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Further explorations of illness uncertainty: Carers' experiences of Parkinson's disease

Running Head: Carer uncertainty of illness

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Abstract

Objective: Dominant models of illness uncertainty define uncertainty as ‘an inability to determine the meaning of illness-related events’. Recent research has shown patient uncertainty to be multidimensional encompassing personal issues indirectly affected by illness. The nature of carer uncertainty has yet to be fully explored. The present study aimed to investigate the nature of illness uncertainty in the carers of patients with Parkinson’s disease (PD).

Design: Eighteen carers of a spouse with PD participated in semi-structured interviews. Transcripts were thematically analysed, statements were coded as uncertain if they reflected ‘a lack of certainty, or a state of limited knowledge, understanding or worry regarding an existing or future outcome’.

Results: The domains of uncertainty expressed by carers closely fitted the five domain framework of patient uncertainty: symptoms and prognosis, medical management, self-management, social functioning and impact. An additional ‘carer-role’ domain was identified.

Conclusions: Carer uncertainty about PD went beyond issues directly related to the illness. The findings have implications for research into uncertainty suggesting that widely used measures may not be accurately capturing the nature of carer uncertainty about chronic illness. The breadth of uncertainty reported has implications for the provision of appropriate support to improve caregiver well-being.

Key words: Uncertainty, Parkinson’s, Carers, well-being, adjustment, beliefs

Introduction

A diagnosis of chronic illness causes significant disruption to family life particularly in unpredictable and incurable conditions. Evidence suggests that patients with chronic illness face significant uncertainty which negatively impacts on their well-being and quality of life (QoL) (e.g. Eastwood, Doering, Roper, & Hays, 2008; McCormick, Naimark, & Tate, 2006; McNulty, Livneh, & Wilson, 2004; Van Pelt, Mullins, Carpentier, & Wolfe-Christensen, 2006). Uncertainty is considered to be an inherent aspect of life associated with the ability to cope and overcome challenges (Kahneman & Tversky, 1982; Kahneman & Tversky, 1982; Lazarus & Folkman, 1984; Weary & Edwards, 1994). Within the spectrum of chronic illness, uncertainty has been described as a cognitive stressor that challenges patient adjustment (Johnson, Zautra, & Davis, 2006).

The dominant theories of illness uncertainty, the Uncertainty in Illness Theory (UIT) (Mishel, 1981, 1988) and the Revised-UIT (RUIT) (Mishel, 1990) define uncertainty as a ‘cognitive state created when the person cannot adequately structure or categorise an event due to a lack of sufficient cues and thereby cannot determine the meaning of illness related events’ (Mishel, 1988, 1990). The UIT was developed in hospitalised patients with acute illness but reconceptualised to explain the experience of living with continuous uncertainty in chronic or recurrent illness. Mishel (1981) proposes four key factors that characterise illness uncertainty: i) Ambiguity about the state of the illness, ii) Complexity regarding treatment and healthcare systems, iii) Lack of information about diagnosis or seriousness, iv) Unpredictability about the course and prognosis. The RUIT explains how uncertainty develops and can lead to adaptive outcomes, where uncertainty is viewed as an opportunity or becomes integrated into the patient’s life, or maladaptive outcomes where uncertainty is viewed as a danger.

High levels of uncertainty when perceived as a threat lead to poor coping and adjustment (Mishel, 1990). Consequently there is a need to identify factors which make some individuals more vulnerable to perceiving uncertainty as a threat rather than an opportunity. This will inform the development of interventions to both reduce uncertainty and enable patients to cope with persistent uncertainty. Interventions including cognitive reframing, information giving, and doctor-patient communication have been trialled in breast and prostate cancer with positive effects (Braden, Mishel, & Longman, 1998; Mishel, 1997; Mishel, 2014; Mishel et al., 2002).

However the validity and comprehensiveness of Mishel's (Mishel, 1988, 1990) definition of uncertainty as 'an inability to determine the meaning of illness related events' across all illnesses and patient groups has been called into question in several qualitative studies (Brashers et al., 2003; Cleanthous, Newman, Shipley, Isenberg, & Cano, 2013; Kasper, Geiger, Freiburger, & Schmidt, 2008). Research shows that in patients uncertainty extends beyond illness related events (e.g. progression) to aspects of life which are indirectly impacted by illness (e.g. social functioning) and is condition specific (Cleanthous et al., 2013). In their exploration of uncertainty in systemic lupus erythematosus (SLE) and rheumatoid arthritis (RA) Cleanthous et al. (2013) found five overarching domains of illness uncertainty: symptoms and progression, medical management, self-management, impact and social functioning. However within these domains illness uncertainty was found to vary between the two conditions. Consequently the exploration of uncertainty within the UIT models is unlikely to lead to a comprehensive understanding of all aspects of uncertainty experienced by patients.

The UIT and RUIT propose that uncertainty of illness is not just experienced by patients but also by those who care for them. Despite this there has been limited research to date examining uncertainty in carers. Research that has been undertaken suggests that illness

uncertainty is central to carers' experience of chronic illness and has been associated with carer depression, anxiety and life satisfaction (Maikranz, Steele, Dreyer, Stratman, & Bovaird, 2007; Unson, Flynn, Glendon, Haymes, & Sancho, 2015; Waldron-Perrine, Rapport, Ryan, & Harper, 2009). Much of this research has employed the UIT and RUIT which, as with patients, may not be adequately examining all aspects of illness uncertainty in carers.

Family caregivers provide the majority of care for patients with chronic conditions (Beesley, 2006). In 2015 it was estimated that there were 6.8 million family carers in the UK (approximately 12% of the population), an increase from 5.8 million in 2001 (Buckner & Yeandle, 2015). The economic value of informal caregiving is estimated to be as high as £132 billion per year in the UK (Buckner & Yeandle, 2015). Caring has significant implications for the caregiver. Carers have poorer physical and psychological well-being than their non-caring peers and show increased risk of mortality (Schulz, 1999). In view of the central role played by family caregivers it is important that efforts are made to maximise carer well-being through the identification and amelioration of factors associated with poor well-being and QoL. There is a clear need to explore carer uncertainty of illness to enhance our understanding of carer adjustment to chronic illness and consequently provide new avenues for therapeutic intervention to improve carer QoL.

Parkinson's disease (PD) is a chronic, degenerative neurological condition affecting approximately 127,000 people in the UK (Parkinson's UK, 2015). The disease is characterised by motor symptoms including tremor, slowness of movement and rigidity. PD is a condition with high uncertainty. The cause of PD is not well understood and there is currently no known cure. Prognosis is highly variable with some patients experiencing minimal symptoms for several years and others becoming wheelchair bound within a few years of diagnosis. Treatment regimens differ substantially between patients and require frequent adjustment as a particular drug and dose becomes less effective or side-effects become unmanageable

(Mueller, 2012). Research has demonstrated that patients experience uncertainty over the effects of PD day to day describing frequent ‘good days’ and ‘bad days’. This uncertainty limits the patient’s ability to plan the simplest activities (Hurt, Weinman, Lee, & Brown, 2012). Uncertainty about the future, particularly with reference to finances, living situation and independence were evident in qualitative research (Wright, Hurt, Gorniak, & Brown, 2015). Explorations of the experience of caring for a person with Parkinson’s have highlighted the central role of uncertainty, particularly around patient well-being, disease progression and future caregiving (Martin, 2015; Williamson, Simpson, & Murray, 2008).

This study aimed specifically to test the applicability of Cleanthous et al.’s (2013) five domain model of patient uncertainty of illness in a sample of caregivers. Due to its disabling nature almost all patients with PD have a carer, most commonly their spouse (Schrag, Hovris, Morley, Quinn, & Jahanshahi, 2006). Given the highly uncertain nature of PD and the significant role played by carers, PD is an appropriate condition within which to explore carer uncertainty of illness.

Methods

Participants

Participants were recruited from the Parkinson’s UK Research Support Network, a network of over 500 people with Parkinson’s and their carers from across the UK. Inclusion criteria were a) main carer for partner or spouse with a diagnosis of idiopathic PD, b) able to communicate fluently in English, and c) partner or spouse provided informed consent for the carer to discuss their condition. Ethical approval for the study was granted by City University London School of Health Research Ethics Committee (ref: Staff/12-13/28).

Spouse carers were purposively sampled as they represent the most common form of informal caregiver in Parkinson’s. 18 carers were recruited, sample sizes of at least 12 have

been found to be adequate in homogenous populations with narrow research objectives (Guest, Bunce, & Johnson, 2006) and in theory driven research (Francis et al., 2010).

Procedure

The study was advertised via an email sent to all Research Support Network members across England. Interested participants were invited to email the study coordinator for further information. Both the carer and person with Parkinson's were provided with information sheets and had the opportunity to discuss the study. Both the carer and person with Parkinson's were asked to provide informed consent.

Data collection

In-depth semi-structured qualitative interviews were performed using a topic list and interview schedule derived from a review of relevant literature to identify topics of interest (see supplemental material). All interviews were conducted over the telephone, data was recorded and transcribed verbatim. Interviews were conducted by an experienced research assistant (MB) who was not familiar with the illness uncertainty literature to prevent bias. Use of the term uncertainty was avoided during the interviews. The use of telephone interviews has become more common in qualitative research (Irvine, Drew, & Sainsbury, 2013) with the advantages of allowing greater anonymity when discussing sensitive topics, cost and time effectiveness and enabling access to harder to reach groups (Sturges & Hanrahan, 2004). Telephone interviews have been found to result in similar quality data as face-to-face interviews (Sturges & Hanrahan, 2004). The interviews were conducted between October 2013 and January 2014 and lasted from 48 minutes to 2 hours and 30 minutes.

Analytical approach

The primary focus of the study was to identify themes of uncertainty described by carers when discussing their partner's Parkinson's and compare and contrast these to Cleanthous et

al.'s five domain model developed in patients. Transcripts were analysed thematically using a detailed line by line coding to examine uncertainty domains following Braun and Clarke's (2006) six-step approach. Quotations were coded as 'uncertain' on the basis of 'a lack of certainty, or any state of limited knowledge, understanding or worry regarding an existing or future outcome' (Cleanthous et al., 2013) using QSR Nvivo 10. Each quotation was then compared to the conceptual domains of uncertainty found by Cleanthous et al. (2013). New codes and domains were created where quotations expressing uncertainty did not adequately fit within the existing framework.

Reliability and validity

Instances of uncertainty were coded by the research assistant (MB) and the main author (CH) and high agreement was found across the interviews. The uncertainty domains described are supported by direct quotations to allow readers to evaluate the author's analysis.

Results

The sample characteristics are shown in table 1. All participants were from a white British background.

INSERT TABLE 1 HERE

Uncertainty Domains

The full list of domains and subdomains of uncertainty developed by Cleathous et al. (2013) can be found in Table 2 along with emerging domains and subdomains found in the present study. Each of the domains from the model are examined in detail for fit within the carer sample and extended where appropriate to reflect carer uncertainty.

Symptoms and prognosis

Uncertainty regarding symptoms and prognosis were common in carer narratives. Carers expressed uncertainty about their spouses' current health status. Carers had difficulty judging how severe symptoms were and whether they had deteriorated as they felt they were masked by medication.

"I mean the symptoms he first presented with, about 18 years ago...he may still have some of those symptoms, but the medication has masked some of those." Participant 15

Carers frequently made comparisons between their spouse's health status and other people they knew with Parkinson's viewing them as not as bad as or much worse than others. But the variable nature of PD made them uncertain how well their spouse actually was.

“...he goes to our group now and he doesn’t seem much different; he hasn’t deteriorated much at all, in my eyes, although he’s much younger than us. Whereas [my spouse] does seem to have deteriorated quite a lot.” Participant 1

Uncertainty around interpretation of symptoms was a strong theme throughout the interviews. A lack of clarity was evident around which symptoms were attributable to PD, medication, co-morbid conditions or simply aging.

“I think he has arthritis in his neck which causes him a lot of trouble at night-time, but I don’t think that’s related to his Parkinson’s, I think that’s related to old-age and arthritis – but I’m not sure.” Participant 4

Carers particularly expressed uncertainty around symptom interpretation before diagnosis and only retrospectively attributed symptoms to Parkinson’s. Limited knowledge of the symptoms of PD pre-diagnosis was evident in many participants and contributed to delays in help-seeking and diagnosis.

“The night sweats stick out in my mind and as I say, in hindsight, I think that was probably Parkinson’s.” Participant 3

This lack of knowledge of potential symptoms was seen to continue on beyond diagnosis with many carers still uncertain whether symptoms could be related to PD. This uncertainty centred on non-motor symptoms of PD rather than the more commonly known motor symptoms.

“...she’s [Parkinson’s nurse] pointed out symptoms that we weren’t sure were connected with Parkinson’s if you like; the things that have been coming up...like um, drooling.” Participant 12

There was a high level of certainty among carers that their spouses’ condition would deteriorate; the uncertainty about illness progression was how quickly this progression would

occur and what the nature of the deterioration would be. This was a major concern for almost all carers.

“Well I’m sure they will [get worse]; I mean that’s the nature of Parkinson’s isn’t it? But nobody can predict what will change or how it will change – or when, so...” Participant 3

PD was viewed by carers as having a high level of uncertainty day-to-day or sometimes within a single day. They often described their spouses as having ‘off days’ where their movement was more affected which limited their ability to plan daily activities and keep social engagements.

“Sometimes, for no reason, you can just have a bad day and sometimes he’ll just have a really good day and you can’t really work out why and just go with it really – make the most of the good days.” Participant 5

Several carers described attempting to try and identify patterns or factors which made things better or worse with limited success.

“So it might be me being...trying to be over-vigilant and spot patterns that are not there just yet – not predictive anyway.” Participant 6

Within the symptoms and prognosis domain Cleanthous et al. (2013) found that patients expressed uncertainty about the potential effect of their condition on their life expectancy. However no uncertainty regarding spouses’ life expectancy was expressed by carers. A sub-domain of symptoms and prognosis which appeared relevant to carers but did not appear in patients was uncertainty about the cause of illness. While many carers understood that PD was caused by a lack of dopamine they still expressed distress at not understanding why it should occur in their spouse.

“you know I mean I know there are different theories going on all over the place, but you know, [spouse] never worked in agriculture, he’s never worked with chemicals. So, I don’t know. I wish I did.” Participant 13

Medical Management

Medical management was a central theme in the carers’ discussions of PD. Much of their caring role involved managing medication and medical appointments. Although there was a relatively high degree of certainty that treatment was largely helpful, a considerable amount of uncertainty still surrounded drug treatment. Future changes in drug regimens were seen as very uncertain with approaches to treatment often being described as trial and error. Most carers felt that their spouse would have to change drugs in the future but what that change may be, and whether there actually was another drug to change to, was uncertain. There was an acknowledgement that Parkinson’s varied greatly between patients and individual response to drugs was equally variable. Most carers were certain that the doctors were doing all they could to manage the condition but it was inevitable that drugs would need to be frequently changed until a good response was achieved.

“I think it may be the only option to go on trial and error. You know you can double up the tablets and see if it works.” Participant 1

Some carers expressed hope for the development of a drug which may significantly improve symptoms or completely cure PD.

“And I am hoping, I’m not looking for a cure, but I’m hope – why I’m sort of interested in research, hoping that there’ll be something quite shortly that will stop it in its tracks. So, given, and then if that happens, if he can get some improvement with his mobility, then maybe we can enjoy again a reasonable quality of life.” Participant 5

There was also a lack of certainty around the potential side effects of anti-parkinsonian medication. Carers were unclear whether certain symptoms were caused by drugs, were part of another disease process, were purely psychological, or were due to PD itself.

“I think that causes considerable confusion [medication], in Parkinson's people, in certain people.... there's never much said about the confusion side of it, I don't think. And I think those symptoms sometimes can be very worrying, because you don't know whether it's the start of dementia or it is just the drugs.” Participant 10

There was also concern at the number of drugs some patients were taking and the potential effects on their health.

“I'm hoping that we can reduce the [name of tablet], 'cos I suspect it's not doing anything at all, just damaging his liver.” Participant 12

Carers showed a high level of knowledge about the drugs their spouse was taking and did not express any uncertainty about the purpose of those medications.

It was widely acknowledged by carers that medical professionals had a high degree of uncertainty when it came to managing PD. For some this resulted in negative feelings of being ‘fobbed off’ and left to help themselves, while others believed that medical staff were doing all they could to try and help patients.

“...because of the progression of Parkinson's, because it's so different in everybody, nobody, the best neurologist in the world could not look at [spouse] and tell me what's gonna happen to him. And they would admit that; they can't. There's just no way of knowing.” Participant 13

“When we speak to the professionals, the neurologist, whoever, they never really differentiate between it could be because of sensitive bladder because of prostate or it could be a sensitive bladder because of Parkinson's. They just listen and acknowledge but they don't really

define. ...we still have checks with the cancer specialist every six months, we ask about the tiredness which really came on with a vengeance with the radiotherapy, and they're not sure really whether he's still tired because of that, or whether it's the Parkinson's. To be honest with you they all seem to pass the buck to each other." Participant 4

Some carers questioned how up-to-date medical staff were on recent advances in treatment and how willing they were to be flexible in their approach, incorporating individual patient needs. This inevitably led carers to question whether their spouse was receiving the best available care. In particular carers noted uncertainty around general knowledge of PD and ability to diagnose and manage PD in general practice.

"I don't think the GP's are as educated as they should be. I think there's a bit of an area there lacking. Because they, they all say well it's...and you know I know they can't be specialists in everything but I, I don't think they are totally aware of...of um, of everything."

Participant 10

When it came to formal support from the hospital carers expressed some uncertainty about whether their spouse was receiving all of the support that should be available to them. Carers described local variation in availability of support (e.g. Parkinson's nurse visits) and long waits for appointments to see physiotherapists and speech and language therapists. Carers reported sourcing support themselves rather than being informed about available support by the hospital team or GP.

"We're becoming aware that perhaps he's not had um, everything that he should have; not had all the support that he should have had. That um, I'm now not entirely sure that I know what." Participant 12

Some carers expressed a lack of certainty around how often their spouse should expect to see health professionals (e.g. neurologist, Parkinson's nurse) and what would prompt a visit.

“There doesn’t seem to be any specific time, like we see the specialist every six months, there’s no specific time for the Parkinson’s Nurse..., I think it must have been about a year...” Participant 12

Interestingly the continuity of care sub-domain seen in patients was not found in the carer sample. This is interesting as trust in the doctor, formal support and treatment were common themes yet carers did not seem concerned about the ability to continue receiving treatment from the same specialist or team.

Self-Management

Several carers were uncertain if there was anything their spouse could do to manage their own condition. Some had attempted exercise but found that co-morbidities made this difficult or were unsure if it was having any beneficial effect.

“I really don’t know what could be done to improve it. That we’re not already doing. I have no answer to that one.” Participant 18

The self-management domain was not found to be a strong theme amongst carers. The sub-domain personal control over prognosis was not present at all in the participant responses. Potentially carers felt that there was little that patients could do to manage their own condition and held a strong medical model of the condition and treatment. An alternative explanation may be that when discussing uncertainty carers were focused on their own role, rather than that of the patient, and consequently the concept may have lacked salience to carers. Carers may be aware that their role is likely to extend and therefore though self-management by the patient maybe desirable, in reality it may appear unrealistic.

Impact

Future financial stability was a source of uncertainty for many carers, mainly due to potential nursing costs both at home or in residential care.

“I’m concerned about if he had to go into a nursing home, how that would be financed”

Participant 5

Forward planning was perceived as difficult by carers due to the unpredictable nature of PD. Many described being unable to plan short term events such as lunch dates, while others described uncertainty around longer term planning with retirement plans being constantly revised as the condition progressed.

“You never know how he’s going to feel from one day to the next. Er, so if people ask you out to their place for lunch or something, he’ll/you’ll say well look at this point in time he can manage it, but I don’t know how he will be in a week’s time.” Participant 15

“We’ve got a five...we had a 20-year plan for seeing the world, it’s a five-year plan now and we’re sort of changing the order in which we do things to um, do the perhaps more difficult things sooner rather than later.” Participant 12

Again the unpredictability of the condition led to uncertainty about the impact of PD on the patient’s mobility and functioning in the future. Most carers felt that PD had been increasingly impacting upon their spouse’s functional ability but did express some uncertainty around whether they had taken over household chores because they had taken on a caring role rather than the patient being unable to perform these activities.

As may be expected due to the different demographic profile of the current sample (married, older adults, higher proportion of males), within the impact domain, reference to uncertainty around having and raising children was not made and few references were made to uncertainty around sustaining a partner and finding and maintaining a job.

Social Functioning

Finally the social functioning domain appeared less central in carers' narratives and wasn't well supported by the data. Only one participant expressed uncertainty over disclosure of diagnosis to others and only two participants reported uncertainty around the social reaction of others to PD. This again may be a consequence of the age of the sample; a large proportion were already retired at diagnosis or were nearing retirement which would reduce anxiety about revealing a condition. Furthermore, long term conditions are much more common amongst older adults and may not be met with the same stigma that they might in a younger age group. It is also likely that friends and relatives would have come into contact with other people with PD and consequently had some understanding of the condition. The majority of uncertainty statements in this domain were about accessing social support but carers did not differentiate between support for themselves or the patient. Only two carers explicitly referred to social support for the patient.

INSERT TABLE 2 HERE

Carer Role

As may be expected, in addition to the five domains of uncertainty found in patients by Cleanthous et al. (2013), a sixth 'carer role domain' was found in the present sample. While the domains discussed above refer to the carer's uncertainty about the patient and their condition, the carer role domain specifically refers to carers' uncertainty about their own role and the impact of the condition on themselves. Participants were uncertain if they would be able to access support for themselves as carers, emotionally, financially and practically from both formal and informal sources. There was some uncertainty around whether friends and family would be willing to provide more help and support if it was needed and indeed whether they would be physically able to help out.

“There’s people in the block of flats we know who I’m sure if asked would help. Although of course many are of a certain age and probably not that able in many ways.” Participant 9

Many carers were unclear about the services that were currently available to them and mentioned finding out about services accidentally rather than being informed.

“I’m not sure what help I can get to carry out that role. Or even stop carrying out that role. But that’s the big thing isn’t it? If somebody needs medical attention, will it be there?” Participant 6

This was closely linked with spouses’ uncertainty about their ability to cope in the longer term. This encompassed coping with the physical challenges of an increasingly disabled partner and the emotional challenges brought with progression of PD. Many carers expressed worry and guilt at the idea of their partner having to move to residential care if they were unable to provide adequate care at home.

“I’m just worried that I won’t be able to manage at some stage. And as I say I wouldn’t like her to go into a home; I’d like to look after her for as long as possible.” Participant 1

Uncertainty about the ability to cope was not simply viewed as a consequence of advancing disease but also as a potential consequence of the spouse themselves experiencing long term health problems.

“You are ageing along the same, you’re not getting any younger, and you’re just hoping that your health er, will be such that you can continue in that caring role for as, as long as needs be. Because that, you know that, that really plays on your mind, you know, what-if, what-if? What if I wasn’t around? What if something happened to me? What if I couldn’t do what I’m doing? And um, that really is, very concerning.” Participant 14

Carers talked specifically about the expectations of them as a carer. They expressed uncertainty about what the role might entail in the future and if they would be able to fulfil

those expectations.

“Well there’s a certain trepidation ‘cos you don’t really know what the future holds. But you know you... that’s, that’s the role isn’t it so...” Participant 3

Finally carers expressed uncertainty about how the role had impacted upon their lives and whether this would change in the future with potential illness progression.

“Well I’ve had to cut back on what I do anyway, but I’m not sure whether that’s gonna be permanent.” Participant 1

Most carers discussed the progression of PD and increasing disability as a certainty with the uncertainty surrounding their ability to adapt to and cope with this change.

Illness Duration

The median duration of illness was 10 years. In order to explore potential changes in uncertainty with disease progression we divided the sample into two groups based on disease duration (up to 10 years (N=8) and over 10 years (N=10)). Carers in the shorter duration group reported more uncertainty around their role as a carer and the availability of support for them in the future (mentioned by 3 participants in the shorter duration group and 1 participant in longer duration group). They also expressed more uncertainty about the ability of the patient to self-manage their condition (mentioned by 5 participants) than the longer duration group (mentioned by 2 participants). These findings may be a reflection of the greater experience of the longer duration group both of the disease and as a carer.

Discussion

Consistent with the findings of Cleanthous et al. (2013) illness uncertainty in carers did extend beyond events directly related to illness (e.g. illness progression, treatment) to personal events indirectly affected by the illness (e.g. social support, financial concerns, forward planning). A new carer domain of illness uncertainty was found which involved

uncertainty around the requirements of the caring role, ability to cope with the role, support received and the impact of the role. The nature of uncertainty experienced by carers was also found to change with illness duration.

Interestingly the uncertainty domains of self-management, social functioning and impact were discussed less frequently by carers than the medical management, and symptom and prognosis domains. These findings may suggest that carers are not fully considering the social implications and impact of illness on the patient because they are considering the condition from their own perspective rather than the patient's. However the dominance of medical management is in many ways not surprising as, for example, medication plays a central role in the management of Parkinson's disease and missed doses and changes in medication can have profound effects on symptoms (Grosset, Bone, & Grosset, 2005). Therefore, it may be expected that medical management would feature heavily in carer narratives about PD. It remains unclear whether these domains are actually less important in PD or less important for carers regardless of the condition. PD patient interviews and carer interviews in other conditions would help to illuminate whether these differences such as self-management were a product of the condition or the carers emphasis on medical management.

Within the symptoms and prognosis domain carers did not express any uncertainty regarding their partner's life expectancy. There are a number of potential reasons for this finding, the participants in this study had an average age of 65 years and life expectancy issues may have been less salient in this older sample compared to younger patients. Alternatively, participants may have felt uncomfortable speculating about their partner's life expectancy. Finally, there was a high level of certainty among the sample that PD was progressive and

had a general downward trajectory, consequently they may have been certain that PD would impact on their partner's life expectancy.

Despite the differences in uncertainty identified between the present sample of carers and the patient samples used by Cleanthous et al (2013), the Cleanthous model appears useful for conceptualising carer uncertainty. The domains of the model were all identified in the carer sample with only one new domain, carer role, arising. Differences in the subdomains were evident however the Cleanthous model already allows for these to vary with illness type. Consequently we believe that the Cleanthous model provides an adequate basis for the development of a carer specific model of uncertainty of illness.

The present research identified a range of areas of illness uncertainty experienced by spouse-carers. Some of these areas of uncertainty are potentially amenable to change through education and communication interventions. Information giving interventions have seen success in decreasing uncertainty e.g. (Mishel et al., 2002). Improving access to information about the symptoms of PD, particularly non-motor symptoms and potential side effects of medication could help carers to more easily differentiate between symptoms of PD, other conditions, and side-effects of medication. Improved knowledge may enable patients and carers to engage in appropriate self-care and seek appropriate help. Improving knowledge in GPs may also enhance support and diagnosis (Abbott, Naismith, & Lewis, 2011).

The identification of a carer role domain of uncertainty highlights the importance of supporting informal carers in their role. Providing clearer signposting to formal support for both patients and carers could alleviate significant uncertainty. Indeed it has been suggested that carers could be better supported by helping them to anticipate the effects of the caring role to reduce uncertainty (Unson et al., 2015). The findings further suggest that uncertainty

may change over the course of a disease and interventions may need to be tailored to individual patients.

Although some elements of uncertainty described by carers can be addressed through improved information and communication, some are an inherent aspects of the condition and rather than attempting to provide certainty, interventions should seek to help carers to cope effectively with illness uncertainty. Mindfulness interventions or Acceptance and Commitment Therapy may be effective methods of increasing tolerance of uncertainty (Wright, Afari, & Zautra, 2009). A trial of mindfulness in PD is currently underway and will explore the impact of mindful exercises on tolerance of uncertainty and may indicate whether this approach would be useful for PD carers (Bogosian et al., in press).

It must be acknowledged that the findings of the present research are limited to the sample studied. All participants were spouse-carers and all from a white British background. Consequently the uncertainty experienced by non-spouse carers such as children, friends or other family members and those from other cultural backgrounds may differ considerably to that found in the present study. Furthermore we cannot be certain the findings are applicable across all caregivers or to what extent carer illness uncertainty may vary across conditions. However the findings do suggest that using Mishel's (Mishel, 1981, 1988, 1990) narrow definition of illness uncertainty fails to capture central elements of carers' uncertainty. It is essential that the nature of uncertainty experienced by carers is fully understood so that appropriate support and interventions can be provided to improve well-being.

The present study aimed to explore the nature of illness uncertainty in carers of people with Parkinson's. While the data allows us to draw conclusions about the domains affected by uncertainty, questions still remain regarding the impact of different aspects of uncertainty on carers and carer well-being. It is likely that some forms of uncertainty are more problematic than others, require greater coping resources, and have a greater impact upon well-being. Understanding how carers experience and manage uncertainty is important for the development of interventions aimed at the reduction, or acceptance, of uncertainty and would be usefully explored in future research.

The findings have implications for our understanding of the nature of illness uncertainty in caregivers. Uncertainty clearly extends beyond factors directly related to the illness and encompasses a range of factors associated with carrying out the caring role. Further exploration of uncertainty of illness in patients with PD and across a broader range of carers would strengthen the findings of this study, inform potential intervention strategies, and enable the development of a comprehensive model and measure of illness uncertainty in caregivers.

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Table 1. Sample characteristics

Characteristics	Carer n = 18
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Gender, n (%)	
<i>Female</i>	10 (66.6)
<i>Male</i>	8 (44.4)
Age	
<i>Mean (SD)</i>	65.4 (5.1)
<i>Range</i>	56-73
Years Since Spouse's Diagnosis	
<i>Mean (SD)</i>	10.3 (6.0)
<i>Range</i>	2-24
Years of Education	
<i>Mean (SD)</i>	15.8 (3.4)
<i>Range</i>	7-20
Years Married	
<i>Mean (SD)</i>	35.2 (14.6)
<i>Range</i>	2-50
Employment status, n (%)	
<i>Employed</i>	1 (5.5)
<i>Retired</i>	17 (94.5)
<hr/>	

Table 2. Uncertainty domains and subdomains from Cleanthous et al. (2013) and emerging domains and subdomains in PD carers

Domains	Subdomains	Explanation
Carer uncertainty about the patient:		
Symptoms and prognosis <i>Illness characteristics, course and progression</i>	Interpretation of health status	Judging how well they are
	Symptom interpretation	Interpreting and labelling physical sensations, distinguishing from other conditions, SEs etc
	Life expectancy*	Unsure of effect on LE
	Illness progression/future	Unsure of future severity
	Predictability of health status	Judging and predicting short and long term health status
	Cause†	Uncertainty around the cause of Parkinson's
Medical management <i>Ability of drs to manage the condition and formal care received</i>	Treatment (necessity, effectiveness and side effects)	Uncertainty over treatment regime, are medications absolutely necessary, unsure of purpose of medication
	Trust in doctor	Consultants knowledge and ability to help their condition, drs uncertainty over prognosis and around initial diagnosis
	Formal support	From the hospital e.g. phone lines etc, uncertainty around the meaning and importance of medical tests
	Continuity of care*	Uncertainty about being able to see the same doctors in future, to get care abroad
Self-management <i>Ability to manage the condition oneself</i>	Personal control over prognosis*	How much control they have over their prognosis
	Management of condition	Is there anything more they could do, what they should or should not

		do, how much to push themselves
Impact <i>Impact of the illness on the patient's life, mostly future based</i>	Occupational/financial	Maintaining, finding a job, financial stability
	Having and raising children*	Physical stamina, fertility
	Finding/sustaining a partner*	Burden of chronic illness on a relationship, finding a new partner with a diagnosis
	Forward planning	General life planning, short term and long term, attending events etc
	Functionality/mobility	Future mobility and day to day physical mobility – impact on ADLs
Social functioning <i>Impact on social aspects of life</i>	Social support	Can close friends and family be counted on and how much support can be expected
	Social reaction	Uncertainty of understanding of the condition, invisibility of symptoms
	Disclosing diagnosis	Employers, social circle, potential partners, expectation of negative implications
Carer uncertainty about themselves		
Carer role† <i>Uncertainty around the potential requirements of the role, ability to cope with the role, support received and impact of the role</i>	Accessing support†	Ability to access support (formal and informal), support provision in the future
	Coping†	Ability to cope physically and emotionally, particularly with future illness progression
	Impact†	Impact of the carer's new role on their life and the marriage
	Role requirements/expectations†	Being good enough, what is expected of them

†Emerging domain/subdomain *Subdomain not found in PD carers

Supplementary Material

Topic List

Dimension	Items
Symptoms (past – present – future)	cause - diagnosis condition-specific unpredictability progression ambiguity
Treatments (past – present – future)	effectiveness side-effects trial&error link with symptoms relapse & change ambiguity concerns
Clinician Interaction	Trust/competency Interpretation of behaviour continuity of care how to approach when to visit the doctor being taken seriously communicating effectively provision of care support preferred degree of involvement satisfaction with care
Impact (past – present – future) (Patient)	Recreational activities functionality productivity social status interpersonal relationships

	<p>employment</p> <p>financial</p> <p>psych. Impact</p> <p>physical limitations</p> <p>living arrangements</p> <p>life changes</p> <p>adjustment</p>
Impact (past – present – future) (Carer)	<p>Recreational activities</p> <p>productivity</p> <p>social status</p> <p>interpersonal relationships</p> <p>employment</p> <p>financial</p> <p>psych. Impact</p> <p>marital relationship (role changes)</p> <p>living arrangements</p> <p>life changes</p> <p>adjustment</p>
Self-management (past – present – future)	<p>present & future</p> <p>what can & can't do</p> <p>what should and should not do</p> <p>exercise</p> <p>life-style issues</p>
Caring role (past-present-future)	<p>Present & future</p> <p>Ability to cope (Psychological & physical)</p> <p>Commitment</p> <p>Care decisions</p>
Information	<p>source / word of mouth</p> <p>acknowledgment of uncertainty in the</p> <p>clinical setting</p>

	quality & style of information received deficiency of information
Social Support	Family reactions Reliability Security Actual level of support preferred level of support acceptability
Understanding/comprehension condition	of ambiguity, complexity, unpredictability and deficiency of information regarding: symptoms, current state of illness, treatment/treatment efficiency

Always keep in mind:

less

more

uncertainty

Interview Schedule

Use the main questions to initiate conversation, if the person spontaneously mentions the issues you don't need to ask about them directly. Only use the questions to probe areas that have not been covered.

Introduction

I'm interested in your experiences as a carer of someone with Parkinson's and what you understand and think about their Parkinson's. I'll be asking you quite broad questions. There are no right or wrong answers, I'm just interested in your thoughts.

****Remember to keep in mind certainty, uncertainty throughout e.g. if someone says back problems caused by husband to quit work probe how sure they are that this was related to PD****

Symptoms:

Could you tell me a little about the symptoms that they experience?

(have they changed over time, do you think they will change in the future?)

Are you clear about which symptoms are related to their Parkinson's? (ask about change over time, how sure are you that they are related to PD?)

What do you think is the cause of the symptoms they experience (Parkinson's)? (ask about change over time)

Are their symptoms predictable? (ask about change over time)

Treatments:

Could you tell me a bit about the treatment they currently receive for their Parkinson's? (ask about change over time)

How effective do you think their treatment is?

Do they experience any side effects? (How sure are you that these are side effects?)

Do you have any concerns about their treatment?

How does the doctor decide what treatment is most suitable?

Have they had to change their treatment? (will they have to?, what might cause a change?)

Clinician Interaction:

Can you tell me about the care your partner receives for their Parkinson's?

How satisfied are you with the care?

If your partner needed to see a doctor or nurse to discuss an issue do you know who to approach and how to get hold of them?

When you do see the doctor/nurse are you able to ask the questions you want to and get the help that you need?

Are you always clear whether or not you need to ask for help with an issue?

Do you feel that the doctors/nurses are always able to give you the right advice?

Do you feel they have enough support? (How much support would you prefer?)

Impact (patient):

How has Parkinson's affected your partner's life? (has it always had that impact, might it change in the future? (how sure are you?))

Has it affected their work at all (did it in the past?)

[do they get as much done, has it affected their relationships, recreational activities finances, social standing, living arrangements (ask about past, present and future)].

How has Parkinson's affected them psychologically?

How has Parkinson's affected them physically? (only asked if not covered previously e.g. symptoms)

Have they had to make changes to their lives?

How much do you feel they have adjusted to the impact of Parkinson's? (will they in future?)

Impact (carer):

How has their Parkinson's affected your life? (Has it always impacted on you? Do you think it will impact on you in the future?)

Has it affected your work at all?

[do you get as much done, has it affected your relationships, recreational activities, finances, social standing, living arrangements (ask about past, present and future)].

How has Parkinson's affected you psychologically?

Have you had to make changes to your life?

How much do you feel you have adjusted to the impact of Parkinson's? (will you in future?)

Has Parkinson's affected your relationship with your partner?

Self-management:

What kinds of things can your partner do to try and manage their Parkinson's themselves?
(What could they do in the past, what might they be able to do in the future?)

Are there certain things that they should avoid doing that will make their symptoms worse?

Are there things that they should do to make their symptoms better?

Are there any changes to their lifestyle that they have made (or could make) which would help them to manage their Parkinson's e.g. diet, exercise etc.

Caring role:

Can you tell me a little bit about your role as a carer for your partner? (have you always done that, do you think that might change in the future?)

How easy do you find it to cope in your role as a carer (physically and emotionally)?

Do you feel you know what is expected of you as a carer or what might be expected of you in the future? How do you feel about that role?

How confident do you feel to make decisions about your partner's care?

Information:

Where do you get most of your information about Parkinson's?

What are your thoughts about the information you have received from the doctor/nurse about Parkinson's? (and the way you received the information?)

Can doctors/nurses always be certain about the information they give you in clinic?

Do you feel you get enough information about Parkinson's?

Social support:

Do you get much support from others? (would you if needed?)

Would you prefer more support than you get now? (or less?)

Is the support that you get reliable?

How secure do you feel they you would have the social (and/or financial) support you needed?

How easy do you find it to accept support?

How did your family react when your partner was diagnosed with Parkinson's?

Understanding/comprehension of condition:

Overall, how well do you feel that you understand your partner's Parkinson's?

(ask in detail about the following if not already covered above)

How well do you feel your understand their symptoms (whether they are definitely due to Parkinson's, when they are likely to occur, how best to manage them)?

How well do you understand their treatment? (how effective it is, how effective it might be in the future, what the side effects are)