencies meant that family-caregivers had to be an advocate for the PwP and co-ordinate all aspects of care, described under the theme of *assuming a project manager role*. A consequence of this was that they were never off duty, described under *managing a constancy of demands*, which led to *perceiving personal loss*.

3.3.1. Assuming a Project Manager Role.
In the absence of co-ordinated services, many family-caregivers assumed the roles of advocate and "project manager," planning, organising, and directing external agencies to deliver the best

possible care for the PwP. This demanded family-caregiver time, energy, and knowledge in liaising with multiple agencies, including communicating with those responsible for the formal care provision, for example, care agency managers, so that care was delivered, and service deficits were effectively addressed. Given that many participants were from the postwar generation, deference to medical professionals was evident, however, the driving needs of the PwP meant many became proficient in communicating with medical professionals and health agencies to ensure needs were met, and if met also improved the life of the family-caregiver.

Perhaps because they were "on-hand" and enmeshed in the PwP everyday needs or they felt that tasks were inappropriate for other family members, spouses often filled gaps in daily care provision, for example, attending to hygiene needs if a formal carer failed to keep an appointment. In contrast siblings and adult offspring often relied on care agencies to facilitate their external employment or to fulfil other responsibilities. Siblings and offspring, rather than spouses, described the need to "monitor" the care provided, sometimes describing external care as substandard with care staff arriving late, or missing visits, not understanding Parkinson's, or not respecting the PwP: "*they (carers) constantly talk over him*" (1071), or delivered inadequate care:

"I expect the same standard as I give him. And it is not always the case and so, because I am here I can see what they are giving him as a care company and what I give him and it is not the same and it is frustrating. So, I am constantly chasing them and that is tiring" (1095).

For several, decreased income and increasing demands on the household budget needed to be managed, including purchasing assistive equipment or supplies unavailable through other routes. Financial complexity associated with progressing symptoms meant family-caregivers assumed the role of "financial director" navigating external financial systems often without much support or guidance, and for some managing finances for the first time due to the PwP reduced ability to continue managing these. Such financial systems included applying and managing grants, for example, to create a walk-in shower-room, navigate the benefit system to claim disability living allowance, and manage payment of formal carers:

"They put in the money every month and we put in money every month into that account and all the carers get paid from that account. So, it is a separate account for the care" (1106).

These "project manager" activities were in addition to being sole initiators and facilitators for any social contact, or responsibility for responding to medical emergencies requiring external help, and to managing all areas of household life, which previously might have been managed as a couple or by the PwP him-/herself, putting increased demand on family-caregivers:

"And there is still the shopping and the washing, everything that they do not, that they do not class as being, because he is with family it is our duty. Our duty as a family is to look after him. (. . .) It is a nightmare it really is, it takes up all my time dealing with stuff" (1095).

3.3.2. Managing a Constancy of Demands. Providing care and support for the well-being of someone with late-stage Parkinson's at home meant that family-caregivers increasingly felt they were "never off duty," especially those living with the PwP as *"It is twenty-four hours because you are always on the lookout for one thing or another"* (1059). The constancy of demands was present throughout the day and night, with symptoms such as sleep disturbance and nocturia negatively impacting the family-caregivers sleep and therefore daytime function. Some caring activities were planned, for example, helping the PwP to dress or eat, but at all other times family-caregivers had to be mindful of spontaneous needs, for example, helping with toileting, or preventing or responding to falls; and the unpredictability of symptoms also made planning difficult. In addition, the progression of the condition meant there was a significant change in pace, where certain tasks became increasingly time consuming, for example, swallowing difficulties in the PwP meant feeding or administering medications consumed a substantial amount of family-caregiver time. Consequently, family-caregivers became less free to fully pursue their own activities or household tasks.

The interview data revealed that other members of the family described feeling similar pressures resulting in some being torn between the PwP and their children's needs, for example, with a burdensome constant responsibility, described by a sibling:

"I have got to be the driver and I cannot mentally and physically take that on. Umm otherwise my life gets consumed. I feel that it gets consumed now" (1095).

3.3.3. Perceiving Personal Loss. The constancy of demands, deficits in service provision, and increased care requirements meant family-caregivers experienced losses in multiple areas of their lives. For some, there was a loss of "space," independence, friendships, ability to travel, spontaneity, interests, "the life they had," and a "loss of self":

"A sense of loss of my own life, I would love to regain a sense of myself, it is the mantra which I repeat to myself, (. . .) you are also entitled to a life, although your life will have been changed irreparably because you have a partner you care for, (. . .) but you too are entitled to something that is yours too. Achieving that is very difficult" (1021).

For spouses, there was a diminishing of the relationship with the PwP. This was due to the reduced physical ability to share activities and socialize together but also changes in behaviour and loss of cognitive ability in the PwP altered the relationship dynamic. For example, impaired memory

meant that decisions previously made as a couple had to be taken by the family-caregiver alone. There was an ambivalence about how much it was appropriate to discuss with wider family members and friends, which increased feelings of isolation. The sense of isolation was further increased when friendships and contacts with other family members were reduced or lost, often due to the burden of additional tasks limiting time and capacity, the PwP becoming anxious when the family-caregiver left home, or difficulties arranging and paying for alternative care for the PwP to facilitate time away from home.

Despite reduced contact, solace and support were often provided by wider family members with examples of them providing advice, assisting with household maintenance, information provision, and grandchildren being the following: "*part of keeping me sane*" (1059). Nevertheless, not all were nearby and were considered to have their own lives to lead:

> "Unfortunately, our daughters, are all away, the help that they can give me is not as much as if they were able to do if they were living closer. But, you know they have their own lives. They do what they can. (...) I wouldn't expect them to be spending all their time here, that would not be right" (1064).

The interview data revealed that a loss of self was compounded by invariably adapting their own needs around those of others. Inadequate service provision meant a loss of career for some and the associated connections, satisfaction, identity, and financial renumeration. The latter having a negative impact on household finances. In addition, some family-caregivers had their own health issues, often ignored and sometimes exacerbated by the stress of their situation.

In order to mitigate the losses experienced family-caregivers felt it was important to maintain external interests and contacts where possible, facilitated by long-term friends and family members who understood if last minute changes to plans were necessary:

> "I was going to meet one of my friends and I had to say, "you know, I cannot do that tomorrow (PwP) is not so brilliant at the moment" (1064).

4. Discussion

Through the three themes, developed from the interview data for the present study, of the following: (1) ensuring continuous support is vital to providing care at home, (2) perceiving unmet service provision needs, and (3) advocating and co-ordinating all aspects of care take their toll, the study extends the existing literature by presenting the lived experience and perception of service needs and provision from the perspectives of family-caregivers of those with late-stage Parkinson's. It elucidates how the lives of caregivers significantly and detrimentally changed as their multifaceted caregiving role expanded with the progression of Parkinson's symptoms in the person they cared for and in relation to service provision. Whilst there is a growing understanding

of the impact of caregiving on various aspects of caregivers' lives across the stages of Parkinson's [5–7, 34, 35] little qualitative work has been conducted on the experience of caregiving specifically in relation to service provision in late-stage Parkinson's.

As findings show there are multiple sources of professional and informal support available to caregivers, and that good relationships with service providers were important in ensuring both delivery of good care together with support and guidance which could be seen to enhance a sense of control and empowerment. A necessary consequence of being a family-caregiver was "becoming an expert" about the personal needs of the PwP, and the multiple aspects of Parkinson's management and care options, necessitating the learning of new skills often with minimal guidance, and highlighting the importance of ongoing information. This subsequently provided a sense of control in an otherwise out-of-control situation, and the importance of empowering careers through increasing their knowledge and focusing on their assets has been reflected in the findings of interviews with health care professionals [36].

Family-caregivers perceived that care provision was fragmented, inefficient, and inflexible, and that services were overstretched. This subsequently led to negative outcomes for those with Parkinson's and stressful ramifications for family-caregivers as they took on additional tasks and responsibilities. In addition, there was a lack of required community-based specialist knowledge, as also reported in multiple sclerosis (MS) [37], and whilst PDNS, OTs, and social workers were cited as having significant supportive roles their availability varied geographically. The importance of regular access to specialist health care echoes findings from the Swedish CLaSP qualitative substudy cohort [14]. The need for more ready access to appropriate expertise and the need for specialist community psychiatric support is similarly indicated as in line with earlier literature reviews participants described behavioural and neuropsychiatric problems as being particularly difficult to manage [38, 39], and quantitative findings from all site data of the European CLaSP cohort showed that neuropsychiatric features were most strongly associated with caregiver burden [40]. A lack of community-based support and difficulties in PwPs attending clinical appointments, geographical differences in local healthcare provision, and a paucity of community-based specialist neuropsychiatric support may mean that telemedicine, previously explored in Parkinson's [41] and accelerated by the Covid-19 pandemic [42] could improve access to specialist services for family-caregivers of PwP in the late stages.

Elucidated by our findings, the demands of daily life and an uncertain disease trajectory and ambiguous future meant family-caregivers primarily focused on the present rather than future long-term care or end of life. This was unless hospice facilities were used, which perhaps reflects the difficulties in gauging when and who bears responsibility for introducing such conversations [43], suggesting a need for improved inclusion for palliative care and future planning conversations in consultations involving family-caregivers. Nevertheless, when the future was discussed, home-based

care was the preferred long-term option, as reported elsewhere [44], although institutionalised care was seen as inevitable; a well-reported outcome for other chronic conditions but particularly Parkinson's [45] and echoed by PwP themselves in the accompanying article from the CLaSP study [27].

Mirroring the carers UK report [13], our findings confirm the high levels of informal care family-caregivers provide; however, in contrast to other studies describing the extent of practical nursing and medical activities undertaken everyday by caregivers [10], our findings provide insights into the extent the family-caregivers role interfaces with service providers when the condition is more advanced. This is often due to fragmented care necessitating the family-caregivers to either provide care or oversee, co-ordinate, and manage the multiple care providers to ensure the best care provision. This is of interest given that many cares will be older and may have their own deteriorating health to consider.

Previous questionnaire data have shown that caregiver strain exists across all stages of Parkinson's however accumulates as the disease progresses [46]. In this current data, the increasing disabling symptoms in PwP meant family-caregivers not only managed constant care demands and management of services but also had to be continuously vigilant to ensure safety, as reflected in other quantitative findings [4] and data from all sites of the European CLaSP cohort where caregivers reported spending 7.6 (+8.2) hours per day supervising the PwP [40].

Findings describe the multiplicity of the family-caregivers remit as many took on new roles such as managing family budgets including complex financial matters such as payment of formal carers and navigating the benefits system to claim disability living allowance. Some financial and household tasks might previously have been managed as a couple or by the PwP, providing an insight into the evolutionary and dynamic nature of caregiving responsibilities and reshaping of the family-caregivers' lives, as reflected in the dementia literature [47]. The tasks taken on by caregivers were acknowledged and appreciated by those with Parkinson's in this substudy reported elsewhere [27] and they described the reshaping of roles as both positive and negative.

The needs of the family-caregivers became subsumed with the needs of the PwP, so that support to deliver good care or good care provision, perceived as receiving a tailored service and responsive and approachable professionals, resulted in indirect benefits for themselves. For example, facilitating a wheelchair could result in positive consequences by being able to be social as a couple away from home. The importance of such "normalcy" in those with Parkinson's and other long-term conditions has been reported [48, 49]; however, family-caregivers are often cited as the facilitators of this "normalcy" in Parkinson's [50] and dementia [51] rather than being able to pursue that goal for themselves and consequently experience many losses, including social and employment, in their own lives.

In contrast to other studies [52, 53], participants did not discuss esteem-based benefits of caregiving such as giving their lives meaning or pride in their successes as caregivers. Instead, they spoke about losses in multiple internal and external life domains. Caregiving demands had significant social and personal impact consequently eroding personal time and challenge the management of their personal lives and routines, as reported in other advanced chronic illnesses [54]. Loss of personal relationships included with the PwP and friends, but relationships' losses were also societal when unable to interact with the world through careers, travel, and interests. As reported in MS [55] family-caregivers described neglecting their own interests and needs, thus compounding a sense of isolation and loss. Given the extent of personal loss and reliance on the family-caregivers' capacity to cope, a means of evaluating family-caregivers QoL, support networks, and mental well-being is advisory.

4.1. Implications. The study findings have several implications for clinical practice. Findings show the need for the multiple service agencies to provide more joined-up care and to help project-manage care, and support family-caregivers in meeting the constant demands and overcome the personal losses they experience. The importance of including family-caregivers in all consultations as increasing symptoms dictate home-based care, relying on family-caregiver resilience particularly as preparation for the future and end of life becomes more significant. During such consultations, family-caregiver well-being should also be evaluated and appropriate support considered. Given the significant role of family-caregivers in the provision of care, future care delivery will need to accommodate a potential gap due to the rise in one-person-households. Future longitudinal research could, for example, focus in more detail on the experience and perception of preparation and support for end of life care.

4.2. Strengths and Weaknesses. The sample was from a group who were providing care for those with significant disability meaning they are not easily recruited to research, thus providing important insights. The sample size was determined based on the saturation principle [32], implying that the inclusion of participants ceased when no new information was gained from additional interviews. Recruitment approaches meant that urban and suburban settings were included, with a variation of socioeconomic contexts within a 50-mile radius of London, England, providing an informative picture of residence and service availability. Despite this, there was a lack of ethnic diversity limiting the transferability of the findings to such population segments. Similarly, there are limitations to transferability of findings as although a purposive sampling approach was taken, the majority of characteristics were based on the person with Parkinson's rather than the family-caregiver. Despite Parkinson's being over-represented in men, the inclusion of predominantly female caregivers is problematic, especially as females tend to assume important supportive roles in marital relationships and in illness. However, the cohort offers some breadth as it includes spouses, adult offspring, and siblings.

Notwithstanding, the team of authors represents clinical and scientific expertise including neurology, nursing, psychology, occupational therapy, and gerontology. Following the rigorous methodology, researchers collaborated in an iterative process during the process, which served well to ensure the validity and trustworthiness of findings.

5. Conclusion

In the absence of appropriate and comprehensive accessible service provision, this study illustrates how family-caregivers are key providers of personalised care, and co-ordinators of multiagency care. As illustrated by the findings, this primary role is an ongoing personal challenge for family-caregivers, even whilst managing their own health problems or competing demands. Health and social care service providers need to be aware and responsive to the demands on family-caregivers and offer appropriate collaborative support for their crucial input to continue.

Data Availability

The coding frameworks developed from the qualitative data used to support the findings of this study are available from the corresponding author upon request. Due to the confidential nature of qualitative data, the source data (i.e., interview data or transcripts) are not available as in order to protect participant privacy consent has not been given for individual data to be shared outside the direct research group.

Additional Points

What is known: The ageing population with the increased prevalence of Parkinson's and the complexity of advanced Parkinson's imply that there is an increasing demand for health care and social service provision. Family-caregivers have a central role in facilitating care at home. As a consequence of taking on a breadth of caring duties to manage and respond to advancing Parkinson's symptoms, there is a considerable burden on family-caregivers which can negatively impact their lives and well-being, and the marital relationship. What this study adds: Family-caregivers of those with Parkinson's assume not only caregiving roles but also the "project-management" of many aspects of health care and social service delivery. The study provides insights into the extent the family-caregiving role interfaces with service providers, including when family-caregivers perceive the need to assume an advocacy role and to manage and monitor external care providers. The lives of family-caregivers are impacted not only by the symptoms of the person they care for but also by the structure and provision of care.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

Supplementary Materials

Supplementary Table 1: This includes exemplar quotes. (*Supplementary Materials*)

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Hindawi

Research Article

Like a Wave in Its Variable Shape, Breadth, and Depth: A Qualitative Interview Study of Experiences of Daytime Sleepiness in People with Parkinson's Disease

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Introduction. Daytime sleepiness is a common nonmotor symptom in Parkinson's disease (PD) which is associated with decreased quality of life and perceived health. However, experiences of daytime sleepiness in people with PD have not been explored. The aim of this qualitative study was to explore experiences of daytime sleepiness in people with PD. *Materials and Methods.* Five women and seven men (42–82 years) with PD for 1.5 to 21 years and excessive daytime sleepiness (i.e., a score of >10 on the Epworth Sleepiness Scale) participated in the study. Data were collected through individual, semistructured, face-to-face interviews and analyzed with qualitative content analysis. *Results* Three themes of the experience of daytime sleepiness were revealed: (1) *not an isolated phenomenon,* (2) *something to struggle against or accept,* and (3) *something beyond sleepiness.* *Conclusion.* Daytime sleepiness is a complex nonmotor symptom in PD which manifests itself in several ways. Some experiences are similar, for instance, the attribution of daytime sleepiness to PD and its medical treatment. Differences depend on how sleepiness manifests itself, affects the person, and impacts daily life, as well as whether it causes feelings of embarrassment. Some participants needed to struggle against daytime sleepiness most of the time, and others had found a way to handle it, for example, with physical activity. However, sleepiness may also be used to benefit the person, for example, if they allow themselves to take a power nap to regain energy. The health care professionals can easily underestimate or misinterpret the prevalence and burden of daytime sleepiness because people with PD may describe daytime sleepiness as tiredness, drowsiness, or feeling exhausted, not as sleepiness.

1. Introduction

Parkinson's disease (PD) is an incurable neurodegenerative disorder with motor and nonmotor symptoms [1, 2]. Among the most disabling nonmotor symptoms are disturbed sleep and reduced vigilance [1]. The most common sleep-related symptoms are insomnia and poor sleep quality, and about 75% of people with PD experience such symptoms [3]. Studies suggest that sleep disturbances in people with PD are caused by disease-mediated effects on brain wakefulness [4].

Excessive daytime sleepiness (EDS) is another common symptom affecting approximately 55% of people with PD [3]. EDS is associated with sleep disorders such as insomnia, REM sleep behavior disorder, restless legs syndrome, and

periodic limb movements [5]. However, there are conflicting results concerning the connection between quality of nighttime sleep and EDS [4]. Some research indicates that the pharmacological treatment of PD may contribute to EDS and sleep disturbance [4], while other studies suggest that EDS in PD is a separate, disabling nonmotor symptom [6, 7].

The American Academy of Sleep Medicine [8] defines EDS as "the inability to stay awake and alert during the major waking episodes of the day, resulting in periods of irrepressible need for sleep or unintended lapses into drowsiness or sleep. Sleepiness may vary in severity and is more likely to occur in sedentary, boring, and monotonous situations that require little active participation." EDS is commonly identified and quantified with self-reported rating scales, for instance, the Epworth Sleepiness Scale (ESS) [9]. The ESS is an eight-item rating scale that assesses the propensity to doze off or fall asleep during various daily activities [9, 10]. Scores range from 0 to 24 (higher scores indicate more severe daytime sleepiness), and scores of >10 suggest abnormally high levels of daytime sleepiness [11].

EDS can be present prior to the diagnosis of PD [3], which suggests that it is related to the disease itself. However, the results of longitudinal studies on the progression of EDS in people with PD have been inconsistent. For example, some studies show rapid worsening of daytime sleepiness in the early stage of the disease [12, 13]. In contrast, a 10-year follow-up of people with PD found that daytime sleepiness was generally stable over time [14].

Previous studies of people with PD show that EDS can negatively impact perceived health [15]. Typically, studies investigating the prevalence and severity of EDS in people with PD have used a medical definition [9] of EDS [12, 13, 16, 17]. One qualitative study explored the burden of EDS in people with obstructive sleep apnea and found that EDS could affect health-related quality of life and daily functioning [18].

Despite the extensive research and numerous publications on the prevalence, severity, and impact of EDS, there is still a lack of knowledge about the subjective experience in people with PD, as well as what it means to live with daytime sleepiness. For example, it is not known whether people with PD and physicians are discussing the same phenomenon when they talk about sleepiness during daytime. When physicians ask about EDS, it is possible that people with PD imagine that their sleepiness must be extremely severe to fit the definition. There is also a lack of knowledge about how people with PD describe their sleepiness. Do they talk about sleepiness, tiredness, weakness, or something else? Scales such as ESS measure the propensity to fall asleep during daytime during the past month [9, 10] but not the burden of sleepiness. Maybe the burden of sleepiness is more difficult to accept and handle in daily life and has more consequences than the propensity for EDS. Do people with PD have strategies to cope with or handle this kind of sleepiness? Do they know that daytime sleepiness is a nonmotor symptom in PD, or do they think it is a natural part of aging? All these gaps in knowledge can lead to health care professionals underestimating or ignoring this nonmotor symptom in PD. More knowledge is therefore needed to understand the impact of daytime sleepiness in PD.

To the best of our knowledge, there are no qualitative studies about how people with PD experience EDS or daytime sleepiness. The aim of this study was therefore to explore the overall experiences of daytime sleepiness in people with PD.

2. Materials and Methods

2.1. Design. In this qualitative study, semistructured individual interviews were conducted with participants diagnosed with PD who had EDS according to ESS (>10 points). Data were analyzed with qualitative content analysis using an inductive approach. The interviews took place between June and October 2016 ($n = 12$). The study followed the Consolidated Criteria for Reporting Qualitative Research (COREQ) [19].

2.2. Sample and Recruitment. This study is a part of a larger prospective study that aimed to investigate the relationship between daytime sleepiness and nonmotor and motor fluctuations in people with PD [20]. Participants in the prospective study were recruited from a hospital outpatient clinic in Stockholm, Sweden, that specializes in PD and other neurological movement disorders. To be eligible, potential participants had to have a clinical diagnosis of PD verified by DaTSCAN and meet the criteria for EDS (>10 points on the ESS). Potential participants were excluded from the study if they had a diagnosis of severe cognitive impairment or dementia or severe untreated depression or were unable to understand Swedish.

Of the 53 participants in the original prospective study, 22 scored >10 on the ESS, which indicates excessive levels of daytime sleepiness [11]. A purposive sample of 12 participants with an ESS score of >10 (five women and seven men) of differing ages, PD duration, and PD severity were invited to participate in the current face-to-face interview study, and all accepted the invitation (Table 1).

2.3. Ethical Considerations. All participants provided their written informed consent before inclusion in the original prospective study [20]. The informed consent form included the information that they might be asked to take part in a qualitative interview study about experiences of daytime sleepiness. The study was conducted in accordance with the Declaration of Helsinki and was approved by the ethical review board at Karolinska Institutet, Sweden (Dnr. 2011/1866-31/4 and 2015/761-32).

2.4. Data Collection

2.4.1. Process and Tools. At an outpatient hospital visit, participants in the prospective study completed questionnaires on daytime sleepiness [9], sleep quality [21], fatigue [22], and symptoms of anxiety and depression [23]. In all questionnaires, except the one that assesses fatigue, higher scores indicate more pronounced symptoms. Motor symptoms were assessed with the Unified Parkinson's Disease Rating Scale, motor scale (part III) [24]. Disease

TABLE 1: Sample characteristics ($n = 12$)[a].

Variable	Participants
Gender (female/male) (n)	5/7
Age (years)	65.0 (61.0–75.75; 42–82)
Time since PD diagnosis (years)	6.5 (2.6–10.75; 1.5–21)
Hoehn & Yahr stage of PD in ON (I–V)[b,c,d]	2.5 (I-II; I-III)
Hoehn & Yahr stage of PD in OFF (I–V)[b,d]	III (III-IV; III-IV) ($n = 7$)
ESS daytime sleepiness score (0–24)	14 (13–19; 12–23)
PSQI, sleep quality score (0–21)[d]	10.0 (9.25–13.0; 9–14)
FACIT-F, fatigue score (0–52)[e]	34.0 (26.75–37; 12–47)
HADS, depression score (0–21)[d]	9.0 (7.25–12.5; 6–15)
HADS, anxiety score (0–21)[d]	13.0 (10.25–15.0; 9–15)

[a]Data are median (q1–q3; min–max) unless otherwise noted. [b]Range, I–V (I = mild unilateral disease; II = bilateral disease without postural impairment; III = bilateral disease with postural impairment, moderate disability; IV = severe disability, still able to walk and stand unassisted; V = confined to bed or wheelchair unless aided). [c]As assessed during the "ON" phase. [d]Higher scores = worse. [e]Higher scores = better. PD, Parkinson's disease; ESS, Epworth Sleepiness Scale; PSQI, Pittsburgh Sleep Quality Index Profile; FACIT-F, Functional Assessment of Chronic Illness Therapy—Fatigue scale; HADS, Hospital Anxiety and Depression Scale.

severity was classified in accordance with Hoehn and Yahr staging [25].

A semistructured interview guide, developed by AH and PH, was used to guide the interviews to get an overall picture of daytime sleepiness. The questions included the following: (1) Can you describe your experience of daytime sleepiness? (2) Can you tell me how daytime sleepiness affects your daily life? (3) Can you tell me if daytime sleepiness affects your Parkinson's symptoms? (4) Can you tell me how you cope with daytime sleepiness? (5) Can you tell me what words you use to describe this sleepiness? Probing questions (e.g., "Can you tell me more?") were used to follow up and deepen responses. If the participant found it difficult to describe his or her experience of daytime sleepiness, the interviewer asked the participant to describe a situation when he or she felt sleepy during the daytime.

At the start of the qualitative study, two pilot interviews were conducted to test the interview guide. No modifications were needed, and the pilot interviews were therefore included in the study. Eleven interviews took place at the outpatient clinic and one in the participant's home, in keeping with the participants' preferences. Each participant was interviewed once. The interviews at the clinic were conducted in a separate room during office hours. Only the participant and interviewer (AH) were present during the interviews. AH had provided care for three of the participants but had not previously met the others. Before the start of each interview, AH repeated the aim of the study and confirmed the participant's willingness to take part in it.

In this study, we simplified the AASM definition in deference to the study population who may have had difficulties keeping a long definition in mind. Daytime sleepiness was thus defined as a "subjective experience of sleepiness in daytime and a tendency to fall asleep or nod off during the daytime without previously planning to go to sleep" to also capture the episodes of sudden onset of sleep.

To minimize the risk of confusion about EDS and daytime sleepiness, which sometimes are used synonymously, we chose to speak about daytime sleepiness with the participants instead of EDS. Before the start of the interview, "tiredness" was explained as a lack of recovery and "sleepiness" as a need for sleep to feel recovered. This was done to explain the differences between these phenomena. These explanations were available in writing during the interview.

The interviews lasted between 10 and 35 minutes and were audio-recorded and transcribed verbatim. To limit inconvenience to participants, the transcripts were not returned to them for comment, but all were invited to contact AH after the interview if they wanted to add, change, or clarify something.

3. Data Analysis

Qualitative content analysis was used to analyze the data. This kind of analysis made it possible for the research group to describe the content of concrete descriptions while remaining close to the text [26]. It also enabled interpretation of latent meaning at a higher level of abstraction. The researchers thus worked simultaneously close to or distant from the text, providing more concrete (close) descriptions and more abstract (distant) interpretations [26]. An inductive approach was chosen because little is known about how people with PD experience daytime sleepiness. Before the analysis started, AH and UÖ reflected on the content of the transcripts to facilitate the choice of approach. The analysis was then conducted by AH and CS using the process described by Graneheim et al. [26, 27]. First, the text was read several times to get a sense of the whole. Then the text relevant to the aim was divided into meaning units, which were converted into shorter condensed meaning units that were labeled with a code. The codes were sorted into conceptual categories on the basis of their similarities and differences and then abstracted into themes (Table 2). During the analysis, AH and CS worked close to the original text by moving back and forth between the text, meaning units, codes, categories, and themes. This was done to maintain their awareness of the context and the essence of participants' experiences of the phenomena. AH and CS collaborated closely during the analytical process to reach consensus about the substance of the content. During this process, the analysis was discussed with PH and SF until all the authors reached consensus.

AH is a registered nurse specialized in PD. She has more than 20 years of experience providing care for people with PD and has a special interest in sleep disorders. CS is a registered nurse specialized in primary health care with expertise in sleep disorders. PH is a registered nurse specialized in PD and has a special interest in outcome measurement and the experience of living with PD. UÖ is a registered nurse specialized in oncology with expertise in cancer-related fatigue. SF is a professor of neurology and has been clinically active as a neurologist for more than 35 years.

TABLE 2: An overview of the analytical process, including examples of meaning units, codes, categories, and themes.

Meaning units	Condensed meaning units	Codes	Categories	Themes
I was never sleepy in the daytime before my PD diagnosis (Participant 4)	Never sleepy before PD	Disease	Related to the disease	
[Daytime sleepiness] comes, but it depends on how much I slept at night, so how prepared I am to take that sleepiness can be different (Participant 12)	Quality of nighttime sleep is essential	Sleep quality	Related to internal factors	Not an isolated phenomenon
But there is a difference (drive car/boat) because I drive a boat a lot. I never get tired then [....] I have never had this kind of problem when I have driven a boat (Participant 3)	The enjoyability of situation can play a role	Situational	Related to external factors	
I have to move when I get sleepy [....] I could run around for hours doing things. And that feels good (Participant 4)	Fight against sleepiness with physical activity	Activity	Coping strategies	
Yeah, it actually sucks to fall asleep whenever. It's uncomfortable [....] I think it's uncomfortable, [....] I don't take it as something negative, so it's a conflict for me (Participant 3)	Conflicting thoughts about daytime sleepiness	Conflict ignore	Attitude	Something to struggle against or accept
Take a nap to refresh and restart my body and brain (Participant 12)	Restart body and brain	Power	Coping strategies	
I can't take it [...] this isn't me (Participant 1)	This isn't me	Self-image	Personality change	
Yes, losing focus—that's at the root of all of it. The sleepiness gets worse then. Lose the thread, and it's hard to find my way back [....] It can be so embarrassing that I prefer to refrain from talking then (Participant 11)	Losing focus is so embarrassing that I prefer to refrain from talking	Shame	Negative feelings	Something beyond sleepiness
I see tiredness as a larger concept that has different facets [....] as being tired and sleepy—sleepy-tired [....] and this tiredness manifests itself as sleepiness (Participant 11)	Sleepiness is a larger concept than only sleepiness	Larger concept	More than sleepiness	

3.1. Methodological Considerations. In this qualitative study, credibility, dependability, and transferability were used to discuss the study's trustworthiness [27]. Interviews were generally brief, which may be seen as a weakness. However, people with PD can have difficulty concentrating because the disease negatively impacts their cognitive functions. They may therefore prefer brief and focused interviews. Clear and focused communication between an interviewer and participants can strengthen the informational power of a study. Thus, fewer participants may be needed in focused than in unfocused dialogs [28]. Other aspects of the study also illustrate its information power. For example, open-ended questions were asked to a purposefully selected group of people with PD who experienced daytime sleepiness but had varied clinical and sociodemographic characteristics [29]. The interviews brought to light a variety of experiences, characterized by both similarities and differences. These experiences were illustrated by citations, which potentially strengthens credibility. In addition, the interviewer (AH) was aware of her prior understanding and used an open and curious approach, adding probing questions to the prepared interview guide [29]. The authors acknowledged their prior understanding (of, e.g., PD and sleep problems) to minimize the influence of prior understanding on the analysis and the interpretations. On the other hand, it is difficult to get a deeper understanding of a phenomenon if the researchers are not familiar with the topic [30]. To increase dependability, AH and CS, who conducted the main analysis,

repeatedly discussed and reflected on the findings in light of their knowledge, and all authors contributed to discussions during the analysis. When they assess transferability, readers should bear in mind that the sample consisted of patients recruited from a clinic specialized in PD that was located in an urban area of Sweden. The written interviews were not returned to the participants for correction (so-called "member checking"). Maybe this would have increased the credibility of the results of this study, but out of respect for the participants' medical condition we decided to refrain. Member checking is usually done in two phases: the first is to ask participants to review the transcripts and the second is to ask them to review the first or final data analysis. However, criticism of the latter phase is that the researchers base their interpretations on several interviews, which may lead to misunderstanding when researchers and participants bring different perspectives to the analysis [30]. The sample size ($n = 12$) may seem to be low in this study but, in qualitative studies, rich content in the interviews is more important than the number of participants. In this study, the material was judged to be rich enough to fill the knowledge gaps identified about experiences of this nonmotor symptom.

4. Results

The analysis revealed three themes that illuminate the experiences of daytime sleepiness in the daily lives of people with PD. They experienced daytime sleepiness as (1) *not an*

isolated phenomenon, (2) *something to struggle against or accept*, and (3) *something beyond sleepiness*. These themes express the experience of daytime sleepiness as like a wave in its variable shape, breadth, and depth. People with PD experience daytime sleepiness in varied ways that depend on its impact on their personal lives. Daytime sleepiness could be a driving force, and napping could provide a refreshing new start during the day.

In the quotations below, three dots indicate a pause in speech. Three dots in square brackets mean that we have omitted one or several words, and four dots in square brackets mean that we have omitted a sentence or more. Words added to the text to clarify the speaker's meaning are also indicated by square brackets.

4.1. Not an Isolated Phenomenon.
This theme captures participants' experiences of daytime sleepiness and how other phenomena influenced this experience. They described it as a part of something bigger rather than an isolated phenomenon. Participants experienced daytime sleepiness to be related to PD itself, as a part of the disease rather than a specific motor or nonmotor symptom. They could attribute it to medical treatment for PD and link daytime sleepiness to certain situations, both monotonous and active situations. Additionally, participants experienced that the quality of nighttime sleep could have an impact on daytime sleepiness.

Participants described the relationship between daytime sleepiness, PD, and medication in several ways. "I was never sleepy during the daytime before my PD diagnosis," said one (Participant 4). "Medication makes me very sleepy, I can hardly stay awake," said another (Participant 12). Participants described themselves as not more tired and sleepy than others of their own age. However, it was more difficult to them to resist sleepiness during boring or monotonous situations than it had been before their PD diagnosis.

Some situations, certain places, and monotonous activities could prompt sleepiness, which could even feel irresistible. However, similar activities might not lead to the same sleepiness if the person enjoyed them. An example was driving a motorboat (enjoyable) as opposed to driving a car (monotonous).

Daytime sleepiness fluctuated during the day. For example, they could feel alert in the morning but hardly able to wake up later or sleepy all day. Sleepiness was most obvious to them in the evening or in passive situations, such as resting after an activity, but it also occurred during more active situations, such as during meetings and when driving. Sleepiness could also be like a barrier that they had to overcome to feel more alert.

"I don't exactly go to the doctor and lie down. So, there I become alert. So it's like some kind of barrier that somehow lets go, and so I become alert, but I really want to sleep for a while" (Participant10).

They also connected their ability to handle daytime sleepiness to the quality and length of their nighttime sleep. If they had slept well at night, they could tolerate daytime sleepiness better. One person (Participant 12) said, "It

[daytime sleepiness] comes, but it depends on how much I slept at night, so how prepared I am to take that sleepiness can be different."

4.2. Something to Struggle against or Accept.
This theme captures participants' experiences of daytime sleepiness as something to struggle against or accept or to find strategies to cope with. Some participants felt exhaustion, fatigue, and an overwhelming need to fall asleep. They might have to struggle with sleepiness several times a day. Others had found a way to accept their sleepiness, for example, by finding different kinds of activities that could push it away or by seeing benefits in sleepiness. At the same time, participants could have conflicting feelings about and varied experiences of daytime sleepiness.

One participant described overwhelming sleepiness as "Like cotton in my head. I never feel awake" (Participant 5). Sleepiness could even be paralyzing, something that took over life and was nearly impossible to fight against. Another described it as follows:

"Now I'm going to knit and lay it [knitting materials] on the sofa, but then I don't have the energy to do it after all. I just sit there [....] Then I just fall asleep" (Participant 1).

As noted, one way to gain acceptance was to figure out strategies to push away or reduce sleepiness with activity. For example, during meetings or other passive situations, it was not enough to change position. Participants needed more vigorous activity to counteract sleepiness, such as rising from a seated position or walking around.

Physical activity was a common way to handle and resist daytime sleepiness. One strategy was to plan activities or keep moving most of the day because it felt like physical activity could push away sleepiness. One person said, "I don't fall asleep while I am walking around" (Participant 11). Another said, "I have to move when I get sleepy like that, and then it feels good, then I feel like I could run around for hours doing things" (Participant 4). At the same time, some very active participants could unexpectedly fall asleep when they sat down. Another had a hard time staying active because the sleepiness was so hard to resist. It was like he had become "stuck in a sleep corridor" and had to fight to keep himself awake. It was difficult to affect sleepiness only with mental activity unless it was interesting or engaging in some way, in which case the person could feel more alert and forget about their sleepiness.

Participants could also accept the feeling but not allow it to disturb their daily life. Even when the experience of daytime sleepiness did not disturb daily life, the feeling of losing control during sleepiness was uncomfortable. Another way to talk about daytime sleepiness was to minimize it but acknowledge that it was a potential danger, for instance, while driving. Participants also expressed a contradictory experience, such as some level of acceptance coupled with a feeling of worry.

"Yeah, it actually sucks to fall asleep whenever. It's uncomfortable. But for me personally, I think it's uncomfortable, but take, I don't take it as something negative, so it's a conflict for me [...] [I] don't take what happens so seriously,

but really, it's uncomfortable to disappear without wanting to" (Participant 3).

Napping could also be seen as positive, a way to feel refreshed and restart the body and brain. Participants said that they "have all the right to rest or sleep for a while" (Participant 8) and that it was fine "to prioritize a nap" (Participant 6) or "take a nap to refresh and restart my body and brain" (Participant 12).

"I love to take naps. I love to be allowed to fall asleep. So, I really look forward to it, and therefore I don't want to book up these times. Instead, I want, I rush home to manage to sit down for this afternoon nap" (Participant 6).

4.3. Something beyond Sleepiness. This theme captures descriptions and consequences of daytime sleepiness in participants' daily life. The phenomenon was complex to describe. None of the participants described their sleepiness as only sleepiness. Instead, they talked about a hard-to-resist mixture of tiredness, sleepiness, and fatigue. Daytime sleepiness could affect self-image. Participants could feel that their sleepiness made them less valuable in the eyes of others and repeatedly brought up losing control when they talked about sleepiness. Even mental and physical functions could be worsened by daytime sleepiness. Several said that daytime sleepiness limited their daily lives, both privately and professionally.

Some participants described their experience as tiredness rather than sleepiness. Some explained that "sleepiness" was not enough to describe the sensation. It was too mild a term. They felt sleepy, but the feeling was part of a larger tiredness, something beyond sleepiness. It was a combination of sleepiness, tiredness, and fatigue and was therefore difficult to name. One described it as drowsiness: "I'm drowsy all the time. I never [fully] wake up during the day [....] No matter how much I rest, it doesn't go away" (Participant 5). Another said: "Sleepiness can be an aspect of tiredness. So, I see tiredness as a larger concept that has different facets. So, I would describe it as being tired and sleepy – sleepy-tired. I don't know what I should say. Sleepy-tired, but I'm more, more tired than before, and this tiredness manifests itself as sleepiness" (Participant 11).

Participants' self-images could change for the worse. One said, "I can't take it... this isn't me" (Participant 1). They described feeling lazy, uninterested in others, and worried that other people noticed their sleepiness (e.g., during meetings): "I'm not aware that I'm dropping off [....] And it's disturbing for the others too, if a person sits and sleeps. It's impolite" (Participant 9). They were also worried about losing control when they felt irresistible sleepiness.

Tiredness and sleepiness could also be experienced as a physical sensation. One person described it in the following way:

"I would like to take this off... like a coat. Some days, it feels like a big bird taking me in it its powerful claws and hugging my body so hard. Hugging and not letting go. Then the day after, it can be like it lets go, and I feel much better for a while [....] Some days are just completely lost" (Participant 1).

Daytime sleepiness could affect even mental functioning. One of the participants said, "When I feel sleepy, I'm not able to solve the most rudimentary mathematical problems like plus and minus" (Participant 11). Another (Participant 12) described losing his judgment when he was in the process of falling asleep. Daytime sleepiness could lead to loss of focus and thus to feelings of embarrassment and could be described like:

"Yes, losing focus – that's at the root of all of it. The sleepiness gets worse then. I lose the thread, and it's hard to find my way back. And it can be really hard when you're sitting and discussing something. It can be so embarrassing that I prefer to refrain from talking then. You lose concentration or forget a memory or whatever it is. I don't know what it is" (Participant 11).

5. Discussion

To the best of our knowledge, this is the first study to explore the experiences of daytime sleepiness in people with PD. Twelve people with PD and EDS (according to the ESS) were interviewed. They experienced daytime sleepiness like a wave in its variable shape, breadth, and depth rather than as an isolated, single phenomenon. The phenomenon was something to struggle against or accept or cope with and could be mastered by different strategies for some participants. It was bigger than just feeling sleepy, something beyond sleepiness.

Participants related sleepiness to having PD and to the treatment they received for the disease. Research shows that PD and its treatment can affect the basic diurnal variation of sleep and wakefulness [4]. They did not relate their sleepiness to the progression of PD over time, which is consistent with the results of a previous longitudinal study that found that EDS did not deteriorate during disease progression [14]. However, other studies have found that EDS increased in severity during the progression of PD [12, 13, 31].

None of the participants expressed the idea that their motor or nonmotor symptoms were directly linked to sleepiness. This finding was unexpected, as several studies [14, 20, 32, 33] have found a correlation between EDS, motor symptoms, and nonmotor symptoms (e.g., depression and anxiety) in people with PD. However, such associations are not necessarily causal and have in general not been particularly strong [6, 14, 32].

According to our findings, the burden of sleepiness could vary during the day and over time. Participants could feel sleepier or even fall asleep during monotonous situations and in the evening. Those who found it easier to accept the sleepiness and used strategies to cope with it may have had a higher level of perceived resilience than those who described their sleepiness as more severe and overwhelming. Few studies have investigated the role of resilience in the experiences of people with PD. One that did found that resilience correlated with nonmotor symptoms like depression, fatigue, and anxiety, as well as with having an optimistic personality [34]. In this study, we did not explore these phenomena or their relationship with sleepiness, but future studies could investigate this topic.

Participants who described their daytime sleepiness as severe and something to struggle against most of the time also described physical symptoms and difficulty thinking during episodes of sleepiness. Their experiences of sleepiness may be related to fatigue or a combination of fatigue and daytime sleepiness. Both fatigue and daytime sleepiness are common nonmotor symptoms in PD, and although their definitions differ, the two phenomena appear to overlap [35, 36]. Daytime sleepiness is characterized by feeling sleepy and at risk of falling asleep, whereas fatigue is characterized by a lack of energy and exhaustion linked to physical and cognitive impairment [37]. If these feelings were experienced simultaneously, it could be difficult to distinguish one from the other.

PD may be a risk factor for social isolation because of symptoms such as impaired communication, including reduced facial and bodily expressions [38]. Daytime sleepiness may further limit social contacts and thus increase isolation. In this study, participants who felt that sleepiness reduced social contacts with others also worried that their family members could become isolated. Moreover, participants associated daytime sleepiness with being seen as lazy and less valuable in the eyes of others. These feelings of embarrassment may have contributed to their altered self-image, which might lead to even greater social isolation [39]. These findings are in line with those of a previous study that explored the experiences of EDS in people with obstructive sleep apnea [18].

Participants in this study found it difficult to describe their feelings of sleepiness. For example, they called their sleepiness "tiredness" because they found that "sleepiness" was too limited a term to express the feeling. They explained their sleepiness as something beyond feeling sleepy, like a combination of sleepiness, tiredness, and fatigue. This underscores the importance of previous recommendations that clinicians ascertain what people with PD mean when they say they feel "tired," "fatigued," "sleepy," "groggy," or "drowsy" [40]. Additionally, many participants had not discussed their daytime sleepiness with their health care practitioner because they did not know that it is a common symptom in PD. This finding is consistent with that of a cohort study that suggested that people with PD may connect daytime sleepiness to poor nighttime sleep and therefore may not bring it up during medical consultations [41].

5.1. *Relevance to Clinical Practice.* Although daytime sleepiness is a well-known symptom in PD, the results of this study illustrate that it also is a hidden problem. Daytime sleepiness is not an isolated nonmotor symptom; it is more complex and has several dimensions related to PD and its medical treatment. It has consequences for daily life and can be difficult to resist and accept. There are several reasons why daytime sleepiness is still a hidden problem in people with PD. One is that people with PD do not always identify their sleepiness as a symptom, rather than a part of the process of aging. Another is that they can use unspecific and mild-sounding terms such as "tiredness" and "drowsiness" to

describe what they are experiencing. On the other hand, when clinicians ask about EDS, it is possible that they use the medical definition of "severe daytime sleepiness" and that, as a result, people with PD imagine that their sleepiness must be extreme to fit the definition. Thus, there is a risk that clinicians and patients may misunderstand each other, resulting in an underestimation of the presence and burden of daytime sleepiness in people with PD. It is important for clinicians to give people with PD the opportunity to describe their experiences of daytime sleepiness by asking in multiple ways about what the patient means when he or she mentions tiredness and/or sleepiness and whether these experiences have consequences in the patient's daily life. It may also be important to invite family members to the discussion to obtain a broader picture of the daytime sleepiness and its impact on everyday life [42]. Instruments such as the ESS can be used to detect daytime sleepiness and assess its severity, but they do not assess the burden or consequences of daytime sleepiness. They should be used as a complement to, rather than a substitute for, discussions with patients about their experiences.

The participants in this study found that physical activity could both relieve and induce sleepiness. Thus, recommendations about physical activity should be individualized, preferably in consultation with a physiotherapist. Self-management education might also help people with PD manage daytime sleepiness. Such education is available in many countries [43], including Sweden, where the National Parkinson School provides a scientifically evaluated educational program to help people with PD and their partners live and cope with the disorder [44].

PD is a very individual disease. In people with PD experience of similar symptoms, both motor and nonmotor, a difference may exist between the individuals and how clinicians assess these. For example, a clinician can assess a tremor as discrete, whereas the patient experiences it as severe and troublesome. A person-centered care approach can help clinicians better understand how each patient experiences his or her symptoms and their impact on daily life [45]. Such an approach could even lead to better adherence to medical treatment and opportunities to facilitate care and self-care based on the individual's needs [45]. People with PD and daytime sleepiness need to be approached individually because of the multidimensional expression of this bothersome phenomenon.

Further research is needed on the topic of daytime sleepiness in people with PD. We investigated people who scored more than 10 points on the ESS, the definition of EDS. There is no qualitative data about the burden and consequences of symptoms of daytime sleepiness on daily life in people with PD who score 10 or fewer points on this instrument. Future studies should investigate whether such people have similar experiences of daytime sleepiness. There is also a need for an instrument to measure the burden of daytime sleepiness, perhaps in the form of a virtual analog scale to measure the burden of sleepiness. Such a scale could potentially be added to the ESS.

6. Conclusions

Daytime sleepiness is a complex nonmotor symptom in PD which manifests itself in several ways. Some experiences are similar, for instance, the attribution of daytime sleepiness to PD and its medical treatment. Differences depend on how sleepiness manifests itself, affects the person, and impacts daily life, as well as on whether it causes feelings of embarrassment. Some participants needed to struggle against daytime sleepiness most of the time, and others had found a way to handle it, for example, with physical activity. However, sleepiness may also be used to benefit the person, for example, if they allow themselves to take a power nap to regain energy. Health care professionals can easily underestimate or misinterpret the prevalence and burden of daytime sleepiness because people with PD may describe daytime sleepiness as tiredness, drowsiness, or feeling exhausted, not as sleepiness.

Data Availability

The protocol and qualitative analysis data used to support the findings of this study are available from the corresponding author upon request. Deidentified participant data are not available for legal and ethical reasons. The prospective study is included in a doctoral thesis about daytime sleepiness in people with PD [46].

Conflicts of Interest

The authors declare that they have no conflicts of interest.

Authors' Contributions

A. H., P. H., and S. F. contributed to the design of the work. A. H. collected the data. A. H., C. Ö., and C. S. contributed to the data analysis and interpretation. A. H., with help from C. S., drafted the manuscript. P. H., U. Ö., and S. F. critically revised the article. A. H., P. H., U. Ö., S. F., and C. S. approved the final version for publication.

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Research Article

Occupational Therapy for People with Early Parkinson's Disease: A Retrospective Program Evaluation

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Purpose. Clinical practice guidelines establish that occupational therapy (OT) services are indicated for people with early Parkinson's disease (PD). However, OT is uncommon compared to other rehabilitation services. This study describes the development and evaluation of a proactive, consultative OT program for people with early PD as a part of an integrated care approach. *Materials and Methods.* The program was developed by an occupational therapist adapting practice guidelines for people with early PD. Retrospective program evaluation occurred at an outpatient rehabilitation clinic. The consultative OT program for early PD includes a 90-minute evaluation with instruction in self-management techniques, individually tailored exercises, and follow-up recommendations. The program was evaluated with the RE-AIM framework. Postprogram semistructured interviews provided patient-reported program effectiveness and satisfaction. *Results.* In 2018, 23 individuals used OT out of 77 people with early PD who attended the proactive rehabilitation program. Most individuals ($n = 16$, 69.6%) were within Hoehn and Yahr stages 1-2 and were seen within 3 years of PD diagnosis. Participants presented with deficits in hand strength (60.0 ± 23.4 pounds) and dexterity (right hand 30.0 ± 8.0 seconds) and reported complaints about basic and/or instrumental activities of daily living ($n = 15$, 65.2%). Semistructured interviews ($n = 16$) revealed that most individuals (75%) reported high satisfaction. Of the 10 who recalled a home exercise program, 60% reported continued adherence. Consultative OT was delivered with fidelity in 22/23 individuals (96%). After one year, only two occupational therapists at one clinic had adopted the program, and the program is maintained in the organization. *Conclusion.* Occupational therapists reached people in the early stages of PD when a specific program was tailored to evaluate and target their specific needs. Motor activity deficits noted in individuals with early PD support future scaling and sustainability efforts of OT within this population. Quality improvement suggestions are discussed for future implementation and clinical trials.

1. Introduction

Occupational performance, or engagement in life activities, is impacted by Parkinson's disease (PD) symptoms, including tremor, bradykinesia, weakness, poor dexterity, fatigue, gait impairment, apathy, depression, and cognitive deficit [1, 2]. An integrated team approach is needed to address these deficits, including contributions from occupational therapists. Clinical practice guidelines (CPG) recommend occupational therapy (OT) services for people with PD in all stages of the disease [3–5]. Yet, data reveals that people with PD rarely use OT [6–8].

Occupational therapy traditionally addresses occupational performance and participation in activities of daily living (ADLs), including self-care, leisure, and work [9]. Occupational therapists are equipped to identify the complex interactions between personal factors, environmental factors, occupational factors, and performance of daily tasks that impact people with PD [10]. Occupational therapy treatment approaches include improving occupational

performance by restoring impaired skills or body functions, as well as providing compensatory strategies, including the use of assistive devices, to help the person with PD to adapt to the environment or activity/task [11]. Self-management is enhanced through education on task performance and routine development, as well as patient and care partner training [12]. Furthermore, OT may help improve engagement in life roles within the home, community, and work environments [1].

Several CPG state that OT should be considered early after diagnosis with PD, to provide assessment, education on PD symptoms, and early intervention if ADL difficulties are present [3–5]. Initiating access to OT early after diagnosis can empower people with early PD (PwEP) to improve their disease self-management with more knowledge about the disease course and symptom management [13, 14]. Improvements in self-perceived performance were reported in one example of an individually tailored OT intervention for people across the stages of PD (62% of participants with Hoehn and Yahr stages 1-2) [15]. Early interventions in OT have been proposed to also address quality of life and health management for secondary prevention [16]. Although there is research supporting OT interventions [15], research regarding specific interventions in PwEP and research addressing barriers to health services for these individuals remain scarce [17].

Our purpose is to describe the development and evaluation of a proactive, consultative OT program for individuals in the early stages of PD. We will describe the results of this retrospective program evaluation using the RE-AIM framework, a tool to aid measurement and understanding of evidence-based, implemented programs [18]. RE-AIM is an abbreviation for (1) reach of the program to PwEP and participant characteristics; (2) effectiveness through feedback from PwEP; (3) adoption at the organization; (4) implementation fidelity of the delivery of care; and (5) program maintenance. We conclude with suggestions for future research and quality improvement in OT services for PwEP.

2. Materials and Methods

2.1. Setting. In 2016, a proactive, consultative rehabilitation program was implemented for PwEP by physical therapists at the Shirley Ryan AbilityLab (SRAlab, formerly the Rehabilitation Institute of Chicago) [19]. Although the program initially focused on PT, PwEP and clinicians expressed interest in OT and speech therapy (ST) services. In late 2017, formal programs in OT and ST were added. Program evaluation interviews and retrospective chart reviews were performed in 2019 for all patients seen in 2018. These methods were determined to be "not human research" by the Northwestern University Institutional Review Board due to the focus on retrospective evaluation of a current clinical program operating using clinical best practices. Due to the observational nature of the evaluation data, we used the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) checklist to guide reporting (Supplemental Material 1) [20].

2.2. Participants. Data were extracted from individuals if they were referred to the proactive PD rehabilitation program for any discipline in 2018 from the University's Movement Disorders Center or the SRAlab Interdisciplinary Parkinson's Disease and Movement Disorders Rehabilitation Screening Program. Individuals with a different movement disorder were excluded from analysis ($n = 2$). In total, 77 PwEP were referred to the proactive PD rehabilitation program, 23 of whom were seen in OT. Of the 77 individuals who utilized the program, 66 were contacted to participate in the program evaluation phone interviews (exclusions: seen in the program prior to 2018, $n = 8$, language barrier, $n = 2$, and error, $n = 1$). Of the 23 OT clients, 16 individuals agreed to participate in the interview (1 opted out following a mailed study letter, and 6 could not be reached with three attempts). These individuals provided verbal consent for audio recording of the interview for program evaluation, future quality improvement efforts, and deidentified dissemination activities.

2.3. Implemented Intervention. After a provider referral, PwEP underwent OT, PT, and/or ST using the proactive delivery model. This consultative model includes, at a minimum, a 90-minute evaluation and intervention session with each professional to whom they were referred. This model was shown to be successful in PT [19] and includes a low dose of intervention, focusing primarily on education and development of home programs. An episode of care in the proactive PT model has typically been accomplished in less than four visits [19]. The proactive model was then adapted in OT by a lead occupational therapist (JJ) with 10 years of experience and 7 years of working with people with PD. These adaptations included the addition of OT-specific outcome measures, documentation tips, educational handouts, and other resources. An evaluation plan was developed that focused on functional performance and identification of common motor and nonmotor problems related to PD. The goal of the prolonged initial session was to obtain a comprehensive battery of outcome measures for future comparison and provide immediate exercise recommendations to reduce the need for frequent follow-ups in this mostly independent population. When deficits were noted or reported, an individually tailored plan was created with the patient and therapist through patient empowerment and shared-decision making. Supplemental Material 2 provides a list of the typical evaluation and intervention items considered by the occupational therapist. Evidence-based OT treatments for PwEP primarily address self-management, functional independence, and meaningful occupations [11, 16]. The lead occupational therapist informally trained another occupational therapist in this model of care.

2.4. Data Sources. Retrospective data were collected from all PwEP who came through the proactive rehabilitation program (PT, OT, and/or ST) in 2018. Data were extracted from the referring providers' note (neurologist or interdisciplinary clinic screening note) and OT documentation in the electronic medical record (EMR) and managed using a tool

created in Research Electronic Data Capture software hosted at the University Clinical and Translational Sciences Institute [21, 22]. Program evaluation phone interviews were completed in 2019 for PwEP seen in 2018.

Reach was measured as the proportion of PwEP who accessed the OT program. Demographics, PD-specific characteristics, and OT evaluation measures are described. Demographics include age, gender, employment status, and insurer. PD-specific characteristics include time since diagnosis and PD severity reported using the Hoehn and Yahr (HY) scale [23]. OT evaluation measures included grip strength, pinch strength, and the Nine-Hole Peg Test. Grip and pinch strength have published normal values for older community-dwelling adults [24]. The Nine-Hole Peg Test is a validated measure of dexterity and upper extremity coordination in PD [25–27]. Additionally, patient-reported basic and instrumental ADL status was categorized by the occupational therapist as independent, modified independent (increased time or use of an assistive device), or requiring assistance.

Program effectiveness was measured using a phone survey and semi-structured interview 12–18 months after the initial evaluation (mean 15 months). Effectiveness was operationalized using the self-reported level of ADL function in everyday tasks after completing consultative OT by rating current status as "improved," "maintained," or "declined." Program satisfaction was measured using a 10-point rating scale. PwEP were also asked if they recalled and continued to use their OT home exercise program (HEP) and other recommendations. The interview guide included multiple choice and open-ended questions, which are shared in the supplementary material (Supplemental Material 3) and provide greater detail of the questions and potential responses/ratings from respondents. Interviews were recorded, and responses were categorized by clinically trained study team members who were not involved in the patient's care (AS; JM). To reduce the risk of bias, an independent auditor (research assistant) not involved in the clinical work checked data entry for accuracy.

Adoption was assessed by the number of occupational therapists trained in the proactive OT model, the number of sites within the organization adopting its use, and the number of referrers to the proactive OT program. These data were extracted from administrative data.

Implementation fidelity was measured by describing the extent to which the number and frequency of visits match a proactive, consultative model (e.g., 1–4 visits spread out to facilitate independent exercise completion). Fidelity measures include treatment information, education, and provision of an OT HEP supported by best practice recommendations [11].

Maintenance of the program is described as the number of PwEP seen in the early OT program in 2020 based on administrative data, although the program evaluation focused on the first year of the program (2018).

2.5. Statistical Analysis. Demographics, OT assessment, and phone interview data are presented with descriptive
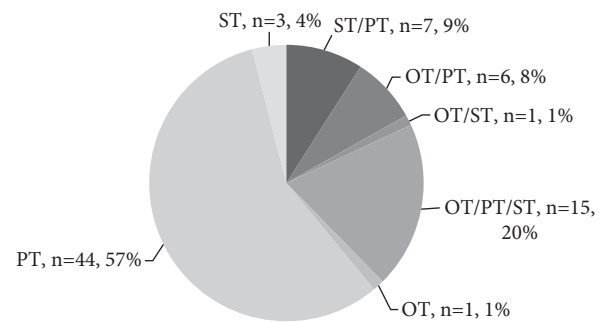

Figure 1: Distribution of all PwEP seen in 2018 ($n = 77$). Number of disciplines seen by each patient. *Note.* OT = occupational therapy; PT = physical therapy; PwEP = people with Parkinson's disease; ST = speech therapy.

statistics. The categorical outcome data were presented as N (%) and continuous data as means ± standard deviations. We compared PwEP who received OT to the other recipients of the proactive rehabilitation program (PT and/or ST) using Chi-square tests for categorical data and t-tests for continuous data, after checking normality assumptions, at a significance level of 0.05.

3. Results

3.1. Reach. Of the 77 people seen in the proactive rehabilitation program during 2018, 23 PwEP (30%) were seen by OT. Out of 23, 22 were seen in combination with at least one other discipline (Figure 1). Demographic and PD-specific characteristics of PwEP who saw an occupational therapist under the consultative delivery model are listed in Table 1. There were no differences found between the group of people who accessed OT compared to those who accessed PT and/or ST, except that there was a significantly smaller proportion of OT users with unknown ethnicity listed in the medical record ($p = 0.0213$). Table 2 describes OT evaluation measures and treatment information. The OT evaluation measures are presented in comparison to average normative data from healthy age-matched men and women and suggest that mild impairments were present in the PwEP participating in OT. In all test variables for both men and women, the values of grip and pinch strength were consistently weaker, and the time to complete the Nine-Hole Peg Test was consistently slower in the study sample compared to health age-based matched sample data. The only exception to this is key pinch strength on the right for women, with the study sample averaging 13.6 (±1.5) pounds pinched versus 13.4 (±2.8) pounds pinched in the healthy, age-based matched sample. Additionally, most individuals ($n = 20$, 87.0%) were documented at a level of modified independence for at least one basic ADL. The top five reported problem areas were upper extremity dressing ($n = 19$, 82.6%), eating ($n = 18$, 78.2%), lower extremity dressing ($n = 18$, 78.2%), bathing ($n = 16$, 69.6%), and grooming ($n = 16$, 69.9%). The two most frequently reported problems with instrumental ADLs were writing ($n = 9$, 39.1%) and keyboarding ($n = 7$, 30.4%).

TABLE 1: Demographic and Parkinson's disease-specific characteristics ($n = 23$).

	All ($n = 71$)	OT ($n = 23$)	PT and/or ST without OT ($n = 48$)	Statistical significance
Gender (% male)	44 (62%)	15 (65.2%)	29 (60.4%)	0.6966
Age, year	66 ± 9	68 ± 11	65 ± 8	0.1085
Race				
White	26 (36.6%)	12 (52.2%)	14 (29.8%)	
Black	1 (1.4%)	0 (0%)	1 (2.1%)	
Asian	3 (4.2%)	1 (4.3%)	2 (4.3%)	0.3755
Others	12 (16.9%)	2 (8.7%)	10 (21.3%)	
Unknown (declined to answer)	28 (39.4%)	8 (34.8%)	20 (42.6%)	
Ethnicity				
Hispanic	0 (0%)	0 (0%)	0 (0%)	
Non-Hispanic	44 (62%)	19 (82.6%)	25 (54.4%)	0.0213*
Unknown (declined to answer)	25 (35.2%)	4 (17.4%)	21 (45.7%)	
Insurance				
Medicare with secondary insurance	27 (38%)	11 (47.8%)	16 (33.3%)	
Medicare only	3 (4.2%)	2 (8.7%)	1 (2.1%)	0.1598
Private insurance only	41 (57.7%)	10 (43.5%)	31 (64.6%)	
Employment status				
Full time	16 (22.5%)	7 (30.4%)	9 (18.6%)	
Part time	1 (1.4%)	1 (4.3%)	0 (0.0%)	
Retired for age	23 (32.4%)	8 (34.8%)	15 (31.3%)	0.1476
Retired for disability	2 (2.8%)	1 (4.3%)	1 (2.1%)	
Unemployed	1 (1.4%)	1 (4.3%)	0 (0.0%)	
Missing	28 (39.4%)	5 (21.7%)	23 (47.9%)	
Hoehn and Yahr stage				
1	11 (15.5%)	4 (23.5%)	7 (33.3%)	
2	26 (36.6%)	12 (70.6%)	14 (66.7%)	0.4565
3	1 (1.4%)	1 (5.9%)	0 (0.0%)	
Missing	39 (54.9%)	6 (26.1%)	33 (68.8%)	
Time since Parkinson's disease medical diagnosis				
1–12 months	41 (57.7%)	12 (52.2%)	29 (60.4%)	
12+ months–3 years	14 (19.7%)	4 (17.4%)	10 (20.8%)	0.2744
3+ years	12 (16.9%)	5 (21.7%)	7 (14.6%)	
Missing	4 (5.6%)	2 (8.7%)	2 (4.2%)	

*Significance of <0.05.

3.2. Effectiveness. Sixteen of the 23 PwEP who engaged in the OT program completed interviews. Over a year after participation in the consultative model of OT, the level of function in everyday tasks was self-reported as "maintained" in 11 respondents (68.8%), "improved" in one (6.2%) respondent, and "declined" in four respondents (25.0%). Twelve (75.0%) respondents were highly satisfied with the OT program. Ten (62.5%) respondents were able to recall their HEP, but 4/10 stated that they no longer follow the exercises. Nine (56.3%) respondents recalled and described OT recommendations they had utilized, the most common being ADL tips, writing strategies, and computer keyboard modifications.

3.3. Adoption. Two occupational therapists conducted proactive evaluation and OT intervention sessions in 2018. All proactive OT sessions were conducted at a single site, even though the proactive PT program spread to two other affiliated outpatient clinics in the same timeframe. Six physicians (neurologists and physiatrists) referred PwEP to the OT program in 2018, out of 15 total referrers to all disciplines providing this consultative model.

3.4. Implementation Fidelity. All PwEP in the OT program were within five years of diagnosis. Three OT delivery patterns were used: (1) 17 (74%) PwEP completed one 90-minute session with no additional follow-up, (2) five (22%) PwEP completed 1–3 additional follow-up visit(s), and (3) one (4%) individual had a restorative bout of OT with seven follow-up visits. Either all PwEP were recommended to attend a re-evaluation in 6–12 months to document functional status and monitor HEP, or the occupational therapist documented that the individual would contact their physician if further treatment was needed.

Table 2 presents results on the assessment tools used and education provided during the initial consultative visit, reflecting implementation fidelity. All PwEP were educated in OT and the role of this provider as an integrated healthcare team member. The outcomes of specific assessment measured are presented in 23 PwEP (100%) who performed grip strength and the 9-Hole Peg Test and 20 PwEP (87%) who performed pinch strength testing. In this table, 19 (83%) PwEP received a proactive home exercise program, including fine motor coordination, upper extremity active range of motion, and strength. Additional common topics of education were home safety modifications

TABLE 2: OT evaluation measures and treatment information ($n = 23$).

OT evaluation measures (average (SD))	Study sample (men = 69.7 ± 11.2 yrs; women = 65.9 ± 10.7 yrs)		Comparison to healthy age-matched samples from literature [24, 26, 27]	
	Men ($n = 15$)	Women ($n = 8$)	Men	Women
Hand strength ($n = 23$)				
Grip left (lbs.)	71.1 (19.6)	42.0 (17.3)	83.8 (17.6)	50.7 (11.2)
Grip right (lbs.)	70.7 (19.2)	40.6 (18.9)	88.2 (18.3)	52.9 (11.7)
Finger strength ($n = 20$)				
Key pinch left (lbs.)	18.8 (4.8)	12.5 (1.9)	20.9 (5.0)	12.6 (2.5)
Key pinch right (lbs.)	19.1 (4.5)	13.6 (1.5)	22.3 (4.3)	13.4 (2.8)
Palmar pinch left (lbs.)	16.1 (5.1)	11.2 (3.6)	19.2 (4.4)	12.7 (2.9)
Palmar pinch right (lbs.)	16.5 (5.0)	12.1 (3.5)	19.5 (4.7)	13.5 (3.2)
Nine-Hole Peg Test ($n = 23$)				
Left hand (seconds)	30.3 (6.1)	27.7 (6.8)	22.3 (3.71)	21.4 (5.66)
Right hand (seconds)	29.9 (8.5)	29.8 (7.5)	21.2 (3.29)	19.9 (3.15)
Chief complaints reported to OT ($n = 23$)				
Fine motor coordination and control	14 (60.9%)			
Upper extremity strength and endurance	10 (43.5%)			
Basic and instrumental ADLs	15 (65.2%)			
ADLs	3 (13%)			
Tremors	11 (47.8%)			
Balance and mobility				
OT education information (implementation fidelity) ($n = 23$)				
Proactive OT education topics				
Role and purpose of OT	23 (100%)			
Home exercise program	19 (82.6%)			
Adaptations and modifications	16 (69.6%)			
Condition information	8 (34.8)			
Equipment and device use	7 (30.4%)			
Barriers to education				
Cognitive deficits	2 (8.7%)			
Language	1 (4.3%)			
Memory deficits	1 (4.3%)			

and expected PD symptoms in anticipation of disease progression. Less commonly, patients received education on equipment aids to promote occupational performance (30.4% of the time).

3.5. Maintenance. The program was sustained for the duration of 2018 and continues to reach PwEP into 2022 with adaptations to the initial program. Adaptations have included new organizational leadership support of OT program champions and mentorship opportunities. In 2020, programmatic reach grew by 47% from 23 to 34 PwEP. Over time, adoption spread to 2 sites, 4 occupational therapists, and 11 referrers.

4. Discussion

This program evaluation offered insights into a novel opportunity for occupational therapists to reach individuals with early stage PD. Most individuals with early PD chose to engage in this consultative model of care with just one 90-minute session focused on assessment, education, and exercise prescription. People with early PD who attended this model of OT presented with impairments in dexterity and hand strength compared to the similarly aged healthy population. Additionally, 87% of PwEP reported difficulty

with at least one ADL. The observed impairments in hand strength and dexterity may be related to the self-reported difficulty or slowness with ADLs, suggesting the need for greater use of OT interventions early after diagnosis [28]. It is important for these mild deficits to be addressed through exercise, education on compensatory strategies that can improve related functions, and long-term monitoring.

At this time, there is a lack of clarity around which treatments will best target the mild deficits noted in people with early PD. Foster and colleagues hypothesize that individuals in the early stages "may benefit greatly from interventions that promote the integration of self-management habits and other healthy performance patterns...into daily life" [11]. Future work on assessments and OT treatments for PwEP should adopt a framework, such as the Person-Environment-Occupation-Performance (PEOP) Model, or use other lifestyle management theories to better guide a person-centered approach [10, 29]. An OT framework would help to target this population with mild impairments and unique occupational performance concerns, which may include disease self-management, tasks required for job retention (e.g., keyboarding), and traditional ADLs. In this current work, a clinician adapted the evidence to their current organizational workflow and documentation constraints. Applying the PEOP framework could support a

more comprehensive evaluation and plan of care. However, the program setup would take additional time and resources, such as addition of new measures to the electronic medical record template. Occupational therapists who work with people with PD are key stakeholders to apply these frameworks and should be supported through compensated time or other incentive programs for the development of novel consultative programs for PwEP.

In this described program, OT provided baseline functional assessments, individually tailored exercise programs and environmental modification recommendations, and basic and instrumental ADL management strategies. The occupational therapist recommended treatment of observed deficits. The majority of PwEP in the OT program (74%) required a low-dose delivery model, with only one 90-minute consultative session. This delivery model had high program satisfaction, suggesting good value without the need for a burdensome commitment. People in the early stages of PD may be more interested in this consultative model versus a more extensive approach for a variety of factors, such as the individual's mild disease severity, insurance, time, or cost. Additional research is needed to better understand the best delivery models and assessments of low-dose, consultative models of care [30].

Improving reach, adoption, and effectiveness of OT for PwEP are opportunities for clinical and research improvement. We found OT to be the least common area of rehabilitation, which aligns with data from larger patient registries [8]. Similar to Roberts and colleagues, we found that OT was initiated most frequently in the context of multidisciplinary rehabilitation rather than a solo entry point into the rehabilitation system [8]. We also found that follow-up visits were rare within the first year, despite the documented impairments supporting the need for OT. Incorporating relevant screening measures in hand strength, dexterity, or daily living self-assessments such as the Movement Disorders Society revision of the Unified Parkinson's Disease Rating Scale [31] in neurology clinics may promote earlier referral and treatment in OT. In addition, further study on successful interventions to improve hand strength and dexterity in PD could improve education provided to the physician and PwEP regarding the need for proactive OT programs, as well as OT delivery itself.

Additional opportunities to improve proactive OT delivery include exploration and application of OT interventions as they relate to employment and self-management. Our data suggest that our initial proactive OT program implementation focused on traditional areas of basic and instrumental ADLs. However, approximately 35% of PwEP in this program were working full or part time, and computer use strategies were a commonly recalled tip from participants. Proactive OT can tailor interventions to address employment, including the psychosocial implications of working with a chronic condition, work-related skills, and workplace productivity. Energy conservation techniques and self-management education may help manage symptoms to help PwEP stay in the workforce longer [32, 33]. Further research to advance PD-specific self-management training programs may improve proactive OT delivery [34]. For instance, it has been proposed that OT can play a role in developing a health plan to promote a healthy lifestyle in older adults using methods that could apply to PwEP [29].

4.1. Limitations. The results of this program evaluation have limited generalizability. First, the sampling strategy for this single-site evaluation introduced recall and selection bias due to the small sample size, delayed follow-up, and lack of control. Second, the high level of variability in clinical documentation made it difficult to extract data from the EMR. Additional organizational support for OT-specific program development and facilitation could improve implementation fidelity in the future. Some documentation had evaluation items missing, which could be due to clinician time constraints or patient barriers, such as cognition or language. Third, our cohort was fully insured, primarily white, and English-speaking, which may limit generalizability to more diverse clinics. Lastly, the nature of a single consultative visit did not allow for follow-up assessments to document the clinical effectiveness of the proactive OT model beyond satisfaction. Future research should include regular follow-up assessments in order to compare the results of this intervention to other OT interventions.

5. Conclusion

This OT program evaluation revealed that even individuals with early PD have key occupational performance and participation issues that can be addressed by occupational therapists. While the use of OT early after diagnosis was lower than other disciplines, satisfaction remained high for those who participated. Future quality improvement efforts, aided by programmatic support or implementation research funding, are recommended to improve the reach of this program.

Data Availability

The data used to support the findings of the study can be obtained from the corresponding author upon reasonable request.

Conflicts of Interest

The authors declare that they have no conflicts of interest.

Authors' Contributions

Alan Sadural and Jillian MacDonald are co-first authors.

Acknowledgments

Supplementary Materials

Supplemental Material 1. STROBE statement: completed checklist for Strengthening the Reporting for Observational Studies in Epidemiology. Supplemental Material 2. Occupational therapy (OT) evaluation and intervention: a list of typical evaluation and intervention items from the consultative OT session. Supplemental Material 3. Phone survey and semistructured interview discussion topics. (*Supplementary Materials*)

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