Palliative Care in Parkinson's Disease: Developing a needs assessment tool

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Thesis Abstract

Title: Palliative care for Parkinson's disease: developing a needs assessment tool

Background: Parkinson's disease (PD) is a common, life-limiting, neurodegenerative condition. Despite calls for improved access to palliative care, provision is lacking, due in part to poor understanding and recognition of palliative care needs. Where services exist, negotiating the chronic-palliative interface is challenging.

Aims: **1)** To establish the palliative needs of people living with PD.

2) To adapt and clinimetrically test a palliative assessment tool for PD.

Method: A sequential mixed method design was adopted:

Aim 1: Systematic review and synthesis of qualitative evidence and primary qualitative study (focus groups and semi-structured interviews). Combined findings used to adapt a palliative care assessment tool for Parkinson's disease.

Aim 2: Face and content validity tested by expert panel. Construct validity examined in 50 people with PD and their carers. Inter-rater reliability examined in broad range of clinicians using video consultations.

Results:

Aim 1 – Four lines of argument were identified: i) Information tension, ii) care tension, iii) inter-personal negotiations, iv) intra-personal negotiations. The response to diagnosis, carer vigilance and presence of disease milestones were highlighted.

Aim 2 - Construct validity was good (tau B > 0.6) for two constructs, moderate (>0.4) for five and fair (>0.2) for the remainder. Reliability as measured by kappa was moderate (kappa >0.4) for four, fair (>0.2) for five and poor for four constructs. Two constructs with poor kappa are explained by extremely high percentage agreement.

Conclusions: Adaptation of the NAT:Parkinson's disease has been successful, with clinimetric properties supporting use by a range of clinicians. Future studies should examine utility within integrated services.

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<u>Authors declaration</u>

I confirm that this work is original and that if any passage(s) or diagram(s) have been copied from academic papers, books the internet or other sources these are clearly identified by the use of quotation marks and the reference(s) is fully cited. I certify that, other than where indicated, this is my own work and does not breach the regulations of HYMS, the University of Hull or the University of York regarding plagiarism or academic conduct in examinations. I have read the HYMS Code of Practice on Academic Misconduct, and state that this piece of work is my own and does not contain any unacknowledged work from any other sources. I confirm that any patient information obtained to produce this piece of work has been appropriately anonymised.

Academic outputs related to thesis

	NUMBER	DETAILS
Publications **Peer reviewed articles**	**1**	Richfield E, Alty J, Jones E. Palliative care for Parkinson's disease: A summary of evidence and future directions. Palliative Medicine 2013: 27(9); 805–810
Published abstracts	**4**	Richfield E, Adams D, Jones E, Johnson M, Campbell C. Specialist palliative care for Parkinson's disease: experiences of a novel integrative service. Age Ageing (2014) 43 (suppl 1): i7.doi:10.1093/ageing/afu036.28 Richfield, E., Flemming, K., Badger, N., Johnson, M.; What are the supportive and palliative care needs of people with Parkinson's disease: Systematic review and synthesis of qualitative research? [abstract]. Movement Disorders 2014;29 Suppl 1 :453 Richfield E, Girgis A, Johnson M. Assessing Palliative Care in Parkinson's Disease - Development of the NAT: Parkinson's Disease. Palliative Medicine 2014: DOI: 10.1177/0269216314532748 Adams D, Richfield E, Johnson M, Jones E & Campbell C. A specialist palliative care service for Parkinson's disease: Review of end of life outcomes. 10.1136/bmjspcare-2014-000654.307 [abstract]
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National	**4**	**International Poster presentations:**
		Movement Disorders Society International Congress, June 2014, Stockholm
		European Association of Palliative Care World Congress, June 2014, Lleida
		National oral presentations
		British Geriatric Society (BGS) Movement disorder section – Birmingham, January 2015
		End of Life in Parkinson's disease – Scarborough, November 2014
		"Parkinson's 2014" – London, September 2014
		Norwich and Norfolk Palliative Care Conference – Norwich, April 2014
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Palliative Care in Parkinson's Disease:

Developing a needs assessment tool

Chapter 1 Introduction

1.1 Introduction to Parkinson's disease

Parkinson's disease is a degenerative neurological condition, affecting up to 274 per 100,000 of the population in the UK(1). The disease is more common in older people, with an estimated prevalence of 1% in those aged greater than 60 years, and up to 1 in 10 of residential or nursing home residents(2). Moreover with an aging population, the prevalence of PD is expected to rise so that, by the year 2030, it is estimated to be more than double that of 2005(3).

The traditional model of Parkinson's disease as a primary motor disorder has led to improved therapeutic strategies and better control of movement for many patients. Subsequently there has been an increased emphasis on the previously neglected non-motor features of the disease. These non-motor features affect many aspects of a patient's physiology, contributing significantly to the disease burden for patients, care-givers and family members, with the combined impact of motor and non-motor features being felt across all life domains. It is vital that new and existing PD services develop to reflect this deeper understanding of the disease, incorporating holistic approaches to care and focusing on the patient beyond their motor symptoms, to improve the experience of those living and caring for PD.

PD is a disease that remains incurable and the requirement for palliation, if not specialist palliative care, has been recognised for some time. For example, the pragmatic approach to disease staging described by Macmahon and Thomas(4) described the final stage of PD as "palliative", during which time the response to dopaminergic therapy wanes, medication may be tapered, and emphasis of care is switched to palliation. In keeping with the chronic nature of PD, this stage is estimated to last on average 2.2 years(5).

More recently there has been a move to extend palliative care approaches, to promote quality of life throughout life threatening illness and a drive to improve access to

palliative care for non-malignant conditions. These changes challenge health professionals to apply palliative principles, where necessary, throughout the disease course and are incorporated in the NICE guidelines for Parkinson's disease(6).

1.2 What is palliative care?

Palliative care has been defined by the World Health Organisation:

> *"Palliative care is an approach which improves the quality of life of patients and their families facing life-threatening illness, through the prevention, assessment and treatment of pain and other physical, psychosocial and spiritual problems.(7)"*

In the past there has been, at least amongst more traditional medical specialities, the tendency to utilise palliative care only when "active" medical treatment - which is to say treatment aimed exclusively at cure, had been exhausted. This view promoted a dichotomous approach, whereby clinicians would focus on actively treating the disease before referring to palliative care once these treatments had failed and death seemed inevitable. The costs associated with this approach are clear, with patients and families ill prepared for death and opportunities to alleviate physical, psychological and emotional suffering potentially missed.

Subsequently the emergence of a new paradigm, in which health care professionals are expected to adopt a more holistic approach at all stages, has altered expectations. The incorporation of palliative care beyond the terminal phase of disease is embodied in an extended version of the WHO definition which states that palliative care:

> *"…is applicable early in the course of illness, in conjunction with other therapies that are intended to prolong life, such as chemotherapy or radiation therapy, and includes those investigations needed to better understand and manage distressing clinical complications.(7)" (World Health Organisation)*

No longer the sole preserve of specialist teams, a palliative approach can now be applied in conjunction with, and complementary to, traditional models of care. This trend may be most apparent in medical specialities such as Medicine for the Elderly, where palliative principles form part of the UK training curriculum, and a holistic perspective to care is encouraged.

It is important here to clarify the distinction between a "palliative approach" and Specialist Palliative Care (SPC). Using our example of Elderly Medicine; trainees would develop an understanding of palliative principles and their application, but would not have recourse to more advanced palliative techniques which remain the preserve of palliative care specialists. So, for example, they may feel comfortable engaging in discussions related to end of life care, but not have the skillset to conduct formal Advance Care Planning (ACP); they may use standard medications for the management of terminal symptoms, but require support when these are unsuccessful and a more tailored approach is required. Thus we can appreciate that a "palliative approach" may be applied by all health professionals, where appropriate, according to their skillset, while Specialist Palliative Care is a more clearly defined entity, and both are encompassed by the broad term of palliative care.

This model is borne out by the National Council for Palliative Care which provides the following definition:

> *"Palliative care is provided by two distinct categories of health and social care professionals:*
>
> • *Those providing the day-to-day care to patients and carers in their homes and in hospitals*
>
> • *Those who specialise in palliative care (consultants in palliative medicine and clinical nurse specialists in palliative care, for example)*
>
> *Those providing day-to-day care should be able to:*
>
> • *Assess the care needs of each patient and their families across the domains of physical, psychological, social spiritual and information needs*
>
> • *Meet those needs within the limits of their knowledge, skills, competence in palliative care*
>
> • *Know when to seek advice from or refer to specialist palliative care services"*

(National Council of Palliative Care(8))

1.3 Palliative care in chronic disease

There is increasing recognition that the principles of palliative care, traditionally associated with malignant conditions, can and should be applied to many areas of

medicine, particularly chronic, life shortening diseases. The movement to identify patients with chronic disease, who would benefit from palliative care, has led to the evolution of SPC services aimed at patients with conditions such as dementia, heart failure, chronic obstructive pulmonary disease and degenerative neurological conditions.

The need for Specialist Palliative Care in Parkinson's disease was first explored using qualitative methods, by Hudson et al, who demonstrated that the challenges faced by PD patients and families are similar to those faced by palliative care cancer populations(9). In interviews with patients, carers and health professionals, five themes were identified:

- Emotional impact of diagnosis,
- Staying connected – highlighting difficulty with communication with the person with PD, but also between caregivers,
- Financial hardship,
- Managing physical challenges,
- Finding help in the advanced stages of disease.

They concluded that palliative care should be extended to PD populations, but, that using prognosis as a trigger for palliative care referral is suboptimal in a cohort of patients with complex chronic disease, who have an uncertain disease trajectory. This challenge is discussed further below.

Further evidence for inadequate palliative care provision for patients and families living with PD is provided by work in the UK looking at place of death. PD patients in the north of England were more likely to die in residential or nursing care and less likely to die at home than non-PD controls from the same population, while none of the PD patients in this study died in a hospice(10).

This last point contrasts with work in the US where 53 per cent of caregivers reported that their relative with PD received hospice care in the final 2.5 weeks of life. Moreover those who received hospice care were more likely to have received adequate analgesia(11).

The implied deficiencies in care in the UK represent a challenge to health care providers and commissioners responsible for Parkinson's disease.

1.4 Palliative care needs in PD

A better understanding of the palliative issues facing patients and carers with PD is required if SPC provision is to be extended effectively.   Following the work of Hudson et al cited above, qualitative studies have tended to focus on the experiences of informal carers.

One Canadian study highlighted a perceived lack of support from health care services(12). The theme *"missing information"* applied to poor communication and provision of information at the time of diagnosis, and also to a lack of direction and support in accessing social services and funding. Carers strongly emphasised the need for a multi-disciplinary approach to PD care, as well as more guidance and support around the progressive nature of the condition.

When reviewing this work it is important to consider the setting, a tertiary centre in Canada, where the authors report that 90% of patients do not have government funded services. This makes it difficult to draw parallels with a UK population with far greater access to state healthcare provision. None the less, similar themes emerge from a later study in Northern Ireland where carers identified a lack of communication from the time of diagnosis, the financial burden of the disease and difficulty accessing social support and funding(13).

The same authors went on to analyse end-of-life experiences by interviewing carers of patients who had died with PD(14). Poor communication from professionals was again highlighted, this time relating to a failure to direct patients towards available services, or help carers prepare for the later stages of disease. Carers indicated that they did not feel prepared for bereavement, and that supportive services stopped at the time of death, adding to a sense of isolation.

Thus despite being grounded in different health cultures, there are common echoes across these three studies of patient and caregiver experience. The triangulation of these results is important and suggests that suggests that the findings of these individual qualitative studies may be applicable beyond the narrow confines from which they were drawn.

However, one important criticism which cannot be overlooked when considering the validity of this body of work is the relative under-representation of people living with Parkinson's disease. Most work exclusively addressed the experience of caregivers, while the study which sought to define both caregiver and patient experience included only 3 people with PD, one of whom had severe dementia, prohibiting direct participation. Studies of palliative care needs, focused at the end of the disease trajectory, will inevitably encounter difficulties in recruitment and inclusion of individuals with advanced disease. However future studies must seek to overcome these practical and ethical challenges if conclusions are to influence practice.

An understanding of the qualitative evidence relating to palliative and supportive care in PD is central to this project. Chapters 2, 3 and 4 set out the methodology, methods and results respectively of a systematic review and qualitative synthesis, exploring this topic fully.

A quantitative approach has also been used to assess caregiver perception of specific symptoms and decision making at end-of-life. Conducted in the US, researchers used assessment tools completed at interview with carers of patients who had died from PD at least 6 months previously(11). They found that bulbar symptoms, such as dysphagia and communication difficulty were common, while 42% of carers reported significant pain, of whom a third felt that their loved one had not received analgesia in the last month of life.

It is also notable that, in this north American population, 48% of carers felt patients had been unable to make decisions in the last month of life, and that the vast majority of patients, 92%, had a living will or advanced directive in place. Anecdotal evidence in the UK suggests that use of advanced care directives is far less frequent. In a condition such as PD where cognitive impairment is often prominent in the latter stages, failure to promote the use of advanced care planning, and advanced directives is likely to represent a significant loss of patient autonomy.

The same tools were subsequently used to gather information on patients who had died from Amyotrophic Lateral Sclerosis (ALS or Motor Neuron Disease) in whom palliative care is a more established discipline(15). Comparison of perceived patient experience for these two neuro-degenerative disorders revealed similar levels of suffering at the

end-of-life. However PD patients were perceived as more likely to experience confusion, be less alert and less aware of imminent death(15). Despite these findings PD patients on average spent shorter times in hospice care.

There are clear differences between this US population and that in the UK. None the less a demonstration of similar levels of need between patients with Motor Neurone Disease and PD is a significant finding, adding weight to the call for SPC services directed at Parkinson's disease. Moreover the prominence of confusion, pain, and impaired mental status in PD patients suggests an important gap in service provision for people with PD.

1.5 Supportive care needs in PD

In addition to the body of work describe above there have been attempts to analyse the supportive care needs of PD patients and their caregivers. Kristjanson et al used a postal questionnaire to assess supportive care needs in four neuro-degenerative conditions: Multiple Sclerosis, Parkinson's Disease, Motor Neurone Disease and Huntington's Disease(16). This study design had the advantage of being able to both identify supportive care need and cross reference findings in PD with other similar conditions.

The questionnaire was generated through semi-structured interviews with patients, carers and health professionals, and was supplemented by the use of established tools to assess quality of life, anxiety and depression and caregiver health. Unfortunately no detail is provided regarding the findings of the semi-structured interviews and the data, which would add significantly to the body of work described above, appears to be unpublished.

At first glance the results are surprising. When assessing the requirement for assistance with activities of daily living, patients with PD, on average, scored all aspects as requiring either 1 (no help) or 2 (occasional help). While practical activities such as housekeeping, finances and transport were scored higher than physical activities such as washing and toileting.

This trend is repeated when asked to assess the importance of supportive services, where only *information provision* was ranked as important by both carers and patients. In addition the study found little evidence of caregiver distress, with low scores for

caregivers on the General Health Questionnaire, and appears to contradict the view that significant unmet need is present in the PD population.

However the findings of this study, suggesting relatively low levels of supportive care need as assessed by patients and caregivers with PD, are probably explained by the study design, and the introduction of systematic bias. Restrictions placed on the authors by the local ethics committees meant that newly diagnosed patients and those with severe disease were excluded from the trial, which has an obvious impact on the external validity of the findings. In addition the low response rates (25% of patients and 19% carers) meant that they recruited far fewer individuals than intended, and is likely to have introduced bias, given that those with high care needs may have been less likely / able to respond. Finally, surveys were distributed through the disease associations, the Australian equivalent of Parkinson's UK. It is likely that individuals in touch with these organisations will also have had access to, and be more aware of other services and benefits, and therefore less likely to have unmet needs.

Therefore although these findings offer a counterpoint to the other literature regarding the requirement for increased palliative services in PD, they should be interpreted with caution.

Several review articles also highlight common issues confronted in advanced PD, and suggest management strategies(17-19). These demonstrate the wide range of issues which may require SPC input, alongside specialist neurological review. Highlighted issues include:

- Physical issues such as freezing, "on – off" phenomena, falls, dystonia, dysphagia and pain,
- Neuro-psychiatric issues such as dementia, anxiety, and hallucination,
- Sleep disturbance,
- Autonomic dysfunction,
- Communication difficulties,
- Ethical issues such as nutritional and ventilatory support at end of life, advanced care planning, and capacity assessment.

It is apparent that many of these issues can be dealt with effectively in specialist neurological clinics, which in the UK often include Elderly Care physicians who are familiar with a more holistic approach. However the qualitative data presented above, would tend to suggest that this is not uniformly the case at present.

1.6 Models of Care

An integrated model of care, whereby clinicians are able to address palliative issues as their expertise allows, while referring to SPC services, from the time of diagnosis, those individuals that require specialist input, seems both feasible and desirable(20-22). Such a model would be dependent on professionals managing PD on a daily basis being able to identify unmet palliative needs and reliably discriminate those which require specialist intervention, from those which can be met within their own sphere.

To allow this model to function, particularly in a resource limited health care environment, the respective roles of chronic care and specialist palliative care and, moreover, the transition from one to the other, appears pivotal to the development of sustainable services, particularly as palliative care strategies are extended to large cohorts of patients with chronic illness. This is discussed by Lanoix, in the context of PD in Canada(23). She considers the different goals and methods of chronic versus palliative care, characterising the former as aiming to improve functionality in addition to quality of life, with a heavy reliance on informal or family care givers, which shapes the way in which patients access services. According to Lanoix's model, the attitude towards the informal caregiver appears central. It describes a situation whereby chronic care services rely upon, and benefit from, but fail to fully acknowledge informal caregivers, in contrast to palliative care which seeks to support and nurture the caregiver as much as the patient. The proposed advantage of the palliative approach being that caregivers are able to participate more fully in the advanced stages of the disease, acting as both carer and advocate for patients whom may be less able to advocate for themselves as the illness progresses. Moreover she argues that failure to adequately support carers in the earlier stages of the disease may leave them unable to cope with participation in the latter stages, even when support strategies are subsequently put in place. Thus part of an

effective palliative strategy for a patient with PD would be to identify and eliminate obstacles to caregiver wellbeing throughout disease course.

1.7 Accessing palliative care: Negotiating the chronic / palliative interface

As outlined above it is evident that palliative techniques can and should be applied to patients with many chronic, life limiting conditions. Meeting this requirement solely through specialist palliative care services is neither practical, through volume of work, nor desirable ethically, as all doctors should aspire to best practice in end of life care, as enshrined by the GMC(24).

Managing the interface between chronic care and specialist palliative care services will be key, not only to the sustainability of specialist services, but to raising the standard of care for all patients by enhancing skills in non-specialist practitioners. In order for provision to grow the way in which patients move between specialist and non-specialist services must be clearly defined. In this regard developing PD services can learn from the experiences of other chronic conditions where services have already started to evolve.

In the UK a "prognosis based" system, whereby patients with any disease, felt to be approaching the final stage of their illness are identified for increased palliative input, is already in operation as part of the government's Gold Standard Framework. This can be problematic as it is notoriously difficult to accurately anticipate the final stages of illness, and clinicians are often reluctant to predict death. This so called "prognostic paralysis" has been identified as a barrier to the application of palliative approaches, and so professionals are encouraged to ask the question *"Would I be surprised if this person died in the next 12 months"* – where the answer is no, a process leading to palliative interventions should be initiated(25).

This system has obvious advantages in terms of simplicity, allowing professionals from many health backgrounds the opportunity to initiate discussion of palliative strategies. Unfortunately the simplicity of the approach is also a major disadvantage, whereby any unmet needs occurring prior to the last year of life are likely to be overlooked. This is particularly pertinent in PD, where we have demonstrated that palliative issues may occur at an early stage and dementia is frequently present(26), requiring earlier intervention if patients are to be fully involved in decision making(27).

An alternative approach, such as that proposed in Congestive Heart Failure (CHF), uses so called "sentinel events" as a means of identifying patients in whom assessment of unmet palliative need should take place(28). In essence, the occurrence of significant episodes such as admission to hospital with decompensated disease, are used as "red flags" to highlight underlying disease progression and worsening prognosis. This is a more nuanced version of the prognosis based system, and could be effective in any condition where significant milestones, associated with worsening prognosis, are readily identifiable.

Finally, a "fully integrated", problem centred model has also been advocated(20). In this system patients can move fluidly between chronic care and SPC services as their needs change. In order to function effectively this would require sensitive, reliable methods of screening for unmet need, an understanding of when and how to apply any screening tools, and the ability to discriminate those needs which require SPC input. It also anticipates flux of patients in both directions, between chronic and SPC services, at multiple time points during their disease course. The increased subtlety of this method comes at the expense of simplicity, and would undoubtedly increase the complexity and difficulty of implementation.

1.8 Obstacles to palliative care in PD

Obstacles to palliative care provision in PD may be structural, in terms of the availability of specialist services, but also psychological in terms of patient and carer perception of palliative care. Patients may associate hospice care with cancer, or be resistant to the idea of "palliative care" because of connotations of terminal decline or end-of-life(14). These obstacles are likely to be particularly pertinent to a model of care which tries to incorporate SPC services early in the natural history of PD, and may require some rebranding in order to make them accessible and acceptable to patients.

1.9 Conclusions

The case for increased palliative provision in Parkinson's disease is compelling. It is driven by broader definitions of palliative care, and changing expectations, at a governmental and individual level, of what constitutes good care for chronic life limiting conditions.

Following on from this there is limited evidence in relation to specific palliative care needs in PD. Provision of information from diagnosis through to bereavement, communication, caregiver support and advanced care planning appear to be areas which would benefit from SPC involvement. Physical issues such as pain and swallowing, as well as the recognised motor and non-motor aspects of PD become more problematic in advanced disease.

Dementia is common in PD, particularly in the latter stages, and highly pertinent to the discussion of palliative care needs. Strategies which identify need on the basis of prognosis are likely to result in far lower rates of patient participation in decision making, with loss of autonomy. As such an integrated model of care, with a flexible approach which is able to rapidly escalate at times of high demand, and taper off as specific issues are addressed, should be the aim for evolving PD – SPC services.

For this to function effectively, a systematic approach to identification and triage of palliative care need is required, this does not currently exist for PD.

1.10 Aims and Objectives of thesis

Having recognised this gap, it was felt that a *clinician* operated tool which could be used by non-palliative specialists, in everyday practice, would be most appropriate. The Needs Assessment Tool: Progressive disease cancer, NAT:PD-c (see appendix 1), was identified as a solution which had already been developed for cancer and other non-malignant disease (see chapter 5). The NAT:PD-c is a short, clinician completed assessment tool, which was developed for just this purpose in cancer[29] and has subsequently been adapted for heart failure[30]. Consisting of a single page, it can be applied rapidly in everyday clinical practice, without increasing consultation times[31]. The format has two distinct phases, encouraging the clinician to first explore their level of concern relating to the presence of unmet palliative care need in each of 13 domains and then to triage this need, according to whether they can address it themselves (palliative approach) or whether it requires referral to a specialist service.

1.10.1 *Aim*

The overall aim was to produce a new, practical method of identifying those patients who would benefit most from palliative care, in order to address the barrier to service development discussed above. The specific objective was to adapt the NAT:PD-c for Parkinson's disease and establish the clinimetric properties of the adapted tool.

1.10.2 *Objectives*

1- To adapt the NATPD-c for use in Parkinson's disease by:

i) Systematic review and qualitative synthesis of the literature pertaining to palliative and supportive care in PD (chapters 2-4)

ii) Conducting a new piece of qualitative research, seeking to build upon and address the gaps within the existing literature base (chapters 2, 5 - 6)

2- Examine face and content validity of the adapted tool by consulting experts in the field (chapter 10)

3- Examine clinimetric properties, namely:

i) Construct validity (chapter 8-10) and,

ii) Inter-rater reliability (chapter 8, 11-12)

Therefore the two research questions for this project were:

Question 1

"What are the palliative and supportive care needs of people living with Parkinson's disease?"

Question 2

"What are the clinimetric properties of the NAT: Parkinson's disease"

A mixed methods approach was adopted for the three distinct phases of the project. These sequential phases of the project are finally drawn together in a discussion chapter (chapter 13) and the implications of the project as a whole in terms of individual clinical practice, service structure and development are discussed in the final chapter (chapter 13) along with suggestions for further research developments which could build upon this work.

Chapter 2

Methodology: Qualitative phase

2.1 Introduction

The first phase of the project was the systematic review and synthesis of qualitative literature, followed by a primary qualitative study. This chapter examines some of the methodological debates underlying qualitative research and demonstrates how these influenced the methods used in each section of the project. This is important, not only in establishing the basis upon which this phase of the project was conducted, but also to promote a reflective approach to the project as a whole – without first setting out the methodological underpinnings of the project, it would be impossible to reflect on the way in which these choices influenced the research findings.

2.2 The fundamentals: beliefs about knowledge

To answer questions about the natural or social worlds around us, we must first understand the types of questions we can ask, and the way in which we will evaluate the "truth" of the answers we receive.

These fundamental questions can be thought of in two distinct, but related categories; namely Ontology and Epistemology.

2.2.1 Ontology: Questions of structure

The term Ontology is defined as:

> *"The branch of metaphysics dealing with the nature of being.(32)"*

In research terms it leads us to question the nature of the world we wish to study and leads to the question "What can we know?"

This will depend on the extent to which we believe the structures of the world we experience to be *externally* determined. Are there naturally occurring categories and

orders which exist and which are present regardless of individual experience or constitution? Answers to this form a continuum, from a highly ordered, externally determined world at one extreme, to an internally constructed and individually experienced one at the other(33).

Figure 1: Illustration of Ontological position

(Definitions adapted from Bryman – Social research methods(33))

Objectivism   Constructivism

| Holds that there are natural occurring structures and phenomena, which exist outside of, and are thus common to, the experience of different individuals. Implies that these phenomena can be known and studied. | Holds that phenomena are individually experienced and that social structures are constructed by the individual, according to their past experiences, beliefs etc. Implies that it is not possible to study or know a single common world. |

Another important step is to decide whether or not the beliefs, assumptions and rules, which we determine for the *natural* world can also be applied to the *social* world, or whether this requires a new, distinct line of argument(34, 35).

From a personal perspective, rather than apply a single ontological position to all aspects of life, it makes greater sense to consider the qualities of a particular area of study and determine, on the basis of this, the degree to which we believe it can be known, studied and generalised.

This concept of a fundamental difference between the natural and social worlds is important and will determine, to a large extent, the answer to our second key question *"How can I know the world?"* which is called epistemology.

2.2.2 *Epistemology: Questions of approach*

The term Epistemology is defined as:

> *"The theory or science of the method and ground of knowledge. It is a core area of philosophical study that includes the sources and limits, rationality and justification of knowledge.(36)"*

It naturally follows on from the ontological considerations discussed above and has a number of key positions, described below.

2.2.2.1 *Positivism*

The term positivism is often used to describe a particular method, developed for study of the natural world, the principles of which are now very much engrained in our consciousness; to the extent that, for many, they constitute the "scientific method".

Positivism, as a way of studying the world, is often aligned with an objectivist worldview; that phenomena can be examined and proven in a way which is free of cultural and social values(34, 37). In turn, a positivist position holds that we can ascertain the likely truth of our hypothesis through our failures to disprove it. This *empirical method* is the cornerstone of most scientific practice and the marker of good practice(35). It allows us to judge the credibility of a piece of research, by evaluating the degree to which practitioners have managed to achieve "value free" examination of the hypothesis in question – studying in isolation from the social, political and historical contexts which may introduce bias.

2.2.2.2 *Realism*

A Realist epistemology accepts many of the tenants of Positivism – for example the existence of an external reality and suggests that the social world can be studied in a similar manner to the natural world(38).

2.2.2.3 *Idealism*

An Idealist position rejects the principle that the social world can be *known* in any objective way and, instead, suggest that there are multiple versions of social reality,

dependent of the perspective of the individual(34). This implies an epistemological subjectivity – we can only know the world through the prism of our own self (experiences, bias, beliefs etc.) and this knowledge will thus differ from that of any other individual.

2.2.2.4 *Post-modernism*

To take a step further along the continuum, is to suppose that there is no common reality; that the social world which an individual inhabits is constructed entirely of their own subjective experiences. In research terms, this suggests that no one opinion, or version of reality is any more valid than another. It implies that attempts to study the social world are futile, given that the claims to truth or knowledge of a researcher are of no greater validity than those of any individual recounting their own experience(39, 40).

These alternative ways of knowing the social world, which seek to account for the contextual nature of the world, to a greater or lesser degree, can also be viewed on a continuum. The degree to which one aligns oneself with the constructivist end of the ontological spectrum, is likely therefore, to determine the epistemological choices one takes. (figure 2)

Figure 2: Epistemological spectrum

Knowledge depends on the senses, the gathering of facts and testing of hypotheses. Scientific discovery should be *value free*.	The external world cannot be objectively *known*, rather there are multiple realities, dependent on the individuals perception. NB – does not discount existence of an external world.
Positivist	**Idealist**

Critical Realist	**Post-modernist**
Suggests that the external world *can* be known, but may not be directly observable. Researchers may estimate a phenomenon (or generative mechanism) by studying its effects.	Disputes the existence of an external reality – "just different versions of different experiences". No individuals' claim to knowledge is any more valuable than another.

The discussion above is a simplified account of complex philosophical issues intended only to illustrate the point that there is no single, absolute, approach to studying the social world.

2.2.2.5 *Subtle Realism*

To conduct applied health and social science research, with the aim of achieving any body of knowledge of value beyond individual musings, we must allow ourselves to accept something as "true enough" to be considered knowledge. This compromise is embodied by Hammersley's concept of *Subtle Realism(38, 41)*, where the objective is to represent, rather than reproduce reality and we are asked to judge knowledge claims as true enough, based on their credibility and plausibility.

2.3 The role of reflexivity

Reflexivity has been defined as:

> *"qualitative researchers' engagement of continuous examination and explanation of how they have influenced a research project."(42)*

Reflexivity allows the researcher to demonstrate rigour and enhance credibility, through identification and analysis of the ways in which the researcher has influenced the research findings. For example, the more constructivist the position, the greater the importance of reflexivity – or the more reflexive the required approach; assuming that one wishes to make credible truth claims(35).

2.4 Qualitative research and theory

What is theory? Silverman suggests the following definition of theories:

> *"Ideas which arrange sets of concepts to define and explain some phenomenon"*

and goes on to suggest that:

> *"Theory provides both a framework for critically understanding phenomena and a basis for considering how, what is unknown, might be organised(43)"*

In the context of social research, theory refers to attempts to explain or understand a

phenomenon to create a level of understanding, at one step removed, from the primary data gathered in research, suggesting that such theory *may* have application beyond the immediate environment from which the primary data was generated(34).

Finally, with regard to the difference between theory and hypotheses, Silverman reminds us that hypotheses are derived from theory and represent something that should be examined, tested and, if necessary, rejected. Theory, on the other hand is not something which can be rejected – existing in its own right and can only be judged more or less useful in any give situation, rather than proven to be *untrue(37)*.

2.5 Qualitative and Quantitative research paradigms

It is impossible to discuss research methodology without addressing the methodological differences between qualitative and quantitative research and, more latterly, the attempts to bridge this divide in the form of mixed methods research.

The apparent incommensurable paradigms of qualitative and quantitative research, is not simply methodological, but, as might be expected, ontological and epistemological(35). Convention would align quantitative research with an Objectivist ontology and, thus, a Positivist epistemology. Likewise, qualitative methods are seen to represent Constructivist, Idealist principles(44). These assumptions have obvious implications for the type of research question that can be addressed.

2.6 Mixing methods of research

The evolving epistemological debate in response to a pragmatic need to address a variety of research questions has allowed the qualitative / quantitative divide to be blurred. If, as later iterations of realism would have us believe (e.g. critical realism, subtle realism)(38), there is an external reality, with truths that can be approximated, whilst accounting for our own unique perspective, then we can see that the two paradigms are not incommensurable. This more flexible philosophy opens the possibility of mixed methods research(45).

2.6.1 *The attraction of mixed methods*

There are several attractions to adopting mixed methods in Applied Health Research (AHR), the most fundamental being the ability to draw on the strengths of both paradigms and the types of knowledge they produce. This potentially allows researchers to work both inductively and deductively within the same project, to address questions of effectiveness in tandem with a deeper understanding of why, or why not, an intervention might be effective in different social groups(46).

Comparison of the two sets of findings allow researchers to validate their research – the support offered by similar findings from different perspectives serving to counterbalance the bias that may be inherent in any single method operating alone. This might usefully be thought of as "methodological triangulation"(47) and is distinct from the use of the term in chapter 4 (qualitative synthesis) where data collected using qualitative methods, in different settings, is triangulated to offer support as to the validity of the findings – data triangulation (see section 2.11.3 below for discussion of triangulation).

Bergmann suggests that mixed methods strengthen mono-method research practice and the evolution of mixed methods:

> "…forces researchers and theorists to return to more fundamental questions in relation to research design and how it connects to research questions, data collection, data analysis, and interpretation of findings."(48)

2.6.2 *Practical applications of mixed methods*

Some have argued that it will, inevitably, be the case that any mixed methods project has one "primary" methodology, which is augmented by the other. Strategies, whereby both paradigms may act as the "primary" and "auxiliary" methodology, have been described(46). However, it is often easier to see how a qualitative piece of work can be assimilated within a primarily quantitative project than *visa versa*. It may be that this is due to the inherently more flexible nature of qualitative work, in terms of sampling strategies for example, or possibly that there are simply more examples of this type of work, with successful research designs often being replicated(49). It is important to

design and conduct mixed methods research in a way that designates and acknowledges the roles played by each research paradigm.

2.6.3 *Models of mixed methods research*

Cresswell and Plano-Clark conducted an analysis of the research literature and identified four main research designs(50), which can in turn be characterized by the timing of the qualitative and quantitative elements of the project, as either concurrent or sequential (figures 3-7 below – adapted from Cresswell et al)(46, 50).

In the triangulation design (figure 3), the qualitative and quantitative components are conducted simultaneously and then integrated at the stage of analysis. As discussed above, this *triangulation of method* allows researchers to enhance the validity of each component. This model appears to value both paradigms equally and appears to represent a true *mixing* of methods.

Figure 3: Concurrent designs - Triangulation
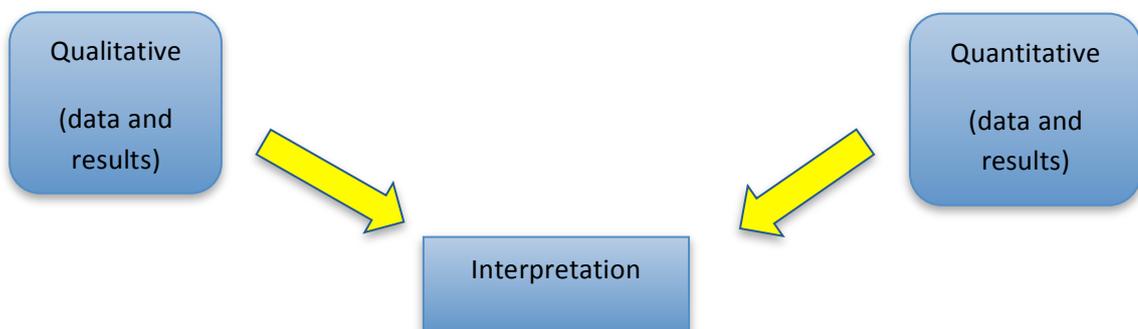
By contrast, the concurrent embedded design (figure 4), might be thought to represent the "augmentation" discussed above. One component, usually qualitative, is conducted during the operationalisation of the main study (for example an RCT). This may, for example, be used to provide information regarding the acceptability of an intervention, the results of which would relate to, but be separate from the main trial results. The potential privileging of one method over the other, certainly appears to be a risk in this instance(50).

Figure 4: Concurrent designs – Embedded
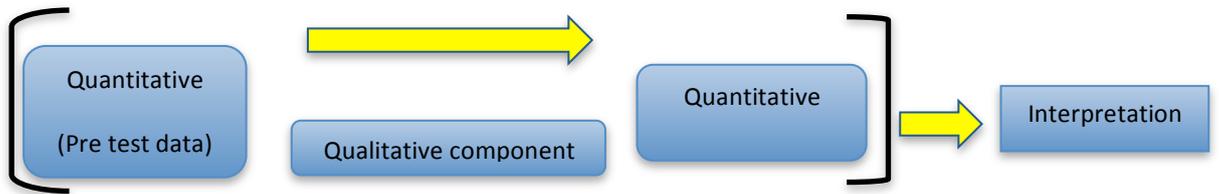
In the sequential designs, each paradigm represents a separate phase of the study. The Explanatory model (figure 5) is similar to the concurrent embedded design discussed above, where the qualitative phase, seeking to add in-depth explanation of quantitative results, is conducted after the results of the main study are available.

Figure 5: Sequential designs – Explanatory

By inverting this sequence, it is possible to create a study that aims to explore, quantitatively, the results of a primary qualitative study (figure 6). For example, qualitative work may be used to generate hypothesis for testing, or to contribute to the development of psychometric tools, which require validation. Those with concern for the degradation of qualitative research principles will note that the language employed in this design tends towards a positivist epistemology, and this will inevitably be necessary to conduct this type of research.

Figure 6: Sequential designs – Exploratory
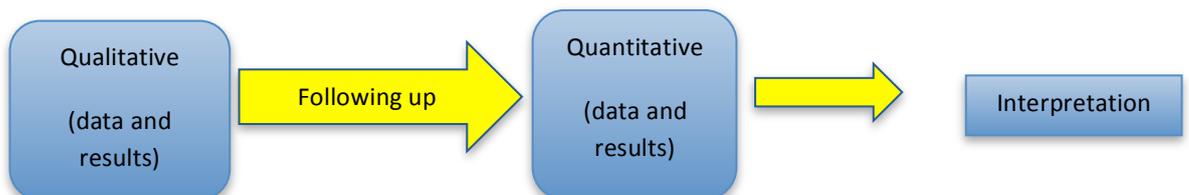
Finally, the sequential embedded design incorporates both of the above, so that qualitative work is used in the planning and analysis stages, to augment the quantitative study (figure 7).

Figure 7: Sequential designs – Embedded
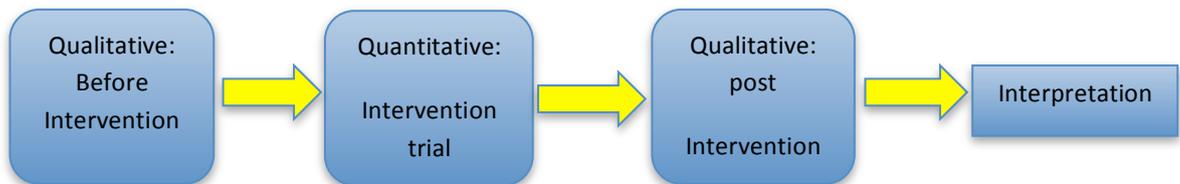
2.6.4 *Mixed methods: Conclusions*

In summary, mixed methods research has advantages for AHR practitioners, drawing on the strengths of each research tradition. It is possible to configure a programme of research so that one method takes precedence, being augmented by the other, or, to simultaneously conduct two pieces of work, which, having been conceived jointly are then brought together at the time of analysis.

In recognising the potential contributions of mixed methods research, particularly in AHR, where a combination of the explanatory (qualitative) and the exploratory (quantitative) in the same topic is highly desirable, the importance of protecting the mono – research traditions from which it has evolved, must also be recognised. This may best be achieved through careful research design, so that each paradigm is used effectively and given equal credence within the overall project.

2.7 The application of methodology to this thesis

Having discussed the methodological considerations of qualitative research in general terms, it is important to examine the application of these principles to this particular research project. This section sets out the philosophical underpinnings of the research enterprise for both the qualitative (discussed below) and quantitative (discussed in chapter 8) components, examining the role of theory - in particular the balance between inductive and deductive research and, should set the scene for subsequent reflection, in relation to the influence these decisions had on the research findings.

2.7.1 *Ontological position*

In terms of ontology, it is assumed that a common, external, reality does exist in relation to the social world; while acknowledging the inevitable influence of personal characteristics on the manner in which this social world is experienced. This would allow for the notion that different aspects of the social world may be more inclined to a shared experience and, thus, more commonly held as *true*, than others.

2.7.2 *Epistemological position*

The epistemology most aligned to this view of reality, is that of subtle realism, as proposed by Hammersley(41). In this, the existence of external reality is accepted, but it is recognised that even in good social research, findings will approximate, rather than exactly represent this reality, due to the nature of individual experience. The corollary of this position is to highlight the role of reflection and triangulation, in order to account for the influence of the researcher and validate the research findings, respectively(38). In rejecting both the positivist claims to truth, through objective research, and the constructivist claims, through the validity of individual experience, we must seek credibility through transparent reporting and by drawing comparison with other bodies of work.

2.7.3 *Approach to mixed methods*

The philosophical positions outlined above are consistent with the use of mixed methods research, without privileging one research paradigm over the other.

In terms of approach, the primary study design is most comparable to the sequential exploratory model described by Cresswell (figure 5)(50). Here we have the qualitative components (systematic review and qualitative synthesis and primary qualitative study) contributing to the production of a quantitative needs assessment tool. In addition, secondary analysis of the quantitative data, examining the patterns of unmet need revealed in the process of construct validation (chapter10), means that some steps have also been taken to combine the qualitative and quantitative arms of the study at the analysis stage. This draws on the triangulation design (figure 2) – where, although the data were not collected concurrently, the approach to combining data sets at the

analytical stage is the key aspect.

2.7.4 *Application of theory*

Whilst recognising that macro-level theory is inherent in daily social life and, therefore will inevitably colour social research, no attempt is made at in depth analysis of grand theory in the context of this project.

It is, however, the impact of mid-range theory that is of most relevance to this discussion. Working as a geriatrician with an interest in PD and palliative care, there is an implicit belief that physical disease also has identifiable social, emotional and spiritual manifestations and that these can, potentially, be ameliorated through effective interventions aimed at symptom relief and support of the individual.

Completing the systematic review and qualitative synthesis (chapter 4) prior to the collection of primary qualitative data (chapter 5-6) also provided a framework of mid-range theory, relating to the issue of palliative care in Parkinson's disease. In particular, the themes describing the dynamic process of information tension and care tension and the theory of intra- and inter- personal negotiation as a means to resolve these tensions, will, inevitably, have informed the data collection and analysis to a degree.

2.7.5 *Induction versus deduction*

The recognition of this mid-range theory and, in particular, the detailed theory developed during the qualitative synthesis, means that it is impossible to claim a purely inductive methodology for this piece of work. Rather, there is a mix of inductive and deductive work – examining the theories developed in the synthesis, while allowing room for new theories to develop, where supported by the data. Whilst this means rejection of a purely inductive qualitative approach, it is a realistic reflection of social research, which is rarely, if ever conducted without some prior theoretical assumptions(33). It also provided excellent opportunity to triangulate findings (triangulation of theory) with those from related studies, helping to enhance the validity and credibility of the research.

2.7.6 *Selecting a research question*

In writing the above discussion of philosophical and theoretical perspectives as they relate to research, it is clear that the research process itself has involved a process of self-reflection and a shift in ontological and epistemological positioning. It has also led to an acceptance that one's philosophical positioning need not be fixed, but rather that there are different types of knowledge and different ways of knowing the world, depending on the question asked. Highly constructivist positions do seem appropriate for examining certain elements of our social world, offering understanding in their own right, but these are less useful in the context of applied health sciences. If we seek to draw conclusions which can have meaning outside of the strict confines of the research, then I believe that we need to acknowledge a shared social reality, and the existence of some "truth" external to the individual experience. This is reflected in the make up of the research question, which seeks to identify unmet palliative and supportive care need, with the inherent assumption that, while individual experience will differ, there will be shared experience and, moreover, that understanding this helps us to influence the experience of others, through practical interventions.

Having selected a research question and a mixed methods approach, it was important to decide on the qualitative methodologies to employ.   In the following sections key methodological issues are summarised, with the rational for their use in this thesis justified at the end of each one. This is done firstly for qualitative literature synthesis and secondly for primary qualitative research.

2. 8 Methodological issues in qualitative literature synthesis

2.8.1 *The role of qualitative synthesis*

Qualitative synthesis is a useful research tool because it introduces the rigour of systematic review to the field of qualitative enquiry, offering the opportunity to address complex AHR questions - for example those relating to patient need or experience, which are not readily answered by quantitative methods, in a systematic manner(51, 52).

 Popay et al suggest that the potential of qualitative synthesis lies in its ability to explore broader questions than those addressed by reviews of clinical effectiveness, to appeal to

policy makers and to establish qualitative enquiry in a research field which is traditionally dominated by the randomised controlled trial (RCT)(53). This potential for qualitative synthesis to raise the status of qualitative research in health care is reflected by Dixon-Woods, who described the incorporation of qualitative synthesis into the clinical evidence base as an *"…acknowledgement of the explanatory power of non-quantitative forms of evidence"(52).*

However, while the synthesis of qualitative literature may be desirable, and offer considerable opportunity to applied health researchers, it also presents several technical and philosophical dilemmas, as outlined below:

2.8.2 *Should qualitative data be combined?*

The concept of qualitative synthesis is relatively new and criticised by those who feel that it is too positivist an approach with the implication that a common "truth" can be identified outside of the social constructions of the individual, seeming to run against the founding, constructionist, principles of qualitative enquiry(54, 55). This presents a problem in AHR where the ability to transfer research findings beyond their initial setting is key, if research is to have an impact on policy and practice, a point addressed by the Health Development Agency (HDA) in the foreword to their guidance on qualitative synthesis. They argue that opposition to qualitative synthesis, based on epistemological objections, is counter-productive and that a more pragmatic approach which acknowledges, but does not prioritise, philosophical differences is necessary if qualitative research is to be fully utilised(54).

2.9 Approach used in this thesis

From a personal perspective the opportunity to compare, contrast and explain, through synthesis, the findings of research from different geographical and temporal settings serves to enhance the credibility of qualitative research in general and should be embraced.

2.10 Assessing Quality

One of the key methodological questions posed by qualitative synthesis is how, if at all, to assess the quality of the primary research.

In systematic reviews of effectiveness, the process of quality appraisal has a very formal structure(56). The exclusion of papers which are methodologically weak is important in maintaining the credibility of the overall review findings, by minimising the introduction of bias. The inclusion and exclusion criteria by which primary research papers will be judged are established *a priori,* as are methods for dealing with studies judged to be "poor quality"(56). Further to this it is self-evident that any synthesis of literature, be it qualitative or quantitative, is beholden to the quality of the primary data. Thus, in order for the systematic review to be considered robust, reviewers must evaluate the quality of their selected primary literature and give transparent explanation for the way in which this influenced the subsequent review findings.

The application of these principles to qualitative synthesis has been the topic of several papers (53, 57-59) and the discussion can also draw on the more general literature addressing quality and credibility in qualitative research.

Drawing on Hammersley's previous work, Cutcliffe and McKenna describe three broad attitudes towards the quality assessment of qualitative literature(60).

1) The first position states that quality should be judged using the same criteria that have become established in quantitative research. This offers the advantage of uniformity of method and is likely to be readily accessible to an AHR audience. However, quality criteria designed to assess quantitative work, may be inappropriate for qualitative research, with the risk that studies which are weak on the production of theory (contributing little to the evolution of knowledge), but which fulfil technical quality criteria, are over privileged(53).

2) The second approach takes the polar opposite view. Stemming from post-modernism, it implies that any evaluation of quality is meaningless, due to the absence of any fixed external criteria. Bearing in mind the earlier philosophical discussion (section 2.2), this view is not compatible with research aimed at

practice or policy. Indeed it would tend to discourage research of any kind, given the assumed absence of an external, shared reality(40).

3) Cutcliffe and McKenna's third approach describes a middle way, calling for the construction of specific criteria for the evaluation of qualitative research, which reflect the nature of the discipline. It is in this area that we can most usefully focus.

2.11 Assessing Credibility

Terms such as validity and reliability have become part of the quantitative research landscape and provide a language through which to assess the credibility of a researcher's claims. In discussing assessment of qualitative research, Guba seeks to identify the "aspects" of good quality and the key terms which are used to refer to these in qualitative and quantitative research respectively(61) (see table 1).

Table 1 **Terms used to describe quality of research** *(Cochrane Guidelines, adapted from Guba and Lincoln(62))*

Aspect	Qualitative Term	Quantitative Term
Truth value	Credibility	Internal Validity
Applicability	Transferability	External Validity or Generalisibility
Consistency	Dependability	Reliability
Neutrality	Confirmability	Objectivity

Various techniques have been put forward to evaluate and establish the credibility (Internal Validity) of primary qualitative research.

2.11.1 *Reflexivity*

Reflexivity lies at the core of credibility in qualitative research. Whilst all research is subject to interpretation to a degree, the inductive and interpretive character of qualitative methods mean that the setting of the research, and the prior experience of the research team, are likely to significantly influence findings. Thus setting out the context of the research project and offering a critique of its impact, is held to be central to establishing credibility(63).

2.11.2 *Peer debriefing*

This has been recommended by several authors as a means of testing reliability and enhancing credibility(64-66). If several reviewers reach similar conclusions, or generate overlapping themes from data, then these conclusions may be considered more reliable. Conversely, peer debriefing may lead a researcher to question their own conclusions; such re-evaluation enhances the integrity of emergent theory.

2.11.3 *Triangulation*

 Triangulation involves enhancing validity by comparing the conclusions reached from different perspectives - for example research in different populations (data triangulation), or undertaken with different methods (method triangulation)(67). However, the underlying assumption of an external "truth", means that it has been criticised for being too positivist in its approach(61).

2.11.4 *Simple counts*

Simple counts are advocated by Silverman, in certain circumstances, to increase the credibility of qualitative findings(67). He suggests that providing readers with counts allows them to assess the frequency with which a particular theme emerged from the data, thus guarding against "anecdotalism" and the temptation for a researcher to "cherry pick" quotes or themes which suit their pre-held assumptions. The danger once again comes from the traditionally positivist outlook of many stakeholders and commissioners, such that provision of counts may detract from more detailed, inductive, analysis. Moreover qualitative studies, which are based on purposive not probabilistic

sampling, cannot yield valid statistical conclusions(66); so counts must be used with caution and with effective communication to stakeholders to avoid misinterpretation.

2.11.5 *Member checking*

Member Checking is held by Guba and Lincoln to be important in satisfying ourselves that research findings are truthful(61). Given that we seek to represent, to greater or lesser extent, the experiences of the research participants, it is suggested that credibility is enhanced by seeking affirmation of results from the participants themselves. If they recognise and concur with the themes described, it seems reasonable that this should support the credibility of our findings. However, the corollary of this would suggest that where participants disagree, or do not recognise, the theory emerging from the research, that this would reduce its credibility. This would seem to be an unreasonable conclusion. If qualitative enquiry aims to be inductive and interpretive, it stands to reason that participants may not recognise the conclusions drawn(68). Hence we have the concept of an analytical heirachy, where the understanding of the researcher, through the process of anaylsis, is separate to and builds upon, the understanding of the individual participant(69).

In addition, a simple narrative description of the data, being less interpretive, is likely to be acknowledged as "accurate" by participants, but is unlikely to contribute much to the development of theory. Moreover, if we accept that the interpretive nature of qualitative analysis means that not all themes will be recognisable to the research participant, what level of agreement is necessary to infer credibility? If one subject agrees with the findings, is this more or less credible than four subjects? As Cutcliffe points out, we are once again approaching a positivist method of evaluating credibility(60). Rather, researchers should be transparent about who did and did not agree with the generated theory.

2.11.6 *Deviant case analysis*

This refers to the way in which authors identify and seek to explain cases which do not fit with their emergent theories. Are they the exception that proves the rule, or do they point to flaws in analysis? Has the process of analysis sought to explain and account for

the deviant cases? For as Silverman points out there is nothing intrinsically deviant about cases, only that they may not fit with the theory we are trying to put forward(70) These questions are useful and provide the opportunity to assess the depth of analysis and thus the rigour of the researcher's method. A robust analysis will recognise and address deviant cases, while absence of such process may cause us to question the credibility of the conclusions presented(71).

So far we can conclude that concerns about the quality of qualitative research are important if we seek to synthesise the literature. The techniques described above can be used to assess credibility and may aid the development of more transparent quality appraisal for primary qualitative research.

2.12 The risk of quality appraisal

However, the prioritisation of quality assessment and the potential for non-specialists to apply methodological checks to qualitative work is not without risk, particularly that of "check list" appraisal.   This in turn can lead to *"Technical Essentialism"*, whereby researchers keen to establish credibility, seek to signpost the use of certain techniques when reporting their research(72). As Barbour points out, this does not mean that the techniques have been used appropriately and may come at the expense of more detailed analysis and reflexivity within the research(72).

It should also be recognised that quality of reporting may not always match, or truly reflect, the quality of methodological practice. Lessons from quantitative study tell us that reporting is often less than satisfactory, a fact that prompted the development of the CONSORT statement(73) and thus effective evaluation of qualitative studies for systematic review is likely to be reliant on improved, uniform, methods of reporting(74).

A number of quality assessment tools have been developed, as detailed in the Cochrane supplementary advice on qualitative synthesis, however there is as yet no consensus regarding the most appropriate choice, or even uniform application, of quality assessment amongst reviewers(75, 76).

Dixon-Woods assessed the effect of using three different approaches to quality appraisal of primary research papers, for inclusion in a qualitative synthesis(77). Interestingly, the

level of agreement between reviewers about whether articles should be included in the synthesis was relatively low, regardless of whether they used an unstructured opinion based approach, or a structured check list. However, in some cases there was significant variation from the same author, depending on which approach was adopted. Another interesting finding from this study was the evident tension, when reviewing qualitative literature, between quality of method and quality of emergent theory – as measured by potential contribution to the field of study. This was neatly represented in the comments from one reviewer discussing the decision to include a paper which was felt to be of poor methodological quality:

(Excerpt from Dixon-Woods et al 2007(77))

> *'A real dilemma. Flaws in conduct of data collection (inexperienced researchers),*
> *few details of methodological process, superficial analysis. But it is highly relevant.'*

The study also noted that the use of systematic "check list" type appraisal methods prompted the analysis of technique, but at the risk of over-emphasising research which may be flawed in other, less objective ways, such as interpretative analysis.

To summarise, there is little agreement around the best methods of appraising quality. It seems reasonable that quality assessment be used to trigger greater technical evaluation of qualitative research to promote transparency and good reporting of method. However, adopting a check list type approach is likely to over-emphasis the methodological process at the expense of theory generation, and may encourage an environment of technical essentialism amongst researchers(72). What is clear, when conducting a qualitative synthesis, is that the final report must be explicit about the way such quality judgements influenced the findings.

2.13 Using Quality Assessment in Research Synthesis

The discussion of quality assessment cannot be viewed in isolation, but rather in the light of how the results of appraisal will be used to influence the literature synthesis. The role of quality assessments in reviews of effectiveness, such as those carried out by the Cochrane collaboration, is often to sift out low quality studies in order to reduce their influence on final recommendations. As such strict criteria, for example covering

methods of group allocation, would be set *a priori*, and studies found to be deficient excluded(78). The Cochrane Qualitative Methods Group cite an example of such an approach used in qualitative review, but emphasise the concerns already discussed, that the uncertainty surrounding assessments of quality risks excluding many studies which would contribute significantly to development of theory(62).

The second suggestion is a system of weighting, similar to that once adopted but now out of favour in quantitative meta-analysis. To date, this idea has not been used in practice, primarily because while we may be able to "weight" articles according to reported methodology, it is difficult to do so for their contribution to theory development(62).

The need to undertake literature synthesis in the midst of these methodological debates has led to the adoption of a pragmatic approach, whereby the influence of studies judged poor quality is assessed in a sensitivity analysis, by estimating the impact of poor quality studies on the review findings. An example of this is described by Thomas and Harden, who, having made the decision not to exclude studies on the basis of quality assessment, performed a sensitivity analysis and concluded that those of poor quality made relatively little contribution to theory(55).

2.14 Approach used in this thesis

Given the disagreements regarding how to appraise quality and the difficulties with study reporting, such that well conducted research may appear less credible due to poor quality reporting, the decision was taken to assess the quality of primary studies, but not to exclude studies on the basis of quality when conducting the qualitative synthesis (chapter 4). The justification for this being that the risk of inappropriately excluding data which adds significantly to theory generation was felt to be greater than the risk of biasing results through inadvertent inclusion of poor quality studies.

2.15 Methods of Data Synthesis

2.15.1 *Positioning the research: Idealist vs Realist*

Multiple approaches to qualitative data synthesis have been described, often building on and adapting previous methods. These have been summarised by Dixon-Wood in her

report for the Health Development Agency(57), and once again the range of approaches stem in part from the variety of epistemological positions adopted by their advocates.

In another review, Barnett-Page discusses the different approaches to synthesis in terms of their approach to six aspects of synthesis production, from the literature search, to the final synthetic product(58). Viewing them as predominantly Idealist (constructivist) or Realist (positivist) she places them in two broad groups, and suggests that practitioners may like to select from within these groups, depending on the aims of their synthesis and the type of outputs required. In general terms, those from the "Idealist" group will produce a more conceptual output, which may not be immediately applicable for policy makers, while the "Realist" end of the spectrum aims to produce theory with direct application to policy. The broad groupings are displayed in table 2

Table 2 **Philosophical positioning of synthetic methods**

Idealist (constructionist end of spectrum)	Realist (positivist end of spectrum)
Meta-Study	Thematic Synthesis
Meta-narrative	Textual narrative synthesis
Meta-ethnography	Framework synthesis
Grounded theory	Ecological Triangulation
Critical Interpretive Synthesis	

(Created from work of Barnett-Page 2009(58))

Whilst this dichotomisation of methodological approaches is somewhat artificial, it has pragmatic advantage in helping to crystallise the choice facing practitioners seeking to carry out qualitative synthesis. The "Realist" group seeks to more closely approximate the systematic reviews of effectiveness seen in quantitative enquiry, while the "Idealist" model represents more of the features of primary qualitative research, in terms of iteration, and interpretation.

The issue of interpretation within a *synthesis* of primary research is an important area of differentiation. Quantitative systematic reviews seek to regulate process so that interpretation is minimised and, in theory, different reviewers could repeat the steps of the original author and reach the same conclusions(78). However it is acknowledged in qualitative synthesis that interpretation is present at the synthetic level and new theory, beyond that present in the primary studies, should emerge(79). Interpretation, whilst it is a perceived weakness in effectiveness reviews, may be considered strength in qualitative synthesis and attempts to limit interpretation risk devaluing the process as a whole.

Finally, the discussion of epistemological position, which appears to so influence the methodological choice, should not be limited to the individual researcher but also applied to the research question. Viewing synthetic methodologies on a continuum from "Idealist" to "Realist", a question which is narrow and well defined with the intention of directly influencing policy, may suit a methodology from the "Realist" end of the spectrum(58). Likewise a broader question, for example one which, as in Critical Interpretive Synthesis (CIS), may be subject to iterative refinement during the project(80), will be more suitable to an "Idealist" approach. Of course researchers are likely to select research questions which dove-tail to their own epistemological and ontological positions – thus an "Idealist" will tend to select broad, less clearly defined questions of study. Nonetheless, it is important to select the method that will best answer our intended question, rather than that with which we feel most closely aligned from a personal perspective.

2.15.2 *Choosing a synthetic method*

Some key elements of qualitative synthesis will vary according to the chosen methodology and researchers should considered their preferred approach to these in advance, alongside the specific requirements of the research question and setting. These are:

2.15.2.1 *Literature searching:*

In traditional systematic reviews this process is well described and all relevant literature, as defined by pre-established search criteria, should be included in the review(78).

However, some methods of qualitative synthesis allow an iterative approach to literature search and data selection, analogous to the theoretical sampling employed in primary qualitative research. This may be particularly useful where very large numbers of primary papers are identified by the database search. In Grounded Theory, as applied to qualitative synthesis, the use of an iterative search strategy seeking theoretical saturation allows the researcher to limit the number of primary papers included in the review without, it is suggested, compromising the outcomes(58, 81). A similar approach to selection of primary studies is adopted in Critical Interpretive Synthesis, where a practitioner may sample from within a large body of primary studies(80, 82). The acceptability of this approach may depend on the intended audience, being unlikely to appeal to those from a traditional systematic review background.

2.15.2.2 *Generation of theory: Interpretation vs Integration:*

This is a key area. Methods such as Meta-Ethnography, Grounded Theory and Critical Interpretive Synthesis all aim to build on the findings of the primary research and move beyond them, to create new, higher level theory(81-83). This is an interpretive process and, by its very nature, means that the new explanatory theories are several steps removed from the original data. Whilst this generation of new explanatory theory seems highly attractive, it inevitably comes at the cost of transparency, as the process of interpretation will be specific to the individual researcher, or research team.

Alternative approaches to qualitative review, such as Content Analysis apply a more integrative approach, seeking to aggregate and describe the findings of primary literature, rather than transform in the generation of higher level theory(56). These methods may be more palatable to some, particularly where clear "answers" to "questions" are required, but are less true to the traditions of qualitative enquiry.

2.15.2.3 *Methodological choice of primary studies – "lumping or splitting":*

It has been suggested that because the methodological approach of qualitative researchers has an influence on their findings, qualitative syntheses should only include studies from a single methodological genre, or alternatively, as in Meta-study, studies of a similar methodology should be grouped together within the over-all synthesis(83). Whilst

this may appeal to hard-line theorists, it undoubtedly has implications for the completeness of a review, especially in areas where there are few published studies. Thus by limiting the synthesis to primary literature of a single methodology, a significant proportion of the data is likely to be omitted and thus introduce significant bias. In addition, the lack of uniform reporting of methods is again important, meaning that it may often be difficult to identify the methodological approach of authors, or that they themselves may wrongly attribute a particular method to their work. These problems with reporting are likely to be emphasised in AHR, where word limits in scientific journals mean that discussion of theoretical issues is often limited.

2.15 Required level of transparency:

Transparency and ultimately reproducibility are certainly desirable in systematic reviews. The degree to which these are prioritised will influence the choice of method for the conduct of a qualitative synthesis. The tension between transparency and interpretation, with generation of higher level theory, is at the heart of the dichotomy of synthesis methods, set out by Barnett-Page(58).

A pragmatic approach is required to navigate the multiple controversies surrounding theoretical application. No method is so established that it must be followed rigorously. Borrowing characteristics from more than one method may offer the best hope of producing synthesis which is at once critical and interpretive, whilst offering the practical applications which must be at the heart of AHR. Finally it should be emphasised that the introduction of interpretation at the level of synthesis, anathema to systematic reviews of effectiveness, must surely require reflexivity on behalf of review authors in order that the process be as transparent as possible.

2.16 Approach used in this thesis

2.16.1 *Positioning the research question*

The question of palliative and supportive care need in Parkinson's disease is broad, and as such it better suited a more *Idealist* approach, as opposed to the narrow questions favoured by the *Realist* methodologies. Further to this a scoping search, discussed in the introductory chapter (chapter 1), suggested a range of primary literature, displaying

geographical and methodological diversity, implying that the chosen style of synthesis must be one that allowed primary studies of all methodologies to be included. This was important as subdividing a relatively small group of primary studies (n =16) into methodological groups, was likely to produce a fragmented, rather than inclusive, review of the topic.

The purpose of the systematic review was primarily to contribute to the adaptation of a palliative needs assessment tool (NAT:PD) for use in Parkinson's disease and, in so doing, to inform policy and practice in an area of healthcare which is witnessing rapid changes. As such, it was important to be as comprehensive as possible and to include all of the primary studies identified by the literature search, rather than sampling from within it.

Interpretation is a strength of qualitative synthesis, which should be recognised and embraced by researchers, in as transparent a way as possible. Selecting a highly integrative method, whilst likely to appeal to a non-qualitative audience, risked abandoning a fundamental aspect of the project.

2.16.2 *Selecting a synthetic method*

With this in mind meta-ethnography seemed to be an appropriate approach. It promotes interpretation, but does so in a structured manner, allowing a high level of transparency as one moves through the stages of synthesis.

2.16.3 *Application of Meta-Ethnography in this thesis*

This review used Meta-Ethnography to analyse the data drawn from primary studies, but made two important changes to the original method, both of which have precedent in the recent literature and serve to increase the utility of the method for HSR research, by allowing its application to a broader range of primary work.

2.16.3.1 *Methodology of primary studies*

In their original publication, Noblitt and Hare suggest that meta-ethnographies be restricted to primary research using the same methodology(84). In practice this has

several difficulties, not least the failure of many researchers to adequately describe and apply a specific methodology within their work. It is also likely that multiple methodologies will be present within the literature base and, therefore a strict adherence to the authors' original description would mean either; excluding some studies, conducting multiple syntheses of small numbers of studies, or conducting several parallel syntheses within the same review - with no guidance on how these should be combined. The requirement to include only primary studies from the same methodological genre has been challenged by Campbell et al, who provide worked examples of meta-ethnographies conducted within a methodologically diverse group of primary research articles(83). This approach offers several advantages to HSR researchers and best met the requirements of this review.

2.16.3.2 *Suitability of studies for Reciprocal Translational Analysis (RTA)*

The process of RTA is the cornerstone of meta-ethnography, representing the process by which reviewers identify the most comprehensive theoretical explanations, seeking to explain all of the phenomena described, from within the primary literature. The RTA addresses each individual study consecutively, with new constructs emerging where necessary, to ensure that all aspects of the data are explained. In order to do this Noblitt and Hare suggest that it is necessary for the primary studies to be "sufficiently" similar, as it would not be fair to perform RTA on studies from opposing perspectives(84). Where such opposition is present, they suggest using a related technique they refer to as "refutational analysis".

The systematic review described in chapter 4, encountered primary studies which looked at the experience of either patients, carers or both together. Whilst it is difficult to define how similar studies need to be to allow RTA to take place, it seems reasonable that the perspective of the carer may be sufficiently different to that of the patient, to require separate RTA's. In addition, the number of studies included in the systematic review (16) presented a logistical challenge if all were to be included in a single RTA. This is highlighted by the fact that Noblitt and Hare's original description of RTA includes only 3 ethnographic studies(84).

One solution, as described by Daker-White et al whilst conducting a review of therapy adherence in chronic disease, was to split the literature base according to studies which seemed suitably similar, conducting RTA for each group, before amalgamating the findings(83). This seems a reasonable approach in cases such as this, where natural divisions exist within the primary studies but which are not sufficient to demand *refutational analysis.*

Therefore, having considered the characteristics of the primary studies to be included in this review and building on the precedents described above, the decision was made to divide the primary studies in to three natural groups *(1- patient only, 2- carer only and 3- both patient and carer)* and conduct three RTA's initially, before drawing parallels between them and combining them to produce a *"Lines of Argument"* analysis (see chapter 4).

2.17 Methodological issues in primary qualitative research

The umbrella term "qualitative research" covers a number of different research disciplines, most of which are aligned to a particular philosophical or theoretical tradition. It is possible to group disciplines in broad methodological categories, to aid selection. One such approach is to categorise based on the ultimate goals of the research, for example the generation of new theory, the identification of meaning through language, or the accurate reporting and understanding of a particular culture(85).

However the division between methodologies is not necessarily clear cut because the approaches themselves are not always well defined or intended to be fixed and prescriptive as with quantitative paradigms. For example, when discussing their later approach to grounded theory, Corbin and Strauss recognise that different researchers may have slightly different aims and may wish to use the text as a guide and that it was not intended to be "used rigidly in a step by step fashion"(86).

Notwithstanding the above discussion, the key questions considered when selecting a primary qualitative methodology for this project were:

2.17.1 *Nature of the research question*

The project had a specific aim, to identify the palliative and supportive care needs associated with PD, and to apply these findings in generating and validating a new clinical tool. Thus the focus was on understanding the experience of individuals, as it relates to palliative and supportive care, and to identify common themes, across participants, which could be used to generate theoretical understanding and be applied more widely. Approaches that attempt to capture and interpret meaning in this way include Grounded Theory(33, 68), Thematic Analysis and Content Analysis(33, 87).

Semi-structured interviews and focus groups were felt to be the best method of data collection, in order to optimise the number and variety of participants, while working within a limited time frame (see chapter 5). This excluded approaches such as ethnography, life history and narrative analysis. However it is easy to see how, in another setting, these approaches may enhance our understanding of palliative and supportive need, for example through observing patients and carers in their daily activities, or exploring in detail the entire journey with PD to understand the likely fluctuant nature of palliative needs.

The second consideration was to treat the dataset as a whole, identifying common themes, as opposed to dividing the data, for example by early and late stage disease and using different analytical frameworks for each. This inclusive approach is best served by a so called "code and retrieve" methodology (Grounded Theory, Thematic Analysis), where a single analytical frame is developed from the raw data, and then applied as a means of handling the data during analysis(69).

The points above distilled the methodological debate to a choice between Grounded Theory and Thematic Analysis, more specifically a type of thematic analysis known as Framework Analysis. Despite terminological differences, these approaches share a number of important steps, for example the structured approach of coding, the creation of analytical frames which are then applied to the transcripts and the encouragement of a constant comparison technique. There are however differences, in particular the approach to prior assumed knowledge about the phenomenon of study and the clarity of data handling, which favoured the use of framework analysis in this study (see table 3).

2.17.2 *Desire for transparency*

In AHR, outputs need to be understood by colleagues and funders who may not have a background in qualitative research. Framework analysis, a version of thematic analysis, was developed with this issue in mind, offering a systematic approach to data handling which encourages interpretation and explanatory analysis, but is also structured in a manner that allows the process to be traced back to the raw data(69, 88).

2.17.3 *Dealing with a priori assumptions*

The structure of this project was such that a systematic review and synthesis of qualitative literature was conducted before the primary qualitative work. It would therefore be unrealistic to suggest that a researcher could approach the qualitative work without any pre-held assumptions. Whilst the objective is of course to discount preconceptions as much as possible during the conduct of qualitative research, framework analysis does allow the formation of the thematic framework to take account of *a priori* issues, as opposed to grounded theory, where these issues should be "bracketed"(33, 89). In this regard, framework analysis appeared to offer a more honest approach, in the context of the project as a whole, where these issues could be acknowledged.

Table 3 – Comparison of Grounded Theory and Framework Analysis

	Grounded Theory	Framework analysis	*Application to thesis*
Desired outcome	Generating new theory, may relate to the topic of study "substantive theory" or have external application "formal theory".	Theory generation not the sole aim. May be used to answer specific questions, or identify needs.	While it was hoped that the project would generate new theory, there was also a specific aim – adaptation of the NAT Parkinson's disease, favouring Framework Analysis.
Approach to understanding	Highly inductive – asked to suspend pre-held beliefs and assumptions	Criticised for being overly deductive, but advocates believe it can be inductive or deductive depending on the question asked.	The qualitative synthesis (chapter 3-4) made it impossible to approach the primary qualitative study without pre-held assumptions. Framework Analysis allows for the existence of prior understanding, which was crucial for the integrity of this research.
Initial Data handling	Data fragmented through coding…*"freeing the researcher from description and forcing higher levels of abstraction"* Strauss. Some advocate an intensive, line-by-line approach.	Coding or "indexing" used to divide the data. Nature of coding less prescriptive.	Both methods advocate a "code and retrieve" method, as adopted in this project.
Analytical framework	Coding frame – Emerges from data as codes are compared and checked against transcripts in process of "constant comparison".	Analytic frame evolves in similar manner to coding frame, but, crucially, can take account of pre-held assumptions.	Framework analysis allowed the findings of the qualitative synthesis to be acknowledged and incorporated in to the analytical framework. This was crucial to the integrity of the research, where "bracketing" of prior understanding would have been very difficult in practice.
Analysis	Concepts developed following initial coding are subsequently used to form categories – sequential levels of abstraction.	Charting – May be based on themes emerging from the data, or include pre-formed categories, depending on the level of induction desired. Grids, which chart participants or groups against these categories allow analysis and comparison between groups.	Adopting a Framework approach allowed prior understanding to be incorporated in analysis – see above. The use of grids promoted transparency, allowing higher-level theories to be traced back to the original transcripts.

2.18 Approach in this thesis

Thus, for the reasons given above, framework analysis was chosen for the analysis of primary qualitative work in this thesis. The analytical framework used is displayed in appendix 2 and a step by step guide to this technique can be seen in appendix 3.

2.19 Conclusion

The conduct of qualitative research is heavily predicated on an understanding of the underpinning theories described above. Decisions regarding selection of research questions and methods of study cannot be finalised until the researchers have an understanding of the ontological and epistemological positions which will inform their research. This will reflect not only the philosophical choices made, but also the very utilitarian issue of research outputs, which need to be appropriate to the scientific milieu in which the project was conceived.

Having considered these important questions and illustrated the way in which the answers informed methodological choices, the following chapters illustrate their application, through the conduct of qualitative research, first in the form of systematic review and qualitative synthesis (chapters 3 and 4) and then primary qualitative work exploring palliative and supportive care need in Parkinson's disease (chapters 5 and 6).

<div align="center">**Chapter 3**</div>

<div align="center">**Systematic review and synthesis of qualitative literature: Methods**</div>

3.1 Introduction

In chapter 2 some of the philosophical differences between qualitative and quantitative research were discussed. In quantitative research the process of systematic review allows researchers to comprehensively draw together the evidence on a single topic, in a structured and comprehensive manner, creating a higher level of evidence, which can be used to produce recommendations for policy and practice(56). As such, systematic review has been championed as a method for producing good quality evidence in areas where individual studies may be too small, or their results too disparate, to guide practice effectively(56). A process initiated by the work of Cochrane has become increasingly refined, such that effectiveness reviews are now governed by strict methodological guidelines, most prominently those issued by the Cochrane collaboration(78). These developments serve to enhance the transparency and repeatability of the review process, which in turn, increases the confidence of clinicians and policy makers who incorporate the findings in to their practice.

While some of the analytic techniques used in reviews of effectiveness, such as meta-analysis, are clearly not suitable for qualitative data, the ability to draw together a number of studies, to produce higher levels of evidence, which enhance quality and academic rigour, is equally desirable(51, 52, 79). Thus, as qualitative methods have become established in health service research (HSR), there has been an increasing drive to develop new techniques which allow primary qualitative studies to be combined, in a manner reflecting systematic reviews of effectiveness; a process referred to as "qualitative synthesis"(56, 75, 90).

3.2 Systematic review protocol

3.2.1 *Review question*

What are the palliative and supportive care needs of patients and caregivers with Parkinson's disease?

3.2.2 Review objectives

The objectives of this review was to establish the current understanding of palliative and supportive care needs in respect to Parkinson's disease, with the results being used to inform the adaptation of a palliative need assessment tool (NAT:PD-c)(29) for use in Parkinson's disease.

3.2.3 Inclusion criteria

3.2.3.1 Participants

The review included all articles investigating the palliative and supportive care needs of patients with idiopathic Parkinson's disease, or their carers, using standard definitions of palliative and supportive care(7, 91) and taking "carer" to be any person who acts in an unpaid care role for an individual with Parkinson's disease. All ages and disease stages were included.

3.2.3.2 Outcomes

The review included all studies which examined palliative or supportive care needs in the target population, using the definitions provided by the WHO and NCHSPCS (National Council for Hospice and Specialist Palliative Care Services) respectively:

WHO definition of palliative care:

" ..an approach to prevent and treat physical, psycho-social and spiritual problems, which impact on quality of life. These are not directed at cure, but may operate alongside curative approaches, and at all stages of chronic life limiting disease from the time of diagnosis."(7)

NCHSPCS definition of supportive care:

'…helps the patient and their family to cope with cancer and treatment of it – from pre-diagnosis, through the process of diagnosis and treatment, to cure, continuing illness or death and into bereavement. It helps the patient to maximise the benefits of treatment and to live as well as possible with the effects of the disease. It is given equal priority alongside diagnosis and treatment.'(91)

3.2.4 Exclusion Criteria

3.2.4.1 Participants

The experience of paid / professional caregivers is not the focus of this review, and such studies were not included. The review focused on idiopathic Parkinson's disease, and did not include studies of the other Parkinsonian syndromes, such as Progressive Supra-nuclear Palsy (PSP) and Multiple System Atrophy (MSA), except where the data relating to idiopathic PD could be extracted separately.

3.2.4.2 Language

Study selection was restricted to English language publications only, due to the limitations of finance and time of the study group. It is acknowledged that this increases the risk of publication bias.

3.2.4.3 *Reviews*

Review articles, or those based on author opinion were excluded. However, hand searching of references was undertaken to ensure all primary data sources used for such articles have been included.

3.2.4.4 *Case Reports*

Case reports were also excluded.

3.2.5 Database searching

Search strategies were developed iteratively in conjunction with an information scientist and applied to the following databases: Cochrane library, Medline – OvidSP 1946 to 14/09/2012, EMBASE, CINAHL ,Web of Knowledge – (Social Science Citation Index (SSCI) and ISI conference proceedings), between 14th and 15th September 2012.

The grey literature was accessed through searching of conference abstracts and contacting experts in the field. The search terms can be seen in box 1-4.

3.2.5.1 *Search Terms*

Search terms for the first concept: Parkinson's disease are displayed in box 1.

This is in keeping with the search string used by the Cochrane movement disorders special interest group.

Box 1

Search terms for Parkinson's disease: Medline

Parkinson's Disease was explored as:

exp Parkinson Disease/

parkinson$.tw.

The concepts of palliative and supportive care were explored using the terms shown in box 2:

Box 2

Search terms for Palliative and supportive care: Medline

```
exp Palliative Care/

Palliat$.tw.

(palliative treatment or palliative medicine). tw

exp Terminal Care/

terminal care.tw.

exp Social Support/ or exp "Quality of Life"/

supportive care.tw.

end-of-life.tw
```

The example search string for Medline on the Ovid platform is shown in box 3, identifying a total of 1072 articles.

During the development of this search string the effect of terms relating to "needs" and "disease burden" was assessed. Including these terms with the above search string, using an AND operator, led to a very restricted search outcome with only 17 hits. Including the terms in the palliative care section with an OR operator led to a loss of specificity, with over 10,000 articles identified. For this reason these terms were not included in the final search string.

3.2.5.2 Qualitative filter

Using filters for qualitative studies can be difficult due to the wide range of descriptions used for qualitative work, meaning that highly specific search terms may lack sensitivity. Flemming et al described the use of a broad, free text search filter for qualitative studies, which was as effective as more complex qualitative filters(92). The search strategy they described is displayed in box 4. Applying this qualitative filter to the example search string in Medline produced 175 results.

Box 3 – Example search strategy on Medline platform

1 exp Parkinson Disease/ (42170)

2 parkinson's disease.tw. (40662)

3 exp Palliative Care/ (36465)

4 palliat$.tw. (40699)

5 (palliative care or palliative medicine).tw. (11593)

6 exp Terminal Care/ (38072)

7 terminal care.tw. (1301)

8 supportive care.tw. (6852)

9 exp "Quality of Life"/ or exp Social Support/ (141259)

10 end-of-life.tw. (8862)

11 3 or 4 or 5 or 6 or 7 or 8 or 9 or 10 (228369)

12 1 or 2 (55192)

13 11 and 12 **(1072)**

Box 4 – search terms for qualitative filter

14. findings.af.

15. interview$.af. or interviews/

16. qualitative.af.

17. or/14-16

3.2.6 Study selection

The results of the database search were initially screened by title and abstract. This was carried out independently by two members of the team (ER and NB), with any disagreement resolved by a third reviewer (MJ). Articles assessed as definitely or possibly relevant were retrieved in full text form and reviewed again for inclusion using the study eligibility form (appendix 4). This was once again conducted separately by two reviewers (ER and NB), with referral to the third reviewer (MJ) for any disagreement.

3.2.7 Quality assessment

The quality of primary studies was not used to exclude studies from this review, as the risk of excluding important studies which make significant contribution to new theory generation, was considered to be greater than the risk of including studies with potential flaws in application or reporting of method.

3.2.8 Data extraction

Primary studies which met the criteria for inclusion in the review were re-read; the findings were coded and tabulated on a data extraction form (see appendix 5). The form was designed so that coded data could be tabulated according to the underlying theme from the NAT-PD to which it best corresponded. Data which did not correspond to any NAT theme was tabulated under a new heading. This was done in order to show, from the first stages, the relationships that exist between the primary qualitative data and the NAT. In turn this allows the process of NAT adaptation to be traced back, through the qualitative synthesis, to the primary papers in the initial literature search.

3.2.9 Data synthesis

Data extracted from the primary papers was synthesised using meta-ethnography, for methodological reasons discussed in chapter 2. The data synthesis was conducted in the following steps:

Step 1 – Data extracted from the primary papers, along with quotes where appropriate was coded and matched against corresponding domain from the NAT. Where data did not correspond to an existing NAT domain it was displayed under a new heading. This produced 166 codes.

Step 2 – Reciprocal Translational Analysis (RTA). Primary papers were grouped according to participants, in to one of three RTAs (Patient / carers / patients and carers). Starting with the extracts for the first paper in the RTA, concepts were created to explain the observations within the coded data.

Step 3 – The initial concepts developed to explain the data extracted from first paper were then applied to the second. Where the existing concepts were not sufficient, new concepts were created. These were all then taken forward and applied to the third paper, and so on, until all the extracted data from the primary papers was covered. This produced 74 concepts across the three RTAs conducted. The contribution of the data codes from each paper to the formation of explanatory concepts, was mapped using grids to ensure that the process could be traced back, at all times, to the raw extracted data. (see example grid for paper 2 – appendix 6).

Step 4 – The initial concepts created in each RTA were used to develop explanatory constructs (referred to as RTA constructs) in the same manner, where the aim was to ensure that all of the existing concepts were explained, with as few RTA constructs as

possible. Again the contribution of initial concepts, to RTA constructs was mapped using grids (see example grid for RTA 1 – appendix 7).

Step 5 – In order to develop over-arching themes for the data set, the RTA constructs were submitted to a Lines of Argument analysis. Here the objective was to discover any analytical themes that linked data across the RTAs. This was the step in the synthesis with the greatest degree of interpretation and produced four lines of argument, as discussed in chapter 4. The contribution of constructs to each line of argument was again mapped and is displayed in tables 7-10 in chapter 4.

3.3 Conclusions

Qualitative synthesis, as an output from systematic literature review, is a great opportunity for applied health researchers. By taking the strengths of both disciplines it is possible to address broad questions, beyond the scope of traditional reviews of effectiveness, in a structured and comprehensive manner. Practitioners must openly acknowledge the decisions taken regarding issues such as quality appraisal and the degree of interpretation included in their analysis, and these issues are addressed above.

In developing the protocol for this review, it was necessary to understand the theoretical underpinnings of qualitative synthesis and, importantly, to recognise those areas in which the methodology is still developing (see chapter 2). This allowed the review protocol to incorporate a number of adaptations to meta-ethnography as it was originally described, including papers from all methodological disciplines and conducting three simultaneous RTAs, in order to produce a synthesis which meets the demands of applied health research whilst remaining theoretically robust. The findings from the systematic literature review and subsequent meta-ethnography are discussed in the following chapter (Chapter 4).

Chapter 4

Systematic review and synthesis of qualitative literature: Results

4.1 Introduction

Chapter 3 set out the methods for a systematic review addressing the *question "What are the palliative and supportive care needs of people living with Parkinson's disease".* This chapter presents the findings of that systematic review and a synthesis of the qualitative evidence.

4.2 Results of database search and study selection

Database searches identified 943 articles, which were reviewed by title and abstract. This led to the selection of 27 full text articles, of which 15 were considered eligible for inclusion(9, 12-14, 93-103). Two further articles, published in abstract form were considered potentially eligible, but we were unable to obtain full text manuscripts from the original authors and the abstracts contained insufficient detail to allow inclusion(104, 105). A flow diagram of study inclusion is displayed below in diagram 1:

A table summarising the reasons for exclusion of full text articles can be found in appendix 8.

Studies were grouped according to whether they included patients (n=5), carers (n=4) or both (n=6) and information was collated regarding the country of origin, research methodology, qualitative method, disease stage of participants and recruitment setting. This is summarised in appendix 9.

Figure 8: PRISMA Flow diagram for study selection

Identification

| Records identified through database searching 1324 | Additional records identified through other sources (n = 7) |

Screening

Records after duplicates removed
(n = 771)

Records screened
(n = 771)

Records excluded
(n = 744)

Not qualitative research, not addressing Parkinson's disease or palliative care, not English language.

Eligibility

Full-text articles assessed for eligibility
(n = 27)

Full-text articles excluded, with reasons
(n = 12)

Included

Studies included in qualitative synthesis
(n = 15)

Reasons for exclusion:

Not qualitative = **4**

Unable to extract PD specific data = **3**

Paper does not explore palliative / supportive care need = **2**

Non-English language = **1**

Author could not provide additional information = **2**

4.3 Analysis: Reciprocal Translational Analysis

Three separate Reciprocal Translational Analyses were conducted, one for each group of participants (Patients / carer / both). The codes from the primary data were used to form concepts, which were in turn the foundation for "RTA constructs". These constructs can be traced back to the primary data from which they emerged and seek to offer the most economical explanation of the phenomena described by these studies. A full description of this process can be found in chapter 3. The tables below illustrate the relationship between the initial concepts formed by the coded data and the RTA constructs.

As would be expected there are considerable areas of similarity in the finding from each group of studies (patient, carer, both). In these cases, an RTA construct developed in the first RTA may also have been used to explain the phenomena described in the next set of studies, although in some cases, extra layers of complexity will have been added, at the construct level. This process is referred to in the analysis below.

For practical reasons the RTA's were conducted in ascending order of size (1st Carers, 2nd Patients, 3rd combined patients / carers) to allow familiarity with the analytical techniques to develop, using the smallest data sets first. This is important when considering the formation of RTA constructs, particularly those which appear in all 3 analyses.

4.3.1 *1st* RTA Carer only studies

In this section of the study there were 39 codes taken from the primary literature, which informed 16 initial concepts, reduced through the process of RTA to 5 core constructs – see table 4.

Table 4 – RTA of Carer only studies

RTA constructs	Initial concepts
Subjugation of Carer needs	Need for support (Formal and Informal, Spiritual)
	Occurrence of Trigger events
	Loss of "life role"
Care Tension	Care as "duty"
	Guilt Re: accepting help
	Benefits of care
Financial Hardship	Loss of Job
	Cost of care
Poor Knowledge	Disease specific knowledge
	Care Specific knowledge
	Service Specific Knowledge
	Information Tension
Clinical dissatisfaction	Biomedical focus of consultations
	Doctor as prescriber
	Poor communication between HCP's
	Negative experience of diagnostic process

4.3.2 2nd *RTA Patient only studies*

Analysis of the patient only studies yielded a great deal of data, with 69 initial codes leading to the identification of 36 concepts from within the primary studies. These were subjected to the process explained by 6 RTA constructs – see table 5.

Table 5: RTA of Patient only studies

RTA Construct	Initial Concept
Being Diagnosed	Emotional response to disease: diagnosis
	Desire for information: diagnosis
	Dissatisfaction with clinicians: recognition
	Dissatisfaction with clinicians: communication
	Protecting family
Negotiating function	Re-negotiating activities
	Emotional response to disease: Physical
	Desire for information: practical knowledge
	Accepting medication
	Medication Anxiety
	Sexual difficulties
Reconstructing Self	Reconstruction of self
	Changing role: Fear of failing
	Changing role: Finance / work / Home
	Milestones of Independence
	Emotional response to disease: Physical
Addressing the Future	Addressing the future: Temporality
	Addressing the future: Maintaining Hope
	Addressing the future: Planning (lack of)
	Desire for information: -Formal -Prognostic -Practical
	Information Tension
	Fear for the future
	Not all information is equal
	Changing role: Fear of failing
	Financial Hardship
	Downward Comparison
Being Cared For	Emotional response to disease: Physical
	Sexual difficulties
	Changing role: Finance / work / Home
	Changing relationships: Better / worse
	Soliciting support: -Asking for help -Receiving help -Negotiating help
	Accepting medication
Clinical Dissatisfaction	Dissatisfaction with clinicians: Gaining recognition
	Understanding individual experience: of symptoms
	Getting Diagnosed: Communication
	Clinician as prescriber

4.3.3 3rd RTA Combined patient / carer studies

The final RTA, of studies involving both patients and carers, identified 58 codes from the primary data, used 22 concepts, which were the basis of 7 RTA constructs. Whilst several of these constructs had already been encountered in the preceding RTA's, there are also 4 new constructs, which either add to, or contrast with those from the patient and carer only analyses. Table 6 demonstrates the RTA constructs and the new concepts, not previously encountered, which contributed to them.

Table 6: RTA of combined patient / carer studies

RTA constructs	Initial concepts
Reconstructing self	Emotional response to disease (others): Physical
Renegotiation of self (carer)	Changing roles: Carer
Solidarity: **Peer support**	Solidarity
	Support: Solidarity with others (patients and carers)
Clinical satisfaction	Being focus of concern (knowledge / intervention)
	Positive attitudes to HCP's: Respect
	Lack of concern (lack knowledge/preparation)
	Patient involvement in care
Changing relationships: Unity and distance	Changing relationships
Information Tension	Respect: Being focus of concern (knowledge / intervention)
	Patient involvement in care
	Barriers to information
	Wanting but not wanting" Information tension
	Barriers to information
	Humiliation: Lack of information
	Fear of the future: Unclear disease trajectory
	Neglect (carers): Lack of information
	Availability of services in advanced stage
Negotiating care	Negotiating care
	Negotiating care: Carer approach
	Negotiating care: Emotional / Practical
	Respect: Being focus of concern (knowledge / intervention)

Note: "initial concepts" displays only those concepts unique to this RTA.

4.4 Discussion of RTA constructs

4.4.1 *Subjugation of carer needs (RTA 1) / Re-negotiation of self: Carer (RTA 3)*

This construct includes the neglect of physical, social and psychological aspects of carer wellbeing, which is captured by terms used within the primary literature, such as "loss of self" and "helplessness".

The RTA suggests two periods of flux which require a change in social role for carers. The first and probably most obvious occurs early in disease course, as they begin to assume the role of carer, and have to make adjustments both in their own life and in their relationship with the patient - for example taking on tasks which were traditionally completed by their partner, or becoming the main bread winner within the relationship. The second period of change occurs upon bereavement, marking the end of the role as informal carer and necessitating a further period of social adaptation. This is substantiated by the subsequent construct *"Renegotiation of self: Carer"* (RTA 3), where it is evident that the process of re-definition may need to take place at the end, as well as the beginning of the care journey, with carers having to adjust to their new "non-carer" status upon bereavement. In addition, evidence for a relative lack of support for carers in this post bereavement period is identified within both of these constructs, by the two studies which included bereaved carers(9, 14).

> *"I knew he was deteriorating but I didn't expect him to die too soon."*
>
> ***Carer quote (14)***

These findings support cross sectional work, conducted in North America by Goy et al. which found that carers often felt unprepared for bereavement and highlighted the need for greater post-bereavement support (11).

The concept of "trigger events" refers to the occurrence of specific features of the disease, such as hallucinations or falls with physical injury, which may precipitate care home admission, or signal the end of established care arrangements. The breakdown of a care relationship as a result of these triggers may be related to the previous subjugation of carer needs, such that there is little reserve to deal with the extra demands on carers

at these times. These events appear to highlight the need for carer support which has the ability to identify and react to times of increased stress.

4.4.2 *"Care Tension" /"Being cared for" / "Negotiating care"*

The construct of "Care tension", derived from the carer only studies, describes the conflicting emotional reaction to caring found within the primary literature. Despite the prominence of negative aspects of care, such as those described above, one study in particular demonstrated the positive aspects of the carers experience, including the benefits of being able to care for loved ones, maintain existing relationships and physically demonstrate love(95).

> Re: Benefits of care – **carer quotes(95)**
>
> *"being able to help"*
>
> *"being able to still be together"*
>
> *"feeling appreciated"*

The theme of care as "duty" was also common. Conflicting attitudes to care appeared to exist, whereby carers could recognise their own need for support and assistance, but paradoxically, viewed this support as a failure, or a dereliction of their "duty". This impression of internal conflict was no more pronounced than in the decision to admit patients to institutionalised care, with studies describing the dynamic tension between obligations to the patient, to self and to other family members.

> *"It was, and still is, something—a decision that I (caregiver) agonise over every day. Every time I go there, I leave in tears because it breaks my heart. But I also know that my first obligation is to my husband and to my boys and to myself, and because of that, she's (mother with Parkinson's disease) three minutes from me—I see her every day."* **Carer quote(100)**

Thus we see emerging evidence for a "care tension" in which competing positive and negative emotions relating to the care role sit alongside various duties and responsibilities, both to the patient, family members and self. Negotiating this tension

may be important to the wellbeing of carers and subsequently, to the stability of patient / carer relationships.

Informed by the patient only studies, the construct of "being cared for" centres on the process of transition that is required in order for individuals to be able to ask for, receive and negotiate care. It draws on many primary concepts including; the emotional response to physical symptoms - both of the patient and their relatives, the effect of change in social role - encompassing employment, domestic duties, physical and sexual functioning, the process of accepting medication, the ability to understand, solicit and negotiate physical support and, the impact of these factors on the pre-existing patient / carer relationship. One of the key features of this construct being the manner in which supportive care is solicited, accepted and negotiated.

Within the primary literature patients and carers appear to describe types of care which can be either positive or negative in its implementation. The ability to be passive as a carer, allowing the patient to maximise their own independence before intervening appears to represent a positive model of care, whilst an interventionist approach - doing things for the patient - may be less so, even whilst achieving the same physical goals(93). Consequently, patients may find that certain approaches to care encourage and promote their independence, while others diminish it.

> *"Well, nowadays if I have trouble doing something and she comes along, she will finish it for me, which is alright sometimes, but she does it such a way that [it] bothers me . Like "get out of the way and let me do it". She makes it sound like she could do it better and easier herself, so I don't bother anymore."* **Patient quote(93)**

The researcher comments that: *"How help is provided is often key to people's ability to accept it."*

The construct "Negotiating care", developed in RTA3, shares many features with those described above, but also adds to the emerging theory of care negotiation.

One important addition is the development of the idea that informal (non-paid) care does not simply materialise, but is itself arrived at through a process of negotiation. For example, we see that the way in which carers are treated and the information they are

given, may influence their ability to negotiate care(103). Captured by the primary concepts "lack of concern" and "Neglect (carers): lack of information, the synthesis suggests that in cases where information provision was lacking, some carers felt unprepared, both for the practicalities of caring and for the future. Conversely, the experience of "being the focus of concern" elicited positive feelings towards clinicians and gratitude for interventions which supported individuals in their care role. This suggests that the process by which informal carers assume their new role can be positively influenced by professionals in the early phase of disease and highlights the potential link between information flow and successful negotiation of care.

> *"I did not receive any written document from anyone, how it [the disease] could influence both her and myself as a relative when it comes to being a relative one could never have imagined I have no idea how I'm going to behave to help X ....I have no experience of nursing, and to help someone to dress, you maybe do it backwards."* **Carer quote(96)**

It also appears that the care dynamic is influenced by individual characteristics and attitudes. In a study focusing on barriers and facilitators of function, patients describe positive (Determination / Resilience / Humour) and negative (Apathy / Laziness / Embarrassment) attitudes which influenced their function(96). Likewise carer characteristics could promote or impede independence, the key appearing to be the degree to which practical considerations such as time and circumstance could be sacrificed to help optimise patient function, as illustrated in the following quotes from separate carers in the same study:

> *"Sometimes it is a function of not getting to the bathroom on time. I told him if it happens, it happens, no big deal."(103)* **Carer quotes 1**

> *"Time is an issue. If we are running late for lunch I would do more for him to get out on time. He takes so long!"(103)* **Carer quotes 2**

From this analysis we can begin to see evidence that a negotiation is taking place – be it overt or covert, between carer giver and care recipient. This is likely to be influenced by physical (level of patient functioning), emotional (response to disease and acceptance of

help), inter-personal (pre-existing patient/carer relationship) and practical (availability of time, resources, support and knowledge) components of the disease.

This is worthy of consideration by health care professionals. It may be that effective palliative and supportive care can target and optimise this process of care negotiation, with the goal of turning the vicious cycle of care provision, carer strain and loss of independence, in to a virtuous circle of care negotiation, maximal independence and the strengthening of carer-patient relationships.

Further to this is the impact of caring on the pre-existing relationship (often, but not always, spousal) between patient and carer and the transparency with which this new "care" relationship is formed.

4.4.3   Changing relationship: Unity and Distance

Informal carers are often spouses or partners and it seems inevitable that the development of PD and the process of adjustment which is necessary on behalf of both patient and carer will alter the dynamic of their previous relationship. What is apparent from the qualitative data presented here, is that this effect can be positive as well as negative and that despite the challenges presented by this progressive disease, for some couples, relationships are strengthened. This theme is best captured by a study exploring patients' and carers' experience of support, which described three patterns of relationship change(103):

1. From Unity to Unity (United couples, remaining so in the face of PD)
2. From Unity to Distance (Couples moving apart as a result of PD)
3. From Distance to Unity (Couples becoming closer as a result of PD)

This is a further factor in the care dynamic, leading on from and related to, the process of care negotiation described above.

Might it be that by helping couples to conduct these negotiations consciously and thus identifying specific areas for support – be that practical, emotional or existential – we can help promote a virtuous circle where the care dynamic strengthens or maintains the relationship? This would require overt recognition of care negotiations as a complex,

dynamic process - but one which may be influenced for the better, rather than an inevitable and inexorable accumulation of carer strain as disease progresses, with its accompanying problems.

4.4.4   Poor Knowledge

This was a particularly rich construct, being common to the majority of carer only studies (RTA1) and can be considered as a number of related themes.

4.4.4.1 *"Disease specific knowledge"*

Refers to a lack of knowledge around diagnosis, prognosis and disease progression. Carers were described as being surprised by the prognosis of PD, the speed of decline and the onset of the terminal phase. They also described, from the time of diagnosis, a lack of disease specific information and as a result often sought information from informal sources such as the internet or patient support groups.

4.4.4.2 *"Care specific knowledge"*

Refers to the carers' preparedness, or lack thereof, for the practical tasks of caring. Primary studies demonstrate frustration at the difficulty in accessing practical information, for example information about medications. This lack of care specific knowledge may leave carers feeling unprepared for the new roles to which they are acclimatising and as suggested above, this may influence their ability to participate in care negotiations.

4.4.4.3 "*Service specific knowledge*"

Further to this is the concept of "*Service specific knowledge*", relating to an awareness of available health care services and the coordination of information transfer between services. Carers often referred to having discovered services on an ad hoc basis, rather than in a coordinated manner. For example, whilst they may have known about the existence of palliative care as a speciality and the hospice movement, their pre-conceptions about the availability and appropriateness of these services for PD was a major obstacle to access(14).

Finally we suggest that there is evidence for an "information tension" emerging from the primary data, a theme which is developed below. The literature certainly describes

carers' frustration at the lack of information provided in the early stages of disease(13, 96, 100), however, researchers also commented on the apparent variability in this desire for information (13). Whilst participants lacked information relating to prognosis and were surprised by the speed with which their loved one declined, they also felt unable to discuss end of life issues. Thus there is an apparent tension between the desire for prognostic information and the wish to avoid difficult conversations, or potentially bad prognostic news. This information tension is echoed strongly in the other aspects of our review and may represent a key target for palliative and supportive care services.

4.4.5   Addressing the Future

The construct "*Addressing the Future*" (patient studies - RTA2) incorporates several important issues. It is heavily influenced by the degree to which people with PD desire information about their disease and builds on the earlier theme of "information tension".

Primary studies often described an altered view of the future. For some people, a lack of information regarding the likely progression of their PD made viewing the future difficult:

> *"I don't know how the disease will progress, so it's frightening and I guess I was pissed off too."* **Patient quote(94)**

While for others, not knowing what to expect was a more positive experience:

> *"…not worry about it and I just go ahead and live" "I feel very fortunate that this kind of awakened that kind of thinking in me"* **Patient quote(94)**

While a third category appeared more conflicted:

> *I don't speak too much about the future,…but you've always got this worry 'Oh am I getting worse?' 'What if ..?* **Patient quote(97)**

> *As long as possible he should stay at home, and we will help each other. It would be terrifying for him to move elsewhere. If I just can go on …The future, I don't want to think about that."* **Carer quote(101)**

We know that prognosis in PD may be extremely variable with differences, for example, according to clinical phenotype - tremor dominant vs PIGD (Postural Instability and Gait

Disorder), or age of onset(106). As a clinician one may view this uncertainty of prognosis as a frustration. However, it appears that in some ways it may be used by patients to maintain a sense of hope, by allowing a range of possible futures - that is to say best and worst case scenarios in terms of disease progression. This is closely related to the primary concept of "temporality" where patients, aware that the future may be uncertain, choose instead to focus solely on the present(93, 94, 99). In contrast, there is clearly another group of individuals who use information as a means of coping, seeking to remove uncertainty where possible, exemplified in the quote below where uncertainty leads directly to anxiety about the future.

> *"...some place I could go to learn about it and learn possibly how this thing progresses. . .. I think if someone could address this a little bit so that you felt well, maybe this isn't going to be as bad as it feels like right now. It would be helpful. But something in a positive vein."* **Patient quote(99)**

Thus it is possible to see how an uncertain future, with the possibility of good as well as bad clinical outcomes, may for some patients help to maintain hope, by contributing to a sense of temporality, while for others become damaging.

An individual's approach to the future also has significant implications for the gathering of knowledge, be that formal (bio-medical understanding) or informal (practical disease knowledge) in nature, about the disease. Undoubtedly the provision of insufficient information, particularly in the diagnostic and early stages of disease, was a common finding(12, 97, 102); however, a sense also develops that not all information is equal. Whilst positive prognostic information may be welcome, less optimistic prognostication may make it difficult to project the range of possible outcomes on to our future selves which, as described above, appears central to the maintenance of temporality and hope, at least for some individuals. Interestingly, this phenomenon appears to exist for informal as well as formal (clinical) sources of information, as demonstrated by the varying attitudes expressed towards support groups (9, 96, 99, 101). Here the description of "downward comparison", whereby meeting individuals with more severe disease caused anxiety and alarm about the future, meant that many people found such groups

unhelpful(99, 101). Anecdotally this is a common phenomenon amongst PD patients with early disease and is supported by this synthesis of the qualitative literature.

> *"...it was the worst thing I could have done . . . I couldn't help observing all the ones in the last and final stages of Parkinson's which is quite unbearable."*
> ***Quote(9)***
>
> *"Everybody else there was 10, 20 years older than I am. Very shaky, lots of them in wheelchairs, couldn't really talk. I became totally depressed and just said, I'm not ready for the support group stuff."* ***Patient quote(99)***

It is useful here to reflect on the evidence suggesting that carers were often surprised by the rate of decline in the later stages of disease and that both patient and carer were often unprepared for the terminal stages(13, 14). This indicates a potentially negative side to "temporality" as a means to addressing the future, mitigating as it does against successful planning and preparation for advanced disease. It may certainly be difficult to participate fully in activities such as advance care planning and preparation for disease progression if a sense of temporality is key to one's maintenance of hope and wellbeing. Moreover, the high prevalence of cognitive impairment in PD, occurring relatively early in disease course(26, 107)' is also an important factor, meaning that continued postponement of discussion around prognosis and advanced disease are likely to result in loss of autonomy over decision making – a point highlighted in service development work reviewing the early experiences of a dedicated specialist PD palliative care service(108).

Thus, the synthesis describes a complex mix where studies illustrate the co-existence of fear and anxiety for the future, the desire for more information, and the importance of temporality in the maintenance of hope. These competing forces can best be thought of as a dynamic "information tension", see below, and may present challenges to clinicians and patients from an early stage. Resolving this tension may be a vital target for palliative care in general and specialist palliative care services in particular.

4.4.6 Information Tension

The concept of information tension, discussed above, was developed throughout this analysis and was introduced as a construct in RTA 3 (combined patient / carer studies), where it was enhanced by the identification of some additional facets:

4.4.6.1 Humiliation: Misperception of disease

Refers to a lack of understanding of PD amongst those who do not have the disease(96). This may lead to misperceptions, both of the disease and of the individual. Thus the "tension" with regard to the dissemination of disease specific information may not be confined to those directly experiencing the disease (PD patients and carers) but also to wider society.

4.4.6.2 "Wanting but not wanting"

"Wanting but not wanting " was a major theme from a North American study(12). They describe the apparent desire of participants for more information regarding prognosis, apparently existing alongside reticence and ambivalence within the same individual participant. They describe this phenomenon as *"wanting but not wanting"*. In the light of what has been learned from the previous RTA discussions above, a plausible explanation of this phenomenon is the juxtaposition of the need to obtain information *(Biomedical explanations of disease, prognostications and tailored practical information)* with the simultaneous desire to maintain hope, through temporality and the construction of multiple possible disease outcomes. Aligning these contradictory desires may lead to the ambivalence of "wanting but not wanting" described by the primary authors.

Moreover, it seems likely that an individual's requirement for information may oscillate, according to the relative importance of each of these opposing desires, at any given time. This presents a clear challenge to clinicians and those involved in the support of patients and carers living with PD. Enhanced information provision, whilst important and addressing some of the concerns identified by the qualitative research, would clearly, in isolation, fail to address the more nuanced theme of information tension (see discussion chapter 13).

In exploring this construct we should also recognise that conflicted feelings regarding the transfer of prognostic information are unlikely to be restricted to patients, but also

experienced by clinicians. To this end, previous work on end of life discussions suggests that clinicians are often worried about destroying hope or causing distress and thus rely on intuition to determine when to initiate end of life discussions(109).

4.4.6.3 *Fear of the future: unclear disease trajectory*

Finally it is important to acknowledge and address an apparent fault line in this emerging theory of information tension. An Australian study(9) discussing the emotional impact of diagnosis, identifies prognostic uncertainty as a source of anxiety which may trigger fear about the future. This appears to directly contradict the developing theory around temporality and the utility of multiple possible outcomes in maintaining hope.

There are several potential explanations for this finding. It may be, as has been suggested above, that not all information is equal. It seems reasonable that failing to receive confirmation of a good prognosis may induce anxiety, while, failing to receive confirmation of a bad prognosis supports the edifice of hope. Thus a lack of prognostic information provision may have both positive and negative consequences. Equally might it not also be the case that one's position on this subject changes according to time and more specifically disease progression?   The study describing "wanting but not wanting" involved participants with late stage disease, while the latter Australian study comprised a range of disease stages, which may explain the different approaches to this issue. Finally we must remember that qualitative enquiry is heavily influenced by the context in which it is undertaken, its participants and its practitioners. It is clear that the dissemination of information regarding PD and specifically prognosis is complex. Given this, it is perfectly reasonable that conflicting positions be reflected within the primary literature, depending on the views of individual participants and the interpretations of the researcher. The lack of uniformity only serves to highlight the dynamic nature of this construct and the difficulties presented in managing the "Information tension".

4.4.6.4 *Barriers to information*

This construct represents a perceived power imbalance between doctor and service user (patient / carer) which is identified as a potential barrier to the flow of information.   This is another facet of information tension, making the passage of information between clinician and patient / carer, even more complicated.

In conclusion, Information tension appears to be an important and complex issue with competing forces at work within individuals on both sides of the consulting room, which significantly impact the patient and carer experience of PD from the time of diagnosis. Resolving this tension is likely to require excellent communication skills, empathy and time. Neurology and Elderly care teams will be able to resolve many of these issues, but may require varying levels of support from expert palliative care services, depending on the skills and confidence within their own teams.

4.4.7 *Being Diagnosed*

This construct is heavily influenced by the process and importance of receiving a diagnosis and the subsequent response, both emotional and practical.

Studies indicated the importance of receiving a diagnosis, to validate troublesome symptoms which patients may often have been experiencing for some considerable time prior to being diagnosis. This time lag, between symptom onset and diagnosis, was sometimes associated with a feeling that symptoms should have been picked up sooner and that doctors failed to give appropriate recognition to features of the disease in the pre-diagnostic stage(93).

Receiving a diagnosis triggered a strong emotional response, as we might anticipate, with descriptions including anger, fear, uncertainty and devastation. In a study of younger patients a common theme was "unfairness", as PD was viewed as a disease of old people, so that it was unexpected and unfair to develop it at a younger age. The destruction caused by receiving a diagnosis of PD was captured by the phrase "dropping the bomb"(99).

The manner in which the diagnosis was conveyed was seen as important and represented an area of significant dissatisfaction. In general a lack of support at the time of diagnosis was noted, with information about the disease either not offered, or not remembered. One author commented that the "Human significance [of diagnosis] was passed over(93)".

As we might expect the studies often describe a strong emotional response to receiving a diagnosis of PD. These include anger and uncertainty(93, 94, 97), fatalism – particularly

for religious individuals(98, 99) and a sense of injustice(93, 97). Following acceptance of the diagnosis, individuals often sought understanding, through the acquisition of biomedical knowledge(93, 98, 99). With regard to this quest for understanding, the desire for information at this early stage appeared to vary, suggesting it may be difficult for clinicians to correctly judge the amount of clinical and prognostic detail to offer. This is in keeping with the idea of information tension, described above.

4.4.8 Negotiating Function

Negotiating function is a very strong emerging theme, drawing on all of the reviewed studies in this group. The primary concept *"re-negotiation of physical activities"* captures the daily challenge of maximising function in the face of fluctuating physical capabilities. Patients must balance the competing demands of their disease and their lives, manipulating schedules and medication regimes in order to maximise function at any given time(94, 97-99). This complex process necessitates the acquisition of information about the illness, which may be *biomedical information*, or perhaps more importantly, *practical information* - referring to the application and personal response to therapeutic strategies within one's own disease experience. This for example, may include knowledge of medication and an understanding of one's own response to particular medication scheduling, or understanding the availability of support services and how best to utilise them on an individual basis. Likewise it may mean that certain activities are sacrificed in order to maintain others.

> *"Well, I didn't notice it until 5:00 p.m. Then I just kind of froze up. Had a freeze up, they call it. I just couldn't move. . .. You just have to find out the hard way that that's what the medication is doing for you, you know. It's kind of a rude awakening."* **Patient(99)**

To conduct this negotiation successfully may require time and support. It may also rely on the acceptance of medication, something about which many patients appear anxious, but which can become an empowering process, arming individuals with the ability to better negotiate with their disease.

"We Parkinson's patients are scared or apprehensive about taking too much medicine for fear that it will, down the road, make us totally unable to function with our legs." **Patient(99)**

"If the medication is wearing off I might take the next dose a bit earlier." **Patient(97)**

Finally it is clear that this process of negotiation will also be influenced by an individuals' emotional response to the physical aspects of PD. Terms such as "shame" and "embarrassment" were common place and will influence the decisions taken by an individual during the negotiation of physical function. For example; does one prioritise the minimisation of dyskinesia at the expense of freedom of movement, due to embarrassment, or embrace dyskinesia as the symbolic representation of freedom(94).

This day to day and, possibly hour by hour, process of negotiation may be key to understanding the need for bio-medical and practical information regarding PD and its treatment. Moreover, helping patients to confront the inevitable emotional reaction to physical symptoms and medication use may maximise their ability to control their own disease, promoting autonomy and patient centred care.

4.4.9   Reconstructing self / Re-negotiating self

We have seen that the early stages of PD may produce strong emotional responses, both to the diagnosis itself and to the physical manifestations of the disease. As time passes many patients are also forced to adapt their activities, which may lead to a change in social role. For example, the loss of a job due to progressive disease, a change in financial circumstances or the inability to participate in previous social activities, may all require the adoption of new domestic roles. These ideas are incorporated in to the RTA construct *"Reconstructing Self"*. This involves adapting to the disease and integrating the diagnosis and its implications in to one's self-identity.

This may apply not only to current life roles, but also to future expectations. This was illustrated by a study in to the experience of young women with PD, where the reconstruction of self included an appraisal of one's ability to fulfil future female roles, for

example grandparenthood(97). Thus PD affects the view of our future as well as current selves, with an adaptation of hopes, fears and expectations for the future.

It is also possible to envisage this reconstruction of self as an on-going process, rather than a single period of assimilation. The primary literature suggests the emergence of what we have termed "Independence Milestones", which may mark significant points in one's journey with PD, being related to lifestyle and functional declines, rather than disease progression per se (93, 97). These may include, for example, giving up paid employment, giving up driving or requiring assistance with personal cares, and each may necessitate a further process of disease assimilation and reconstruction of self.

This theme was also present in RTA 3 (combined patient / carer studies), which contributed an additional element to the construct, with the primary concept *"Emotional response to disease (others): Physical"*.

The synthesis describes above an individuals' own emotional response to the physical aspects of PD influences view of self, and in turn the re-negotiation of self which appears to be an inevitable part of the disease. A new aspect to this is the role played by the emotional reaction of *others* to the physical manifestations of disease. This is described by one participant thus:

> *"We have had a rather large circle of friends, it has now diminished. I think that the main reason is that our friends are not capable of handling the situation. They probably want to come and see us, but don't know how to behave."* **Carer Quote***(101)*

Thus the process of re-negotiating self-image may need to account not only for one's own emotional response to the disease, but also that of our significant others.

4.4.10 Clinical dissatisfaction / dissatisfaction with clinicians

The construct *"clinical dissatisfaction"* was developed in the carer only studies (RTA 1), drawing on themes common to many of the primary papers. Consultations were frequently seen to have an overly biomedical focus, with little time for discussion of the care role, to impart practical advice, or discuss psycho-social aspects of disease. There

was also the concept of "Doctor as prescriber", with a perceived prioritisation of medication adjustment during clinical consultations.

> "…just for the clinicians to look more at the whole person, not just questions about Parkinson's." **Patient Quote(12)**

The gaps in practical knowledge experienced by patients and carers, described above, were echoed in the problems with inter-disciplinary communication, which meant that participants described a lack of coordination in care, having to search out services themselves, which was a further source of dissatisfaction.

> *". . . it was frustrating, very frustrating because you were the liaison with the health people, with the GP and you were at them to constantly to go back and say this is not working"* **carer quote(14)**

The diagnostic process was frequently described as difficult. This included delays in achieving a diagnosis, the perception of knowledge gaps among clinical staff and the dissatisfaction with the manner in which the diagnosis was communicated. There are clear links here between the handling of the diagnostic period, particularly communication, and the availability of information and preparedness for caring set out above.

The construct "Dissatisfaction with clinicians", from RTA 2, provided a direct parallel with the carer only studies. Clinicians were often felt to be primarily prescribers of medication, rather than providers of holistic care. One study in particular described a failure to address the patient experience of specific symptoms, thus creating a uniform approach to management rather than one which was patient centred(102). There were consistent expressions of frustration at the delay in obtaining recognition for symptoms – as distinct from receiving a correct diagnosis – and disappointment with the communication of the diagnosis, with for example, the diagnosis being given without an accompanying relative(97) or without the time being taken to offer explanation.

Quotes: Being diagnosed

"…I was shocked; in maybe 12 minutes of his total time seeing me, he diagnosed me with an illness, gave me no hope [and] told me to take some medicine, period. And then he dismissed me." **Patient quote (93)**

''I was more or less told you will be given some tablets and to go away and get on with it'' **Patient quote (8)**

"I don't know how other women have felt when they have been told but I found it very upsetting, felt very isolated with no backup." **Patient quote (97)**

"....the doctor said that there are diseases that are much worse, he [the doctor] did not think that it was anything special at all, but the situation I have today could not be any worse..." **Patient quote(96)**

This appears to be of particular significance given the strong themes around emotional response to diagnosis, information gathering and subsequent negotiations which we have described above. The importance of the diagnostic process and manner in which this is conveyed, has long been recognised in malignant disease and is embedded in clinical practice(110). It may be that similar consideration is required for the diagnostic process in PD. Supporting this process may be a further role for palliative and supportive care services.

4.4.11 *Clinical satisfaction*

The emergence, from RTA 3 (combined patient / carer studies) of a new construct indicating *satisfaction* with clinicians offers an important contrast to the ideas above, providing insights regarding the aspects of professional conduct which promote satisfaction.

The primary concept; *"Being the focus of concern"* appears key to feeling satisfied with clinical services. Within this, the receipt of practical support and interventions, the provision of disease specific knowledge and a client centred approach - moving consultations beyond discussion of the physical manifestations of disease - were all noted to promote dignity and make people feel respected(101, 103). This in turn engendered

positive feelings towards health care professionals and the clinical consultation. This point is re-enforced through contrast with the earlier description of features which caused dissatisfaction, including the idea of "doctor as prescriber", poor communication of diagnosis and failure to recognise the patients' own priorities in treatment.

Thus we can see positive and negative aspects of clinical interactions which are likely to shape not only an individuals' attitude towards clinicians, but also influence their own feelings towards the disease and their ability to navigate the negotiations (function, care, self) which characterise the disease experience.

4.4.12 Financial Hardship

This concept is in many ways self-explanatory, with financial difficulty relating to the loss of employment, either by the patient due to disease progression, or the carer due to fulfilment of the care role. It also played a role in care decisions, for example influencing the availability of supportive care or access to care homes(100). Clearly this dynamic will change according to the health system in which one operates, where access may be limited to a greater or lesser extent, according to ability to pay.

4.4.13 Solidarity: Peer support

The construct of Solidarity illustrates the potentially important supportive role played by others living with and caring for PD. In practical terms this often takes the form of local support groups, or carers associations.

This apparently straight forward idea is complicated however, by the concept of *"downward comparison"* which is encountered frequently throughout the literature(9, 101). Thus individuals describe the deleterious effect of meeting people with more advanced disease, generating anxiety about the future. There appears to be a process of informal information gathering, whereby people learn about the disease through encountering other patients, which has the potential to remove the protective "temporality" which we described earlier.

The occurrence of downward comparison means that for some individuals to benefit from support groups would require a new approach to addressing their fears for the future and helping them to manage the flow of information available to them. Therefore the

paradox of support groups is also inextricably linked to our earlier construct of "Information tension".

4.5   Development of Lines of Argument analysis

4.5.1   Introduction

Although for the purpose of clarity the constructs arising from this synthesis have been presented as separate entities, it is clear that they are interwoven and represent a complex network of experiences, which interact with each other. One way of illustrating this point is by developing a higher level of theory, described by Noblit and Hare as "Lines of Argument analysis". Here the aim is to identify over-arching themes, arising from the RTA generated constructs.

We suggest that four lines of argument can be seen within the data. The terms "tension" and "negotiation" are frequently used to help illustrate the dynamic nature of the relationship between constructs few, if any, of which can be viewed in isolation.

The proposed lines of argument are:

- Information tension
- Care tension
- Intra-personal negotiations
- Inter-personal negotiations

The tables below illustrate the contribution of individual constructs to these lines of argument.

4.5.2   Information tension

Discussed in detail above, this is arguably the central theme in the analysis of palliative and supportive care experience in PD. From the time of diagnosis there is a tension involving patient, carer, clinicians and other support services (for example non-clinical support groups) regarding the flow of both biomedical, prognostic and practical information. This tension operates not only between, but also within individual patients, carers and clinicians. It will influence many aspects of the disease experience, encompassing; *practical* (e.g. ability of patients and carers to negotiate an effective care

structure), *clinical* (e.g. ability of patients and relatives to plan for future care), *psychological* (e.g. opportunity to address fear and anxiety relating to disease progression), *existential* (e.g. maintenance of hope) and *spiritual* issues. It follows that as information tension is key to the experience of PD, it also represents an important target for palliative and supportive care interventions.

Table 7 : Line of Argument – Information tension

Line of Argument	RTA Construct
	Re-negotiating self
	Information Tension
	Negotiating care
	Solidarity
	Clinical satisfaction
	Changing relationship: Unity and Distance
	Renegotiation of self (carer)
Information Tension	Being Diagnosed
	Negotiating function
	Reconstructing Self
	Addressing the Future
	Being Cared For
	Dissatisfaction with clinicians
	Subjugation of Carer needs
	Care Tension
	Financial Hardship
	Poor Knowledge
	Clinical dissatisfaction

4.5.3   *Care tension*

This line of argument incorporates the competing forces which influence, for better or worse, the care structure which surrounds the patient / carer dyad. In brief it includes the degree to which an individual patient can accept and define their care needs, the type of care which informal carers are able and willing to offer, the degree to which clinical organisations prepare and support individuals for the care role and the level of access to supportive services. These issues are in turn dependent on other elements of disease

experience, not least, the successful resolution of information tension, but also the ability of individuals to assimilate and adapt to the practical, spiritual and existential impact of PD. Acknowledging the presence of each of these competing and, at times opposing, forces may help to produce care structures which are best suited to meeting the needs of all involved.

Table 8: Line of Argument – Care Tension

Line of Argument	RTA Construct
	Re-negotiating self
	Information Tension
	Negotiating care
	Solidarity
	Clinical satisfaction
	Changing relationship: Unity and Distance
	Renegotiation of self (carer)
Care Tension	Being Diagnosed
	Negotiating function
	Reconstructing Self
	Addressing the Future
	Being Cared For
	Dissatisfaction with clinicians
	Subjugation of Carer needs
	Care Tension
	Financial Hardship
	Poor Knowledge
	Clinical dissatisfaction

4.5.4   *Inter-personal negotiations*

These often result from and are defined by the tensions described above. If we consider the construct "negotiating care" as an example, the process of negotiation whilst often covert, is central to establishing care structures. Similar inter-personal negotiations appear to take place regarding the flow of information between clinician, patient and carer (in all directions). The implications of this process of negotiation may be far reaching, affecting for example, the degree to which patients / carers are satisfied with their clinical care, or the way in which PD changes the pre-existing relationship between

patient and carer. It may be that making these negotiations more transparent could improve the experience of service users and clinical staff alike and help to meet some of the palliative care needs associated with PD

4.5.5   *Intra-personal negotiations*

In contrast to the negotiations above, which concern the way in which individuals and services interact, this line of argument addresses the way in which individuals navigate their own, internal experience of PD. For example, in patients this may include; **practical** (function / care), **existential** (role, self-image) and **spiritual** (maintaining hope, fear of disease progression) negotiations. This process is inextricably linked to the lines of argument described above and would appear to be present from diagnosis, if not before, supporting individuals through this process requires good palliative care skills and may include a role for SPC.

By way of illustrating the interaction between constructs, let us take the last construct *Solidarity: Peer support* (4.4.13). An individual may use *temporality* (4.4.5) as a means of maintaining hope for the future; as such, they are more likely to experience the phenomenon of *downward comparison*; which in turn may lead them to reject services such as support groups. This will influence the *negotiation of care* (4.4.2) and the relationship between patient and carer, as well as the ability of the carer to access support. From this point it is possible to see how a change in one construct may have far reaching consequences, requiring a shift in the dynamic relationships between constructs throughout our theoretical model.

Table 9: Line of Argument – Inter-personal negotiations

Line of Argument	RTA Construct
	Re-negotiating self
	Information Tension
	Negotiating care
	Solidarity
	Clinical satisfaction
	Changing relationship: Unity and Distance
Inter-personal Negotiations	Renegotiation of self (carer)
	Being Diagnosed
	Negotiating function
	Reconstructing Self
	Addressing the Future
	Being Cared For
	Dissatisfaction with clinicians
	Subjugation of Carer needs
	Care Tension
	Financial Hardship
	Poor Knowledge
	Clinical dissatisfaction

Table 10: Line of Argument – Intrapersonal Negotiations

Line of Argument	RTA Construct
	Re-negotiating self
	Information Tension
	Negotiating care
	Solidarity
	Clinical satisfaction
	Changing relationship: Unity and Distance
Intra-personal Negotiations	Renegotiation of self (carer)
	Being Diagnosed
	Negotiating function
	Reconstructing Self
	Addressing the Future
	Being Cared For
	Dissatisfaction with clinicians
	Subjugation of Carer needs
	Care Tension
	Financial Hardship
	Poor Knowledge
	Clinical dissatisfaction

4.6 Conclusions

4.6.1 *Summary*

A synthesis of the qualitative evidence examining supportive and palliative care in Parkinson's disease and 18 fundamental RTA constructs was conducted. Whilst each construct can be described discretely there is as much to learn from the interplay between them. The Lines of Argument analysis, represented by four over-arching themes, seeks to address this, the themes – described as two "tensions" (information tension and care tension) and two "negotiations" (inter-personal and intra-personal) reflecting the state of dynamic equilibrium in which the constructs exist.

4.6.2 *Implications*

Parkinson's disease is a complex, chronic, neuro-degenerative condition. This research suggests that an appreciation of the constructs described above, and the nature of their inter-action, is crucial for clinicians seeking to adopt a truly patient centred approach to disease management.

It is not uncommon for clinicians to avoid or discourage discussions of prognosis, because of the potentially negative impact of these discussions on patients and their sense of hope(109). Aside from the obvious paternalistic concerns, in this proposed model of disease, acts of omission may be just as damaging, if not more so, than acts of commission. What is crucial is to have an appreciation of the dynamic nature of the patient / carer experience. Thus, an understanding of the concepts and themes found within the qualitative research, may be highly relevant in day to day clinical practice, for those seeking to adopt a palliative approach, as well as those involved in specialist palliative care provision. These implications are discussed further in chapter 13.

The qualitative synthesis described above was used in the adaptation of the NAT:Parkinson's disease (chapter 7) and was followed by the primary qualitative study, described in the next chapters (5 and 6).

Chapter 5

Qualitative study: Methods

5.1 Introduction

The finding from the systematic review and qualitative synthesis described in chapter 4 established that, while relatively small, there is an established literature base looking at palliative and supportive care needs in PD. Drawing this together provided some insight to the experience of people living with PD and how this relates to their potential unmet palliative care need.

The next phase of this project aimed to expand this knowledge base, using qualitative methods to further explore the palliative and supportive needs of people with PD, in relation to the entire PD journey and including the patient voice, as a development to previous studies which often involved carers or bereaved relatives. A secondary objective was to capture some of the experience of people with PD and dementia, who logically should represent a significant proportion of palliative care need in PD, but have, to date, had little representation in the studies described.

The following chapter sets out the methods used in this piece of primary qualitative research.

5.2 Setting

The Scarborough PD clinic, based in north Yorkshire, is the locality referral clinic for movement disorders in a population of approximately 230,000 people. It takes mixed referrals from primary and secondary care, with no selection according to age, disease severity or co-morbidity, avoiding the potential recruitment bias, introduced in tertiary centres, where referrals are split between Neurology and Elderly Care services according to age and stage of disease.

A database of all PD patients had been compiled for the area, and this constituted the sampling frame from which the study population was drawn. Potential participants were identified, using purposive sampling, from within this cohort of patients.

5.3 Eligibility criteria for patient participants

While the traditional model of PD holds that palliative care needs are focused in the final, palliative, stage(5, 111), we were keen to avoid this assumption regarding which patients should experience palliative care needs, in the selection process. It was therefore important to include participants from as broad a range of the disease as possible, as reflected in the inclusion and exclusion criteria, below.

Inclusion: All patients attending Scarborough Movement Disorders clinic,

- Over the age of 18 years,
- With a clinical diagnosis of Idiopathic PD,
- At any stage of the condition.

Exclusion:

- Patients in whom there is diagnostic doubt,
- Patients unable to provide informed consent,
- Patients with significant communication difficulties, such that they are unable to participate in interviews.

As mentioned above, the levels of dementia in PD, particularly in the latter stages are high(26, 107) and it was important to try and capture the experience of this group where possible. This was reflected in the above criteria, where patients with early stage dementia, retaining sufficient mental capacity to give informed consent, and the communication skills to meaningfully participate, were eligible to participate fully. However, this did not address the experience of those with more advanced dementia, who we were unable to include. In order to rectify this, carers of patients with advanced dementia, were offered interviews, asking them for their perception of the patient's needs, in addition to reflecting on their own experience.

This process of accessing the views of a non-capacitous individual, through the perception of a close relative / caregiver, has precedent in clinical practice, where it is frequently

used during best interest decision making. In addition, a similar approach has been used in palliative research where, for example, Goy et al use caregiver interviews to access the caregiver's perception of the end of life experience of their relative with advanced

 PD(11, 15).

It was felt that this pragmatic approach provided the best balance between maximising the generalisability of the research, by including as broad a range of participants as possible, while ensuring that patients' rights were preserved through strict adherence to the requirement for informed consent at all times.

5.4 Recruitment

Potential participants were selected from the existing database, which includes all patients within the Scarborough movement disorders service. The consultant responsible for this service was asked to guide the selection of appropriate participants, to ensure that the study represents all the important diversity within the service. In addition, appropriate participants were also identified at the movement disorder clinic.

This purposive sampling method, allowed us to specifically selected patients according to pre-defined characteristics; age, gender, disease stage and presence of informal career, see figure 9. The aim was to include both older and younger patients (the mean age of onset for PD being 68), carers of different gender and with non-spousal relationship to the person they cared for, and to capture the experience of patients and carers for both early and later stage disease.

Once identified through the database, Potential participants were sent a letter of invitation and a participant information leaflet (PIL) by post, with a reply slip and stamped addressed envelope. Those who registered an interest in participation were offered the opportunity to meet, face to face, to discuss the project further and ask questions. Those who did not wish to participate could either indicate this on the reply slip, or simply not reply. The letter of invitation forewarned recipients that a single reminder letter would be sent at 2 weeks, but that no further contact would be made after this point if they choose not to respond.

Those participants identified through clinic were approached by a member of the clinical team and offered a PIL, after which the recruitment process was identical to that set out above.

Figure 9: Sampling strategy for focus group participants

Focus group 1 – Patients Early (H+Y 1-2) **Up to 6 participants**

Varied according to:
 	
    1- **Age**
 	
    2- **Gender**
 	
    3- **Presence of informal caregiver**

Focus group 2 – Carers Early (H+Y stage1-2) **Up to 6 participants**

Varied according to:
 	
    1- **Age**
 	
    2- **Gender**
 	
    3- **Relationship to patient**
       (Partner / Relative/ Other)

Interviews – Patients and Carers **Late (H+Y 3-5)** **Up to 12 participants**

Varied according to:
 	
    1- **Age**
 	
    2- **Gender**
 	
    3- **Presence of informal carer**

5.5 Sample size

Qualitative studies use theoretical rather than probabilistic sampling strategies, meaning that the sample size is not intended to provide a basis for statistical analysis but, rather, to ensure that the range of participants is sufficient for the theoretical aims of the study(66).

This study targeted two focus groups, with a maximum of 6 participants in each and up to six patient / carer interviews.

5.6 Data collection

Data collection was undertaken using a combination of focus groups and semi-structured interviews. Focus groups were employed for those patients with early stage disease, defined as Hoehn and Yahr stage 1 and 2(112), who were likely to be less frail and have fewer communication difficulties, such as hypophonia, which could limit the participation in a focus group of those with more advanced disease. Separate focus groups were formed for patient and informal carers. It was important to limit the carer focus group to those caring for people with early stage disease, not only to assist subsequent comparison across groups during analysis, but also to avoid exposing participants to the potentially complex needs of later stage PD, particularly given the risk of downward comparison described in the systematic review (chapter 4, section4.4.13).

For those patients with more advanced disease (Hoehn and Yahr stages 3 - 5) who may have struggled to participate in focus group discussions, or where patients required the support of a carer during the research process, semi-structured interviews were offered, with the carer present if desired. If the patient chose to be interviewed alone the carer was also invited to be interviewed separately, but the patients' eligibility was not contingent on the carer's participation.

This approach to qualitative interviewing has been used previously in older people with cognitive impairment, where it was found that participants often preferred joint interview with their carer.(113)

Finally, as discussed above, carers of patients with dementia were invited to interview individually, to discuss their own experience of PD and their perception of the palliative and supportive needs of their loved one.

Focus groups were strictly limited to 1 hour in duration, with refreshments provided. Groups were conducted in a room within the education department of the hospice, which had good disability access and low background noise. Although the use of the hospice in this capacity may have caused concern, a preliminary patient and carer discussion group conducted as part of service development, indicated that the hospice education department was an acceptable venue for participants.

Focus groups were facilitated by the primary investigator (ER), following specific training in running and moderating focus groups, and supported by a fellow PhD student with training in qualitative methods, in order to assist with running the group.

The group was set up in such a way that participants with hearing or communication difficulties could be positioned closest to the facilitator in order to maximise their participation. Groups were scheduled for mid-morning or mid-afternoon, to coincide with the usual period of maximum PD medication effect and, where relevant, participants were asked to attend in the "on" phase.

Interviews were conducted in a location of the participant's choice, usually their own home, although one participant did prefer to be interviewed in a private room within the education centre.

All discussions were audio-recorded and transcribed, in an anonymous format, for analysis.

5.7 Data Management

The personal data of participants was held in paper form, in a locked filing cabinet, in the research office at Scarborough General Hospital, the key held by the principle investigator (ER).

A backup copy of the personal data file was kept on the hard drive of the university computer system, which is password controlled, with access only by the principle

investigator (ER). Personal data was not stored on laptop computers or mobile data sticks.

Audio recordings of focus groups and interviews were destroyed at the end of the study. Anonymised transcripts will be kept in locked, fireproof containers in the university archive for a period of 5 years. Access to these files will be restricted to members of the original research team and may be used in subsequent analysis, which has been stated in the patient information leaflet.

5.8 Data Analysis

Data analysis was conducted using the approach of framework analysis previously described in chapter 2 (section 2.18).

Initial coding was undertaken, independently, on two transcripts, one focus group and one patient / carer dyad, by two members of the research team, ER and MJ. Coding was then compared and a provisional analytic framework produced. This framework was then tested during coding of subsequent transcripts, with adjustments made where new themes were not covered. The final analytical framework (see appendix 2) was then applied to each of the transcripts in turn, coding sections of data which corresponded to each part of the framework, in the computer package Nvivo.

Forming the analytical framework in this way, and then applying it to the raw transcripts, encouraged a process of constant comparison.

Once this initial process was complete, framework matrices were produced for each of the high level codes in turn: Diagnosis, Emotional response, Care and carer, Health beliefs, Being supported and Viewing the future. These matrices were populated wherever there was coded data, using a summary and where necessary quotes. This ensured that the subsequent analysis could be traced back to the original text.

5.9 Study approvals

The study was approved by the Regional Ethics Committee (REC Number: 12/YH/0332) and the Research and Development board and was included on the NIHR study portfolio (Portfolio number: 107841). See appendix 10

5.10 Conclusion

The protocol for the primary qualitative research phase of this study used purposive sampling to ensure, as far as possible, that participants were able to represent the full spectrum of PD. Efforts were taken maximise the opportunity to participate and to minimise any associated participant burden and the protocol was approved by the regional ethics committee and local research and development departments.

Having established the theoretical and methodological positioning of the research (chapter 2) and the methods employed, the next chapter will set out the findings.

Chapter 6

Qualitative Study: Results and Analysis

6.1 Introduction

As described in the previous chapter, this qualitative study was conducted using a combination of focus groups and semi-structured interviews and the data analysed using framework analysis (see chapter 2). The chapter below sets out the results, starting with the characteristics of participants, followed by the formation of the analytical framework and finally the framework analysis. These results are presented according to the main themes of the framework matrix, to assist the reader in tracing the analysis back to the original data, in order to optimise the transparency of the analytical process.

6.2 Participant characteristics

Focus groups were conducted for participants with early stage disease, defined as Hoehn and Yahr stage 1 or 2. The characteristics of participants in each group are set out below:

Patient focus group (5 participants)

    4 male patients, all with female (spousal) informal carers

    1Female patient with no informal carer

Carer focus group (4 participants)

    Gender: all female

    Relationship to carer: Spouse, although 1 had also cared for a sister with PD.

Interviews were conducted for participants with later stage disease, defined as Hoehn and Yahr stage 3 or above. Three interviews were conducted in patient / carer dyads, while one was a lone patient (see table 12). No participants asked to be interviewed separately from their carer or spouse.

Despite several scheduled appointments there were no successfully completed interviews for carers of people with dementia.

Table 12: Interview participants (Advanced disease)

Interview 1	Female patient **P1** / Male carer (spouse) **C1**
Interview 2	Male patient **P2**/ Female carer (spouse) **C2**
Interview 3	Female patient **P8**/ Male carer (spouse) **C3**
Interview 4	Female patient **C9**, no informal carer

6.3 Formation of the analytical framework

A description of the steps involved in production of the analytical framework can be found in the methods chapter (chapter 5). The framework had 6 main themes, consisting of 16 constructs and 115 primary codes. The full analytical framework can be found in appendix 2.

6.4 Results of framework analysis

Where quotes are cited in the text, focus group representatives are identified by their title (also indicating gender) and the first letter of their first name, for example: **Mr T**, while interview participants were identified as a patient (P) or carer (C) with their participant number and gender indicated, for example: **C1 Male**

Six themes arose from the data and are discussed here in order. At the end of each theme or subtheme findings, an analysis will be presented.

6.5 Findings Theme 1: Diagnosis (Identifying symptoms / Receiving a diagnosis)

6.5.1. *Identifying symptoms*

An interesting feature of PD as a disease is that, while the well known diagnostic criteria are based on the motor features (bradykinesia, rigidity, tremor and postural instability), it is often the non-motor features of PD which emerge first. This important period, where non-motor features of the disease are present without the diagnostic motor features, has become known as the pre-motor (sometimes termed pre-clinical) phase of the disease.

This lead pre-motor phase is of interest to researchers looking at neuro-modulatory medication, but also has a practical importance for the study of care needs, in that patients may well experience symptoms for months, or even years, before they display the diagnostic features of PD.

This phenomenon was clear in the qualitative data, with patients and carers describing symptoms which had been present for some time prior to diagnosis:

Patient focus group

Mr Tr: *I think back to issues I'd had ten, fifteen years prior to being diagnosed and obviously I had signs of Parkinson's but undiagnosed for a long, long time. The only thing that alerted anybody to it was a left hand tremor.*

In addition, once motor symptoms do emerge they can be subtle and previous studies have shown that diagnostic accuracy, particularly amongst non-specialists is often low. This is reflected in the common experience amongst participants of having undiagnosed motor symptoms for a number of years, having been incorrectly re-assured about motor symptoms, or managed for the wrong condition:

P9 Female: *I knew that something wasn't right and I'd been to my GP a time or two and she had said, oh I think it's just a reaction to something else. I don't think you've got Parkinson's or anything like that.*

Patient focus group

Mr T: *I pointed out (to GP) and I said "Oh by the way I've got slight movement in me left hand". "Oh" he said "it's nothing"*

The sense of relief at diagnosis was not limited to patients, but was also experienced by their carers. It was evident that while some patients were very aware of their symptoms prior to diagnosis, others had either not recognised them, attributed them to something else – such as an old injury, or chosen to ignore them.

Patient focus group

Mr T: *I just noticed this slight tremor …… and a few months later, I was ignoring it because I thought it was nothing really, thought maybe it was old age, which can come quite easily.*

In these instances the role of the significant others, often spouse, in appraising early features of disease and initiating medical assessment was greater and may account for the relief felt once a formal diagnosis had been made.

Carer focus group

Mrs B: *I felt for some time before that it (was) probably Parkinson's that he had because his tremor had become more obvious over the months previously.*

Carer focus group

Mrs J: *Yeah, well my husband had been shuffling about for a considerable time, and then the shaking started, and it wasn't until me son's wedding that, well afterwards people commented like has he got Parkinson's like, you know, you know, going to doctors, sort of thing (laughs) under pressure, he didn't want to go.*

C1 Male: *Well, I was pleased that – to know really what it was she was slurring her feet so badly. And although I didn't suspect it was Parkinson's, as soon as he said it was I realised that it was immediately that's was what had been causing her to shuffle her feet.*

6.5.2 *Receiving a diagnosis*

During the data analysis the process of receiving a diagnosis of PD and the implication this had for the patient and carer became a major theme. This theme can be considered to have three aspects: i) delivery of diagnosis, ii) initial reaction to diagnosis and iii) subsequent response to diagnosis.

6.5.2.1 Delivery of diagnosis

It was common for participants to describe dissatisfaction with the manner in which the diagnosis was conveyed. This was often related to the perceived brevity of the consultation and the lack of information given regarding the diagnosis.

> **P8 Female**: …a*nd it was the way I was told. I was just told, 'you've got Parkinson's and go away and deal with it'. It wasn't really done in a very sympathetic way.*

> **P2 Male**: …*at the end of the day she (specialty doctor) just said to me, "You've got Parkinson's," and there was no explanation. All I did was come out of there with no explanation of anything at all.*

> **Patients focus group**

> **Mr T**: *Well I've had no tests and no questions.*

> **Mr D**: *I, I haven't. Yeah, I was just told I had Parkinson's and that was it*

In other instances it was perceived by the patient that the clinician was in some way holding back, trying to "ease the blow" of diagnosis.

> ***Patients focus group – early disease***

> **Mr T**: *Well (specialty doctor) said when they diagnosed me he said "Well you needn't worry, it won't kill you".*

> **Mr D**: *Mm.*

> **Ms S**: *Yeah.*

> **Mr To**: *But that is not quite true is it? I mean…*

> **Mr T**: *Might lead to deterioration which…*

> **Ms S**: *It could…*

> **Mr T**: *…get, get something else, lower your… resistance*

> **P8 Female**: *The doctor said, "Don't worry about it, we can help you." That's all that was said but it doesn't mean anything if they don't expand on that.*

However, the experience of diagnosis was not universally negative and other, more positive, experiences of diagnostic delivery were also shared. In direct contrast to the examples given above these tended to highlight the time taken by the clinician and reflect the positive aspects of their manner and the amount of information offered:

> **P9 Female**: *I went to a very nice neurologist…… who I found quite helpful in his manner in the way he went about things. I felt he'd picked up on everything very well and he was quite certain that it was Parkinson's.*

It is also interesting to consider here the experience of the carer, particularly when not present at the initial consultation. In this instance the diagnosis was delivered to them by the patient, outside of the controlled confines of the clinic and without the ability to ask questions or seek clarification.

> **C2 Female**: *He'd gone on his own, came home and walked through the door and said, "Oh, they say I've got Parkinson's," and he came home with nothing and they hadn't told him anything.*

This geographic and temporal distance from the initial consultation could result in misunderstandings, in terms of the implications of the diagnosis and also denied the opportunity to be present and support their loved one:

> **C2 Female**: *I mean I didn't know – I wasn't even with him because I never imagined he would receive this diagnosis.*

> **C3 Male**: *I was shocked, again because I didn't really know anything about Parkinson's so I thought 'does that mean she's going to die?' I wasn't sure.*

6.5.2.2 *Initial reaction to diagnosis*

The range of initial reaction to receiving the diagnosis can again be viewed in groups. There are those who demonstrated surprise or shock, those that experienced relief and some that suggest fear for the future, particularly regarding future decline.

P8 Female: *I was waiting for something to kick in. I was waiting for it to go worse before it was actually worse.*

P1 Female: *I didn't believe it. I only had one of the symptoms and that was very poor writing, it got very bad…… and I was reluctant to accept it even then because I just didn't feel ill.*

Patient focus group

Mr T: *I was amazed, I thought it was a, some sort of trapped nerve……I wasn't aware of any other symptoms. So it was a bit of a shock, I must admit.*

Carer focus group

Mrs C: *I, we sort of knew there was something and then just very, very slight shake, and I think it was relief really. You know, you, you had a diagnosis, you knew what it was.*

This range of reaction and the way in which it may relate to prior appraisal of symptoms is discussed below.

6.5.2.3 *Subsequent response to diagnosis*

The initial reaction described above was followed by a response to the diagnosis. In particular participants appeared to differ in the degree to which they acknowledged the diagnosis, the way in which they approached telling others and their attitude towards seeking further information.

There was a clear divide between those participants who recognised the diagnosis and confronted it head on and those who made an apparently deliberate decision to ignore it.

Carer focus group

Mrs H*: …he was told, yes it is Parkinson's. And it hit him hard, very hard, the fact that he'd actually got something wrong with him…….and hard for me, because he didn't want anybody to know and he didn't want to talk about it.*

Mrs B: *I think I've been very fortunate from that point of view because (patient) accepted it really well, he, his sentence, when he was asked how he felt about it, was "Well it's really not much different from getting older, is it?"*

Patient focus group

Mr D: *Two months prior to being diagnosed with Parkinson's I was diagnosed with severe prostate cancer. So (sighs) I ignored the Parkinson's, I didn't really take it on board.*

This phenomenon of either acknowledging, or ignoring the diagnosis seems to be echoed in other aspects of the participants' response to being diagnosed. For example some went to great lengths to immediately let friends and even strangers know their diagnosis, while others took the opposite view, even hiding the fact that they had PD from family.

Patient focus group

Mr C: *Well, you know, I can't remember quite how I felt, and I know I was upset. I was also determined that I would tell other people straightaway.*

Ms S: *Actually I didn't tell my daughter……I mean I didn't tell people around me unless they asked, you know, what's wrong with me. You know, in, in a way I just decided (laughs) to, you know, ignore it, as much as I could for as long as I could*

Carer focus group

Ms H: *…and hard for me, because he didn't want anybody to know and he didn't want to talk about it.*

The idea that the response to diagnosis was governed by the degree to which the individual acknowledged it is elaborated further by the varied responses in terms of information seeking. Where some participants immediately sought to gather information, often having to use the internet or books (see section 6.9.3 on information) this was a stark contrast to those who actively sought to ignore the condition.

P9 Female: *But by that time (seeing new consultant) I'd done a lot of reading up myself and I knew what was going on. I was relieved to know what it was.*

P8 Female: *When I first went to see the consultant and he said I had Parkinson's I was shocked because I didn't know what it was and I felt numb. I didn't really feel upset or depressed or sad or anything I just came home and wanted to find out all about it.*

6.5.3 *Analysis*

The failure to correctly recognise symptoms was understandably a cause of frustration, amongst both patients and carers, but also appears to have important implications for the diagnostic process that follows. The feeling of knowing something is wrong, without receiving validation of that fact from health care professionals was a source of anxiety for some, reflected in the relief expressed on finally receiving a diagnosis. This is also linked to the perception of health care professionals (see also section 6.10.1) and the ability to access support (see also section 6.10.2).

Thus, even at an early stage the potential exists for very different experiences of the diagnostic process, between those who have recognised the seriousness of symptoms and may be frustrated or anxious as a result of diagnostic delay and those who are approaching the diagnostic consultation unaware of the potentially serious nature of their symptoms. These groups may have very different emotional responses to the diagnosis and perhaps different supportive care needs stemming from their experience. Further to this, when a spouse or loved one is the primary driver for presentation, having recognised early symptoms, their response and needs may be very different from those of the patient. It is interesting here to reflect on the early emergence of a theme - "vigilant protector" relating to carers, which is expanded upon in Care and Carer (section 6.5.2) and the development of a *care role* and associated *burden of care*, even in the process of diagnosis, long before we may perceive an individual with PD as "needing care".

6.5.3.1. *Receiving a diagnosis*

The manner in which participants experienced the actual receipt of diagnosis varied considerably, but there were common features of those whose experiences were good (time taken, information offered, friendly manner) and those whose experiences were less good (rushed, no understanding imparted, clinical manner). The nature of this

difference is not surprising, suggesting that a patient centered approach which allows time, seeks to assess understanding and reinforce information offered, produces a better patient experience and greater satisfaction with the consultation. What is more surprising is the degree to which patients' experience did not reflect this ideal approach. There may be reasons for this apparent lack of patient centeredness, for example time pressures in busy clinics, but these should not supplant a search for deeper explanations. For example, in the patient focus group quotes above, describing "easing the blow", there is a sense of paternalism, with the clinician offering a palatable half truth while ignoring the known increases in relative death rates in PD cohorts compared with the general population(26, 114, 115). What is more, the patients see through the pretence, recognising from their own experience that death rates are likely to be higher.

The misperceptions that may result from a failure to manage information and ensure a degree of understanding at first consultation are obvious from the quotes above, ranging from a belief that there is no increased mortality associated with PD, to believing the condition to be terminal within months. This failure of communication appears to present a specific challenge in terms of unmet palliative and supportive care need.

Finally it is valuable to reflect on the diagnostic experience of the absent carer. Denied the opportunity to ask questions, or to support their loved one as the diagnosis is given, their own experience is uncontrolled, receiving the diagnosis "by proxy", without the use of professional communication skills or the opportunity to explore its meaning in full. There is also a sense of guilt in the quotes from carer C2 at not being present, which reflects the fact that, even at this early stage, a type of care dynamic exists, as spouse or loved one seeks to care for a newly diagnosed patient. Given the importance of informal carers in PD and the desire to support them, this may be one aspect which could be targeted, to improve their early experience of PD and their relationship with clinical teams.

6.5.3.2 Initial reaction

The quotes above illustrate the range of initial reactions to being diagnosed. They also demonstrated that many participants experienced a long pre-diagnostic phase, with symptoms for months or years before discovering they had PD. This fits very well with

the sense of frustration (at delayed diagnosis) and relief (at validation of symptoms) which participants display. In contrast, those who had not experienced pre-diagnostic symptoms, had failed to recognise them, or had attributed them to another cause, tended to be shocked and surprised when learning that they had PD.

6.5.3.3 Subsequent response

The results also demonstrates very different participant responses to three key issues in the period soon after diagnosis, which may affect the supportive and palliative care needs of those individuals in this period. If, for example, the response is to ignore the diagnosis, then openness to and requirements for disease specific information will be very different to the person who acknowledges the condition and seeks to gain disease specific knowledge as part of a coping strategy. Likewise, the individual who seeks to hide the diagnosis from friends and relations may have specific needs relating to social isolation and stigma. This is not to suggest that one response is right or wrong, but rather to recognise that the needs associated with each position are likely to be highly different.

In discussing these findings it is important to consider the impact on informal carers. Several quotes demonstrate fear and anxiety on behalf of carers, following the diagnosis of loved ones, this seems to particularly be the case where there is a lack of information or a misperception regarding the implications of a diagnosis (i.e. thinking that PD is terminal within months). In addition, although some might have considered informal "care" to begin in the later stages of disease and be largely physical, it is clear that a carer / patient relationship (as opposed to care / spouse) has begun to form even in these early days. This is evidenced by the need to become vigilant advocates for reluctant loved ones in the pre-diagnostic phase, to be able to offer support during the receipt of diagnosis and to gather information in their own right following diagnosis.

These findings have several implications, both for the individual clinician and for the construction of effective services, which are discussed in chapter 13.

6.6 Theme 2 – Response to disease

These first section of results, relating to the carers' emotional response, are presented with the carers for patients with early disease first, followed by those in late disease, in order to allow easy comparison between the two.

6.6.1 *Emotional response of carer – early disease*

Frustration was a frequently cited emotion from carers of patients even in the early phase of disease (Hoehn and Yahr 1-2). Some of this frustration stemmed from the phenomenon, described above, of ignoring or denying the diagnosis. Inevitably this could lead to a perceived failure on behalf of the patient to address, or even to acknowledge, the impacts of PD on everyday life.

> **Carer focus group**
>
> **Mrs H**: *Sometimes I think I, I'm going mad, simply because I'm up against this brick wall who will not accept that his mind is not as fast, he doesn't walk as fast, he doesn't react as, and it's like a brick wall.*

Frustration was also evident in relation to the the lack of motivation, which is a recognised feature of PD, but which could manifest as inactivity.

> **Mrs C**: *Oh I think we lose our temper, we don't want to, but I think it's so…*
>
> **Mrs H**: *Well because I know…*
>
> F: *(…). (talking together)*
>
> **Mrs H**: *…I know, I know that possibly he could do more if he'd just get that bit of oomph. (laughs)*
>
> **Mrs B**: *Motivation.*

However these expressions of frustration and occasionally anger were often tempered by a sympathy for the challenges faced by the patient, a recognition that they were not really to blame and, most importantly by a sense of pride. This was manifest both as pride looking back on past achievements and in current achievements against adversity.

Mrs H: *…..and yet obviously, when you talk to people, it is, it's part of Parkinson's*

Mrs B: *….and there were twelve pieces of scripture that he read, and he read it in front of everybody. And I was so proud…*

Mrs C: **Re: previous employment** *…you know, and his, his brain was wonderful…but I don't think he could do it now.*

Other notable emotional responses from carers of people with PD in early disease included a strong sense of solidarity, both with the patient and with each other. This was best demonstrated in relation to the aforementioned frustrations whereby, despite the patient's refusal to admit the extent of the impact of PD to doctors, the carer recognises that she could never "betray" him by interjecting on his behalf.

Mrs H: *….it is hard for a man, for a wife to sit there and say "Oh yeah, but I mean he's forgotten this and he's so slow" and that, and I wouldn't do it to him…*

Mrs B: *No, I wouldn't, I couldn't do it.*

Mrs H: *…I would never do that. And, but I sit there and (consultant) is sat there (laughs) or (nurse specialist), and I think who is this man that I've brought here? (laughter) It's not the one that was, I got up with this morning, you know, it's, it's really strange.*

Mrs B: *Oh dear.*

Finally, humour was an important part of the emotional response to the adversity imposed by PD. Once again, this could be seen in distinct categories, namely the humour between patient and carer, and the humour between carers – in this case other members of the focus group, which may not necessarily be compatible.

6.6.2 *Emotional response of carer (late Dx)*

Carers for people with more advanced PD expressed feelings of frustration, but also expressed anger more commonly than those in the early disease group. This anger was usually directed at the disease, rather than the person with PD, although on occasion

there was also anger at perceived personality changes which made coping with the condition more difficult.

> **C3 Male**: *I feel angry about it quite often. It just annoys me. When I seem angry with you it's not you personally, I'm sort of angry with the condition you've got.*

> **C2 Female***: I suppose it depends on what a person's personality is to start with, and (patient)'s always been a bit stubborn anyway but that has increased, so he's really stubborn. And we tell him things …….to carry out things for his own safety ……. but he seems to not take much notice of what we say which is very frustrating.*

Once again, these emotions were tempered by the knowledge that the situation was equally difficult for the person living with PD, that they were not personally to blame and, interestingly, that in some ways they were not the same person that they had been at the onset of disease. In one case in particular, where there appeared to be a significant amount of carer strain and difficulty managing the patient / carer relationship, there was a real sense of loss, both personal and on behalf of the patient.

> **C2 Female:** *Life just changes and it's never the same again and it's very frustrating.*

> **C2 Female:** *I'm talking to a brick wall here. It's a very frustrating condition, it's frustrating for the person who has it, we appreciate that, but it's also frustrating to live with.*

> **C2 Female:** *They're not as strong character as they were before. So sometimes it's more like looking after – this sounds awful, but a child rather than an adult.*

> **C2 Female:** *Family life is really affected because at times you feel Parkinson rules the house because everything revolves round it.*

6.6.3 *Analysis*

The brick wall analogy is used by two separate carers in reference to the frustration of trying to understand and be understood by, the person with PD. While this is a common expression, its frequent use in reference to PD is interesting and appears to reflect some of the known physical manifestations of the disease. Hypomimia (paucity of facial

expression) hypophonia (soft voice) and bradyphrenia (slowness of cognitive processing) are all physical manifestations which make communication more difficult(116, 117) and could relate to the "brick wall" metaphor. The "brick wall" also serves as a description of the apparent barrier between carer and the pre-Parkinson's person, adding to the sense of loss.

It is interesting to consider how this emotional response influences the dynamic between patient and carer. The "brick wall" clearly symbolises a breakdown in communication – physical as much as psychological, in a strained patient / carer relationship. The degree of strain is highlighted by patient's comment that he sometimes doesn't call for help after falling out of bed, as he feels he will be blamed:

> **P2 Male**: *I don't like to get them down. Sometimes they tell me it's my own fault because I try to do things when I shouldn't.*

We are able to contrast this with other patient / carer relationships, also in those with advanced disease, where the importance of good communication regarding the physical manifestations of PD was identified by the participants as key to successfully maintaining the care relationship.

6.6.4 *Addressing symptoms*

In discussing the impact of diagnosis a distinction was made between participants according to the degree to which they acknowledged the diagnosis. This pattern can also be seen when examining the way in which participants addressed the symptoms of PD and its impact on their lives.

One strategy appears to involve engagement. Recognising the manifestations of PD, both personally and publically, and taking deliberate steps to minimise their impact, both functionally and socially. In early disease this may involve simply carrying on as normal:

> **Patient focus group**
>
> **Mr D**: *Actually having Parkinson's doesn't bother me, I just get on with it and, you know, make the most of it, it really doesn't.*

> **P9 Female**: *I think I had just decided that I wasn't going to let it beat me. I had just decided that I would have the nice, slow onset*

However in later stages it appeared to involve more a process of gradual adaptation, recognising the effect of physical decline and altering activities accordingly.

> **P9 Female**: *But I don't go for walks as I used to. But there are other things to do. You adapt to what your abilities are.*

A prime example of this continuing in to the later stages of disease is the response to motor fluctuations, with several respondents describing a deliberate change in planning, timing and duration of activities, to account for predictable "on – off" fluctuation.

The alternative strategy involved denial of the impact of symptoms and withdrawal from activities. This was sometimes accompanied by an adverse response to perceived social stigma, or anxiety at not being able to perform socially to previous level.

> **Patient focus group**

> **Mr T**: *The fact that you've got a tremor, you feel, I don't know, that you can't do anything*

> **(C2) Female:** *But (patient) doesn't – he's not happy in large numbers of people*

> **(P2) Male**: *I used to go to the dinners and allsorts, so I don't go to those very often…. it's the embarrassment really of not going*

The analogy used by a carer for this approach was "Peter Pan" – as the patient employed great effort and physical resource to convince others, including the medical team, that the disease was having less impact than was really the case.

Interestingly, the data does not necessarily imply that these approaches are fixed. One participant describes not telling work colleagues about her diagnosis in the early stages, as part of a strategy to "appear healthy" – a strategy which she acknowledged was in part self-delusion. However, as the disease progressed she then acknowledged its impact and made great efforts to adapt activities.

P8 Female: *Because I didn't want to be thought of as an ill person, I suppose. I didn't want to have this condition. I suppose I was a bit burying my head in the sand. I thought the longer I can live without medication – I don't need this medication, I'm a well person*

P8 Female: *Yes. And now it's several years down the line and you get to adapt to it…… So I get on with the life I have, the condition I have and I know what I can and can't do so I don't needlessly try and do something that I know I really can't do*

6.6.5 *Analysis*

The importance of this section of results is in highlighting the different impact on the lives of patients and carers of different coping strategies. The theme does not exist in isolation, but can be linked back to the initial response to diagnosis (section 6.5.2) and forwards to the later analysis on approaches to medication, information and planning for the future). Tracing this seam of evidence, through the different phases of disease, is useful and makes the theme of analysis more robust. The switch in approach described by participant P8 is particularly interesting. It suggests a tipping point, in terms of disease progression, at which point an "ignore" policy is no longer effective / positive, an idea that is linked to the later description of disease milestones. Moreover, it may be that maintenance of an "ignore" policy beyond a certain time, implying embarrassment, anxiety and social withdrawal, has increasingly negative connotations. If this is the case then it also suggests a potentially fruitful target for palliative and supportive care, in that positive coping strategies in early disease stages may help to improve outcomes downstream.

6.6.6 Adjusting roles

The manner in which patients and carers are able to assimilate PD in to their lives and the degree to which this necessitates an adjustment to previous roles is related to the discussion above. There were several examples where carers began to assume both physical and decision making tasks which had previously been the domain of the patient. These were often most noticeable when certain gender stereotypes were involved, particularly involving perceived male activities such as work place conflict or driving.

Patient focus group – discussing anxiety related to conflict

Mr T: *..conflict has been part of my professional life and you just accept it, you know, you deal with issues but now I, I'll avoid getting in a conflict issue with somebody.*

Carer focus group

Mrs H: *…even though he doesn't drive that often, it's always there. But…*

Mrs C: *It's hard for a man I think not, to have that taken off them.*

In another instance a female patient describes the adaptation necessary to go from the person who delivers care, to one who receives it:

P1 Female: *I visited for Victim Support for a lot of years …… I've always liked doing things like that, haven't I? But I feel now people are doing it for me.*

6.6.6.1 *Analysis*

This biographical disruption was also a cause of anxiety for the future, for example regarding financial security and the need to provide ongoing care as the disease progressed.

However, this should not be viewed as a passive process. In a final example, a patient with advanced disease explains how she had always been determined not to be a "Parkinson's person". This participant had adopted the approach of acknowledgment and gradual adaptation, and her approach seems to contrast with others who adapted less well. There is a suggestion here that acknowledgement of PD and its consequences helped to facilitate a greater degree of self-determination. This may be particularly true when it comes to approaching the future and end of life decision making, a topic discussed later (section 6.9.5).

6.6.7 *Fear*

Along with frustration (see above) fear was the most frequently described emotion. For carers this was manifest in a number of ways, the first being fear for the patient, in terms of physical, cognitive and social decline and the associated risks.

Carer focus group - discussing their anxieties

Mrs H: *Probably the mental side, you know…*

Mrs B: *Yes, yes.*

Mrs H: *…that worries me…*

Mrs B: *Yes.*

Mrs H: *…the mental attitude, the mental side of it, I'm frightened how that develops for a safety kind of thing*

Fear was also described in relation to the uncertainty of disease progression and knowing what to expect:

Mrs J: *No, he won't, he, he still won't come to terms with it. He, he's started taking the medication, but…it's frightening me cos I don't know what to expect.*

This was expanded upon by other participants when discussing future concerns about financial security, particularly relating to uncertainty surrounding the continued provision of free state care.

Mrs C: *I think one of the most frightening things is your finance, you know, losing your property.*

Possibly the most interesting cause of fear, from a carer perspective, involved concerns about what would happen to the patient if "something happens to me" so that they were no longer able to provide care.

Mrs J: *But I think, I mean you don't know what's going to happen to yourself as well, you need the children to be able to be…*

Mrs H: *That's my fear is because I know that (patient) is already, that he, you know, I mean I do it all now. My fear is if that happens to me and it, and all this with, my mother had Alzheimer's and, and I think I've seen it all and I try to make, I want to make life easy for them, but it's a bit scary, because…*

Mrs B: *Mm, I agree*

This fear at the prospect of the carer / patient relationship being broken, was also expressed by patients, who recognised the role care played in maintaining wellbeing.

> **P8 Female**: *At the moment I'm pretending that you (carer) might outlive me so that he'll be there to look after me. But the chances are that that might not be the case.*

This is linked to the most prominent cause of fear in patients, which was a fear of the future, particularly associated with a loss of independence. It is interesting to note how one respondent described this fear increasing with each step change in physical condition – this is linked to the later description of disease milestones (section 6.9.1). Within this anxiety regarding future decline, some patients had fears relating to the development of particular, feared, complications such as falls or neuro-cognitive decline.

> **P9 Female:** *the thought of having severe depression that is bad for me because I've been there. The thought that that could occur again through Parkinson's that does frighten me.*

6.6.7.1 *Analysis*

Fear is a prominent response to PD and particularly the experience of disease progression. Patients and carers describe a number of shared fears – i.e. fear regarding the uncertain future, although the emphasis was slightly different for each group, for example patients worried about becoming dependent, while carers worried about no longer being able to meet the increasing care demands as disease progressed – rather selflessly, carers seemed less concerned with the implied escalating care burden, but rather the effect on their loved one if they were no longer able to meet the challenge.

It may be that some of these fears can be alleviated through changes to current practice, for example greater exploration of information needs in order to address uncertainty (See also SLR – chapter 4, section 4.4.4), or by improving self-determination in late stage disease through greater awareness of and access to Advance Care Planning. However, in the context of a progressive condition, there will be some fears which cannot to directly addressed and are likely to require greater access to support services to alleviate some of their impact.

6.7 Findings Theme 3: Care and carer

6.7.1 Carer as individual

This section of the results highlighted the biographical disruption experienced by carers as a result of PD and had two main aspects. The first can be called anticipated sacrifice, whereby the future changes which may be necessary as the PD progressed, in order to accommodate an increased care role, were explored. Examples included the loss of paid employment and changes to accommodation.

> **C3 Male**: *I can't see that I'm going to be able to work till I'm 65 because within maybe a few years somebody's going to have to look after you aren't they, more full time.*

> **P8 Female:** *I can bury my head in the sand and I often think I'm going to be able to do this for the next umpteen years until you retire. But…..the minute you retire I think you'll be helping me.*

> **Discussing the time needed to care**

> **P1 Female:** *No, he had to give up his hobby. He played in a brass band and he can't do that.*

> **Discussing the need to find suitable accommodation**

> **P1 Female:** *You're not keen on that one?*

> **C1Male:** *No not really.*

> **P1Female:** *But you understand why I would like it*

> **C1Male:** *Well, yeah, I feel it was communal living, although you're not quite that way, but I'd feel that way.*

The other element to this biographical disruption, touched on previously, was a sense of loss, on behalf of the carer. This could relate to a loss of the future which they had anticipated before PD, a loss of social activity, or a sense in which they lost their loved one to PD.

C2 Female: *Well, I don't know if it affects everybody but I feel the Parkinson's changes the person's personality, so (patient) isn't the man that I married.*

Some of these points, particularly regarding a concern for future care provision, relate to the concept of the carer as a vigilant protector, described below.

6.7.2 Vigilant protector

The first part of this theme relates to carer vigilance. There were multiple examples within the data of carers being the first to notice physical and cognitive decline, sometimes before the patient themselves. In fact there was a sense that they were in a heightened state of alertness, trying to detect change, both positive and negative, as early as possible.

Carer focus group

Mrs H: *But yeah, and it's three and a half years, they've just doubled up his dose of medication, his tremor's quite bad and he's, walks very, very slowly and I, I have to watch him because mentally he doesn't think there's anything, but he has slowed down. And I have to watch what he's doing, without him even realising….*

Mrs B: *But he doesn't realise, as you said, just exactly what a difference there is in him, you know.*

Mrs H: *That's right, it's, yeah, I mean I can see a huge difference in, in my husband.*

C3 Male: *I knew. I could feel a sort of slight tremor in you but nobody else would be able to tell…*

C3 Male: *I notice that if you get stressed and anxious your symptoms get worse. So if you try and be more calm and with an optimistic outlook and so on you seem a lot better.*

However this vigilance was not passive, but rather it appeared to represent one of the burdens of care, because having recognised a decline there was a responsibility to act on it, in order to protect. This protective role was evident in a number of different ways and was represented within the analytical framework by the construct of carer as guardian

(see section 6.7.2). The first and perhaps most obvious example was physical protection, for example guarding against accidents or falls.

Carer focus group

Mrs H: *And I have to watch what he's doing, without him even realising, if he's on the cooker, if he's on, you know. Today he even, he turned some soup off and he's going down to the cupboard to turn the handle on the cupboard, instead of the gas, and this is a worry.*

There were however more intriguing examples of guardianship, for example socially, helping loved ones avoid difficult situations or stigmas, in one instance a carer has post from Parkinson's UK delivered in her name as her husband is worried about the postman knowing his diagnosis.

Emotional guardianship, as well as fierce loyalty, was demonstrated when carers acknowledged that they were careful how much information to offer to clinicians in clinic, in order not to betray their loved one, even though they felt that the portrayal of the impacts of PD were unrealistic

Carer focus group

Mrs H: *for a wife to sit there and say "Oh yeah, but I mean he's forgotten this and he's so slow" and that, and I wouldn't do it to him…*

Mrs B: *No, I wouldn't, I couldn't do it.*

Mrs H: *…I would never do that….*

Guardianship also extended to a detailed consideration of the future, particularly in instances where the person with PD may not have been ready to engage in with this type of planning. Examples included protecting the shared future of patient and carer, evidenced by carers organising additional help at home, or home adaptations, as necessary evils in order to preserve their current social arrangement. Similarly there were examples of protecting an individual future, which was demonstrated in the desire to plan for future care in the event that the carer were to die, a fear shared by patients

and carers in this study, as discussed above. Finally, there was an evident desire to protect the future of others, particularly children, either by helping them to avoid a care role, or preparing them for it, or hiding the severity of disease.

Carer focus group

Mrs H: *I already do everything and so my big concern there is if suddenly I found I wasn't, and the only one thing that I really have seriously thought about is making the children able to deal with everything if anything happened to me, simply because that does worry me, because I know that (patient) couldn't cope with anything.*

These observations are supported from the patient data, which reflect some of the protective roles performed by carers. To this end there is recognition that the care relationship helps to preserve independence and reduces concern for the future.

Patient focus group - Discussing concerns for the future

Mr D: *I think if anything happened to my wife I might have a different view. She gives me tremendous support…..But I think without good support it, I can understand you being concerned*

Recognition that the carer has an integral role in protecting the patients' future:

Mr C: *…we're really assuming that the worst will happen so that all our planning is, particularly financial planning, that sort of thing, is to, my wife will be left to do it, things like Powers of Attorney and things like that.*

It is interesting at this point to contrast the findings above with the data from the female patients and the sole patient who did not have an informal carer. Some of the vigilant protector traits can be identified in the language of female patients, far more so than the male patients. For example, both female patients, P1 and P8 express concern at the physical impacts of care on their husbands and P8 discusses at various times a need to inform him of symptoms, to ensure that he is aware of their progression and impact.

P1 Female: *But he's got 14 screws in his shoulder, so he's delicate you see as well. So I have to look after him.*

P8 Female: *..and even now I do it. I say, "This is how it is for me" because you've got to understand as much as you can to help me.*

C3 Male: *Yes, I have*.

P8 Female: *I think in the early days it was a difficult thing for him to comprehend. I think sometimes you may have thought that I wasn't telling the truth*

In the latter instance, while the husband still displays some vigilant protector characteristics (see C3 quotes above), this offers an interesting contrast to the carer vigilance discussed above, hinting at a gender as well as a carer dynamic contributing to the vigilant protector role.

6.7.3 *The care process*

The transcripts provided a great deal of data relating to the care process. For patients this area was dominated by the desire to remain independent and the recognition alluded to above, that informal carers could play an important role in helping to maintain independence.

This contributes to a dilemma, because while initially being independent may not require help, a point will come when the provision of assistance helps to facilitate further independence. The important question then appears to be how and when to ask for help.

Patient focus group

Mr C: *It's taken me a while to get to the stage of saying to me wife or daughter "Will you cut that up for me?" And so, so they're really… I just don't really like doing it.*

P8 Female: *And now I have to ask other people to do things and that's just the way it is. I'm not happy about it but I've adapted to it.*

This appears to be a complex question for the individual, and several contributing factors are identified in the transcripts. Firstly that the manner in which care is offered and the insight which the carer has in to the patients' experience appears important

> **P8 Female**: *I think it was probably harder for you (carer) because you've got to deal with me having it and I've got to try and explain to you how it is for me and that's quite difficult to tell somebody what your daily life is like.*

Where good lines of communication existed between patient and care it appeared that provision and acceptance of care could be a natural extension of the pre-existing relationship:

> **Discussing care:**
>
> **P1 Female**: *No, it's – he used to make very good meals. You see I always got back late at night from libraries, which you do. And he used to go early in the morning so I did breakfast and finished that part off and he did the evening part. But we've always found it easy to do that.*
>
> **Interviewer**: *So you're used to doing things for each other working as a team in that sense?*
>
> **C1 Male**: *Yeah.*

Conversely, an example from the early disease group illustrates how lack of communication if, for example, the patient has failed to acknowledge the disease (see above) can inhibit care:

> **Carer focus group**
>
> **Mrs J**: *Oh no, we're having to look ahead, yeah. We don't quite know what at the moment cos he still, you know, he won't, he's not come to terms with it, sort of thing, that's, that's the blooming awkward thing.*

Interestingly, from a patient perspective there also appeared to be a moral component in the decision to ask for help. One patient expressing that they would remain totally independent, without help from anyone, for as long as possible because it was the "right

thing to do", while another identifies that she finds the idea of care easier to accept if she is also able to provide some form of help to others

Discussing the transition from helping others to receiving help

P1 Female: *Yes, I think I sort of feel as if I still ought to be doing something, I don't know what.*

From the perspective of carers, the dilemma appears to be striking the correct balance between taking over a particular activity, at one extreme and a desire to push the patient towards maintaining independence at the other.

Carer focus group

Mrs B: *I really push him, because I think to keep active is really important…*

Mrs J: *And I was taking him shopping, I say "Do you want so and so?" and he'll stand there (laughs) just stand there…..he can't, he can't work out whether he wants it or not. No, I don't take him shopping any more.*

Finally there was evidence that specific symptoms could have a profound effect on the care process, for example the emergence of hallucinations, which is in keeping with previous research showing the contribution of neuro-psychiatric symptoms to carer burden.

6.7.4 *Analysis*

The theme care and caring represents an important part of the findings in this qualitative study. The concept of carer as a "vigilant protector" is drawn from both patient and carer experience and appears to illustrate an important, though perhaps unspoken aspect of the patient / carer relationship. It appears to develop early in the natural history of the disease and one of the important findings is that it is not a passive phenomenon, but rather that bearing witness to the physical and cognitive declines associated with PD carried with it a responsibility to act as guardian. The guardianship of carers extended beyond the immediate relationship with the patient, to infer a protective role of others, particularly family, as well as the responsibility to consider the future as a couple and

individually. This often led to a quest for information about the disease, in order to anticipate decline and assist in difficult decision making, such as initiating changes to the home, or environment to protect the existing care arrangements. In some instances this gives the impression of being a significant burden and cause for anxiety.

It is possible that some of the findings in this area are not solely accounted for by becoming a carer, but also depend to a degree on gender. This is particularly highlighted by the case of two female patients who exhibit some of the vigilant protector traits in reference to their carer.

The other important part of this theme related to the process by which informal care is conducted. From the perspective of patients this appeared to depend on the balance between remaining independent and *asking for help*, particularly the recognition that help offered and received in the right way could facilitate further independence. For carers a similar dynamic was described in the way in which care was offered, needing to strike a balance between the opposite approaches of *taking over* and *pushing*.

Having described the factors which may affect the decision to ask for help, it is then possible to identify within the transcripts positive and negative examples of this care dynamic. This was evident in earlier quotes, where there was an example of a patient with advanced disease who felt unable to call for help after falling out of bed, because of the perception of blame, exemplifying how a lack of mutual understanding inhibits "asking for help" and contributes to a dysfunctional care dynamic. The impression from the whole transcript is that this represents the impact of severe care strain, creating a vicious cycle where the manner in which care if offered leads to reduced ability to accept care and in turn to greater care strain.

Alternatively there are examples, particularly in transcript 4 (P1, C1) and transcript 3 (P8 C3) of how promoting good communication and active understanding of care needs makes asking for and receiving care more acceptable. This links with the findings of the systematic review (chapter 4 section 4.4.2) and implies that it may be possible to support the aspects of a positive care dynamic mentioned above, in order to improve the experience of patient and carer. It is also interesting to trace the patterns which emerge from the data described thus far, which appear to link particular early experiences (i.e.

failure to acknowledge impact of PD) to reduced communication regarding patient experiences and care needs, and subsequent impact on the care dynamic and the manner in which care is offered and received.

6.8 Findings *Theme 4: Health beliefs*

6.8.1 *Health beliefs: disease*

This section of results predominantly relates to participant perceptions and on occasion misperceptions regarding the cause and prognosis of PD. There are a number of relatively rare, single gene disorders which can cause PD. Outside of this group, there are several proposed mechanisms for the aetiology of the disease, ranging from infection, to chemical exposure, to a prion-like disorder. The lack of certainty in this area is reflected in the health beliefs of participants, with a number of people seeking to attribute their disease to previous experience, such as high sports participation or previous head injury.

There was also a range of perceptions regarding prognosis. A number of people expressed the belief that PD does not shorten life, which as was demonstrated earlier may stem from the information offered at diagnosis and represent attempts to "soften the blow" (see section 6.5.2). The relative lack of clarity on this topic was illustrated in transcript 3, where the patient had become fixated with the idea that she had 20 years of good quality life with PD before death or infirmity, while her carer feared that having been diagnosed his wife would be dead in a number of months.

> **P8 Female**: *I've got this idea that after 20 years I'm going to be completely disabled. I don't know why 20 years – I must have read it in the early days and thought 'right, from diagnosis to 20 years down the line, that's it, over'*

6.8.2 *Health beliefs: Medication*

The transcripts revealed the presence of contrasting attitudes towards starting medication, with many participants expressing a wish to delay treatment for as long as possible, but some challenging their clinician in order to start treatment as soon as possible:

Patient focus group

Mr C: *..the doctor says "Well there's this available but it's ……. only good for five years so why not wait to take it until you're perhaps in your eighties". And I said "No, please can I have it now? You don't know, I mean might be run over by a bus in five years' time". (laughter) As I say, which it was I don't know*.

Amongst the people who delayed medication a range of reasons were cited, which can be broadly grouped together. The first group relates to the patients' perception of being medication free, which was equated with being well, for example one participant described how delaying medication allowed her to "pretend" she didn't have the disease:

Mrs S: *I went a few years without any medication, as long as I could, and I think in, until, till about a year ago I just pretend I haven't got it*

Interviewer: *What do you think the reason was for wanting to delay medication for as long as possible?*

P8 Female: *Because I didn't want to be thought of as an ill person, I suppose. I didn't want to have this condition. I suppose I was a bit burying my head in the sand. I thought the longer I can live without medication – I don't need this medication, I'm a well person*

The second reason for delaying medication was the avoidance of side effects, with participants particularly wary of the cognitive side effects of drugs and, as one participant expressed it, not wanting to:

P9 Female: *…pump myself full of something that does nasty things to me..*

Interestingly once medication was initiated, the understandable link between medication and a perception of self as unwell was once again seen as a prime reason to avoid dose escalation or addition of further medications.

Patient focus group

Mr D: *Trying to resist medication for as long as I can and then I'll move to the next stage...its almost like a progression after 2,3 or 5 years and then you move on to the next one, and I'm going as slow as I can*

In the example above there is a sense not only that medication escalation represents worsening disease, but that resisting medication can resist disease progression itself, perhaps offering a sense of control.

The above examples are predominantly taken from the early disease focus group. While similar attitudes towards starting medication were also expressed in the participants with later stage disease, an interesting contrast is provided by the subsequent reactions in this group, once medication became inevitable, with a suggestion from some participants that having resisted medication escalation for as long as possible, the impact on function was such that they wished they had started earlier.

C2 Female: *We'd heard interesting things about medication, lots of stories and we felt we would put off until it was absolutely necessary*

Interviewer: *And how did you feel then when you did have to start medication?*

P2 Male:*Well, I was glad really. I was getting a little bit worse. I thought I need help.* So I got some help with the tablets.

And again regarding Apomorphine, which he was currently taking:

P2 Male: *There again that was put off to us for a long time. I understand it was available. So I just didn't know why we haven't been offered it.*

This illustrates the important perspective available to people in the more advanced stages of disease, being able to assess with hindsight the impact of medication. On several occasions this revealed a sense of medications, both their initiation and waning effect, being important milestones within the person's journey with PD.

Discussing impact of apomorphine:

P9 Female: *Disappointed because I thought it was going to be, not exactly a wonder drug, but I thought it was going to be really helpful……I thought it was going to be much, much more helpful. So we're actually experimenting with trying a slightly higher dose now to see if that's the problem….But I'm not sure which way it's going to go at the moment. But I'm not giving up yet. Carry on, see what happens*

Discussing the waning effect of L-dopa

P8 Female*: I now think this is serious. I'm not going to get better…..Now I'm starting to think if Sinemet's worn off so quickly – only two or three years – then how much more – am I going to have to keep upping the medication fairly regularly? When will it stop working in the future? I have started to think 'what age am I going to be when I'm totally dependent on (carer) or somebody else?'*

6.8.3 *Analysis*

The results relating to health beliefs suggest that there are some misperceptions regarding PD, particularly the aetiology, prognosis and associated mortality. In some instances these reflect the lack of scientific knowledge, particularly the debate around causality which remains an open topic of research(118). In other instances it may also reflect the information offered by health care professionals, particularly regarding prognosis and disease progression. It seems likely that the variable disease course which different people with PD experience discourages open discussion of the added mortality associated with the condition. This becomes more understandable given the apparent misinterpretation of prognostic information, from P8, relating to anticipated decline at 20 years. It is possible that this was a reference to the prominent Sydney cohort, which followed patients for 20 years, reporting high levels of mortality and dementia amongst survivors(26). The quote, where the 20 year mark has become a psychological barrier, is reminiscent of the experience in malignant conditions where clinicians are usually careful to avoid giving direct predictions of longevity, because of the way in which they become a part of the patients' consciousness. This is part of a wider discussion regarding

information dissemination, a topic examined more fully in the later section; viewing the future (section 6.9).

The cancer analogy is apt, as it was common for participants to use cancer as a reference point elsewhere in the study. Several people highlighted a relative lack of public awareness around PD when compared with malignant conditions and others used the care provision for cancer as an example of the care they would like to receive, particularly relating to community services.

The important aspect of this theme related to the significance of medication, both physical and psychological. The conflicting attitude towards starting medication is perhaps not surprising when considered alongside the traditional clinical concerns regarding the development of motor complication with L-dopa and the development of dyskinesia in a relatively predictable fashion(119). This has led to clinical strategies aimed at delaying L-dopa therapy and, as such, many patients appear to be understandably reluctant to start medication.

However as evidence changes this may become a hindrance to good care. It is suggested that, although lower L-dopa doses are associated with fewer motor complications they are also associated with worse quality of life(120). As such, clinicians increasingly advocate earlier initiation of medication(121) and a recent study has suggested that outcomes may be at least equivalent in patients starting L-dopa as first line treatment and may even be improved(122). With this in mind, even in the absence of neuroprotective treatments, an ingrained fear of medications leading to delays in treatment may be detrimental to the long term care of patients.

For patients, medication initiation and escalation appears to strongly represent disease progression and, in some instances, controlling the escalation of medication may be seen as a means of trying to delay disease progression. It is also striking that the fairly predictable progression through medications, which most patients will experience, may represent a series of milestones within the life-course of the condition. As such starting a new medication has a wider, existential quality about it. This may represent time when wider questions are being asked about the implications of disease progression and

prognosis, and could be considered as a time for exploring palliative and supportive care needs (see chapter 13 section 13.5.2.2 on proposed use of disease milestones).

6.9 Findings Theme 5: Viewing the future

6.9.1 Disease milestones

One of the benefits of interviewing people, particularly those with advanced disease, was the ability to form a sense of the overall trajectory of their experience of PD, from the time of diagnosis, through early disease, medication initiation and the subsequent development of more complex symptoms. This produced the impression, even during the data acquisition phase, that the gradual decline of PD may be punctuated by particular events or experiences, which hold great significance and that as patients' reflected back on their journey, these events became milestones by which they measured disease progression. The concept of disease milestones and watershed moments, which can also be found in some of the literature reviewed in chapter 4, was thus incorporated in to the analytical framework, with the results described below.

In terms of disease progression, several participants commented on the gradual decline associated with PD, which was seen as advantageous, allowing them to adapt to their changing physical abilities.

> **P9 Female:** *But I don't go for walks as I used to. But there are other things to do. You adapt to what your abilities are*

> **P8 Female**: *But so far, because it's a slow moving illness, you tend to deal with it as you go along. It's not too bad really*.

However, within this process of adaptation there were particular moments of great significance, which could be identified throughout the transcripts. Examples include:

i)The noticeable progression of physical symptoms, particularly development of bilateral symptoms or axial symptoms:

> **P8 Female**: *With me it's not being able to use my hands very well and balance issues and so on.*

ii) Often characterised by the first emergence of falls:

>**C3 Male:** *I'd say about four years after you had it I can distinctly remember we went shopping in York – it might have been this time of year – and you stumbled off the kerb. That's the first real time I could tell that she had Parkinson's.*

iii) As disease progressed the emergence of motor fluctuations was felt to be a highly significant moment, given the impact on function and the increased importance of medication timing:

>**C2 Male**: *He leads quite an active social life, but when he was experiencing these on off periods he didn't cope very well with that at all.*

iv) The need to move house in order to accommodate current or anticipated physical decline also carried great significance as a marker of disease progression and a time when physical decline had to be acknowledged.

>**P9 Female:** *You see an awful lot of people who are still living in places that are really not very suitable for them with Parkinson's. Having to cope with stairs and things that...so that (moving house to a bungalow) was certainly something that I consciously did.*

See also example of P1C1 given above.

v) However, milestones were not all directly related to changing physical condition, but could also be characterised by functional or social loss, for example having to relinquish a driving license or giving up paid employment:

>**P9 Female**: *So I was medically discharged I suppose, early retirement. That didn't go down well with me at all because I actually loved my job……It wasn't until quite a bit later that I started to think along the lines of I feel a bit cheated because I wasn't that old.*

vi) The other very important milestone, alluded to above, involved medication. This included initiation of medication for the first time, but also the need for multi-drug strategies and, especially for people with later stage disease, medication failure, either

through waning response to L-dopa or lack of effect from advanced therapies such as Apomorphine. See quotes in section 6.8.2 above, relating to health beliefs: medication.

Although the concept of disease milestones really developed from the interviews in later stage disease, when applied to the early disease focus groups it became clear that a similar experience was present, both in terms of milestones already experienced, such as starting medication, but also in anticipation of future events which would constitute a watershed moment for the individual. One carer comments that her partner had been managing well with PD for years, until sudden functional loss:

> **Carer focus group**
>
> **Mrs C**: *I can honestly say, up to this last year, he hasn't much of a tremor at all, yes, I think he's slowed down,…and then it's just hit us like a bolt out the blue.*

While a section of discussion really encapsulated the idea of milestones, participants remarking that on a background of gradual decline, it was the sudden realisation that you are not as good as you used to be, which caused pause for thought.

> **Mr To**: *The thing about Parkinson's is that it's a sneaky little disease, it creeps up on you without you…*
>
> **Mr D**: *Mm.*
>
> **Mr To**: *…really noticing it.*
>
> **Mrs S**: Mm.
>
> **Mr D**: *Yes.*
>
> **Mrs S**: *Mm.*
>
> **Mr To**: *A little……stages there's a little movement and then you realise that you're not as good as you were.*

Patients with early stage disease were also able to envisage future events as highly significant, such as the loss of a carer, or reduced mobility.

Patient focus group Re: a time when discussing the future might be appropriate

Mrs S: *Well for me it'd be when I can't walk any more.*

6.9.2 *Analysis*

The concept of disease milestones was very powerful. It was a means by which participants, particularly those with advanced disease could reflect on and structure their experience of PD, but also emerged as a method for anticipating future decline or losses which may be associated with disease progression. Whilst some milestones appear to be fairly predictable and perhaps part of a common experience, for example those relating to initiating medication or falling, others may be more specific to the individual, reflecting a personal significance associated with a particular event, for example the loss of a social activity or development of a feared complication such as hallucinations.

Recognition of disease milestones may be useful in clinical practice, for example as an indicator of times of greater supportive and palliative care need, where screening may be most beneficial. It is then important to appreciate that while some milestones may be more "generic", meaning that the clinician can have a prior awareness and be vigilant for them, others will be more "person specific" requiring a person centered consultation and good communication skills for identification. This potential application is discussed further in chapter 13.

6.9.3 *Information*

The topic of information has already been touched upon in the results relating to diagnosis, particularly the lack of information provided at this time. It was also a key component of the systematic review (chapter 4 section 4.4.6). The results displayed below are complimentary to this prior work and focus on:

i)   Conflicting attitudes to information,
ii)   Information discord,
iii)   Information sources,
iv)   Information timing,
v)   Barriers to information

6.9.3.1 *Conflicting attitudes to information*

Analysis of the transcripts very clearly displays the differing approaches which participants had to information about the disease. The divide here was between those who desired more information in the early stages of disease, even at diagnosis and those who did not, and this did not appear to depend on disease stage, or whether the participant was a carer or patient.

At one extreme we see a great desire for information regarding PD and its prognosis, even from diagnosis

> **Patient focus group – discussing disease progression**
>
> **Mr To**: *So it is, it's not a, you know, we need to know, I think we need to know more about the disease. We should have been told more.*
>
> **Facilitator**: *Yeah. When do you think you should have had that… information?*
>
> **Mr To**: (Sighs) Yeah, I mean from the, from the very beginning because I think you should know what you're looking for.

While one participant equated the lack of information provision with feeling "fobbed off". This reflects findings in a previous study where a lack of information was equated with a sense of neglect(96)

> **P8 Female:** *Just general lack of information overall I think, in the early days. Just being fobbed off a bit I think.*

At the other extreme was the feeling that more information regarding the natural history of the disease would not be a useful thing, particularly as the disease course is variable and so the things discussed may never occur.

> **Carer focus group**
>
> **Mrs C**: *But if it isn't happening now then it may never happen so why get something in your mind that might happen?*

This contrasted with the opinion of another participant, in the same discussion, who needed more information in order to formulate coping strategies for the future:

> **Mrs H**: *I actually went online when he was diagnosed…*

> **Mrs B**: *Mm, I did, mm.*

> **Mrs H:** *…and tried, as I do with anything, I just, because if I don't understand the illness, I can't deal with it. I have to know ex, all, everything about it…*

> **Mrs B**: Yeah, I'm with you.

6.9.3.2 *Information discord*

From the examples above it is clear that very different approaches to, and requirements for, information exist. This may present a particular difficulty when there are discordant approaches within the patient / carer relationship. There were two examples of this within the study group, the first from the early disease focus groups and the second from the late disease interviews.

> **Mrs H:** *…(patient) had no interest whatsoever in what it was or, you know, it, somebody told him he's got this and "Well they could be wrong" you know, you know (…).*

> **Facilitator:** *So do you think you had, in a sense, a similar experience then where you were wanting more information… and, and actually (patient), in a sense, didn't need to hear, didn't want to hear that?*

> **Mrs H:** *Oh definitely. Keep it away from me, yeah, he didn't want to know because he may not have it.*

> **Discussing information at diagnosis:**

> **P2 Male:** *…All I did was come out of there with no explanation of anything at all. It did seem to me that it was a lapse in information….*

> **P2 Male**: *I think it would've been the thing to do (provide prognostic information), whole history of it.*

> **C2 Female**: *...I also – that's just a personal thing, would rather not know, for me personally, what is in the future. And that's just my personal opinion.*

In these scenarios to meet the information needs of one person would be to clearly contravene the desire of the other.

Finally the transcripts build on a point raised by the systematic review, that not all information is equal and that any single person may have an internal tension regarding the desire for information. One person describing this as the balance between being informed and being hopeful:

> **P8 Female:** *Because everyone's experience is different. It can be difficult to read about things. It's a good thing that you can read about other people's experiences but because everybody's different it also makes it more worrying in a way because you think 'am I going to get all these symptoms in the future?'*

> **P9 Female:** *I think if you look at it in the right way, looking at research and stuff like that it gives you hope that something's going to turn the corner, something's going to happen*

6.9.3.3 *Information sources*

Participants described accessing information from a wide variety of sources, including the internet, magazines, patient literature and peer support groups. There was particular praise for the literature provided by organisations such as Parkinson's UK and for the Parkinson's nurse specialist (PDNS) as a valuable resource. Interestingly, while the quote above describes the lack of information at diagnosis as a "lapse", it was not always expected that doctors should be the source of information, indeed one participant describes why they felt the PDNS was a more appropriate choice for such discussions:

> **C3 Male:** *She's the more human face, I suppose. She seems to have more empathy with you, doesn't she?*

6.9.3.4 *Information timing*

While several examples above express an individual's desire for greater access to prognostic information right at the time of diagnosis, others who wanted information felt that it would be more appropriate as the disease progressed and that they would not have been ready earlier. There was agreement that timing of information would be a very personal issue, possibly dependent on some of the issues such as acceptance of diagnosis, discussed earlier.

> **Carer focus group – discussing whether prognostic information should be discussed at diagnosis.**
>
> **Mrs J**: *Probably a little bit too soon, maybe within the year, sort of thing, so you knew what to expect as things got, you know…*
>
> **Mrs B:** *I think again, as well from diagnosis, sorry…*
>
> **Mrs H:** *No, it's fine.*
>
> **Mrs B**: *…I, I think it's how a patient accepts the fact that they're being told they've got Parkinson's. Now in, in my case, with my husband, he accepted it very well, in your case your husband still hasn't accepted it.*
>
> **Mrs H**: *Still hasn't fully, no.*
>
> **Mrs B**: *You know, so gauging the right time would be very important wouldn't it?*

These results can be viewed alongside the earlier examples of patient suggesting milestones, such as loss of mobility, as appropriate times for prognostic discussion (see section 6.9.1).

6.9.3.5 *Information barriers*

A number of potential barriers to prognostic discussion became apparent during the discussions. One patient with advanced disease, who had expressed a wish for information both at diagnosis and at the time of starting Apomorphine, wondered whose responsibility it was to initiate these discussions:

Patient / Carer interview (P2 C2)

P2 Male: *The longer it goes the worse it gets. [unintelligible 0:14:21 I wonder] if it's going to get any worse than it is now.*

Interviewer: *And is that the sort of thing you've had the opportunity to discuss with anybody?*

P2 Male: *No. I suppose I haven't asked for it have I?*

C2 Female: *No.*

P2 Male: *I'm just wondering if I had to ask them myself.*

In other instances, even initiating a discussion regarding prognosis with a clinician did not give satisfactory results.

Carer focus group

Mrs H: *..whenever he goes to (consultant), he always says "But how am I going to end up?" That is, that is the only thing that's really on his mind "But what is going to happen to me?"*

Facilitator: *…do you think he gets a satisfactory answer to that question?*

Mrs H: *No, no, because he, because he, he, he, all he gets to know is that it affects everybody so differently, and the only thing he's been told by (consultant) is "It won't kill you"….But that's not enough, that's not enough for a man to know. He wants to know…..am I going to be in a wheelchair? Am I going to need somebody to take me the toilet? Am I going to, you know, because he couldn't bear that, and, and he can't seem to get that in his mind because he can't get any answers, and that's the worst thing for him.*

6.9.4 *Analysis*

This is a powerful section of data which supports and builds upon the findings of the systematic review, particularly the theme of information tension. It is evident that people

living with PD have very different, personal and sometimes conflicting requirements for prognostic information. It may be particularly difficult to meet peoples' needs where they are discordant within the patient /carer relationship and these discussions are likely to require advanced communication skills and possibly SPC involvement.   It is also important to highlight that conflicting needs for information may actually exist within the same individual, a finding which corresponds to the previous work of Giles et al. a phenomenon they describe "wanting but not wanting"(12).

It is interesting that many participants describe accessing information from non-medical sources and, where health care professionals are involved, may find the PDNS a more appropriate guide than the consultant. Whether this is cause or effect is difficult to know, but certainly the study highlights instances where medical professional may become a barrier to, rather than a medium for effective discussion of prognosis. It is also interesting to reflect that while all patients will have contact with health care professionals, not all will have access to, or ability to utilise other non-clinical sources of information such as the internet. Thus neglect of this issue by clinicians may put some patients at risk of being disenfranchised.

These results suggest a complex area of unmet palliative and supportive care need, which may be difficult to address. However, failure to respond appropriately to information needs appears to cause frustration and on occasion disaffection with the medical profession. It is also, when considering the natural history of the disease and the prevalence of dementia, a potentially lost opportunity for people to participate in and influence decisions regarding the later part of their illness.

6.9.5 *Planning*

Having described the results relating to prognostic information the subsequent, related, section of "Addressing the future" looks at the results for planning the future. As might be anticipated, participants again described a range of approaches to planning. Some had an approach of "living for today", akin to the concept of temporality (see chapter 4 section 4.4.6.3) where avoiding planning seems to allow the existence of multiple possible futures in relation to progression of disease.

Patient focus group

Mr To: *But it's not necessarily the same for everybody, so. This is one of the reasons why you don't look too far ahead because everybody's different. Everybody's suffering with slightly different things, you know.*

Mr T: *So I tend not to think of… what will be happening in ten or fifteen years' time…. and like I, I might get run over by a bus anyway.*

Other reasons for delaying planning for the future included a fatalistic approach, just letting things to take their natural course

Mr C: *..it wouldn't occur to me to look on the internet for anything. Just let, I suppose I let things just take their course.*

or, despite having fairly advanced disease, feeling too young to be trying to plan at this stage.

P8 Female*: I suppose there would come a time eventually but it's not something – as I said before, because I feel young you do tend to put that off I think.*

The last quote is significant because it was followed by a patient led discussion, where she subsequently acknowledged that she had fears regarding the future, especially being alone, and in her "serious moments" felt that herself and her husband should have plans in place:

P8 Female: *I think, in my serious moments, we should make some sort of arrangement for that because my worst nightmare is to be in a home on my own without you being there. When I'm feeling that way inclined I do think about that.*

Her husband then captured the dynamic between anxiety for the future and anxiety associated with planning, describing the inaction as a trick you are able to play on yourself

C3 Male: *…it's a sort of trick for yourself, to stay optimistic. I suppose I bury my head in the sand and try not to think about all that, not too deeply anyway, at the moment. Having some kind of plan would probably be a good idea but it just seems too….(sentence unfinished by participant).*

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As the couple discussed this topic on their own, they came to the conclusion that some planning, with support from the clinical team may be appropriate:

> **C3 Male**: *It's possible so I suppose really we ought to have …*

> **P8 Female**: *It is something it would be handy to discuss with people because when you're on your own you think what do we do, where do we go?*

This episode really demonstrated the dynamic nature of these decisions and it was insightful to watch this couple work through their shared concerns regarding future care planning, effectively in real time. The other fascinating issue raised regarded the responsibilities of health care professionals. Not only was it felt that clinicians could play a supportive role, but also that they had a responsibility, as experts who could chart the disease progression from a neutral perspective, to raise the topic of care planning at a time which they deemed appropriate. The idea that clinicians should raise the issue of care planning was echoed elsewhere, with one lady who did not think she would personally want to plan until the later stages of disease "leave it to the last minute I think" still open to the idea of it being raised:

> **Interviewer**: *How do you think you would have felt about someone, you know, raising those issues with you?*

> **P1 Female**: *I wouldn't have minded at all, no.*

The carer data revealed similar trends regarding a mixture of those wanting and needing to plan for the future and others very much resisting the idea, suggesting that planning would be an admission of mortality and "scary". However they also reflected carer traits from earlier themes, such as vigilant protector, where carers felt it necessary to take the lead with planning, while discordant approaches to planning could cause tension, similar to the description regarding information needs. One participant described her relief, having heard the stories of others, that she and her husband had both agreed to move house in preparation for worsening mobility:

Mrs B: *I'm, quite honestly, thinking myself very fortunate, listening to you, and, and to you as well......And when I said to (husband) "What do you think about this?" He said "Well we don't know what's going to happen in the future, it's just as well to do it now whilst I'm still capable of doing it" you know. So we did, we, we, you know, bought the flat.*

6.9.5.1 *Analysis*

The concepts of desire for information and approach to planning are clearly closely linked. The tensions which were present in the former are also evident in the latter, with some participants appearing conflicted regarding a desire to plan in order to address anxieties about disease progression, without wanting to admit defeat or give up hope for the future. It again seemed easier for these tensions to be resolved where the carer and patient had similar philosophical approaches to the future, but potentially problematic if their needs were discordant. There was also an impression that this was an active, ongoing process, requiring support and that at least some of those involved would welcome the appropriate intervention of clinical teams. Indeed, more than this, there was a sense that as expert witnesses to the progression of disease they carried a responsibility to be more than passive observers.

6.10 Theme 6: Being supported

6.10.1 *Experience of PD services*

Perhaps the largest element of this theme relates to the experience of being diagnosed and has been covered in detail above. In addition to this participants described their experience of services, highlighting the features of a positive service which included being able to place trust in your clinician, building a rapport through recurrent appointment with the same person and a clinician taking a person centered approach to care.

Discussing trust

Mr C: *I'm very trusting I think. (laughs) Yes, trust, trust my doctor and he advised me.*

Discussing continuity of care

P9 Female: *..But he made me feel as though I was going to be looked after. It was good because really I haven't had anything in the past that's taken me in to see a consultant on a regular basis. I hadn't had that experience.*

The converse of this was the severe distress felt when services broke down, as one carer described her inability to access help as her partner's symptoms worsened, or the difficulties experiences when seeing different doctors at each visit, breaking the bonds of continuity and trust.

Dicsussing breakdown of service provision

Mrs C: *And this last week has just been horrendous. And of course Dr (consultant) is on holiday, the Parkinson nurse, is, she's ill…*

Mrs H: *Oh dear.*

Mrs B: *Oh dear.*

Mrs C: *…this week, rang my own doctor up, who is just wonderful doctor, he's on holiday. So, you know…*

Discussing lack of continuity

C2 Female: *The consultants kept changing at that period. There was nobody based in (*name of hospital*) …. So when we went for a consultation every six months it was a different person and they didn't do or say very much.*

There was very much a sense that participants relied on secondary care services for support and several participants described clinic as a place to deal with physical symptoms, rather than emotional or supportive needs. While some rationalised this as

appropriate given the time and resource available, others were less satisfied with this approach.

Discussing perceived roles of clinical team

C3 Male: *You're a realist as well. You tell me you kind of know that the consultant has only got a certain amount of time.*

P8 Female:*Regardless of the amount of time the consultant has I would always see them as they dispense the medication and the knowledge.*

C3 Male: *Whereas the emotional side of it …*

P8 Female: *And the practical side.*

C3 Male: *You would deal with her [nurse specialist] wouldn't you? You always have done anyway.*

Discussing focus on physical symptoms

Mrs H: *But I sometimes feel when you come to the clinic, it's, you know "How are you? You know, have you, have you noticed any difference?" Not to me, because I just sit as a, you know, and then I "No, absolutely fine".*

6.10.2 *Support*

As mentioned previously, many of the supportive aspects of care appeared to be filled either by the PDNS or by the charitable sector. Several participants mentioned frustration at the gaps between clinic visits and the need for more responsive services.

> **P9**: *We probably wait about eight months between appointments to see the consultant at the moment. It would be really nice that when something happens that you had access in some way. I don't mean going there every other week or something silly. But rather than having to wait until the next appointment to go and discuss something, it'd be nice to have that ability to go and discuss things at the time.*

This also accounted for some of the many positive comments about PDNS, who are often able to offer a more responsive service. As demonstrated by earlier quotes they were sometimes also perceived to be more approachable and to have a legitimate role in supportive care, rather than the focus on physical symptoms characterising medical practice.

One of the points on which participants differed was attitude to support groups. The concept of downward comparison, mentioned in the systematic review, was again prominent and cited as a reason for avoiding groups run for peer support, such as local Parkinson's UK meetings.

> **Mr Tr**: *And anybody in here to, with the local Parkinson's Society meetings?*
>
> **Mrs S**: *No, I haven't.*
>
> **Mr C**: *Yes, I go up there.*
>
> **Mr To**: *No.*
>
> **Mr Tr**: *You'll know then…*
>
> **Mr C***: I've been to all of them.*
>
> **Mr Tr**: *(…). Yes, I went to one of them, I was woefully disappointed…. I found it depressing.*

Downward comparison could also lead participants to avoid social functions where they knew other people with PD would be present:

> **Mr C**: *You were, you mentioned how does seeing other people affect you and I must admit at some of the functions we go to I'll say "Don't let me get like that, we'll stay at home rather than go into company."*

Another interesting aspect to this phenomenon included the presence of "upward comparison" whereby the same physical occurrence; an encounter with somebody who has more advanced PD, could elicit the opposite response, prompting a positive feeling regarding your own position.

Mr Tr: I have a neighbour who's in denial about Parkinson's and he's on, he doesn't have any medication at all and he's got a, a terrible left hand tremor, worse than mine.

Mr D: Yeah. I can put up with this but I'd hate it if I started having like I've seen, I see with (relative).

Upward comparison was also seen in the carer focus group:

Mrs B: I'm, quite honestly, thinking myself very fortunate, listening to you, and, and to you as well…..although I do a lot more now than I did previously, he's doing really, you know.

In keeping with this theme, participants with more advanced disease tended to express more positive attitudes towards support groups and as mentioned previously these organisations often had an integral role to play in dissemination of information and accessing support.

P9 Female: Then a little later on I joined the local Parkinson's group...got a lot of help and support from them.

C2 Female: We joined the local Parkinson group and went to monthly meetings, so mixing with other patients if you like. We received information that way as well. Not always beneficial, is it, but it's a natural thing to compare and exchange experiences, but sometimes it is useful.

6.10.2.1 Analysis

The results contain a number of important points regarding the way in which participants viewed and accessed support services. There was a tendency for allied health professionals, particularly the PDNS and the charitable sector as the main provider of supportive services, rather than the consultant clinic. This is particularly relevant given downward comparison, an apparently strong barrier for some when accessing support groups.

6.11 Discussion

The findings above clearly demonstrate the presence of palliative and supportive care need, from the very first consultation and affecting different aspects of the individuals' experience. Some issues appear to affect both patient and carer, for example a tension regarding the desire for and approach to prognostic information, while others may be more specific to one group.

The striking themes from the analysis include; the manner in which the diagnosis is reached and conveyed, the establishment and maintenance of informal care relationships, different approaches to information seeking and care planning and the way in which support, from health and charitable sectors, is accessed. Although there is no right or wrong way to approach these issues, some approaches do seem to yield greater satisfaction for those involved. It is also possible to link results across categories, trace patterns of need and speculate regarding cause and effect. For example patients who fail to acknowledge the diagnosis during the early stages, have very different approaches to information seeking and care planning, compared with those who acknowledge the disease and appear to use information gathering to assist in coping. This in turn has implications for future care planning, for carers and the degree to which they must become vigilant protectors. Discriminating these groups may be an important aspect of practicing in a patient centred manner, so that supportive care strategies can be tailored for the individual. This will be particularly important and challenging where patient and carer have discordant approaches to key issues, such as information seeking.

6.12 Conclusion

The qualitative analysis broadly echoes the results of the qualitative synthesis, supporting the key concepts of information tension and care tension. They also add depth to the analysis, with a sense of the way in which early disease experience may impact on later supportive and palliative care need, suggesting a need for patient centered care from the outset, delivered by clinicians with excellent communication skills. In so doing they challenge existing practice and encourage the development of reactive, patient centered approaches to care, which may only be possible with a good working understanding of the likely needs faced by different patients.

The knowledge gained through this work and the systematic review which preceded it formed the basis for the adaptation of the NAT:PD-c in Parkinson's disease. This adaptation process is described in the next chapter (chapter 7). The practical application of findings, beyond contributing to the adaptation is discussed in chapter 13.

Chapter 7

Adaptation

Developing the Needs Assessment Tool: Parkinson's disease

(NAT: Parkinson's disease)

7.1 Introduction

The first 6 chapters of this thesis have largely focused on a qualitative exploration of palliative care needs in Parkinson's disease. Whilst this work can be viewed in isolation and has several implications for clinical practice (see chapter 13), the primary objective was to provide the basis for adaptation of the Needs Assessment Tool Progressive Disease-cancer (NAT:PD-c) for use in Parkinson's disease.

This chapter describes the adaptation process, demonstrating how the qualitative work in chapters 2-6 was applied to produce the Needs Assessment Tool: Parkinson's disease (NAT:Parkinson's disease – see appendix 11), a new instrument for the identification and triage of unmet palliative care need in PD. This chapter effectively links the qualitative work described above, to the second half of the thesis, which focuses on the clinimetric testing of the NAT:Parkinson's disease (chapters 8-12).

7.2 Exploring the NAT:PD-c

The Needs Assessment Tool: Progressive Disease-cancer (NAT:PD-c) is a palliative needs assessment, covering both caregiver and patient domains, which was originally developed for use in cancer patients(29, 123). Its primary attractions are that it is user friendly, covering a single side of A4 paper, with an additional page of guidance. The tool is designed to be completed by a clinician after routine consultation meaning that, unlike many clinical assessment tools, it can be incorporated in to routine clinical practice. This statement is supported by the evaluation of the NAT:PD-c, which demonstrated that there was no increase in the time taken to review patients in clinic where the tool was used(31).

Following its initial development for cancer, the NAT format has subsequently undergone successful adaptation for use in heart failure(30), and there is ongoing work on its adaptation for interstitial lung disease and malignancy in primary care. This project focusing on Parkinson's disease, therefore sits within a wider programme to improve the initial assessment and triage of palliative and supportive care needs for people with malignant and, increasingly with non-malignant chronic conditions.

One aim of the adaptation process was to maintain, as far as possible, the original NAT format. The single page assessment tool is divided into the following four sections:

Section 1 – Highlighting those individuals at increased risk of unmet need,

Section 2 – Assessing patient wellbeing,

Section 3 – Assessing the ability of carer and family to care for patient,

Section 4 – Assessing carer and family wellbeing.

Each section has a number of domains, for example – Section 2.1: *unmet patient physical needs*. The assessor is asked to rate their level of concern regarding the presence of unmet needs in each domain as 0-2, where 0 is no need, 1 is some or uncertain needs and 2 is definite unmet need. Where unmet needs are present, assessors are then prompted to triage, according to the ability of local services to address them and to consider referral to specialist services (palliative or other) where this is not possible. As such, once familiar with the tool, a member of the clinical team is able to complete the assessment of each domain within a matter of seconds, providing a rapid bedside assessment of unmet needs accompanied by a documented plan of action.

Where significant levels of concern regarding unmet palliative care needs are identified a further, more detailed palliative assessment will be required in the form of comprehensive specialist palliative review, possibly incorporating quantitative assessment tools. The NAT:PD is not therefore intended to replace available patient-report assessment tools(124-126), but rather, is complementary to them. The implications for this in PD are discussed later (see chapter 13 Section 13.5.4).

In adapting the NAT for Parkinson's disease, it was important to operate, as far as possible, within this tested structure, whilst ensuring that the tool covered all the important aspects of palliative and supportive care need in people with PD.

7.3 Adaptation method

The adaptation was based primarily on the qualitative data provided by the systematic review (chapter 3-4) and primary qualitative study (chapter 5-6). It also drew upon data from cohort studies in PD, to provide a longitudinal view of the disease course.

The first step was to broadly arrange the qualitative data according to the four primary categories of the NAT, for example, data relating to patients' reaction to being diagnosed was placed in **Section 2**: *Unmet patient needs*. Where data was clearly relevant to more than one category this was permitted, for example, data relating to carer burden may be cited under **Section 3**: *Unmet carer needs*, as well as **Section 4**: *ability of carer to care for patient*.

Data which fell outside of the NAT categorisation were grouped separately, under the heading *other*, although this was rarely required given the broad nature of the original categories.

Once the data had been stratified in this way, the domains within each category were adapted individually, along with their supporting information, to ensure that they captured each theme from the qualitative data that had been assigned to them.

This was an iterative process conducted by the primary researcher (ER), with external review by a second researcher (MJ) to ensure agreement regarding comprehensive coverage of the qualitative themes.

7.4 Face and content validation of the adapted tool

Once this process was complete, the face and content validity of the draft instrument were examined, by a range of clinicians involved in the care of PD patients, these included:

 Neurology (2 Consultant clinicians with an interest in movement disorders),

Elderly medicine (1 Consultant and 1 senior specialty trainee with an interest in PD),

Palliative care (2 Consultant clinicians, one with an interest in neurological care),

Specialist nursing (1 PD nurse specialist and 1 palliative neurology nurse specialist).

This process led to a number of further changes, predominantly targeting user friendliness of the tool and comprehensiveness of the user guidance (see chapter 10 clinimetric testing: results).

7.5 Results

Under each of its categories, the NAT format is designed to help clinicians quickly identify the likelihood of unmet palliative care need *(0= none, 1= some / possible, 2 = significant)* and initiate a triage process, so that needs are either dealt with directly by the assessor, by another member of the team, or referred to specialist services. This feature is key to the concept and has been retained in the new tool.

7.5.1 *Section 1: Highlighting individuals at increased risk of unmet need*

The most significant change to previous incarnations of the NAT was the addition of a supplementary "red flags" domain to the first section of the tool, designed to assist in identifying sub-groups of patient who may be at greatest risk of unmet palliative needs.

The decision to use red flags was influenced by the findings of the qualitative studies, for example the concept that early disease experiences may presage subsequent palliative and supportive care need and that "disease milestones", which people with PD used to punctuate their experience of PD, may represent periods of heightened care need. In addition the trajectory of PD, with high levels of cognitive impairment, meant that red

flags may be useful in identifying future need at a time when patients retain decision making capacity, thus maximising autonomy.

 Deliberately placed at the beginning of the document, the red flags are intended to heighten clinician vigilance, rather than act as a direct trigger for specialist referral. For example, although the presence of axial involvement (Hoehn and Yahr stage 3)(112) is considered to place people at greater risk of unmet need, it in no way suggests that these individuals will all require specialist palliative care referral, rather that clinicians may need to have a greater suspicion of unmet need, in all domains, where axial motor symptoms are present.

The specific red flags used within the NAT:Parkinson's are based on studies relating to prognostic markers in PD, longitudinal studies of quality of life and expert consensus. In particular they were influenced by a clinico-pathological study, conducted by Kempster et al, which identifies four key features that appear to herald the last years of life *(Formed visual hallucination, cognitive impairment, recurrent falls and admission to 24 hour care)(127)*. This study is particularly interesting, as these sentinel features of disease progression appear to develop in much the same order, and in the same relation to onset of terminal stages, regardless of the age of disease onset – see figure 10. Although not necessarily advocating a prognostic approach to palliative care evaluation (see chapter 1), it was felt that the pattern of disease evolution described by Kempster, particularly the development of formed visual hallucinations prior to cognitive decline, in all age groups, provided a strong indication that their emergence should trigger a heightened awareness of palliative need. This is particularly relevant given prevalence of dementia in PD and the implications of cognitive decline for patient decision making autonomy. As such formed visual hallucination, recurrent falls and admission to 24 hour care were all included in the red flag section (see box 1). The presence of dementia was not included in this box, primarily because it was felt that greater emphasis should be placed on visual hallucinations, as a herald to impending cognitive decline, which provides the opportunity to intervene whilst the patient is most likely to retain decision-making capacity.

Evidence from a longitudinal quality of life study suggests that the development of axial symptoms, which define Hoehn and Yahr stage 3 disease, was associated with a step

decline in quality of life(128) and this is also the time at which carer strain shows evidence of increasing(129, 130). These findings are supported by the qualitative work presented here, with patients and carers often describing falls and reduced mobility as a cause of concern and increased carer burden. Whilst recognising that many people with H + Y stage 3 disease will not have supportive and palliative care needs which require SPC input, this easily identifiable marker of disease progression does provide a good opportunity to search for and address unmet need where it exists. In particular it may be that this represents an opportunity to identify unmet needs early, when they can be dealt with at a local level, avoiding escalation with the aim of reducing both patient distress and the requirement for subsequent SPC referral. This is related to the concept of regular disease "milestones" at which a palliative screening tool could be applied is expanded upon in chapter 13 and presents itself as a topic for future research. (see chapter 13 section…. 13.7)

Box 1 – Red Flags

Red Flags for increased risk on palliative care need
Recurrent falls (>2)
Formed visual hallucinations (not related to inter-current illness or medication change)
Admission to 24 hour care
Onset of Hoehn and Yahr stage 3 symptoms (Axial instability and bilateral disease)
Failure to attend scheduled clinic appointment

The final red flag was formed on the basis of consensus opinion of the local PD team and affirmed by the external experts who assessed the NAT: Parkinson's for content validity. Anecdotally people with PD generally attend clinic regularly and are loath to miss appointments. When they do miss clinics recurrently, because of pressure on services, they are often referred back to their GP or lost to follow up. It is the experience of the local PD team that these people have often either been admitted to hospital, to 24 hour care, or have had another change in circumstance that has prevented attendance at

clinic. As such this may represent a vulnerable group, experiencing worsening disease or precarious care arrangements, who would benefit from comprehensive review, but often appear to fall through the gaps in the system. The utility of this as a marker of unmet palliative care need is another potential topic of future research (chapter 13, section 13.7).

Figure 10 – Milestones predicting Mortality (from Kempster et al (127)
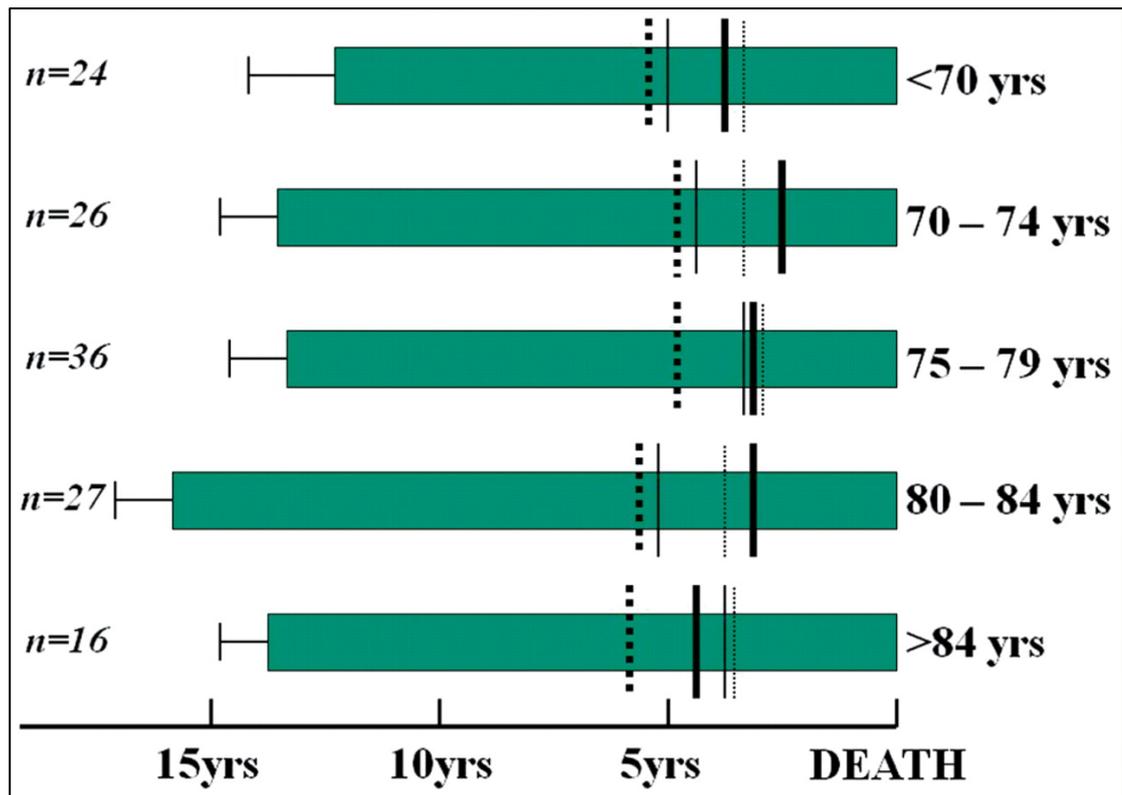
Disease course and disability milestones for the five age-at-death groupings, aligned for time of death. Regular falls = fine lines; residential care = heavy lines; cognitive disability = fine dots; visual hallucinations = heavy dots. Error bars show the standard error of the mean disease duration.

7.5.2 *Section 2: Assessing Patient wellbeing*

Physical symptoms (motor and non-motor) are the defining characteristics of PD and while sharing some features with other palliative conditions (e.g. pain, constipation, fatigue) the supporting documentation now reflects the presence of prominent PD specific symptoms (e.g. freezing, dyskinesia, urinary difficulties, drooling).

The psychological question has been expanded to include neuro-psychiatric issues, the significance of which are increasingly recognised in PD and which contribute significantly to caregiver burden (e.g. hallucination, anxiety, depression and cognitive impairment). The intention was also to reflect the complex emotional and psychological response to a diagnosis of PD, which was strongly represented in the findings from both the systematic review (chapter 4) and qualitative study (chapter 6).

As a direct result of the qualitative work presented previously (chapter 4 and 6) the supporting information relating to the stem: *"health beliefs, social and cultural factors making care more complex"* has been changed, to highlight the difficulty in perception of palliative or hospice care amongst PD patients and carers, as well as the issue of "downward comparison" which may complicate the use of support groups.

Finally the difficulties associated with information management, described in the qualitative work as *"information tension",* have led to the section on information needs being expanded, with specific prompts regarding the prognosis of PD, which was often poorly understood.

7.5.3 *Section 3: Assessing ability to care for patient*

This section now reflects the difficulties posed by neuro-psychiatric symptoms in PD and the contribution of these issues to break down of informal care arrangements, as highlighted by the earlier systematic review and qualitative synthesis.

It has also been expanded to capture the complex dynamic between patient and carer, which defines informal care arrangements and was one of the major themes within the qualitative work, described as *"care tension"*,

The qualitative components of this thesis developed a greater understanding of the importance of information in relation to carers and their ability to care for patients. This was used to adapt the supporting information in this section, particularly around the importance of promoting biomedical understanding, prognosis and service availability.

7.5.4 *Section 4: Assessing carer / family wellbeing*

The spiritual and existential impacts of caring, particularly the need to redefine one's self and one's roles were reflected in the literature review and qualitative study and these have been included in the section on carer wellbeing.

Informal carers for people with PD are often spouses(130). It is apparent from the qualitative evidence presented earlier (chapters 4 and 6) that PD can have both positive and negative impacts on the pre-existing relationship between patient and spouse and that health care professionals concerned with carer wellbeing should be mindful of this dynamic. The prompt: *"Do the family / carer currently feel that caring has a net positive or negative affect for them personally and their relationship with the patient?"* has been introduced to the tool with this in mind.

Finally the importance of the last section, regarding preparation for grief relating to impending or recent bereavement, has been retained in the tool and should not be underestimated as it was an important theme arising from the systematic review of qualitative literature (chapter 3 and 4).

7.6 Conclusions

This chapter described the process of adaptation of the NAT:PD-c for use in Parkinson's disease, based on the qualitative findings from chapter 4 and 6. In particular, "red flags' have been introduced to raise awareness of groups more likely to have palliative care concerns, while the physical and neuro-psychiatric problems associated with PD have been highlighted.

The clinimetric testing of the adapted tool, known as the NAT:Parkinson's disease, is described in the following chapter (8-12).

Chapter 8

Methodologies relating to Clinimetric testing

8.1 Introduction

Having discussed the adaptation of the NAT-Parkinson's disease in previous chapters, the focus of this section is the process of clinimetric testing, which sought to establish the validity and reliability of the new tool for use in everyday clinical practice.

This first chapter addresses the methodological considerations that underpin validation and reliability as concepts and, the transfer of these concepts in to clinical testing. Subsequent chapters will justify the specific methods selected for the two arms of the project, set out the results of the validity and reliability testing and, finally, discuss these results in the context of previous studies.

8.2 Validation

8.2.1 *Principles of validation*

In terms of clinical outcome measures, validation is the process by which we seek to demonstrate that a new instrument does what it claims to do and measures what it purports to measure(131). There are different approaches to validation. The so-called *Trinitarian view*, divides validity in to three separate categories: Content validity, Criterion validity and Construct validity(132, 133). Subsequent revision suggests that, rather than separate entities, these categories should be viewed as facets of a single concept. Under this classification the aim is to determine the degree of confidence with which inferences about a specific population can be made using the new scale. This differs subtly from earlier definitions in that it seeks to validate the inferences drawn from the scale i.e. "Unmet palliative care needs are present", rather then the scale itself. This serves to

contextualise the process and means that further validation studies would be required in new populations(132).

The various facets of validity examine the subject instrument in different ways and, in the course of developing a new assessment tool, researchers will usually need to utilise more than one of these techniques, in order to satisfactorily demonstrate its validity.

8.2.2 *Face Validity*

Face validity refers to the appearance of the tool, and whether it appears to measure the phenomenon of interest. Appearance is obviously a rather subjective criteria and the outcome will depend, to an extent, on the audience to whom the tool is presented. In this regard it has been suggested that face validity should be considered as validity in the eyes of a lay person, while content validity (see below) should be thought of as being validity in the eyes of an expert(134). This approach would work well for a patient completed assessment tool, such as the PDQ-39(135). However, given that the NAT:Parkinson's disease is a clinician completed instrument, where patients and carers do not complete any aspect themselves, it is suggested here that both its face and content validity should be determined by the range of clinical staff who are likely to use it.

8.2.3 *Content Validity*

Content validity is used to assess scales which contain multiple variables. It asks the question - *Does this scale cover all the areas which should be covered, within the construct being measured?*

Content validity is based on the idea that some measures can be *intrinsically valid*. This suggests that, if a scale contains all aspects of the phenomenon of interest (i.e. palliative care need in PD) and contains no irrelevant aspects, then it must have some validity.

This process obviously requires a considerable knowledge of the construct being examined. As such, it would usually be addressed by a group of experts in the field, who would be best placed to examine the content for completeness, as opposed to face validity, which does not require this degree of in depth knowledge(132) (see above).

While content validity may also be subjective, it implies a more forensic examination of the elements within the instrument, than would be needed to establish face validity. These two, related, aspects of validity are defined by the HTA statement on validity assessments thus:

"Face validity examines whether an instrument appears to measure what it is intended to measure, and construct validity examines the extent to which the domain of interest is comprehensively sampled by the items, or questions, in the instrument"

(Guyatt *et al.,* 1993b:624- Quoted in HTA report "Evaluating patient-based measures for use in clinical trials"(136))

8.2.4 *Criterion Validity*

Criterion validity is established through comparison of the new tool or scale, with an established "criterion" scale(131, 132). The criterion scale being, one that is known to accurately measure the phenomenon of interest and acts as the established gold standard measure. In health care, such gold standard measures are most often available for physical parameters, for example, laboratory measurements of blood glucose or cholesterol. In this instance, a new hand held glucose monitor might be assessed against the criterion of the established laboratory measurement.

Clearly, to assess criterion validity, the new tool under examination, must be measuring exactly the same phenomenon as the old, established, criterion measure. It would then, usually have some inherent advantage over the existing test, such as reduced cost, or fewer side effects. An example of this would be the diagnosis of pulmonary embolism, where the gold standard test - pulmonary angiography, has been superseded by CTPA (Computer Tomography Pulmonary Angiography), a less invasive test.

Criterion validity may be referred to as concurrent; where the new scale and the criterion measure are administered contemporaneously, or predictive, when the outcome of the criterion test will not be known for some time(132).

This form of validation allows us to assess, with some certainty, the degree to which a new instrument measures what is purports to measure. However, in areas of medicine

which are more subjective, such as the presence or absence of unmet palliative care need, it may be much more difficult to identify a criterion measure. Indeed, it seems unlikely that we could claim to "know", rather than simply strive to estimate, such complex phenomena. Therefore, it is necessary to find a method of testing validity, which does not rely on the presence of an absolute, gold standard measure.

8.2.5 *Construct Validity*

Described by Cronbach and Meehl in the 1950's, Construct validity offers a solution to this problem, by allowing the assessment of validity, in the absence of a gold standard comparator.

A construct is described as "…some postulated attribute of people, assumed to be reflected in test performance" and would commonly refer to a psychological trait(137). However, it is equally applicable to this study, where the "*postulated attribute*" is unmet palliative care need, which we "*assume to be reflected*" in the results of the NAT-Parkinson's disease.

In criterion validity, both the criterion and the test instrument are assumed to be measuring the same underlying phenomenon(131, 132). This is not the case in construct validity where, instead, the assumption is that the phenomena or constructs measured by the two instruments are different, but related. This may be particularly useful where, as with the NAT-Parkinson's disease, the tool is developed to measure a construct for which no other measure exists.

Based on our existing knowledge, we should be able to predict the way in which one construct will relate to the other; for example, as the motor symptoms of PD progress, it seems reasonable to expect greater palliative care needs in the physical domain. In this way, validity can be established by testing a series of predictions about the relationship the test instrument could be expected to have with other, established, scales(132). In the example used above, it might also be reasonable to assume a positive correlation between physical palliative care need and pain. The validity of the new measure of physical palliative care need would be supported by a positive correlation with scales

measuring pain and motor severity. Conversely, a failure to demonstrate this relationship would call the validity of the new tool in to question.

It is important to highlight two characteristics of construct validity:

Firstly that, unlike criterion validity, the aim here is to provide evidence to *support* the validity of the new measure, rather than *prove* it. Thus the number of tests that could be done to establish construct validity, is limited only by the number of possible associations between the new and the established scales(132). The greater the number of associations demonstrated, the stronger the claims of validity. Cronbach and Meehl envisaged the construct under study existing within a network of other related constructs – the nomological network(137). Each time a connection within the network is demonstrated, the instrument becomes more valid and more is known about the construct under study. This is useful because it also conveys the sense that the constructs under study are not absolute or observable. If they were, it would not be necessary to test construct validity. Rather our understanding of them may change slightly, through the process of validation.

Secondly, it is also important to point out that it is neither expected, nor desirable, for the association between test and comparator scales to be perfect. The amount of correlation that can be achieved will be influenced by the respective reliability of the test and comparator scales, where the greater the re-test error, the smaller the expected correlation.   In addition, an association that is *"too good"* would imply that the two scales are simply measuring the exact same, rather than related constructs, which may in turn call in to question the need for the new instrument(131).

In order to strengthen claims of construct validity, it may be useful to demonstrate both divergent and convergent patterns. That is, to demonstrate that our tool is positively associated with constructs which it should be related to, and negatively associated with those which should not be related(131, 132, 134). In the example given above, palliative needs in the physical domain may be expected to have a positive relationship to motor scales and pain, but less so with non-motor features of the disease. The terms **convergent** and **divergent** validity can be used respectively, to describe this feature of construct validation.

8.2.6 *Summary*

In summary, validity is best described as a single concept, with multiple facets. By the same token, validation is not a single discreet activity, but rather a process, which draws, to a greater or lesser degree, on the concepts described above. Individually, none of these techniques will be sufficient to *prove* validity, but they can be used cumulatively to establish a body of evidence, which supports the valid use of the instrument, in a given situation.

8.3 Reliability

8.3.1 *Why test reliability?*

The score obtained from an instrument, such as a psychological scale, can be conceptualised as containing two elements; the *true* score and the *error* associated with the measurement(138, 139). Likewise, variation in scores for different individuals will comprise the true variability between those individuals, plus the measurement error associated with the test. Assessments of reliability seek to establish the degree to which measurement error, of which there are several types, is a factor in the results achieved by a test. The greater the amount of variability introduced by error, the less reliable the test(138, 140). Put another way, if a test has perfect reliability, then repeated tests will only vary if there are true changes in the subject being assessed.

If validity provides a sense of how closely a new instrument approximates the construct it claims to measure, then reliability relates to the ability to achieve similar results, with repeated measurements(138). It is important to note that a highly reliable tool does not necessarily give the correct result; it may be reliably inaccurate, due for example, to a systematic error in the instrument(138). Likewise an instrument which is extremely valid, offering a close approximation of the phenomenon under study, may be of little use if it has poor reliability, as we can not be confident that the variation in results is due to true change in the study subject, rather than in the measurement process. Thus if there is low reliability it becomes difficult to glean useful data in clinical practice and the tool may be of little use.

Reliability may be adversely affected by variability in the instrument itself, or in the person applying the instrument. Variability due to the instrument is easy to visualise when considering mechanical tools, such as a manual sphygmomanometer, where there will be intrinsic error in the pressure reading within the cuff, or the rotation of the dial, but may be less obvious for verbally administered instruments. For such tools, like the NAT-Parkinson's disease, this type of intrinsic error may come from the nature of the questions; for example, where a question is too ambiguous, the person answering may reply differently each time, without their underlying condition changing. This would lead to variation that did not reflect true change in the subject being assessed. In the same way, variability may be introduced by the person applying the instrument, for example they may concentrate less on repeated measures, or be influenced by previous results(139).

A second source of variability comes from differences between observers. In the examples given above, when different people rate blood pressure using the same machine, variability will be introduced by multiple factors, for example the sensitivity of their hearing or the speed at which they deflate the blood pressure cuff. The same phenomenon may occur in the application of rating scales, as different raters perceive items on the scale differently.

These examples relate to two aspects of reliability, test-retest and inter-rater reliability respectively, and are discussed further below.

8.3.2 *Test-retest reliability*

This examines the variability associated with repeated measurements of the same subject, taken by the same assessor at different points in time(139). In doing so it allows us to estimate the variability associated with the instrument itself. This is akin to calculating the error associated with a new blood pressure monitor, by taking repeated measures in the same individual, with the same operator.

Ideally, to ensure that any variation observed is due to the instrument error, there should be no changes in the subject (in this case a patient with PD) or the rater (clinician), between assessments. Clearly however, this is difficult to achieve and thus the test is

subject to confounding, particularly by change in the underlying health status of the subject. If, for example we perform the second assessment of an individual 2 weeks after the first, any change in their disease over this time may alter their palliative care needs, meaning that at least some of the difference between the first and second assessment is due to true variation. This would tend to reduce the apparent reliability of the tool(136, 138).

One approach to minimising the risk of this occurring is to perform the second assessment immediately, or very soon after the first, removing the possibility of a significant change in health status. However, this in turn creates its own risks of contamination as patients and assessors may remember, and be influenced by their response to the first assessment. This so called *"priming"* of participants may lead to greater levels of agreement than would be present in real life, and hence over-estimate test-retest reliability.

Thus, the ideal amount of time which should be left between assessments will depend on how quickly the underlying constructs change and to what extent the participants are able to recall and replicate the assessment.

In general it is advised that between 2 and 14 days should be allowed between assessments for health measurement scales(140).

8.3.3 *Inter-Rater reliability*

Inter-rater reliability refers to the variability associated with repeated measures of the same subject, with the same tool, by different assessors(139, 140).

Depending on the nature and practical administration of the assessment, the same difficulty with change over time and priming of the subject may be encountered. However, it would be possible here to conduct both assessments simultaneously, reducing the risk of confounding from this particular source.

It is interesting to note that, conceptually, inter-rater reliability incorporates both the measurement error of the tool - that due to repeated assessments as discussed above, as well as the variability associated with different assessors. Another way of expressing this

is to say that as it includes all of the variability incorporated in test re-test reliability (repeated assessment), as well as an additional variation unique to inter-rater reliability (inter-operator). As such, the degree of variation should always be larger; and hence the correlation between assessments smaller, when examining inter-rater reliability than test-retest(140).

The implication of this conceptual difference is that it may not always be necessary to measure both test-retest and inter-rater reliability. It is suggested that for practical purposes, if inter-rater reliability is high this may be sufficient, as test-retest reliability should, by definition, be greater still, as all the variability form the latter is incorporated in the former(140).

The disadvantage of this strategy is, of course, that a low score for inter-rater reliability does not reveal anything about the adequacy of test-retest reliability, and would necessitate a further study to test this concept.

8.4 Conclusion

The above discussion of the theory behind validity and reliability sets the scene for the following chapter, which presents the methods used in the clinimetric testing of the NAT-Parkinson's disease. Although it is tempting to think of validity and reliability as discreet concepts, which can be examined in isolation, it is evident that this is not the case. Each is a multi-faceted concept, which ideally requires a process of examination, rather than a single test. Equally, the results of this process need to be interpreted in the round. An instrument will not be universally valid or reliable. Rather the findings will apply to a specific population and a specific use of the instrument; a point which is particularly important where instruments may be adopted for use beyond that for which they were originally developed.

It is important to recognise that there are margins of error, both in the manner in which they reflect the construct and in the way this theoretical measure performs in repeated clinical practice. The balance between these two qualities and the way in which they can be applied to the population under study, will determine the degree to which a new instrument is clinically useful.

The following chapters have been arranged to cover first the methods (chapter 9) and results (chapter 10) for validity, followed by the same for reliability (chapters 11 and 12), in order that the logical flow of information is not interrupted. These will then be drawn together in the discussions (chapter 12).

Chapter 9

Testing Validity: Methods

9.1 Introduction

In the preceding chapter covering methodology discussion focused on the way in which validity is conceptualised. In this chapter, the approach adopted for each facet examined for this tool (face, content, construct) is addressed in turn, with an emphasis on construct validation, which formed the bulk of the work. In addition the rationale for selecting the study population, and sample size is laid out, along with a discussion of analytical methods.

9.2 Face and content validation

The design of this project, with a qualitative phase preceding and informing the adaptation of the needs assessment tool, has the benefit of promoting face and content validity. Patients and carers were enrolled in both the primary qualitative study (chapters 5-6) and the research articles incorporated in to the systematic literature review (chapters 3-4). The adaptation process then explicitly matched the constructs arising in this work to the themes of the existing NAT:PD-c(29). This served to enhance face and content validity, by ensuring that the new instrument is embedded in and relevant to, the experience of both patients and their carers.

Likewise the decision to adapt a pre-existing palliative assessment instrument, allowed us to build on a framework for palliative needs assessment which had already been validated; supporting the content validity of the NAT-Parkinson's disease as a measure of palliative need.

Following the initial adaptation process, face and content validity were established by reviewing the NAT: Parkinsons disease tool with experts in the field. 8 clinicians involved in the care of people with PD (2 consultant neurologists, 1 consultant and one specialist

trainee in PD and elderly medicine, 1 PD nurse specialist) and in an existing PD palliative care service (2 consultant palliative physicians, 1 palliative neurology nurse specialist) were sent the tool and independently asked to assess its content and appropriateness, for assessing palliative care need in every day clinical practice.

9.2 Criterion validity

At the time this project was conceived and designed, there were no validated measures of palliative care need in PD to act as criterion measures. As such, the main focus of this section of the project was on exploring construct validity (see below).

Since the start of the research, two PD palliative care assessment tools have been described in the literature(124-126). Both of these are adaptations of existing palliative assessments, the POS(125) and ESAS(126) respectively, with the addition of a supplementary section of Parkinson's specific questions. These tools, which both produce summary scores, would certainly have been useful for the construct validation of the NAT-Parkinson's, but could, arguably, not have been used as criterion measures, because they differ in their underlying construct from the NAT-Parkinson's, in that they aim to quantify, rather than screen for, unmet need. In addition, neither is in widespread clinical use at this time, so it is difficult to make a case for them having the status of gold standard measures within the field of study. Nonetheless, it is likely that these tools will be an important part of future research in this area, which could be supplemented by a screening tool such as the NAT (see chapter 13).

9.4 Construct validity

In the absence of an identifiable criterion measure, the main focus of the validation phase of the study was construct validation.

The first step in this element of the study was to identify the key constructs contained within the NAT-Parkinson's disease, in order to identify existing measurement tools which could act as comparators.

Because of the time restriction which were inevitably in place, with the need to obtain approvals from the regional ethics as well as local research and development committees,

the process of selecting comparator tools began prior to the final adaptation, based on the framework of common themes which constitute the NAT in all of its current forms(29, 30).

9.4.1 *Identifying underlying constructs*

The NAT format explores unmet palliative care need under three main themes:

1- Patient wellbeing

2- Ability of carer / family to care for the patient

3- Patient and family wellbeing

Within these themes, it was possible to identify five common, or key, constructs.

1- Physical symptoms (Motor and non-motor)

2- Physical functioning

3- Depression / Anxiety and neuro-psychiatric functioning

4- Patient psychosocial functioning

5- Carer strain

9.4.2 *Identifying comparator tools*

The theory underlying the process of construct validation is discussed in chapter 8 (see section 8.2.5). Given the absence of any other tools designed to measure palliative care need in PD, it was important to select comparator tools that measure constructs with a predictable relationship to those identified in the NAT. Wherever possible, these tools should have been validated for use in PD, because, as discussed in chapter 8, the validation process is specific to the situation in which the tool is used, rather than applying to the tool per se(132).

It was also important to consider practical aspects, such as administration of the potential comparator tools. It is recognised that, because scores on a scale are due in part to the measuring process as well as the underlying construct, a degree of correlation between scales may occur because of similarities in their administration process(132). Therefore, it is suggested that the process is more rigorous if the primary scale and the comparator scale are *"maximally different"(141)*; for example self-administered questionnaire, verses,

observer administered scale(132). The final and by no means the least important consideration, was the implication for participant burden of the decisions taken during instrument selection. While, from a theoretical viewpoint, it would have been desirable to explore the validity of the NAT Parkinson's against many different tools, this would have placed an intolerable and unacceptable burden on participants.

9.4.3 *Comparing available PD tools*

There are many existing rating scales in PD, most of which are designed primarily for use in research, and few of which form part of routine clinical practice, outside of academic centres. The Movement Disorders Society has commissioned a series of reviews, evaluating the quality and utility of rating scales(142).

While some of these tools afford the opportunity to evaluate individual symptoms in great detail, other scales, particularly those measuring quality of life have the advantage of examining several constructs simultaneously, helping to reduce the burden placed on participants. Of the global assessment tools in PD the most widely used are the PDQ-39(135) and the UPDRS (Unified Parkinson's Disease Rating Scale)(143). The latter has been the subject of expert critique by the Movement Disorders Society (MDS)(144), leading to production of a modified version (MDS-UPDRS)(145), which had recently undergone validation(146). These two tools were selected for further evaluation, regarding their suitability as comparator scales.

9.4.3.1 *MDS-UPDRS*

The MDS-UPDRS was produced following criticism of the widely used UPDRS, particularly its lack of focus on non-motor aspects of PD, which have received increased attention in recent years(144). In addition the authors sought to improve some aspects of administration, such as the instructions provided for raters, in order to improve the uniformity and reliability(145). The scale is made up of 4 sections covering:

1- Non-motor aspects of experience of daily living
2- Motor aspects of experience of daily living
3- Motor Examination
4- Motor Complications

It is advised that each section should be used as a "stand alone" score for that element of the disease. Importantly, the validation process did not support the creation of a composite score or "summary index", created by combining the scores for each section, as was the case for the original version of the UPDRS(146). While summary indices are often used in clinical practice, for the purpose of validating the NAT Parkinson's disease the absence of a summary index was not disadvantageous, as correlation with individual sections of the MDS-UPDRS was considered to be of greater interest for the validation process.

During construction of the instrument, the MDS task force used factor analysis to identify the various aspects of PD assessed in each section of the instrument (see figure 11).

Figure 11 Factors identified during development of MDS-UPDRS

Part 1	Depression, Anxiety, Apathy Other Non-Motor functions
Part 2	Fine motor functions Tremor and eating tasks Large motor functions
Part 3	Midline function Rest Tremor Rigidity Bradykinesia: upper extremity (R), upper extremity (L), lower limbs. Postural / kinetic tremor
Part 4	Fluctuations (including off state dystonia) Dyskinesias

There are clear similarities between the factors identified in sections 1 and 2 of MDS-UPDRS and the patient physical and psychological constructs examined by the NAT. These are illustrated in a summary table below (see table 13).

It is worth noting that whilst the physical examination section (section 3) of MDS-UPDRS is the best indicator of physical symptoms and disease stage, it does not focus on

function, which is covered in section 2. As such, section 2 was considered to be more applicable to this study.

9.4.3.2 *Administration of the MDS-UPDRS*

The MDS-UPDRS is markedly different from the PDQ-39(135), and many other HRQoL tools, in terms of administration. Sections 1 and 4 are designed to be completed by the health care professional, with input from patient / carer, while section 2 is completed by the participant and section 3 is dependent on clinical examination. In addition the authors explicitly recognise the high prevalence of cognitive impairment in PD and the impact this will have on administration of the tool. In sections 1 and 2 users are asked to identify who is contributing to the completion of the tool; patient, carer, or patient and carer in equal proportion. This raised the intriguing possibility that the tool may be valid for assessment of patients with significant cognitive impairment, such that carers are the sole respondent. Given the prevalence of cognitive impairment in PD; cohort studies suggest a cumulative incidence of up to 60% by 12 years(107), it is important to consider this group in validation studies. The option of proxy completion of the MDS-UPDRS by carers, provided the opportunity to develop a feasibility arm, exploring the use of the NAT-Parkinson's disease, in patients with dementia (see section 9.10).

9.4.3.1 *Burden of administration*

Based on the information provided by the development task force, it was possible to estimate some of the burden to participants, associated with use of the MDS-UPDRS. The design team deliberately aimed for a final tool that could be completed in less than 30 minutes of clinic time(145). They give the estimated times, broken down by section (see below). However this does not account for the time taken by respondents to complete the questionnaire section (section 2) at home, which is likely to vary according to the nature and severity of their impairments, and the degree to which carers contribute. These figures were used when estimating the burden which would be placed on patients and carers through using the MDS-UPDRS to validate the NAT: Parkinson's disease.

The breakdown of administration times for MDS-UPDRS by section:

- Section 1- 10 minutes

- Section 3 – 15 minutes

- Section 4 – 5 minutes

9.4.4 *PDQ-39*

The PDQ-39 is a well established, PD specific, HRQoL tool which has been well validated and forms the basis of assessment in many PD trials. It consists of 39 questions which cover eight domains within the overall construct of quality of life(135, 147). In contrast to the MDS-UPDRS, the PDQ-39 may be used as a summary index (PDQ-39SI), or the individual sections may be scored separately. The domains identified during development of the PDQ-39, along with the construct from the NAT-Parkinson's, to which they best correspondence, are displayed below in the summary table below (see table 13).

Table 13 Showing themes from comparator tools and corresponding NAT construct

Theme from Comparator Tool	Corresponding NAT construct
MDS-UPDRS part 1	
Depression, Anxiety, Apathy	Depression and Anxiety / Neuro-psychiatric
Other Non-Motor functions	Motor and Non-motor physical symptoms
MDS-UPDRS part 2	
Fine motor functions	Motor and Non-motor physical symptoms
Tremor and eating tasks	Physical functioning
Large motor functions	Motor and Non-motor physical symptoms
PDQ-39	
Mobility	Motor and Non-motor physical symptoms
Activities of Daily Living (ADL)	Physical functioning
Emotional wellbeing	Depression and Anxiety / Neuro-psychiatric
Stigma	Patient psycho-social functioning
Social support	Patient psycho-social functioning
Cognition	Depression and Anxiety / Neuro-psychiatric
Communication	
Bodily discomfort	Motor and Non-motor physical symptoms

9.4.4.1 *Discussion of PDQ-39*

Whilst the content validity of the PDQ-39 was initially felt to be good, particularly as it was derived from qualitative patient interviews, subsequent authors have criticised it for not covering sleep and sexual dysfunction, which may be prominent in PD(148).

The areas covered by the PDQ-39 appear to map well with many of the concepts of the NAT. This offered the obvious advantage, in terms of participant burden, of being able to validate several elements of the NAT Parkinson's with a single comparator questionnaire, whilst allowing comparison with sub-sections as well as an over-all summary index.

9.4.5 *SCOPA-PS*

One area which is not well addressed by either of the tools discussed so far is psycho-social functioning. The SCOPA-PS is a tool designed specifically to examine the psycho-social aspects of PD(149). The tool consists of only 11 questions and is self-completed by the patient, which offered the advantage of participants being able to complete the tool prior to study assessment, but again potentially disadvantaging those with cognitive impairment.

As demonstrated above, some aspects of psycho-social functioning are covered in the general PD quality of life tools. The question here, was whether these constructs could be adequately examined using the PDQ-39 / MDS-UPDRS, or whether a specific psycho-social tool was required.

The authors of the SCOPA-PS recognise that general assessment instruments will cover some aspects of psycho-social functioning , particularly as HRQoL is generally considered to consist of 3 parts (physical, mental and social). However they make the case for a separate instrument, focusing specifically on psycho-social aspects of the disease, as having greater utility(149). Specifically, they state that patients may often struggle to separate physical from psycho-social function, while clinicians, by dint of their profession, tend to somatise problems and may overlook the psycho-social aspects of disease

In the final analysis, it was felt that the presence of a PD specific psychosocial tool would be of benefit and, moreover, that the attributes of the SCOPA-PS, being only 11 questions

and self-administered, meant that the balance between data collection and participant burden could be maintained.

9.5. Assessing carer strain

There has been increasing interest in caregiver strain in PD over recent years, and several groups have identified factors that may be associated with it(150-152). These studies suggest that carer strain is associated with patient quality of life scores(152), disease stage and, more surprisingly, inversely correlated with carer age. This latter finding is thought to be a result of altered levels of expectation in older, compared with younger carers(150).

Despite there being an apparent distinction between "carer burden" and "carer strain", with the latter being the result of the former, these terms often appear to be used inter-changeably in the literature and drawing a distinction between tools which purport to measure one rather than the other is unlikely to be helpful.

Of the instruments designed to measure carer strain or burden both the Zarit Burden Interview (ZBI) and the Caregiver Strain Index (CSI) have been utilised in previous PD studies(153, 154).

The 22 item ZBI was initially developed in carers of individuals with dementia, as a 22 question inventory, which is self-completed by the carer in question(155). It has subsequently undergone revisions. A review by Higginson et al examined the qualities of the shorter versions of the ZBI and concluded that the 12 item, ZBI-12, was the most effective and moreover that it was valid to use the instrument in populations outside of dementia, including palliative care, advanced cancer and brain injuries(156).

The CSI(157) is a 13 item assessment, each item in the original version being scored "yes / No", but subsequently, the tool was updated to utilise a 3 point scale with the anchors "Always / Sometimes / Never". The updated scale demonstrates greater internal reliability (0.9) than the original and also has data to support test-retest reliability, which was not previously available(158)

The ZBI has the advantage of being validated and frequently used in palliative care, while the CSI was developed in a more general population (general elderly carers, rather than dementia carers). In addition the m-CSI had recently been utilised in a number of PD studies, facilitating future comparison across the field(152, 154, 159). As a result the m-CSI was adopted for this study.

9.6 Final Selection of comparator tools

The final selection of comparator tools was influenced by the need to cover all constructs contained in the NAT-Parkinson's disease, as comprehensibly as possible, balanced against the need to minimise the burden of participation.

As such, both the PDQ-39 and MDS-UPDRS were chosen, each of which addressed multiple constructs. The individual domain scores of the PDQ-39, rather than the summary score, were used for validity testing as they often represented a closer approximation of the construct being examined. The non-motor features of PD, which are not well covered by the PDQ-39 were captured in the MDS-UPDRS part 1.

In addition the SCOPA-PS, was selected as with only 11 items it was felt to offer a good balance between assessment depth and burden of completion. Finally, the carer elements of the NAT were assessed using the modified caregiver strain index (m-CSI).

All of these assessment tools, except for part 1 of the MDS-UPDRS, are self-completed, allowing them to be administered as postal questionnaires. This served to reduce the length of time participants had to spend in direct contact with the clinician, as well as favouring Campbell's principle of maximum difference (see section 8.2.5).

Based on the above it was possible to predict, a priori, which comparator tools would be expected to correlate to each construct of the NAT-Parkinson's disease and those where little correlation would be expected. It was also possible in some instances to estimate the size of this correlation. For example, where the totality of a single construct could only be captured more than one comparator tool - e.g. section 2.1 patient physical which includes motor and non-motor symptoms, it was evident the correlation with each comparator tool must be less than perfect. This also allowed convergent and divergent validity to be examined, by comparing the Tau B result achieved for these items against

another comparator tool where moderate or good correlation had not been expected, (see section chapter 10, table 22). These predicted associations are displayed in table 14.

The sub-sections of the NAT: Parkinson's disease relating to financial concerns were not examined as no suitable comparator tool was identified. Therefore 11 of the 13 sub-sections of the tool were examined in the validation process, the protocol for which is set out below.

Table 14 NAT constructs and predicted correlations

NAT: Parkinson's Construct	Comparator tool	Correlation predicted
2.1 Patient physical	MDS-UPDRS 1	Moderate
	PDQ – mobility	Moderate
2.2 Patient psychological	MDS-UPDRS 1	Moderate
	PDQ-Emotion	Moderate
2.3 Patient ADL	MDS-UPDRS -2	High
	PDQ - ADL	High
2.4 Spiritual	PDQ-Emotion	Moderate
	SCOPA	Moderate
2.6 Health beliefs	PDQ-Support	Moderate
	SCOPA	Moderate
3.1 Carer distress	m-CSI	High
	MDS-UPDRS -1	Moderate
3.2 Carer difficulty (physical)	MDS-UPDRS-2	Moderate
	m-CSI	High
3.3 Carer difficulty (coping)	m-CSI	High
3.5 Carer difficulty inter-personal	m-CSI	Moderate
	SCOPA	Moderate
4.1 Carer/family wellbeing	m-CSI	Moderate
	MDS-UPDRS-1	Moderate
4.2 Grief	M-CSI	Moderate

9.7 Validation study protocol

9.7.1 *Setting*

The research was based within the PD service at the Scarborough and North East Yorkshire hospitals trust, which has since become part of the York Hospitals Trust. This service cares for around 450 people with movement disorders, the majority of whom

have PD, and is inclusive of all patients, regardless of age or co-morbidity. This offered an advantage compared with larger tertiary centres, which often have separate general medical and elderly care PD services, which effectively stratify patients according to age or disease stage.

The centre also benefits from, what we believe to be, one of the first specialist palliative care PD services in the country. As such it was ideally placed to conduct research of this sensitive nature, given the local availability of expertise to meet any potential issues arising from, or more likely, identified through the research process.

9.7.2 Eligibility

The inclusion and exclusion for the validity study are set out below.

Inclusion criteria:

- A confirmed diagnosis of Idiopathic Parkinson's disease,

- Age over 18,

- All disease stages from time of diagnosis.

Exclusion:

- Diagnostic uncertainty,

- Patients unable to give informed consent,

- Communication difficulties, such that completion of assessment instruments is impossible.

9.7.2.1 *Discussion of eligibility criteria*

Cognitive impairment is common in PD, and may be present even from the early stages of the disease, with cohort studies showing cumulative incidence of dementia at around 60% by 12 years post diagnosis(107). In addition PD appears to confer a twice normal risk of MCI (Mild Cognitive Impairment) even at the time of presentation(160). Given this, it was essential to consider the inclusion of people with cognitive impairment in our study design. This was one reason for the decision to use participants' capacity to consent,

rather than a cut off score on cognitive assessment testing, as the basis for inclusion in the study. This provides the opportunity for those with mild cognitive impairment, or even early dementia to participate freely, whilst maintaining the primacy of informed consent.

The difficulties of including people with more advanced dementia include the challenge of informed consent and the practical difficulty of data collection.

A potential solution was provided by the properties of the NAT, which is an administrator completed instrument, and the nature of the clinical consultation in dementia.

In daily clinical practice it is common for both patient and carer to be present during consultations and to contribute to a varying degree. As patients develop dementia, their carer may naturally become the greater source of verbal information, whilst acknowledging the need to gather verbal and non-verbal information from the patient wherever possible. The degree to which this "proxy information" from the carer influences clinical decision making will vary according to the residual cognitive functioning of the patient.

A clinician completed instrument such as the NAT, where scores are based on the overall impression of a consultation, has the advantage that the natural discourse described above can be reflected in the assessment. It is therefore feasible that the NAT-Parkinson's disease could be used successfully in a full range of patients, including those with dementia, even at late stage.

Thus it was decided to examine the use of the NAT-Parkinson's in dementia, as a feasibility study, running parallel to the main study group. Carers of patients with PD and dementia were eligible for inclusion, to act as a proxy for the patient, and represent their palliative care needs. The other inclusion and exclusion criteria being the same as the main study (see above).

9.8 Recruitment and Consent

Patients were recruited through the PD service at Scarborough and North East Yorkshire Hospitals Trust. Sequential patients, attending the PD clinic, were invited to take part in

the project, by a member of the clinical team (EJ, ER, LB). If they expressed an interest in participating, they were provided with Participant Information Leaflets (PIL) and received a follow up contact by telephone, or if requested, in person, to answer any questions they may have had about the study. This step was completed by either the clinical research nurses or the principal investigator (ER).

Those patients expressing a wish to participate in the study at the 1 week follow up, then received the self-completed assessment forms by post, and had a time scheduled, at their convenience, for administration of the clinician completed assessments (NAT-Parkinson's disease and MDS-UPDRS part 1).

Participants were asked to bring the completed questionnaires with them when attending the clinical assessment. Completion and submission by hand, of the postal questionnaires was taken as implied consent to this element of the study.

The advantage of this design was to reduce the number of participant contacts and thus reduce the burden of participation, which is particularly relevant in this area of study.

Potential participants who preferred a face to face meeting, with written consent prior to the receipt of any assessment materials, could request this at the time of enrolment (1 week follow up telephone call).

The posted documents included patient assessment tools (PDQ-39, MD-UPDRS part 2, SCOPA-PS) and a carer completed questionnaire (modified CSI). On receipt of the postal documents, patients were asked to give the m-CSI to their carer, if they have one, along with a carer version of the Participant Information Leaflet. It was made clear in the study material that patient participation was not dependent on participation by their carer and in turn, carers were under no obligation to participate. Likewise, it was emphasised to all potential participants that their decision regarding involvement in the study would in no way influence their future care.

At attendance for the clinical assessment, all participants were asked to provide written consent, and were given the opportunity to withdraw from the study, including withdrawal of their postal questionnaires, if they wish. It was also recognised that some participants would require assistance to complete questionnaires and time was allocated

for this, prior to the start of clinical assessment. A flow chart illustrating the progression of participants through the validation study is shown below **(figure 12).**

Figure 12   Flow chart for main validation study

Clinic attendance

Invitation to participate

Decline   **Express interest**   **Unsure**

**Contact number
provided**

PIL provided

Express interest

Telephone follow up (1 week)

Receive self-completed questionnaires

Written consent and administration of clinician completed tools
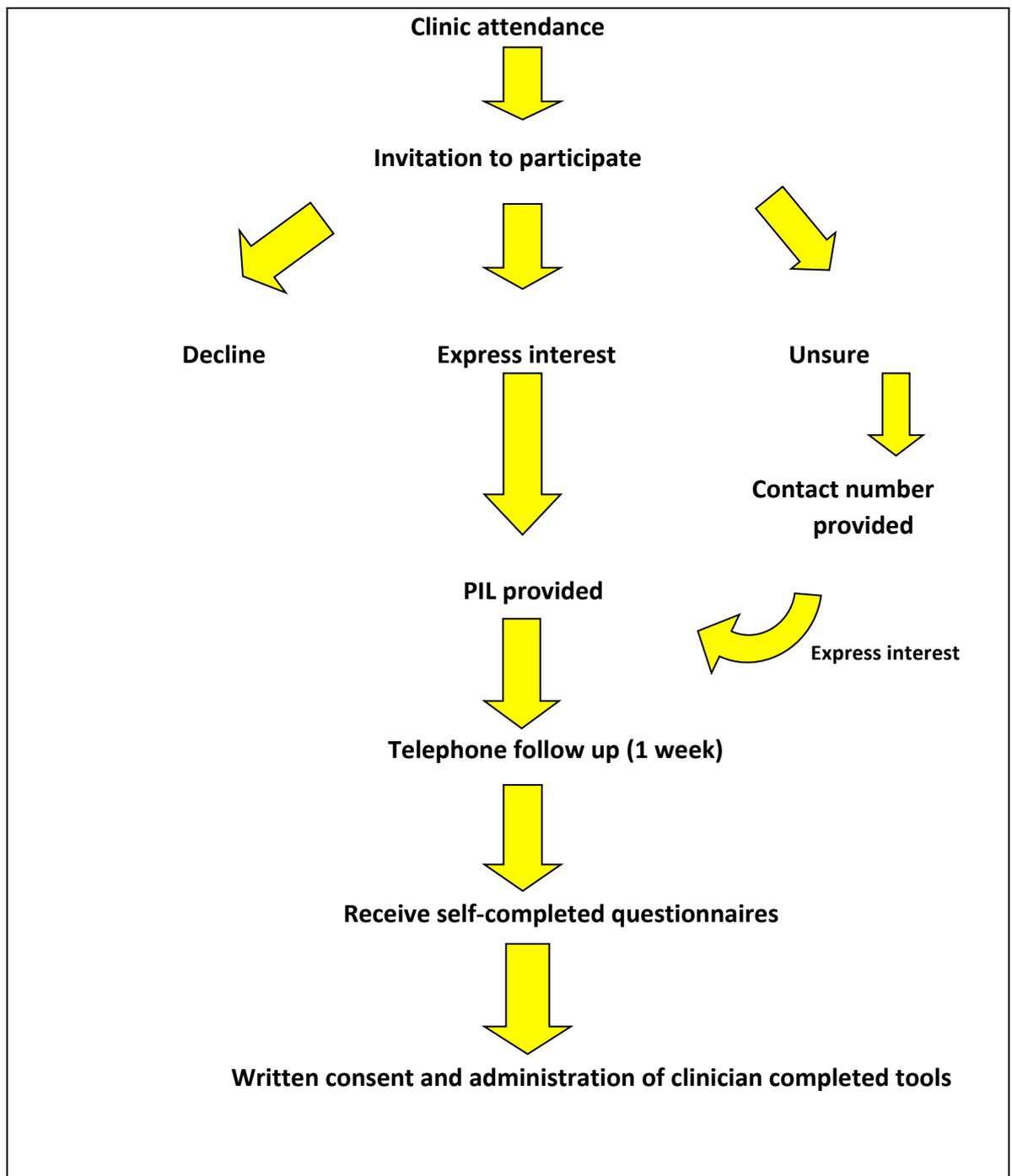
9.9 Data Collection

Stage 1 – postal questionnaires

The participants received the postal questionnaires at least 1 week prior to the clinical assessment. Telephone assistance with completion was available from a research nurse and face-to-face assistance available when participants attended the hospital, before the start of the clinical assessment.

Carers who chose to participate were asked to complete the m-CSI and return it in the separate envelope provided, at the time of the face-to-face assessment. This was linked to the patient completed questionnaires by a unique study code, but responses were of course held in confidence and were not shared with the patient.

Stage 2 – Clinical assessment

The NAT-Parkinson's disease and the MDS-UPDRS part 1 are clinician completed assessments and as such, were completed in face to face interviews with the patient and, where appropriate, their carer.

The MDS-UPDRS part 2 is estimated to take no more than 10 minutes to complete(146). In turn the validation work on the original NAT:PD-c suggested that it could be completed during the course of a routine clinical appointment, and that it did not lengthen the time of the assessment, with the average consultation taking 17 minutes(31). Thus we estimated that an appointment time of 30-40 minutes would be required to complete the assessments, to ensure that participants did not feel rushed and had ample time to ask questions.

Table 15 – Summary of validity testing

Phase of study	Tools administered	
	Patient completed	*Carer Completed*
Phase 1: Postal Questionnaire	MDS-UPDRS part 2 PDQ-39 SCOPA-PS	Caregiver Strain Index (m-CSI)
Phase 2: Clinician led assessment	MDS-UPDRS part 1 NAT-Parkinson's disease	

9.10 Feasibility testing in Dementia

As an adjunct to the main validation process it was intended to test the feasibility of using the NAT-Parkinson's disease to assess need in patients with dementia, where the tool would be completed on the basis of carer, proxy representation, of patient need.

The design of the MDS-UPDRS is such that it can legitimately be used as a carer completed assessment, and thus it was possible to assess validity of the "Motor and Non-motor physical symptoms", "physical functioning" and "caregiver strain" domains of the NAT: Parkinson's disease in this patient group. It was recognised that it would not be possible to assess psycho-social functioning, for which valid comparator in this population could not be identified.

In this feasibility study, phase 1 consisted of the MDS-UPDRS part 2 and the m-CSI, and phase 2 was as described above, with face-to-face completion of MDS-UPDRS part 1 and NAT-Parkinson's disease.

Potential participants were identified from the same PD clinic as the main study. Informal carers of patients who lacked capacity to consent to participation, but fulfilled the other inclusion criteria, were eligible to participate. They were approached by a member of the clinical team and invited to act as a consultee for the patient (usually a spouse or relative) and, if they expressed an interest in so doing, they were provided with the consultee information sheet.

At a follow up phone call, 1 week after the initial approach, potential participants were given the opportunity to discuss the study and ask any questions relating to the study information, or the role of consultee. If they expressed an interest in participation, the primary investigator (ER) visited them, at their convenience, to obtain written consent. This process was conducted in the following steps, in accordance with the Mental Capacity Act:

Step 1 – Carer asked to give their opinion as to what the wishes and feelings of the patient would be towards participation in the research project, if they retained capacity.

Step 2 – Carer asked for their advice as to whether the patient should take part in the research project.

Step 3 – If they indicated that, in their opinion, the patient *would* wish to participate in the research had they retained capacity to consent and that they *should* be included in the project, the carer was asked to provide informed consent for their own participation, as a proxy representative of the patient's needs. (see Figure 13).

It is important to note that the above applies to informal carers. The study did not approach professional or paid carers to act in this capacity.

Figure 13 Flow diagram of recruitment process for feasibility study

Clinic attendance

Initial approach to carer

Decline

Express interest

Unsure

Contact number provide
Express interest

Consultee information sheet provided

Telephone follow up (1 week)

Written consent to participation and provision of questionnaires

Administration of clinician completed tools
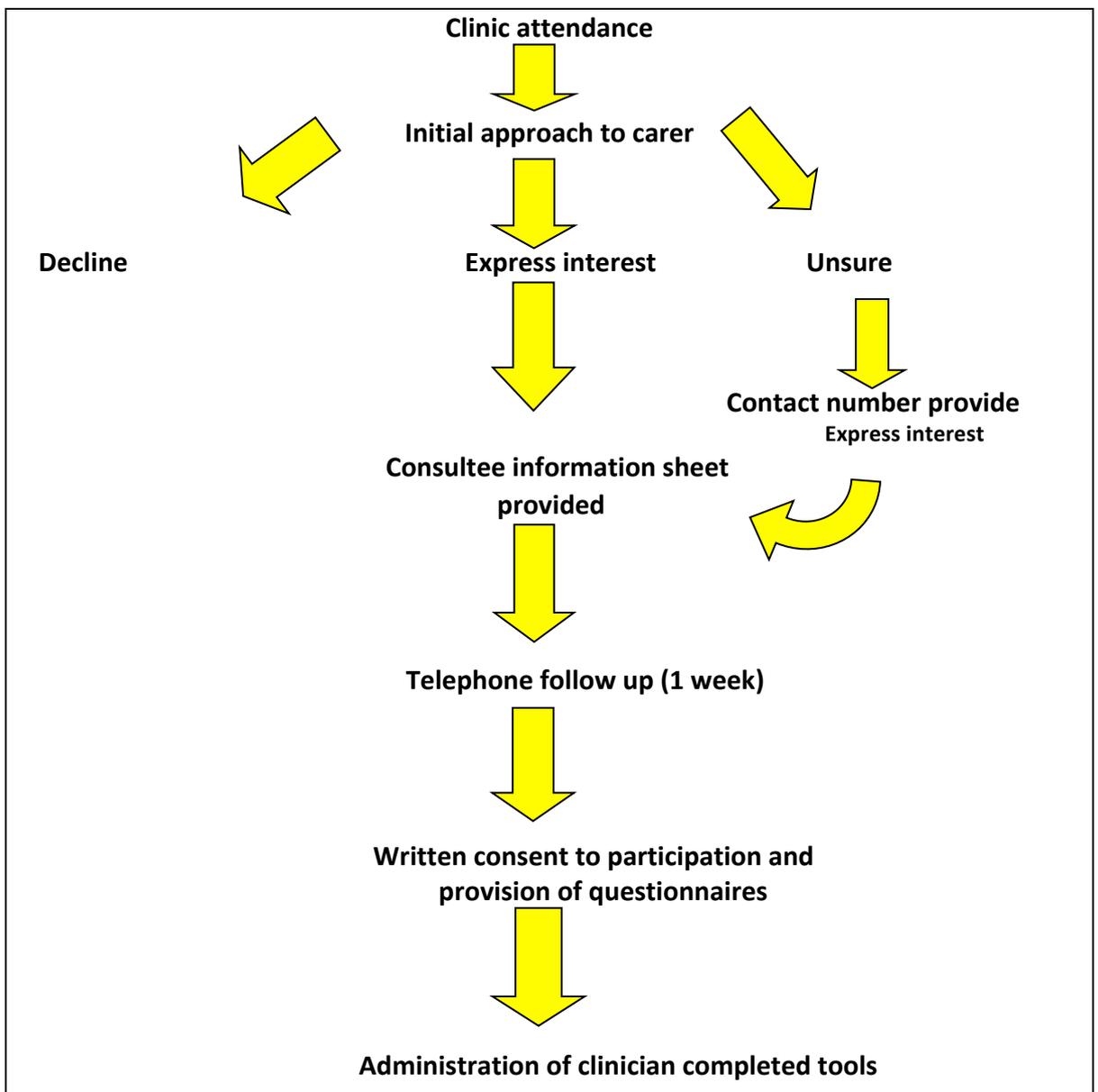
Table 16 – Summary of validation in dementia

Phase of study	Tools administered (Carer completed)
Phase 1: Postal Questionnaire	MDS-UPDRS part 2 Caregiver Strain Index (CSI)
Phase 2: Clinician led assessment	MDS-UPDRS part 1 NAT-Parkinson's disease

9.10.1 *Participant burden*

The burden placed on participants is always an important consideration. This is particularly the case when the participants are patients and carers living with a chronic, degenerative disease. The importance of minimising participant burden was recognized from the outset and had a major influence on project design. The potential sources of participant burden for the validation phase of the project can be considered in three sections; recruitment and consent, completion of postal questionnaires and completion of clinical review.

The recruitment process outlined above, reflects efforts to reduce participant burden, by minimising the number of contacts required with the study team. The best example of this being the use of presumed consent, based on the completion and return of postal questionnaires, to avoid an additional face to face meeting between the first contact and the final clinical assessment.

It was difficult to estimate the time burden placed on participants in completing the self-report questionnaires, not least because of the likely effect of disease stage. Therefore, in order to evaluate whether the burden placed on participants was reasonable, comparison was made with previous validation studies conducted in PD. In a selection of studies published prior to the submission of this protocol, the number of comparator instruments used ranged from 1 – 15, while the total number of items which participants

were asked to complete ranged between 39 – 232(135, 146, 149, 161, 162). The current study required participants to complete a maximum of 3 comparator instruments, containing 76 items in total, well within the range of previously conducted studies, and was judged not to impose an unreasonable burden on participants.

The clinical review inevitably involved some imposition, as participants were asked to attend the outpatients department. Steps were taken to alleviate the strain of this by arranging transport where needed, reimbursing travel costs and providing refreshments. Thought was given to the physical environment, with good wheelchair access ensured and ample time allowed for each visit.

9.11 Sample Size Calculations

Sample size calculation for validation trials can be challenging because this study design seeks to identify *agreement*, rather than a pre-specified *difference* between groups, which is the starting point for power based sample calculations, such as those used in clinical trials. It was noted that the previous incarnations of the NAT concept, the NAT:PD-c and the NAT-HF, were validated using a sample of 50 people, a number which was not overtly justified in the subsequent publications(29, 30). In order to address this issue and ensure that the current study design was robust, advice was sought from a leading statistician (Prof. J. Martin Bland). With his kind assistance computer simulations were created to estimate the population size required to obtain significant results with an analysis of this type. Using the statistical package STATA, simulated datasets were created, based on a population with the characteristics described below. These simulated datasets allowed a series of analyses to be conducted (repeated 1000 times), which in turn were used to estimate the power to detect a significant correlation, where one existed, for a given sample size.

The simulations were performed for the physical domain of the NAT, as it was felt to be the domain where the population characteristics, illustrated below, could be estimated with the greatest precision.

To generate the simulations we first had to estimate the likely distribution of responses to the new NAT-Parkinson's disease, and the equivalent MDS-UPDRS scores.

This is an approximation, and relies on estimation of the following data:

- Standard Deviation (SD) of comparator tool
- Proportion of participants in each rank of the NAT
- Estimated MDS-UPDRS score for each rank of the NAT

9.11.1 *Standard deviations of comparator tool (MDS-UPDRS)*

These were published during the original validation of MDS-UPDRS(146):

Part 1 - Mean 11.5 (SD = 7.0)

Part 2 - Mean 16.0 (SD = 10.0)

9.11.2 *Estimated proportions of participants in each rank of NAT-Parkinson's*

Proportions of participants in each rank of the NAT-Parkinson's were estimated from previous validation of the same tool when adapted for Heart Failure (NAT: PD-HF)(30).

Table 17 Proportions of participants in each rank of the NAT:PD-HF(30)

	NAT rank = 0 (No Concern)	NAT rank = 1 (Some concern)	NAT rank = 2 (Significant Concern)
Domain: Patient physical	N = 26 (25.5%)	N = 57 (55.9%)	N = 19 (18.6%)
Domain: Patient ADL's	N = 43 (42.6%)	N = 45 (44.6%)	N = 13 (12.9 %)

9.11.3 *Estimated MDS-UPDRS scores for each rank of NAT-Parkinson's*

We know that MDS-UPDRS score rises in line with Hoehn and Yahr staging(163).

By estimating which Hoehn and Yahr stages will be represented in each rank (0,1,2) of the NAT-Parkinson's, we can estimate the average MDS-UPDRS score which would be expected to occur, according to rank, as demonstrated below:

Table 18 shows estimated scores for MDS-UPDRS part 1

Table 19 shows estimated scores for MDS-UPDRS part 2

Table 18:   MDS-UPDRS part 1

	MDS-UPDRS score	Estimated NAT-Parkinson's Rank	Derived MDS-UPDRS Score
H&Y Stage 1	7.42 (± 4.54)	Rank 0 *(No Concern)*	7.42
H&Y Stage 2	9.82 (± 5.72)	Rank1 *(Some Concern)*	12.28
H&Y Stage 3	14.74 (± 7.42)		
H&Y Stage 4	19.51 (± 8.15)	Rank 2 *(Significant Concern)*	18.46
H&Y Stage 5	17.40 (± 6.11)		

Table 19   MDS-UPDRS part 2

	MDS-UPDRS score	Estimated NAT-Parkinson's Rank	Derived MDS-UPDRS Score
H&Y Stage 1	8.34 (± 4.20)	Rank 0 *(No Concern)*	8.34
H&Y Stage 2	12.00 (± 7.16)	Rank 1 *(Some Concern)*	15.81
H&Y Stage 3	19.63 (± 7.49)		
H&Y Stage 4	31.57 (± 8.64)	Rank 2 *(Significant Concern)*	34.29
H&Y Stage 5	37.00 (± 2.83)		

This method is clearly dependent on several large assumptions, the largest of which is the assumption that the PD population will have a similar proportion of participants in each rank of the NAT as was the case for heart failure. The justification for this lies in the fact that both are chronic progressive diseases, which have been shown to have palliative care needs comparable to a cancer population(9, 126, 164).

The second assumption, which draws parallels between the staging system of Hoehn and Yahr, and the NAT-Parkinson's disease ranking, was felt to be more robust. It seemed highly plausible that palliative care needs would rise in line with disease progression, as represented by Hoehn and Yahr stage. It is also noted that the steepest rise in MDS-UPDRS occurs between Hoehn and Yahr stage 3 and 4. This is in keeping with the previously discussed deterioration in quality of life after stage 3(130) (see chapter 7) and it seems reasonable that this be equated with the presence of significant concerns regarding unmet palliative care needs, a NAT rank of 2.

Whilst the sample size generated by this method is undoubtedly an approximation, and based on assumptions, this is of course also the case for traditional power calculations used in trials. Following consultation with a statistician and the thesis advisory panel, this was felt to represent the best available method for estimating sample size.

Simulations based on these figures suggest a sample size of 50 participants, as used in the original NAT studies, would have a 98% power to demonstrate at least moderate correlation, where one exists.

9.12 Statistical analysis

Assessment of correlation between the NAT-Parkinson's disease and the comparator tools used Kendall's Tau-B correlation coefficient. The strength of association as assessed by Kendall's tau b ranges from -1 (all pairs disagree) to +1 (all pairs agree). In interpreting the results of Kendall's Tau B, correlations between constructs was arbitrarily evaluated as; Good (>0.6), Moderate (0.4-0.6), Fair (0.2-0.4) and poor (<0.2).

9.13 Study approvals

The study was approved by the Regional Ethics Committee (REC number: 13/YH/0006), the Research and Development board and was included in the NIHR study portfolio (Portfolio number: 12774). See appendix 12

9.14 Conclusions

This chapter has set out the methods used to assess the face, content and construct validation of the NAT: Parkinson's disease. In order to produce a study protocol that offered the correct balance between methodological rigour and participant burden, an innovative approach was required in some areas and, inevitably, some compromises were made. There was also progression, particularly around the theoretical justification of sample size, and the choice of analytical technique, from the work validating previous incarnations of the NAT screening tool, which are felt to strengthen the project as a whole.

The following chapter (chapter 10) presents the results of the NAT-Parkinson's disease validation.

Chapter 10

Testing Validity: Results

10.1 Introduction

Chapter 9 established the methods used for validity testing of the NAT: Parkinson's disease. This chapter presents the findings from this phase of the clinimetric evaluation, starting with a brief description of the changes made during face and content validation, followed by the results of construct validity testing.

10.2 Face and content validation

This was conducted as the last phase in the adaptation process (see chapter 7). Consultation with the expert panel produced a number of minor changes to the original adaptation. The most prominent example was the decision to include missed clinic appointments in the "red flag" section. Other changes involved the wording of the guidance for users, to ensure that this was clear and covered all the desired domains.

In addition, as the tool is clinician administered, these experts were also asked to comment on its face validity. This produced changes to the appearance of the tool, for example streamlining the triage section of the tool and the introduction of question stems, which were used to improve the appearance of the instrument and enhance its applicability to daily clinical practice

10.3 Construct validation

10.3.1 *Describing the study population:*

The validation study recruited 50 patients with PD and, where available and willing to contribute, their carers also completed an assessment of caregiver strain.

The sample included 26 males and 24 females, reflective of the established epidemiology of PD (ref). The patients were recruited from a movement disorders clinic, which takes patients of all ages. This was reflected in the study population, with age ranging from 59 – 89 years (59-86 for men; 60-89 for women). The overall mean age was 74 years (73 for men, 76 for women).

Hoehn and Yahr score is a well established measure of disease severity(112) and was measured in the "on state" at the time of clinic attendance. Scores range from 1 (unilateral, non-disabling disease) to 5 (chair or bed bound without assistance) with all stages represented in the study population (table 20).

The most common Hoehn and Yahr stage amongst both male and female participants was stage 2, which is similar to other studies of Parkinson's disease, but represents a point of difference from a recent study of palliative care in PD (Saleem et al), which concentrated on patients with more advanced disease (H &Y Stage >2). It is felt that the representative make up of this sample is a strength of the study, and serves to enhance the external validity of findings.

Table 20: Hoehn and Yahr stage by gender

		Male	Female
	Stage 1	3	3
	Stage 2	11	11
H+Y	Stage 3	10	5
stage	Stage 4	2	2
	Stage 5	0	1

A little over half the population (56% n=28) had a carer who was willing to take part in the study. Whilst males were more likely to have a carer (65% n= 17) than females (46% n=11) this difference was not statistically significant (p = 0.16).

10.4 Results

The tables below display the Kendall's tau B results for each section of the NAT: Parkinson's disease in turn.

Table 21 – Construct validity results for NAT section 2: Patient wellbeing

NAT: Parkinson's Construct	MDSUPDR part 1	MDS-UPDRS Part 2	PDQ Mobility	PDQ ADL	PDQ Emotion	PDQ Stigma	PDQ support	PDQ Discomfort	PDQ Comm.	SCOPA	m-CSI
Tau B results for NAT section 2: Patient wellbeing											
2.1 Patient physical	0.48		**0.51**						0.28		
2.2 Patient psychological	0.50				**0.55**		0.11				
2.3 Patient ADL		**0.62**		0.56			-0.19				
2.4 Spiritual					**0.42**	0.26				0.39	
2.6 Health beliefs							**0.26**	0.23		0.24	
Tau B results for NAT section 3: Ability to care for patient											
3.1 Carer distress	0.45		0.38								**0.63**
3.2 Carer difficulty (physical)		**0.48**							0.38		**0.48**
3.3 Carer difficulty (coping)											**0.46**
3.5 Carer difficulty inter-personal							0.20			0.30	**0.32**
Tau B results for NAT section 4: Carer / family wellbeing											
4.1 Carer/family wellbeing	0.34										**0.36**
4.2 Grief											**0.28**

Table 22 : Evidence of convergent and divergent validity

(comparator tool in brackets)

Construct	Convergent	Divergent
Patient Physical	0.51 (PDQ-Mob)	0.28 (PDQ – comm)
Patient psych.	0.55 (PDQ – Emotion)	0.11 (PDQ – support)
Patient ADL	0.62 (MDS UPDRS-2)	0.19 (PDQ-Support)
Patient Spiritual	0.42 (PDQ – Emotion)	0.26 (PDQ-Stigma)
Carer Distress	0.63 (M-CSI)	0.38 (PDQ-mob)
Carer Physical	0.48 (mCSI)	0.38 (PDQ-Comm)

10.4.1 *Discussion of results*

This section presents an interpretation of the construct validity findings.

As illustrated in table 21, there was at least fair correlation for all constructs examined. These results are considered below for each of the three sections: 1) patient wellbeing, 2) ability of carer to care for patient and 3) carer wellbeing.

10.4.2 *Patient wellbeing*

Section 2.1 of the NAT-Parkinson's disease includes concern regarding the physical impacts of both the motor and non-motor features of PD. It was necessary to use two comparator scales - PDQ-39 mobility (motor) and MDS-UPDRS 1 (non-motor) in order to adequately cover this construct. Therefore, while we would expect some correlation with both comparator scales, it would be very unlikely for either scale to reach very high levels of correlation.

The results (21) demonstrate moderately good correlation (Tau B range 0.4 – 0.6) with both comparator scales, supporting the assertion that concerns regarding unmet

palliative care needs in the physical domain, are related to the presence of motor *and* non-motor symptoms.

The validity of this construct is also supported by the presence of convergent and divergent patterns (table 21), meaning that the construct correlates well with the predicted comparator scales - PDQ mobility (0.51) and MDS-UPDRS 1 (0.48) and poorly with a scale which where no correlation was anticipated – PDQ communication (0.28).

In conclusion, the presence of a moderately high tau B statistic, and the demonstration of both convergence and divergence, offers good evidence for the overall validity of this construct.

A similar issue was apparent regarding the unmet psychological and neuropsychiatric need of patients; no single comparator scale would comprehensively cover the whole construct. However, correlation with both scales reached the upper end of moderate agreement – PDQ emotion (0.55) and MDS-UPDRS (0.50) supporting the validity of this construct of the NAT: Parkinson's disease.

Further evidence is found in the low correlation to the unrelated scale PDQ social support (0.11), demonstrating divergent validity.

Good levels of correlation were expected between the construct addressing patient ability to perform ADLs and both the PDQ-39 ADL scale and the MDS-UPDRS-2, which includes detailed questions about eating, dressing, hygiene and functional mobility. These expectations were borne out in the analysis, with correlation either high (0.62 MDS-UPDRS 2) or at the upper end of moderate (0.56 PDQ-39 ADL) for both.

Once again the good evidence for validity of this construct is supported by divergence with the unrelated domain, PDQ support (-0.19).

Elements of the spiritual and existential construct are found within the SCOPA-PS (0.39) and the PDQ emotion (0.42) sub-scale. The results demonstrate moderate levels of correlation with the PDQ-emotion (0.42) and fair correlation, just failing to reach moderate, with SCOPA-PS (0.39). The presence of a divergent relationship with the unrelated PDQ-stigma sub-scale also provides evidence in support of the validity of this construct.

The final sub-section for the domain relating to health beliefs is quite wide ranging, covering individual beliefs, as well as the cultural and social aspects of care. The primary comparator scale included at study outset to assess the validity of this section was SCOPA PS in addition to the PDQ social support scale. In actual fact, neither of these scales demonstrated even moderate correlation (Tau B 0.24 and 0.26 respectively).

This finding is disappointing and probably explained by the breadth of the construct being examined by NAT section 2.5. As a result the comparator tools chosen either included additional unrelated areas, or did not cover the breadth of the construct, both of which could explain the low levels of correlation. For example the SCOPA-PS includes some questions on existential issues *"During the past month, have you been concerned about the future?"* and activities of daily living *"…have you had difficulty with work, household or other chores?"* which are covered elsewhere in the NAT. While the PDQ social support sub-scale does not cover aspects of this construct such as attitudes to information. As such, it can be argued that the disappointing result is due to a failure to identify comparator scales, which measured all components of this multi-faceted question

10.4.3 *Ability of carer to care for patient*

As discussed in chapters 4 and 6 carer strain is an important feature of Parkinson's disease and therefore the detection of unmet needs in relation to carer distress is a key component of the NAT-Parkinson's disease. The validity of this sub-section is supported by the demonstration of a good correlation with the modified Caregiver Strain Index (mCSI) (0.63). There is a weaker correlation with PDQ mobility (0.38), a scale where a correlation was not anticipated, but where some correlation does make sense in the context that the physical effort of care is likely to be higher if the patient is less mobile.

This is also consistent with the anticipated finding, that levels of concern regarding difficulty providing physical care were moderately correlated with both the m-CSI (0.48) and the MDS-UPDRS 2 (0.48) - which measures physical dependence and difficulty with ADL's.

Given the breadth of the construct examined by NAT 3.3-carer difficulty coping, the m-CSI was felt to be the scale of choice, whilst acknowledging the imperfect match of

comparator scale to construct. The findings displayed in table 1 support the construct validity of this construct, demonstrating a moderate level of correlation between this item and the m-CSI.

At the outset of the project, the intention was to use the comparator scales m-CSI and SCOPA-PS to assess the correlation for problems with inter-personal relationships (NAT 3.5). Whilst the findings demonstrated a fair level of correlation for these scales (0.32 and 0.30 respectively), they failed to demonstrate at least moderate correlation, indicating that the support for construct validity is weaker than anticipated.

This could be because the question is not valid to assess the construct or, alternatively, that the breadth of the construct means that the chosen comparator tools could not cover it sufficiently well. As such, whilst better correlation may have been hoped for, it may be that the incomplete matching of study and comparator scale account for this result and that use of alternative scales would have yielded better results. This is something which could be explored in future research projects.

There may also be a more fundamental reason for the lower level of agreement. The assessments were made in the context of the *patient's* clinic appointment where the carer may not feel able to fully, if at all, express their concerns, perceiving the focus of the consultation to be about the patient and not them(165, 166). Also it is inevitable that some assessments of carer need, where the carer was not present at interview, were made using the patient as a proxy.

Overall, the findings displayed in table 21 support the construct validity of section 3, assessing *Carer's ability to care*, but the poorer correlation than anticipated for some patient constructs highlights the importance of a specific assessment of carer needs, in the context where there is clear permission for carers to express their needs, and not just the patient's.

10.4.4 *Carer and family wellbeing / Grief*

Both elements, 4.1-*carer and family wellbeing* (0.36) and 4.2-*grief (0.28)*, demonstrated fair levels of correlation which does offer some support for their validity, all be it weaker than for other constructs. Once again, the results in this area may be explained by the

fact that the m-CSI assesses other constructs, in addition to those discussed here, such that levels of correlation are inevitably lower.

10.5 Conclusions

The NAT-Parkinson's disease has been developed as a short screening tool, to identify unmet palliative care needs in people living with PD. It asks clinicians to rate their level of concern for 3 domains (patient wellbeing, ability of carer to care for patient, carer and family wellbeing) which between them contain 13 individual questions. Each question is a construct in its own right, and indicates levels of concern relating to that area of the disease. The tool is not intended as a palliative rating scale and so no attempt is made to produce a summary score.

Construct validity has been examined for 11 of the 13 constructs in turn (patient and carer financial concern not examined), using existing PD rating scales wherever possible. One of the main difficulties in assessing construct validity, where there is no gold standard measure to use for comparison, is selecting comparator scales which are as closely matched to the construct under examination as possible. Failure to do this may lead to a situation where the principle scale (in this case the NAT-Parkinson's disease) and the comparator scale are in fact measuring two different, related constructs. The corollary of this being that measures of correlation are likely to be lower and, in some instances, that low correlation may not truly indicate an invalid construct, but rather a failure to demonstrate construct validity as a result of mismatch between scales.

The analysis of construct validity for the NAT-Parkinson's disease reveals several sections where this mismatching of primary to comparator scale may have been a problem and this is particularly the case for sections that assess broad constructs.

These results must also be interpreted in the light of the project aims and the nature of the constructs examined. The stated intention of the NAT: Parkinson's disease is to identify people with potentially unmet palliative needs, with a tool that can be used in everyday clinical practice, with minimal training, with the expectation that these people may need further, in depth specialist palliative assessment. This inevitably results in the use of broad constructs to represent palliative need. In the absence of a criterion

measure, this approach is expected to produce lower levels of correlation than narrow, highly defined constructs, where comparator tools could be closely matched. This represented a balance between demonstrable clinimetric properties and every day clinical application, as the use of narrow constructs would have produced a highly detailed, unwieldy tool, which would not fit the objectives of the project.

Although there is positive correlation with carer constructs, these are at a lower level than for patient constructs. Given that the carer may not perceive it to be legitimate to raise their own concerns within the context of the patient's clinic visit, or may be constrained by the presence of the patient, then this highlights the need for carers to have their needs explicitly and specifically addressed.

Overall, these results support the valid use of the NAT: Parkinson's disease in a mixed population of PD patients. It is important to note that the ratings were conducted by a single clinician, therefore, in order to justify its use by a variety of raters, which would clearly be necessary in clinical practice, it is necessary to ascertain the inter-rater reliability of the instrument. This is examined in the following chapter (chapter 11).

Chapter 11

Testing Reliability: Methods

11.1 Introduction

The earlier chapter on methodologies (chapter 8) outlined some of the theoretical issues which must be considered when examining the reliability of a new clinical measure. In particular, it highlighted the difference between test re-test and inter-rater reliability, in terms of the origins of the variance which they measure. This is relevant to the decision taken here, to explore inter-rater reliability preferentially (see section 8.3.3). It also discussed the assertion that, in order for a tool to be clinically useful, it should have demonstrated acceptable levels of both validity and reliability, neither in isolation being good enough. With this in mind, having set out the data supporting construct validity of the NAT-Parkinson's (chapter 10), the focus will now switch to the exploration of inter rater reliability.

11.2 Describing the study population

The purpose of this phase of the study was to assess the performance of the NAT-Parkinson's disease, in a broad range of PD clinicians, reflecting those who are likely to use the tool in clinical practice. As such, clinicians were eligible for inclusion if they worked in one of the following disciplines; Neurology, Elderly Medicine and Palliative Medicine, as either a specialist nurses, or as a doctor with specialty training (consultant and specialist registrar level).

11.3 Study design

In the discussions of methodology (chapter 8), some of the chief difficulties with assessment of inter-rater reliability were identified, namely the occurrence of change in the subject over time (between ratings) and "priming", particularly in this case, of the

patient being assessed. Steps were taken to address these in the design for the reliability study.

11.3.1 Practical considerations

From a practical viewpoint it would be ideal to have a study design which allowed multiple clinicians to assess the same individual concurrently, so that we could be certain that they are all assessing the patient in the same state. For many practical reasons this seems almost impossible to achieve in a real life clinical setting. Therefore most study designs will require repeated assessments to be sequential, rather than concurrent and, as such, will encounter the theoretical problems of priming and change in clinical state. In addition, the use of sequential assessments raises significant practical concerns, not least because of the participant burden incurred by multiple separate assessments. It seems inevitable that studies based on sequential measurements will increase participant burden, as a result of longer appointments, or repeated follow up visits. This is particularly pertinent in PD palliative care assessments, where participants are likely to have physical and cognitive impairments, and where each assessment is likely to last 20 - 30 minutes.

Achieving a balance between the desire to produce the most robust data possible, the consideration of participant burden, and practicalities of data collection in a clinical environment, became the key determinant of study design.

11.3.2 Previous reliability testing of NAT-PD-C / NAT-PD-HF

In deciding on the best design for reliability testing of the NAT-Parkinson's disease, it was useful to review the techniques used in clinimetric testing of previous versions of the NAT, designed for cancer and heart failure.

11.4 NAT-PD-C

The original reliability testing of the NAT in cancer (NAT-PD-C) was designed explicitly to reduce participant burden, using videotaped consultations, with actors playing the roles of patient and carer(123). Each of the three videos was based on a different clinical

scenario, designed to bring out a variety of palliative issues. 103 clinical staff watched these 3 simulated consultations and completed the NAT for each one.

This technique not only reduced participant burden, but also avoided the potential bias introduced by changes in a patients' clinical status over time. In addition it nullifies the effect of priming in relation to the patient / carer, as they are only interviewed once, although this may still be an issue for the assessors.

The criticism of this method lies primarily in the artificial nature of the process. The inability for the assessing clinician to influence the consultation, and the use of actors rather than real patients, means that the outcomes of the NAT are unlikely to reflect real life.

It a sense, this technique examines a very pure form of reliability – the amount of variation between assessors, when viewing the exact same consultation – without accounting for the subtleties individual clinicians and patients will add to the consultation, in "real life" which may alter the assessment.

This point is acknowledged by the authors, and cited as one reason that levels of agreement were poor for some of the more complex, person centered domains, such as spirituality.

Later analysis of the NAT-PD-c and of the subsequently developed NAT: PD-HF, an adaptation of the tool for heart failure, used real life clinical consultations to assess reliability and validity(29, 30). In this instance two independent clinicians interviewed patients and carers, separately, on the same day. While several clinicians contributed to the study as a whole, no more than two assessed any given patient.

Applying the discussions above, we see that this method inevitably incurs greater participant burden. The researchers tried to minimise this by completing both assessments on the same visit, which also avoided the possibility of clinical change over time. This method also had the significant benefit of being embedded in clinical practice and thus reflecting the nuances of individual consultation style.

The potential criticism is based in the fact that each patient is only assessed by two different clinicians. Thus while there is "proof of principle" that two separate people with training in the tool can agree, there will be less variation between raters than would be encountered in clinical practice.

11.4.1 Study design for the NAT-Parkinson's disease

It was felt that sequential assessments would not be reasonable in the PD population. This was due to the unique characteristic of PD, as well as the general consideration of time. For example, the administration of medications follows a strict schedule in PD, particularly for those with later stage disease, where motor fluctuations due to changing dopamine levels, can be marked(119). This would not only cause distress to participants, it would also potentially lead to highly significant change in clinical state, even between back to back interviews. Thus the decision was taken to use video consultations when examining inter-rater reliability, which also avoided the problem of priming, with regard to patient participants.

Videos were made, with appropriate consent, during the clinical consultations conducted for the validity phase of the study (see chapter 9-10). This meant that a range of videos were available, some involving patients alone and others the patient – carer dyad. Ten videos were selected for use in the reliability phase, to demonstrate a range of clinical stages, age, gender and presence of carer; thus maximizing the variation within subjects, to enhance external validity.

11.4.2 Characteristics of video consultations

The video consultations were made during the validation phase of the study, by a single rater, in the "on" state wherever possible and reflected the full spectrum of PD severity, from very early disease (the participant in video 9 had been diagnosed within the last 3 months and had Hoehn and Yahr stage 1 disease) to late stage disease (Participant in video 4 was wheelchair bound – Hoehn and Yahr stage 5 and participant in video 6 was awaiting deep brain stimulation). Participants in the video consultations were all able to consent to participation and, as such, are unlikely to have had significant cognitive impairment, although this was not formally measured. The videos were recordings of

actual doctor – patient interaction as part of a routine clinic and as such were not scripted in any way. They ranged in length from 15-25 minutes.

11.4.3 Study population size

The number of video consultations and subsequent clinical assessments required was based on expert opinion following consultation with Professor Bland.

It was important to have a sufficient number of video consultations, to show a range of unmet palliative care need and, a sufficient number of ratings to provide a range of assessor responses, for each video.

The original clinimetric testing of the NAT:PD-C, a version of the NAT developed for use in cancer, used only 3 video consultations, a factor which was acknowledged in the paper to have limited the subsequent analysis(123). We estimated that 10 videos would be required to represent a sufficient range of unmet need and 10 assessments to provide sufficient variety of response.

Importantly, from a statistical standpoint, it is not necessary, or desirable, for each assessor to assess every video, rather, that a range of video consultations be assessed by a range of clinicians. The statistical test used, an extension to Fleiss' kappa, does not consider the identity of the rater(167) – see below.

11.4.4 Characteristics of raters

In order to achieve 100 separate video assessments 34 raters were recruited, in addition to the use of 8 primary assessments from the validation study. Each rater assessed an average of 2.9 videos (range 2-4) at a single sitting, to minimise change over time.

A broad range of participants was sought, from within the frame of clinicians likely to be involved often in the care of PD, to reflect the range of potential users by clinical background, specialty and experience. The 2006 NICE guidelines(6) indicate that patients with PD should be diagnosed and followed up in secondary care and it is therefore assumed that users of the NAT-Parkinson's disease will be drawn primarily from this group, this was reflected in the recruitment of raters. The clinical groups chosen for reliability testing were therefore; i) Elderly Medicine at consultant and senior registrar

grades, ii) Neurology, at consultant and senior registrar grades, iii) PD Nurse specialists, iv) Palliative care specialists, including consultants, senior registrars and palliative neurology nurse specialists.

11.4.5 Recruitment

Participants were initially approached either by e-mail, or verbally, to gauge their interest in participation. If they expressed an interest they were provided with a copy of the Participant Information Leaflet (PIL) and given the opportunity to discuss the study and ask questions. Those who remained interested in participation had a time booked, at their convenience, to collect informed consent and conduct the data collection. These two procedures were usually conducted at a single visit, in accordance with the preference of the participants.

11.4.6 Data collection

In order to reduce the burden of participation on clinicians and, thus, to enhance recruitment, videos were stored securely on the university hard drive, which could be accessed remotely via a secure, encrypted portal. The primary investigator (ER) was responsible for visiting participants at their convenience, conducting a short training session on the use of the NAT-Parkinson's (5minutes) and providing access to the videos, which were password protected. In order to protect sensitive data, participants were not given unsupervised access to the videos and downloading was not permitted.

Data collection was generally conducted individually, or in small groups of 2 to 3, with participants separated to avoid contamination of responses. The exception to this was a large group of participants, based at an educational training day (16 participants leading to 32 assessments).   In this case the session was divided in to two groups of 8 participants, with each group was shown two different videos and the sessions run concurrently, under exam conditions, meaning that participants were unable to exchange views about the outcomes during data collection.

11.4.7 Randomisation

It is important to emphasis that the statistical method used to analyse agreement between rates, does not consider the identity of the raters, only that they are independent. Therefore, it was not necessary to randomly assign participants to the different videos.

11.4.8 Statistical analysis

Kappa is a statistic used to measure the level of agreement between subjects, above that which would be expected by chance. The original version, devised by Cohen and referred to eponymously as Cohen's kappa, measures the agreement between pairs(168). It does not however, allow for partial agreement. This could occur with a tool such as the NAT-Parkinson's, as in the following example:

Say 3 separate assessors, A, B and C, gave scores of 0 (no concern), 1 (some concern) and 2 (significant concern) respectively for a particular domain.

None of them agree completely.

However, the amount of disagreement between rater A and B; who are separated by one category of concern, is clearly less than that between A and C, who are separated by 2 categories of concern.

In order to account for this difference, we may use a weighted kappa, which applies a weighting to the difference between categories of scaled (ordinal) data. The weighting must be expressly stated and may either be determined by expert opinion, or more conventionally applied in either linear, or quadratic manner(169).

The other significant evolution of kappa has been to allow the assessment of multiple raters, rather than pairs. This is referred to as Fleiss' method(167).

In the reliability testing for the original NAT:PD-c, the researchers assessed pairs of raters and dichotomised the outcomes from the tool, in order to use an unweighted kappa. Thus each pair of raters was assigned a value of 1 (agree) or 0 (disagree) and a Kappa statistic calculated on this basis. For an outcome score that ranks each domain in 3

categories (*0 – no concern, 1 – some concern, 2 – significant concern*) this appears to have a significant impact, because it is unable to weight the responses. For example if one assessor scored a question as *1 – some concern*, while the other scored it *2 – significant concern*, the above method would record "disagree" for this pair of raters. It is thus unable to account for partial agreement where this exists.

With this in mind, the ideal analytical technique for the NAT-Parkinson's reliability data, would allow the use of a weighted kappa, in multiple raters. Therefore an adaptation of Fleiss' kappa, which allows weighted kappa to be used in multiple raters was used [adaptation by personal communication, Prof Martin Bland(170)]. The interpretation of kappa and the implication for study findings, is discussed in the chapter 12 (section 12.5).

The raw data was used to construct a separate table for each sub-section of the NAT-Parkinson's, displaying the 10 separate ratings of that sub-section, for each of the 10 videos. This allowed the weighted kappa for each subsection to be calculated, taking in to account the assessments of all videos, rather than for each video individually.

11.4.8.1 *Selecting the weighting of kappa*

Weighted Kappa is used to reflect the fact that for ordinal data, where the order of categories is significant, the difference in agreement between raters selecting categories one step apart (say category 1 and category 2), is smaller than the difference in selecting categories 2 or more steps apart (say category 1 and category 3). To acknowledge this difference, a weighting is added to the difference between categories, allowing us to credit a partial agreement, so that, for example, a score of 1 is given for raters choosing the same category, 0.5 for categories 1 step apart and 0 for scores 2 steps apart. This is a simple example of linear weighting, where the difference between categories is attributed the same weight (0.5).

Although it was initially suggested that weighting should be determined by a panel of experts(171), this is difficult to achieve in practice. Rather, the choice is usually between linear weighting (where the weighting is proportional to the number of categories apart, see above) and quadratic weighting (where the weighting is proportional to the square of the number of categories apart)(169).

In the case of the NAT Parkinson's disease, a decision was taken in consultation with a statistician and the thesis assessment panel, that a linear weighting would be the most appropriate choice. In actual fact, the impact of weighting was relatively low (see chapter 12 results table 23) because it was more common for raters to be within one score, with ratings at the extremes being rare.

11.4.9 *Addressing missing data*

The NAT-Parkinson's disease has three main domains, one relating to the patient wellbeing, one to family wellbeing and one to the ability of the carer to care for the patient. Equally, we know that a significant minority of people living with PD will not have an informal carer, to whom section 3, "ability of the carer to care for the patient", will not apply. During data collection it became clear that raters approached this problem in different ways. Some recorded not applicable (n/a) in section 3, while others left it blank, or scored 0 *"no concerns"*. In order to have a uniform method of recording data and, to ensure that those cases where there was no carer were distinguished from true missing data (where raters failed to enter a score in error), data was recorded in the following ways:

Where the patient in the video did not have an informal carer – In this case, it was felt that the absence of an informal carer, which would be recorded in section 1b "priority referral for further assessment", made section 3 "inactive". Therefore, where the rater had entered *0 "no concerns"*, or left the entire section blank, this was recorded in the data set as Not Applicable (N/A). In one case a participant had recorded a score in a single question of section 3, leaving the rest blank. In this case there was absolutely no question about the presence of a carer and it was felt that this represented an error – supported by the fact that the rest of the section as blank. This data point was therefore recorded as an error.

Where the patient in the video did have an informal carer – in this case, the presence of an informal carer made section 3 "active". Entries of *0 "no concern"*, N/A, or where the entire section was left blank, were recorded in the data set as 0, indicating that despite the presence of a carer the rater had not raised concerns in these areas.

Where a single sub-category was blank – in the cases (two) where part of the section had been completed, with other parts left blank, the participant was assumed to have made a transcription error and the blanks responses were recorded as *missing data*.

These changes tended to reduce, marginally, the value of kappa in the final analysis, compared to a preliminary analysis where all N/A and blank scores were recorded as 0, to indicate no concern registered.

These decisions were arrived at by consensus, following discussion with other members of the research team and are of particular relevance as there are currently two other NAT adaptation projects underway (assessing the NAT format in primary care and interstitial lung disease) so it was important to establish a consistent approach to these issues.

11.5 Study approvals

The study was approved by the Regional Ethics Committee (REC number: 13/YH/0006), the Research and Development board and was included in the NIHR study portfolio (Portfolio number: 12774). See appendix 12

11.6 Conclusions

In examining the reliability of the NAT-Parkinson's disease, the methods chosen reflect a balance between the theory of analysis (chapter 8) and the reality of clinical testing in a vulnerable group of participants. Pragmatic choices were taken to limit, wherever possible, the burden on participants, while maintaining scientific rigor. Examples of this, include the decisions to examine inter-rater reliability in isolation and the use of video consultations.

The results derived from this work are set out in the final chapter of this section (Chapter 12).

<center>**Chapter 12**</center>

<center>**Testing Reliability: Results**</center>

12.1 Introduction

Inter-rater reliability was the second clinimetric feature of the NAT Parkinson's disease to be examined and is of great importance to the practical application of the new tool. While the construct validity testing (see chapter 10) demonstrated that the tool could perform well in the hands of a single, experienced rater, this is of little practical use if the results cannot be replicated across a range of raters, with different clinical backgrounds. Therefore, the purpose of examining inter-rater reliability was to establish the usefulness of the tool, outside of the confines of the study. The findings are presented in this chapter.

12.2 *Kappa for inter-rater reliability*

The tables below contain the unweighted and weighted kappa for inter-rater reliability, displayed by NAT sub-section.

Green denotes a sub-category where the demonstrated inter-rater reliability was "good" (kappa >0.60), Orange denotes "moderate" (kappa 0.40 – 0.59) and blue "fair" (Kappa 0.20-0.39).

Table 23 illustrating kappa by NAT-Parkinson's disease domain

NAT Section	Proportion of observations equal to:			Unweighted		Weighted	
	0	1	2	kappa	SE	kappa	SE
NAT 2.1 Physical	2%	49%	49%	0.39	0.05	**0.39**	0.05
NAT 2.2 Psychological	28%	54%	18%	0.22	0.06	**0.33**	0.06
NAT 2.3 ADL	34%	52%	14%	0.21	0.06	**0.27**	0.06
NAT 2.4 Spiritual	56%	38%	6%	0.16	0.06	**0.21**	0.06
NAT 2.5 Financial	89%	6%	5%	0.48	0.26	**0.54**	0.26
NAT 2.6 cultural	70%	29%	1%	0.11	0.10	**0.11**	0.10
NAT 3.1 Carer distress	59%	37%	4%	0.51	0.81	**0.55**	0.81
NAT 3.2 Carer physical	66%	27%	6%	0.39	0.12	**0.45**	0.12
NAT 3.3 Coping	67%	33%	0%	0.40	0.10	**0.40**	0.10
NAT 3.4 Carer financial	96%	4%	0%	0.03	0.54	**0.03**	0.54
NAT 3.5 Interpersonal	66%	33%	1%	0.18	0.11	**0.19**	0.11
NAT 4.1 Carer wellbeing	74%	26%	0%	0.28	0.12	**0.28**	0.12
NAT 4.2 Grief	92.75%	7.25%	0%	−0.01	0.34	**−0.01**	0.34

12.3 *Discussion of results: Inter-rater reliability*

There are 13 sub-sections, contained within the three domains relating to patient wellbeing, family wellbeing and ability of carer to care for the patient. The results of the inter-rater reliability testing suggest that 10 of these sub-sections demonstrate either moderate or fair reliability, which supports their use by a variety of clinicians in everyday clinical practice.

Two of the remaining sections demonstrate very low kappa values, which at first glance, would appear to suggest that they are not sufficiently reliable to be used in clinical practice. Interestingly though, this is explained by the fact that when there is almost no spread of data points (i.e. the vast majority of raters record the same score) the kappa value will be low. This appears paradoxical, given that, all data points in the same category would surely represent perfect agreement? It can be explained by the fact that kappa is determined by the amount of agreement after accounting for that expected by chance; as opposed to the percentage agreement between raters.

12.3.1 *Percentage agreement vs kappa agreement*

Percentage agreement can be simply calculated by looking at the proportion of scores in each category (0,1 or 2). Taking the example of section 3.4 *"carer financial concerns"* (kappa of 0.03) we see that the percentage agreement is very high at 96%, with all but 4% of ratings being in the category "0".

However, kappa seeks to estimate the proportion of agreement present that would not be expected by chance:

"……the amount by which the observed agreement exceeds that expected by chance alone, divided by the maximum which this difference could be" (172)

As such kappa is *always* less than proportional or percentage agreement.

The difficulties arise at the extremes, because the kappa is calculated using the probability of a correct answer and is at its greatest when that probability is 0.5. At either extreme the kappa falls, unless agreement is perfect. This is illustrated by the Figure 14 below.
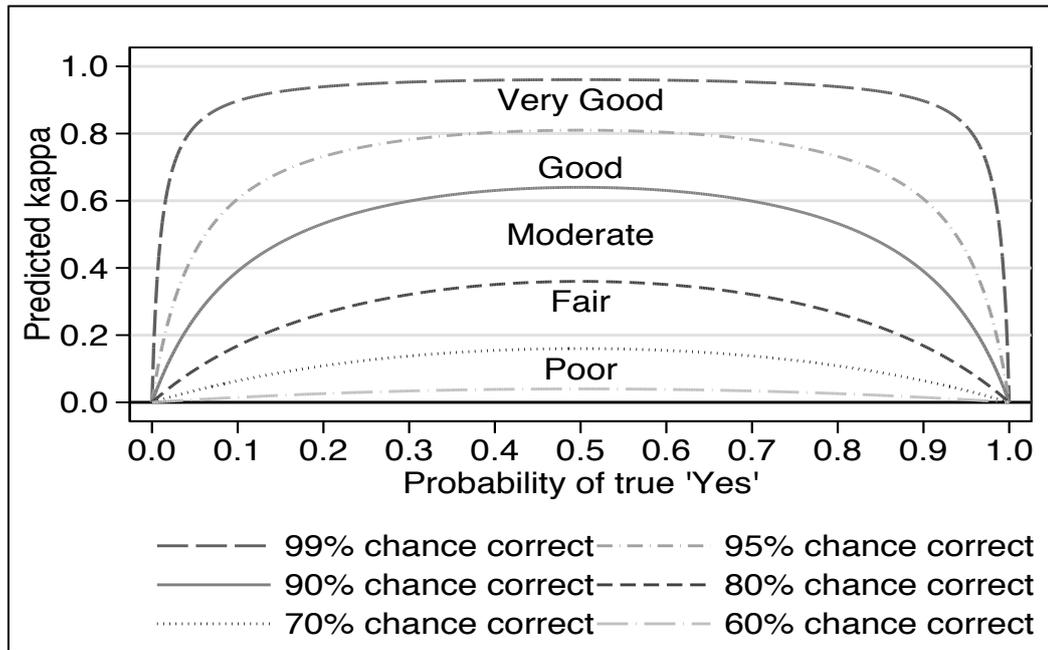
Figure 14 - Predicted kappa for two categories, 'yes' and 'no', by probability of a 'yes' and probability observer will be correct. (From Bland (169))

This principle can be applied to the raw data from the present study, to illustrate why, in some cases, it would be impossible to obtain a high kappa value, despite apparently high agreement.

The table 24, below, shows the scores given for the NAT category 3.4 *"financial concerns"*, by raters 2 to10, against those of rater 1. There are 10 raters (columns) and 10 videos (rows). It is clear that, in terms of proportion, there is almost perfect agreement and the probability of a correct answer (blue) is extremely high. This would sit to the far right of the curve in Bland's graph (above), and makes it impossible to achieve a high kappa value.

Table 24 - Showing scores for NAT category 3.4

Displaying raters 2-10 against rater 1

Correct scores = Blue, Incorrect scores = Red

Rater Number									
1	**2**	**3**	**4**	**5**	**6**	**7**	**8**	**9**	**10**
0	0	0	0	0	0	0	0	0	0
0	1	0	0	0	0	0	0	0	0
0	0	0	0	0	0	0	0	0	0
0	0	0	0	0	0	1	0	0	1
0	0	0	0	0	0	0	0	0	0
0	0	0	0	0	0	0	0	0	0
0	0	0	0	0	0	0	0	0	0
0	0	0	0	0	0	0	0	0	0
0	0	0	0	0	0	0	0	0	0
0	0	0	0	0	0	0	0	0	0

The extremely low kappa values for section 3.4 (percentage agreement 96%) and 4.1 (percentage agreement 93%), can both be explained by this feature of Kappa, where, despite almost all ratings being in the same category (agreeing) the kappa value is very poor. This is something of a paradox and, at such extremes kappa becomes a poor measure of agreement(169).

The presence of a "floor effect"

The high number of "0 – no concern" responses for the constructs related to grief (NAT 4.2) and finance (NAT 3.4) appear to represent a floor effect. That is a clustering of

responses at the low end of a scale, resulting in a positive skew to the data set. (173) In psychological testing this is said to occur when a test or question is too difficult, resulting in a clustering of responses at the low end of a scale with the consequence that the test struggles to distinguish between them. The question thus has poor discriminatory value(174). In the case of the NAT-Parkinson's disease, where the clinicians were responding to the video consultations, there appear to be a number of possible explanations. Firstly, it may be that the 10 videos simply failed to display sufficient variation of need relating to these constructs and hence responses were clustered around zero. Alternatively, it may be that many responders had insufficient experience in assessing needs relating to these areas, particularly grief, and hence the question was in effect "too hard". The implications of this observation are discussed in chapter 13.

12.4 Discussion of NAT 2.6 and NAT 3.5

Section 2.6, addressing concerns relating to health beliefs, cultural and social factors making care more complex, and section 3.5 – difficulty with interpersonal relationships, have a kappa of 0.11 and 0.19 respectively, suggesting little agreement between raters. Some of this result may be attributable to the phenomenon described above, however, while approximately two thirds of responses score "0" in both data sets, the skew in distribution is not as extreme as those categories discussed above (sections 3.4 and 4.2) and, over-all, is not dissimilar to other categories with fair or moderate kappa values. As such, high pre-test expected agreement is unlikely to be solely responsible for these low kappa statistics.

One alternative explanation here is the wide variety of clinicians included in the study. The participants in this phase of the study ranged from Neurologists, to Geriatricians, Nurse specialists and specialist Palliative care clinicians. As such, the level of experience and knowledge in assessment of palliative needs would have varied widely. It seems plausible that these differences would have been less accentuated in the categories covering physical, neuro-psychiatric and ADL based needs, where the "core" medical training and specialist PD knowledge common to all participants would have been sufficient to assess need. However, in the more esoteric categories; covering emotional, spiritual, social and cultural components of unmet need, which are far more familiar to

those with a palliative flavour to their training, is seems reasonable to suggest that the differences between assessors would have been exaggerated.

It would be interesting to test this hypothesis by re-running these elements of the study, using only clinicians from the same specialist background, to see whether agreement is higher.

Alternatively, this result could be viewed as highlighting a training need. The NAT-Parkinson's disease is designed for use as part of an integrated palliative care model, involving clinicians from all of the specialties involved in the reliability testing. Such a model is likely to rely on "up-skilling" of clinicians from each specialty, so that, for example, the specialist PD practitioners (neurology and elderly medicine) become sufficiently familiar with palliative medicine to be able to practice with a "palliative approach", while palliative care specialists enhance their neurological skills in order to be familiar with the complex management of advanced PD. It is possible to speculate then, that examining inter-rater reliability within an established integrated team, is likely to yield much better levels of agreement and, moreover, to be a better representation of clinical practice.

12.5 Interpretation of kappa scores

The fact that kappa takes in to account the amount of agreement expected by chance is important in interpreting kappa scores. In particular the realisation that a low kappa score, say the 0.19 recorded for NAT section 3.5, is still demonstrating the presence of more agreement than would be expected by chance alone. Therefore some commentators have interpreted scores between 0.00 – 0.20 as *"slight agreement"* and only scores less than 0 as poor See table 25 adapted from Bland(169)).   Whilst others would advance a slightly more stringent interpretation the cut off points used for fair (0.21-0.4), moderate (0.41-0.6) and good/substantial (0.61-0.8) agreement are consistent(169).

Table 25 Landis and Kock interpretation of Kappa adapted from Bland

Value of Kappa	Interpretation: Landis and Kock
< 0.0	Poor
0.0 – 0.2	Slight
0.21-0.4	Fair
0.41 – 0.6	Moderate
0.61-0.8	Substantial
0.81-1.0	Almost perfect

12.6 Conclusions

The majority of NAT sections have demonstrated either fair or moderate agreement as measured by a weighted kappa statistic. In 2 instances where kappa failed to produce fair or moderate agreement, the percentage agreement was extremely high, producing a paradoxically low kappa statistic as discussed above. Satisfactory reliability was not demonstrated for two categories of the tool. These both relate to clinical areas that are far more familiar to palliative specialists (NAT 2.6 - cultural concerns relating to patient wellbeing, NAT 3.5 – interpersonal concerns relating to carers ability to care for patient) than either neurology or elderly medicine. There may also be some variability due to the inherent difficulty of assessing carer needs by proxy, where carers were not present for the video consultation.

These domains capture some of the most complex aspects of palliative disease experience and their inclusion in a PD palliative screening tool is therefore considered to be very important. It is likely that the poor inter-rater reliability results arise from the variability of clinical experience between raters in this study. Examining this clinimetric property within an integrated palliative care system, where skill sharing should improve palliative competencies in non-specialists, may yield better results and be an improved on the current study design. This should be considered in future studies.

The last 5 chapters have described the quantitative phase of this sequential mixed methods project, the clinimetric testing of the NAT: Parkinson's disease, and have set out the case for its construct validity and inter-rater reliability. In the final chapter (chapter 13) the qualitative and quantitative phases of the project will be drawn together, identifying the areas where understanding has been advanced, discussing clinical implications, strengths and limitations and finally areas for future research.

Chapter 13

Discussion of findings

13.1 Introduction

This thesis began by identifying a gap in existing practice; the lack of a practical, bedside method for identification and triage of unmet palliative and supportive care need in Parkinson's disease. The objectives of the thesis were to:

1- Adapt the NAT:PD-c for use in Parkinson's disease, using the knowledge gained from i) systematic review and synthesis of qualitative literature and ii) qualitative study exploring supportive and palliative care need in PD.

2- Assess the clinimetric properties of the adapted tool in a diverse population of PD patients and clinicians, specifically examining i) construct validity and ii) inter-rater reliability.

These research objectives produced the following research questions:

1- *What are the supportive and palliative care needs of people living with Parkinson's disease?*

2- *What are the clinimetric properties of the NAT:Parkinson's disease?*

In order to answer these two separate questions the study adopted a sequential mixed methods approach, where the qualitative phase was conducted first, informing the adaptation process and being followed by the second phase; clinimetric testing. This discussion chapter will draw together the two phases of the study, summarise and synthesis the findings from each, and describe their clinical applications. As a guide to the chapter, the key findings of the thesis can be found in Appendix 13, which provides a précis of the findings from each method with a synthesis to demonstrate how, together, they address the research questions identified at the outset of the project.

13.2 Summary findings for research question 1:

What are the supportive and palliative care needs of people living with Parkinson's disease?

The primary and secondary qualitative work demonstrated the existence of supportive and palliative care needs in people living with PD. These are many and effect all domains of life for both people with the disease, and those who care for them. The issues identified in the systematic literature review related to; the process of being diagnosed, the impacts of PD on patient and carer – particularly the care process and how this is structured, preparation for advanced disease and even post bereavement care. Even though the qualitative literature review (for references see chapter 4) found relatively few papers which presented the patient's voice, the primary qualitative work conducted to fill this gap confirmed and further developed these findings.

Palliative and supportive care needs were present from diagnosis and continued throughout the disease duration, although they fluctuated in nature and severity over this time. In particular two areas were characterised as tensions; information (*information tension*) and the need to be cared for or to provide care (*care tension*). For the individuals involved, resolving these tensions represented a series of negotiations, both

intra-personal and *inter-personal* and the results of these negotiations are described as a settlement (care settlement / information settlement). At any given time the decision reached was a result of a balance between competing forces, for example the desire to know more about the disease, versus the need to maintain hope through temporality; or the desire of the patient to remain independent, balanced against the ability of the carer to offer and administer care. As such, the information tension and care tension are never fully resolved, but rather held in a dynamic state offering the best outcomes at any one time and illustrate the crucial role of regular assessment of patient and carer and need throughout the illness.

13.2.1 *Information needs*

Patients and carers often required information. This related to service availability and practical support, to practical aspects of care and how to provide it, as well as to

biomedical information. Timely and sufficient information giving had an important role to play in helping people to live well with PD and, where absent, appeared to contribute to negative outcomes.

The information tension was particularly prominent around prognostication. This was a result of two opposite mechanisms used for the maintenance of hope; temporality and information seeking where, at their extremes, the first mechanism aims to maintain the prospect of an uncertain future, while the second tries to dispel uncertainty.

Information need and information tension are evident from the first consultation and affect the degree to which individuals seek, or avoid prognostic information. Indeed the whole diagnostic experience, not simply the fact of being diagnosed, is important, including the manner in which that diagnosis is delivered, the information offered, the early emotional response and the support available at the time to help promote understanding and adjustment. Clinical approaches which allocate sufficient time to diagnosis and allow information gathering - which could be described as patient centered approaches, contrasted with those that focus solely on delivering a factual diagnosis and this was highlighted as a key aspect of good practice. Moreover these few crucial minutes seemed to influence the reaction and subsequent response of participants to being diagnosed.

Managing this period seems all the more important when considering the way in which the emotional response to diagnosis may resonate beyond the diagnostic period. For example it's influence can be seen in the degree to which information is sought and offered, which in turn affects the ability of patient and carer to achieve the important goals discussed in the qualitative synthesis, such as maintaining independence, establishing effective care structures and planning for the future.

The context in which diagnosis occurred also appeared to influence the subsequent reaction and response. Experience in the pre-diagnostic period, particularly the recognition of motor and non-motor disease features prior to specialist assessment, appeared to alter the expectations of patient and carer going in to the consultation. This in turn could influence their reaction to diagnosis, demonstrated by the contrast between reactions of surprise, devastation and relief. Context also refers here to the people

present during the diagnostic consultation. In other areas of medicine, when delivering bad news, it would be inconceivable not to at least consider the appropriateness of having relatives, family or friends present(110, 175). In PD the diagnosis, being based on clinical evaluation, can be made and communicated at a single consultation. For people whose experience of the pre-diagnostic period has not led them to suspect a serious underlying condition this may be particularly difficult and this may be an area where care could be improved. Thus a greater emphasis on optimising this diagnostic process may be beneficial, not only for promoting a better patient / carer experience of diagnosis, but also in influencing subsequent experience and needs.

The highly individual nature of the response to diagnosis is supported in the primary qualitative study by the cases where patient and carer described completely opposite reactions, in terms of information seeking. These apparently discordant needs within a single patient / carer dyad are likely to present a particular challenge to health professionals and supportive care services. In addition to this, the analysis highlighted the presence of conflicting attitudes to prognostic information in the same individual, supporting the theory of wanting but not wanting described previously by Giles and Myasaki(12), which is a key component to the theme of information tension.

Potential barriers to discussion of disease prognosis were identified in the perceived role of doctors and other health professionals within the secondary care team and the nature of the clinical consultation. Doctors were suspected and at times even expected, to have a focus on the physical and pharmaceutical aspects of PD care, at the expense of a more holistic approach. This was a cause of dissatisfaction and led at least one participant (P2) to wonder whether *he* should have been the one to initiate prognostic discussions earlier in the disease course. The literature also described that physicians were reluctant to initiate prognostic discussion because of a concern that they will remove hope(109), while at the same time significant numbers of patients appear to believe that it is the responsibility of *clinicians* to raise prognostic issues for the first time(176). It is therefore apparent that the unmet needs described above may arise as a result of these two, incongruous sets of health beliefs.

It is also interesting to note the contrasting and overwhelmingly positive attitude towards non-medical health professionals, particularly the Parkinson's nurse specialist (PDNS), in this regard. Thus while it may be important to challenge and develop the approach of doctors, to better reflect the nuanced and fluctuating needs of patients and carers regarding prognostic information, one answer for services seeking to meet this challenge may be to further support and develop the role of the PDNS, who appears to occupy a different and more approachable position within secondary care services, as perceived by service users(177).

Following on from the above discussion, the analysis supports the importance of non-clinical information resources, such as the internet, books and, particularly through charitable groups such as Parkinson's UK, the role of peer experience and support. However, utilisation of these resources also appears to be limited by perceived barriers. In particular the existence of downward comparison, which is heavily supported by the findings of the qualitative study presented here and is expanded upon by the concept of upward comparison.

Despite the barriers described above, there also appeared to be triggers for individuals to reassess their information needs, in the form of disease milestones. The fluctuation in needs from diagnosis to death and bereavement appeared to be marked by particular milestones. The concept of disease milestones, which had initially been used to describe the need for continued re-evaluation of self, was subsequently developed as a structure for the wider way in which participants experienced the disease.

The model of PD as a process of gradual decline punctuated by milestones can be found elsewhere. Evans et.al. use the analogy at a symptom level, describing the milestones as development of i) dyskinesia, ii) Hoehn and Yahr stage 3, iii) gait freezing and iv) dementia(128). The work of MacMahon and Thomas could also be interpreted in this way, with the emergence of reduced response to medication, cognitive impairment and institutionalisation used as a marker for the "palliative phase" of disease(4, 5). The qualitative work described here expands on these ideas, describing the importance not only of symptoms, but also of functional and social loss and life events. As such there appear to be some milestones which are relatively predictable (generic milestones), such

as the physical and psychological symptoms described above, while others will be highly personal (personal milestones). This has implications for clinicians wishing to identify milestones when they occur; generic milestones requiring an understanding of the natural history of PD and personal milestones requiring a sensitivity to individual patient experience.

13.2.2 *Carer tension*

Patients described difficulties in adjusting to being dependent on their carer(s), often focusing on independence, which could be enhanced or suppressed by a variety of patient specific characteristics, as well as the manner in which care was offered and the influence of practical issues such as time and environment.

In the same way that patients had to adapt to the idea of requiring help and balance this with the need for independence, so carers had to learn how to adopt this role and assimilate it in to their biography. The degree to which this process was successful was influenced by the availability of practical and service specific information and was identified as a potential target for palliative and supportive care.

Access to services and support was influenced by a number of things, including external factors, such as the ability of secondary care services to communicate effectively with users, but also by internal factors such as the phenomenon of downward comparison.

These aspects of patient and carer experience all contribute to the care tension, whilst highlighting a number of possible ways in which responsive health care services may be able to support and positively influence the care negotiations which are described here as central to establishing successful care settlements.

Emanating from the qualitative study the representation of carer as vigilant protector was a new theme of understanding. This began with the process of bearing witness to decline, which could begin in the pre-diagnostic period and carried with it a responsibility of guardianship. This guardianship was not limited to the patient / carer relationship, or to physical protection, but extended to family and encompassed a range of physical, psychological and emotional care. The implications of this discovery are two-fold. Firstly it suggests that informal care roles may be present from the very early stages of disease

and may in turn infer an early care burden. Secondly, where carers are acting as vigilant protector, their requirements in terms of information and support may be very different from those of the patient. This is an important aspect of the care tension, which should be considered by health care professional seeking to design responsive, effective support services.

Even though the quantitative study was designed to be sequential and therefore not expected to contribute to this first research question, nonetheless, it was apparent from the adaptation, face and content validation, that clinicians were able to recognise a group of patients who were more likely to have unmet needs, which is consistent with the concept of milestones (as seen with the identification of "red flags"). Further, clinicians recognised the vast range of need described in the tool agreeing that the needs outlined were ones experienced by patients in their clinical practice.

The knowledge generated in this first qualitative section of the thesis contributed to the adaptation of the NAT-Parkinson's disease. Specifically, in page 1 of the tool it triggered the development of a red flags section, the adaptation of the patient physical domain and supported the inclusion of domains covering carer need, grief and health beliefs. The greater contribution however was to page 2 – "issues to consider", where the qualitative data was used to make the tool PD specific. This led to changes in each domain of the tool, or example, a greater emphasis was placed on information flow and its impact of the care dynamic, stemming from the "information tension" and "care tension" described in detail above (chapter 4 and 6).

13.3 Summary findings for research question 2: *What are the clinimetric properties of the NAT:Parkinson's disease?*

The NAT: Parkinson's disease appears to have face, content and construct validity, as a means of rapidly identifying unmet palliative care need across a wide range of domains in everyday clinical practice, and inter-rater reliability for use by a broad range of clinicians, with different specialist training and areas of clinical expertise.

The domain relating to health beliefs (2.6 *patient health beliefs, social and cultural factors making care more complex)* demonstrated poorer construct validity and inter-rater reliability, probably due to both challenges with comparator tools (construct) and clinician experience in assessing this area (reliability).

The inter-rater reliability was also limited for some of the carer domains for similar reasons and it is important to consider that the data was collected using video consultations and a relatively small number of responders (see chapter chapter 11). However, given the wide variation in rater experience and clinical skills (see chapter 11) it is felt that, overall, use of the tool in a range of clinicians is supported by these findings. Of note, the properties are slightly stronger for the patient than for the carer although some carer domains achieved apparently poor inter-rater agreement, as a result of an anomaly in relation to the statistical methods used (weighted kappa).

In summary, the three stages of clinimetric testing have established that the NAT: Parkinson's disease has construct validity and may be reliably used to assess unmet palliative and supportive care need in people with PD and their carers. It can be used by the full range of clinical specialists caring for the condition, with the need for minimal training, to rapidly assess and triage unmet need and, as such, could be utilised by services seeking to improve palliative and supportive care for PD.

13.3.1 Feasibility in people with dementia

The feasibility arm of this study assessed the feasibility of using the NAT: Parkinson's disease in people with dementia, on the basis of carer-proxy representation of need. Unfortunately it proved extremely difficult to recruit participants in to this arm of the study. As such an analysis of unmet needs has not been presented. However on the basis of the three completed assessments, it is at least possible to say that administering the tool in this way appears feasible and is worthy of future study.

13.4 Overall synthesis: *What are the supportive and palliative care needs of people with PD, and their carers, and can these be identified by clinicians using the adapted NAT:Parkinson's?*

This thesis has demonstrated and delineated that people with Parkinson's disease, and those who care for them, have a significant burden of supportive and palliative care needs from diagnosis through to bereavement, and, that the NAT:Parkinson's tool has sufficient clinimetric properties to help these be identified by the clinician. The following section outlines the clinical implications of these findings.

13.5 Clinical implications

Taken as a whole the findings of this project offer a number of challenges to existing practices.

13.5.1 Rethinking the current model of the "palliative phase" of Parkinson's disease

The first challenge comes from the confirmed presence of unmet palliative and supportive care needs throughout the natural history of PD, for both patients and carers. This finding requires an adjustment to the current model of disease which has previously described the natural history in 4 phases, the last of which constitutes the "palliative phase" and is defined by waning response to medication, cognitive decline and the need for 24 hour care(5). This model does not fit with the patterns of need identified by this thesis.

The presence of unmet need implies that a palliative approach to care is appropriate for PD. Comparison of PD to the WHO definition of palliative care confirms that it meets all of the key criteria being; incurable, life limiting(26, 114, 115), with significant impact on quality of life for both patients(128, 178, 179) and carers(130, 180) and causing significant pain and symptom burden consistent with malignant conditions(126, 181). The qualitative synthesis in particular adds to this evidence base, by describing the nature of unmet needs at a personal level for those living and caring for PD. In its entirety this evidence for

unmet palliative need and appropriateness of palliative approaches to care is overwhelming and demands appropriate responses from clinical and support services.

Subsequent challenges relate to the specific findings of the qualitative studies:

1- Firstly the diagnostic experience for patients and carers often appears to be poor.

2- Secondly, information regarding services, practical support and most strikingly disease progression and prognosis is often inadequate. As a result it is more difficult for those living with PD to achieve key objectives such as, promoting of independence, establishing and maintaining effective care arrangements, supporting the role of carers and planning for future care in advanced stages. These deficiencies in turn can be a cause of frustration and dissatisfaction with clinicians and services.

3- Thirdly are the barriers to meeting the needs described above. These include training and skills, as well as practical issues such as consultation time, particularly on the first clinic visit, which may need to be addressed if a palliative approach to care is to be adopted by PD specialists.

13.5.2 Addressing the specific challenges of unmet palliative care need

Having identified the challenges posed by this project it is useful to consider potential solutions and the way in which the work presented here may contribute to them.

13.5.2.1 Addressing specific challenges: diagnosis

The process of diagnosis has been repeatedly identified as a challenge. As a PD specialist I would seek to address the following areas to try and improve care and promote the positive aspects of diagnosis identified in the qualitative studies:

1- Adjusting clinic appointments to ensure sufficient time for first consultation.

2- Building a personal framework for understanding the needs arising from the diagnostic process, this is based on the theoretical frameworks used in the qualitative synthesis and framework analysis.

3- Be aware of the importance of gathering information during the consultation in relation to an individual's prior experience, expectations regarding diagnosis and attitude to information seeking, in order to inform subsequent decision making.

4- Provide explicit invitation to the carer to legitimise their needs and views within the otherwise patient-focused consultation, and consider providing a separate forum for them to express concerns if they do not wish to do this with the patient present.

5- Seek to develop advanced communication skills through formal and informal training and development.

13.5.2.2 *Addressing specific challenges: Information*

Another important area highlighted throughout this thesis is management of the information tension experienced by people with PD. The qualitative findings are useful in this regard to promote an appreciation of the variable and fluctuant nature of information needs, the relationship to maintenance of hope, formation of effective care dynamics and planning for advanced stages. Allied with enhanced communication skills, the concept of disease milestones provides a structure with which to approach this issue, helping to identify times when information needs may be highest. This is a concept which requires further development. Future work with patients and carers may also like to explore ways in which the effect of downward comparison can be negated.

It is also important to utilise the skills of the whole team and particularly the PDNS, who may be seen as the suitable person discuss prognostic information by some patients.

13.5.2.3 *Addressing specific issues: support and care tension*

An understanding of the issues presented here may be used to promote access to support, for example by appreciating the impact of downward comparison when designing literature, clinics and support groups. Similarly an appreciation of the theory, *care tension*, should prompt improvements in the way that practical information and support is offered in order to aid patients and carers adapting to the new care roles and facilitate care settlements which promote independence and minimise carer strain.

Finally, building on the concept of disease milestones and the known patterns of carer strain in relation to factors such as patient wellbeing(152), carer age(150) and presence of neuro-psychiatric symptoms(130), it would be interesting to identify key "red flags" and "milestones" for carer needs, as times for assessment and intervention.

13.5.2.4 *Addressing specific issues: Planning for advanced stages*

The lack of preparation for advanced stages of disease and the potential loss of decision making autonomy due to cognitive impairment are striking features of the synthesis. This also represents an area where existing services appear to perform poorly, at least in the UK, with few patients accessing hospice care in the terminal phase(10, 182, 183). The evidence presented here and elsewhere(176) suggests that a number of individuals with PD would wish to discuss these issues even early in the course of disease, although others would not. People also vary considerably regarding who they think should initiate these discussion, but at least a proportion feel that it should be clinician led(176) – see also chapter 6. Once again an appreciation of disease milestones, combined with good communication skills may be useful here. In addition, the pattern of formed hallucinations emerging prior to cognitive impairment and followed by an apparently stereotyped pattern of decline(127), as identified in the discussion of red flags (chapter 7) may be an important opportunity to preserve decision making autonomy. This is an area which should be examined in the future research.

It is clear that a "one size fits all" approach to timing of advanced care discussions is likely to be inadequate and, in some cases, harmful. However the response to *information tension* should not be to avoid the discussion, but rather to respond to it in a person centred manner, using excellent communication skills. Developing these skills and supporting the process where particular difficulties are encountered, may be a key area for palliative care services.

13.5.3 *Designing responsive services*

There are three stages of assessment which are required to enable palliative needs to be met, these are:

1- Identification of unmet need
2- Triage of unmet need
3- Comprehensive palliative needs assessment

13.5.3.1 *Identification of need*

Effective identification of need requires two things, firstly a heightened awareness of the existence of need amongst clinicians caring for PD and secondly a rapid bedside test that can be incorporated in to everyday clinical practice, with minimal time demands. The first element can be addressed through promotion of the disease model proposed above and wider publication of the evidence for need, such as the qualitative work presented here, while the NAT: Parkinson's disease is now a viable solution to the second.

13.5.3.2 *Triage of need*

The degree to which identified needs are met within local services, either by the person conducting the assessment or another member of the usual care team, will depend on the skill sets available. Thus, by monitoring the number and nature of referrals to specialist palliative care, through the triage system embedded in the NAT, it will be possible to identify areas where local skills need to be enhanced. For example if there are a high proportion of referrals for advance care planning, then particular training focused in this area may be useful. Likewise, specialist palliative care teams may require the support of PD specialists in order to build and develop their PD specific skills. Embedding a symbiotic relationship between the two disciplines in this way may help to promote collaboration and sustainability(27, 184). Fluid movement of patients between specialist PD and Palliative care services, according to need, will be important if integrated services are to be sustainable(23). The process of need identification and triage, described above, offers a solution to this key problem.

13.5.3.3 *Comprehensive assessment of need*

The third step, for those with complex needs which cannot be met within a local service, involves comprehensive palliative care assessment and on occasion specialist palliative care referral. This may utilise other palliative assessment tools, such as POS-PD(124, 125) and ESAS-PD(126), which offer a comprehensive assessment of unmet need, but may be less suitable for the rapid assessment and triage described above. As such these tools are complimentary to each other, occupying different roles in the overall assessment of palliative need.

13.5.4 Using the NAT: Parkinson's disease

It will be important to establish the best way to utilise the NAT:Parkinson's disease in clinical practice. The low burden of use, for both patients and clinicians, means that it would be possible to apply the tool at regular intervals. In addition the disease milestones described in the qualitative study, lend themselves as triggers for needs assessment. One solution would combine these approaches, with a regular palliative needs assessment, perhaps as part of a comprehensive evaluation offered to all PD service users (see below), in addition to screening when milestones are identified.

The challenges and proposed solutions discussed above should not be viewed in isolation, but rather within a wider body of work which is targeting a more holistic, person centered approach to PD care. To this end there is increasing emphasis on the impact of non-motor symptoms(162, 185) and initiatives which produced greater inter-professional working in the Netherlands(186) are now being developed in the UK(187). Given the greater prevalence of PD in older populations(1) the evolving landscape of geriatric medicine with an emphasis on frailty(188), community working and comprehensive, multi-disciplinary assessment is also relevant(189, 190). Any initiatives aiming to reduce the level of unmet palliative and supportive care need should do so within this existing care framework. As such the NAT:Parkinson's disease may be best utilised as one component in a rolling programme of Comprehensive Parkinsonian Assessment, analogous to the concept of Comprehensive Geriatric Assessment (CGA) which has proven benefit in older people(189, 190).

13.5.5 Screening versus assessment and triage

Although at various times within this thesis the term "screening" has been used to describe the early identification and preliminary evaluation of unmet palliative and supportive care need, this is perhaps not the best way to conceptualise the NAT-Parkinson's disease. Screening in health care is a tightly defined activity, appropriate only under certain conditions(191). Screening tools are then evaluated against gold standard diagnostic tests, assessing their sensitivity and specificity, which will in turn determine whether or not they are appropriate for clinical use. The NAT-Parkinson's disease has not been evaluated in this way, indeed, given the absence of gold standard criteria for establishing unmet palliative care needs such an assessment would have been impossible. Interestingly, the subsequent emergence of the POS-PP and ESAS-PD, which could conceivably be used in this way, may allow such evaluation in the future.

In the absence of an established clinical environment where formal screening could take place, the NAT-Parkinson's may be best conceptualised as a tool for rapid assessment and triage of unmet need, as described above. It will be important in future work to establish the most effective times within the disease trajectory for the tool to be applied (see below).

13.6 Strengths and limitations of this work

13.6.1 Strengths: Qualitative Phase

13.6.1.1 *SLR and qualitative synthesis*

The systematic review and qualitative synthesis is a strength of this project, bringing together for the first time a rich but disparate qualitative literature base. Meta-ethnography, an idealist approach as categorised by Barnett-Page (see chapter 2) was selected, primarily because it embraces the strengths of qualitative synthesis; namely the ability to be inductive, to move beyond an aggregation of the previously published work and generate new understanding. Whilst other, realist, approaches have been suggested as more appealing to health service policy makers, as they prioritise integration over interpretation and thus may be more transparent, this was felt to be outweighed by the

potentially lost opportunity to generate new theory in an area which, until now, has been poorly understood.

Given the use of an idealist approach it was important to triangulate and identify common themes from studies across geographical and temporal divides, this is felt to greatly enhance the credibility and applicability of these findings. The framework created is a valuable tool for understanding the palliative and supportive care needs of people living with PD and firmly establishes the presence of need throughout the disease course, in keeping with modern definitions of palliative care.

13.6.1.2 *Qualitative study*

The qualitative study was valuable in confirming and adding to the findings of the qualitative synthesis. The inclusion of a high proportion of patient participants, with both early and late stage disease, meant that it was possible to compare and contrast the experience of different participants, to strengthen the analysis.

Taken together, the two pieces of qualitative work in this project greatly enhance our understanding of the personal experience of PD and how that translates in to palliative and supportive care need. The translation of these findings in to clinical applications demonstrates the strength of the mixed methods research design.

13.6.2 *Strengths: adaptation*

The adaptation is demonstrably grounded in the literature relating to palliative and supportive care need in PD. The inclusion of a "red flags" section adds a new element to the original NAT:PD-c design and the work on question stems and appearance make the tool more attractive for clinicians to use.

13.6.3 *Strengths: Clinimetric testing*

13.6.3.1 *Construct Validity*

The construct validity testing was designed to include the widest possible group of patients and carers, the face and content validation including participants from across

three continents. Since the start of this project, two patient completed assessment tools, POS-PD and ESAS-PD, have been published(124-126). In both cases the assessments were conducted in people with Hoehn and Yahr stage 3 disease and beyond. The inclusion of people at all disease stages is therefore a point of difference and a strength of the NAT:Parkinson's disease as a tool to embed in routine clinical practice by non-palliative care specialists.

Elsewhere the study design sought to build on that of the original NAT;PD-c and address potential limitations. Examples include the use of Kendall's tau B for statistical analysis and computer simulations to justify study population size.

The effort to conduct a feasibility study in people with dementia was a strength of the study design. Although recruitment was poor, this does suggest that future evaluation of the tool in this group is possible.

13.6.3.2 *Inter-rater reliability*

The variety of raters involved in the study is a strength and supports the use of this tool in everyday clinical practice. While higher levels of inter-rater agreement could almost certainly have been achieved if testing had been conducted within a single clinical group, for example, neurologists only, this would not have established reliability across the range of clinicians involved in caring for people living with PD. This enhances the external validity of these findings.

Developments to previous study designs included the use of 10 video consultations of real doctor / patient interaction. As a result, the study has examined the reliability of the NAT:Parkinson's disease to detect an array of real life palliative care need, in patients and carers, throughout the spectrum of disease. In addition the use of multiple clinical raters strengthens the findings in this phase of the project. Finally the use of weighted kappa in multiple raters, an adaptation to Fleiss' method, is an improvement on previous study designs, where the 3 outcome categories had to be dichotomised (see chapter 11).

13.6.4 Limitations: Qualitative phase

13.6.4.1 *SLR and qualitative synthesis*

Due to limitations of resource the study did not include foreign language papers, which is a potential source of bias. The reasons for exclusion of papers at the title and abstract stage are given as broad categories, such as "Study not qualitative", it would be useful in future to record the reason for excluding each study at this early stage, so that more precise information could be given. The decision not to exclude studies on the basis of quality may be interpreted as a limitation, however, given the lack of agreement on this topic in the methodological literature and the desire to include all relevant primary data, this decision is justifiable and in keeping with practice elsewhere see chapter 2 section 2.13.

13.6.4.2 *Qualitative study*

The main limitation in this phase of the project was the failure to recruit current carers of people with dementia. It is notoriously difficult to include people with dementia in research(113) and this is an area which should be addressed in the future.

13.6.5 Limitations: Clinimetric testing

13.6.5.1 *Validation*

The matching of comparator tools to constructs within the NAT:Parkinson's disease was not ideal and this is likely to have reduced levels of correlation. Since the start of this project, PD specific adaptations of the POS and ESAS have been published, which would have been better suited, had they been available at the time.

13.6.5.2 *Inter-rater reliability*

The wide variety of clinical raters, a strength in terms of external validity, is likely to have reduced levels of agreement, particularly for sub-sections of the tool which are more familiar to palliative specialists.

A limitation of kappa lies in the low values obtained when percentage agreement is extremely high, but not perfect, and led to low kappa values for two sub-sections. This may be due in part to a floor effect of the tool itself (see chapter 12) or to a failure of the video consultation used in the testing to display sufficient variation of need relating to these constructs. One way to assess this further would be to analyse these constructs using the construct validity data set, where all data was collected by a single rater (see chapter 9). If the same phenomenon was observed then it is likely that this represents a floor effect, rather than the impact of video consultations. In the event that this is a floor effect, where the question is "too hard" and fails to discriminate between responders, it may be that greater training in these areas, such as the assessment of unmet need relating to grief, may improve performance and produce a greater spread of data points. Equally it would be interesting to see whether the same phenomenon occurred where the responders were all trained in specialist palliative care.

Finally, this study did not examine test-retest reliability. Although much of the variance due to test-retest is also present in inter-rater reliability(140), this is an area which could be examined in future work.

13.7 Implications for future research

Future studies could seek to address the limitations discussed above, particularly the test-retest variability associated with the NAT:Parkinson's disease and the impact of POS-PD(124, 125) and ESAS –PD(126) on construct validation results. In addition, although the qualitative experience of people living with PD is now well understood, the experience of people with dementia remains poorly accessed and future study designs should try to address this.

The development of "disease milestones" has been interesting and lends itself to future study, to explore the utility of milestones as indicators of unmet palliative and supportive care need and thus triggers for assessment and opportunities for tailored intervention. In addition the extension of milestones to the carer disease experience, particularly to

identify times when carer support would be most valuable, is an interesting area to develop.

The ability to rapidly assess unmet palliative care needs, combined with subsequent comprehensive needs assessment using tool such as the POS-PD(124, 125), would allow the unmet needs of people with PD to be quantified, throughout the disease course. This could be done within one of the existing PD cohort studies and would be valuable in identifying high risk groups and times when palliative and supportive care services can be of greatest value. This is an area of study which should be prioritised.

Finally, the NAT: Parkinson's disease was developed in order to improve the integration of specialist PD and palliative care services, with a view to facilitating sustainable service development and improving the experience of patients and carers. With this in mind, further evaluation, to assess the impact of the tool within an integrated PD palliative care service is needed. This should focus not only on outcomes related to patient and carer needs and experience, but also staff training needs and service dynamics.

Future studies could also assess how the NAT:Parkinson's disease and POS-PD or ESAS-PD could be optimally used in the same clinical system: the Nat:Parkinson's disease identifying patients and carers who would benefit from completing the POS-PD

13.8 Conclusions

This project has answered the original research questions: i) what are the palliative and supportive care needs of people living with PD and ii) what are the clinimetric properties of the NAT: Parkinson's disease.

The qualitative work enhances our understanding of the palliative and supportive care needs of people with PD, providing a framework for understanding the experience of patients and carers and challenges health professionals to meet these needs.

The successful adaptation and clinimetric testing of the NAT: Parkinson's disease, with evidence to support both construct validity and inter-rater reliability, means that this tool can be recommended as a rapid, clinician completed assessment for unmet palliative care

need. Its application is not limited to developing PD palliative care services, but could form part of a wider, patient-centered, needs driven approach to care. It is hoped that improved recognition and subsequent palliative interventions will serve to improve the lives and experience of people living with PD.

References

1.   Parkinson's UK. Parkinson's prevelence in the United Kingdom (2009) 2009 [Available from: http://www.parkinsons.org.uk/sites/default/files/parkinsonsprevalenceuk_0.pdf.
2.   British Geriatric Society. Parkinson's disease 2007 [Available from: http://www.bgs.org.uk/index.php/topresources/publicationfind/goodpractice/376-parkinsonsdisease.
3.   Dorsey E.R CR, Thompson J.P, BiglanK.M, Holloway R.G. et.al. Projected number of people with Parkinson disease in the most populous nations, 2005 through 2030. Neurology. 2007;68:384–6.
4.   Macmahon D, Thomas S. Practical approach to quality of life in Parkinson's disease: the nurse's role. Journal of Neurology. 1998;245(suppl. 1): S19-S22.
5.   MacMahon DG, Thomas S, Campbell S. Validation of pathways paradigm for the management of PD. Parkinsonism and Related Disorders. 1999;5:S53.
6.   National Institute for Health and Care Excellence. Parkinson's disease: Diagnosis and management in primary and secondary care 2006 [Available from: https://www.nice.org.uk/guidance/cg35.
7.   World Health Organisation. WHO definition of palliative care 2015 [Available from: http://www.who.int/cancer/palliative/definition/en/.
8.   National Council for Palliative Care. Palliative care explained 2015 [Available from: http://www.ncpc.org.uk/palliative-care-explained.
9.   Hudson PL, Toye C, Kristjanson LJ. Would people with Parkinson's disease benefit from palliative care? Palliative Medicine. 2006;20(2):87-94.
10.   Snell K PS, Lee M, et al. The place of death in Parkinson's disease. Age Ageing 2009;38(5):617-9.

11.   Goy.E.R, Carter JH, Ganzini.L. . Parkinson's disease at the end of life: caregiver perspectives. Neurology. 2007;69:611-2.
12.   Giles S, Myasaki J. Palliative stage Parkinson's disease: patient and family experience of health care services. Palliative Medicine. 2009;23:120-5.
13.   McLaughlin D, Hasson F, Kernohan WG, Waldron M, McLaughlin M, Cochrane B, et al. Living and coping with Parkinson's disease: perceptions of informal carers. Palliat Med. 2011;25(2):177-82.
14.   Hasson F, Kernohan WG, McLaughlin M, Waldron M, McLaughlin D, Chambers H, et al. An exploration into the palliative and end-of-life experiences of carers of people with Parkinson's disease. Palliat Med. 2010;24(7):731-6.
15.   Goy ER, Carter J, Ganzini L. Neurologic disease at the end of life: caregiver descriptions of Parkinson disease and amyotrophic lateral sclerosis. J Palliat Med. 2008;11(4):548-54.
16.   Kristjanson LJ, Samar AM, Oldham.L. Palliative care and support for people with neurodegenerative conditions and their carers. Int Journal of Palliative Nursing. 2005;12(8):368-77.
17.   Elman LB, Houghton DJ, Wu GF, Hurtig HI, Markowitz CE, McCluskey L. Palliative care in amyotrophic lateral sclerosis, Parkinson's disease, and multiple sclerosis. J Palliat Med. 2007;10(2):433-57.
18.   Campbell C, Jones E. Palliative and end-of-life care in advanced Parkinson's disease and multiple sclerosis. Clinical Medicine. 2010;10(3):290-2.
19.   Wilcox.S.K. Extending palliative care to patients with Parkinson's disease. British Journal Hosp Med. 2010;71(1):26-30.
20.   Johnson M.J Gadoud A. Palliative care for people with chronic heart failure: when is it time? J Palliat Care. 2011;27(1):37-42.

21.   The National End of Life Care Programme. End of life care in long term neurological conditions: a framework for implementation 2010 [Available from: http://www.endoflifecare.nhs.uk/assets/downloads/improving_eolc_neurology.pdf.

22.   Watson M. Changing emphasis in end-of-life care. British Journal of Hospital Medicine 2010;71(1):6-7.

23.   Lanoix M. Palliative care and Parkinson's disease: managing the chronic-palliative interface. Chronic Illn. 2009;5(1):46-55.

24.   General Medical Council. Treatment and care towards the end of life: good practice in decision making 2010 [Available from: http://www.gmc-uk.org/guidance/ethical_guidance/end_of_life_care.asp.

25.   Murray.S.A Boyd K, Sheikh.A. Palliative care in chronic illness: We need to move from prognostic paralysis to active total care. BMJ. 2005(330):611-2.

26.   Hely MA, Reid WG, Adena MA, Halliday GM, Morris JG. The Sydney multicenter study of Parkinson's disease: the inevitability of dementia at 20 years. Mov Disord. 2008;23(6):837-44.

27.   Richfield E AJ, Jones E. Palliative care for Parkinson's disease: A summary of evidence and future directions. Palliative Medicine. 2013;27(9):805-10.

28.   Boyd KJ, Worth A, Kendall M, Pratt R, Hockley J, Denvir M, et al. Making sure services deliver for people with advanced heart failure: a longitudinal qualitative study of patients, family carers, and health professionals. Palliat Med. 2009;23(8):767-76.

29.   Waller A, Girgis A, Lecathelinais C, Scott W, Foot L, Sibbritt D, et al. Validity, reliability and clinical feasibility of a Needs Assessment Tool for people with progressive cancer. Psychooncology. 2010;19(7):726-33.

30.   Waller A. Girgis A. Davidson PM . Facilitating needs-based support and palliative care for people with chronic heart failure: preliminary evidence for the acceptability, inter-rater reliability, and validity of a needs assessment tool. J Pain Symptom Manage. 2013;45(5):912-25.

31.   Waller A, Girgis A, Johnson C, Lecathelinais C, Sibbritt D, Seldon M, et al. Implications of a needs assessment intervention for people with progressive cancer: impact on clinical assessment, response and service utilisation. Psychooncology. 2012;21(5):550-7.

32.   Oxford English Dictionary: Oxford University Press; 2015 [On line dictionary]. Available from: http://www.oxforddictionaries.com/definition/english/ontology.

33.   Bryman A. Social Research Methods. 4th ed. Oxford: Oxford University Press; 2012.

34.   Bryman A. Social Research Strategies. In: A. B, editor. Social Research Methods. 4th ed. Oxford: Oxford University Press; 2012.

35.   Hammersley M. Methodology who needs it? London: Sage; 2011.

36.   Stone L. Epistemology. . In: Given.L.M, editor. The Sage Encyclopedia of Qualitative Research Methods. Thousand Oaks, CA: SAGE Publications; 2008.

37.   Silverman D. Using Theories. In: K M, editor. Doing Qualitative Research. 4th ed. Thousand Oak, California: Sage; 2013.

38.   O'Reilly K. "Realism". Key Concepts in Ethnography London: SAGE Publications Ltd; 2009.

39.   Gubrium J.F HJA. The new Language of qualitative method. New YOrk: Oxford University Press; 1997.

40.   Bryman A. The nature of Qualitative Research. Social Research Methods. 4th ed. Oxford: Oxford University Press; 2012.

41.   Hammersley M. Reading Ethnographic Research. 2nd ed. London: Longman; 1998.

42.   Dowling M. Reflexivity. In: L.M. G, editor. The Sage Encyclopedia of Qualitative Research Methods. Thousand Oaks, CA: SAGE Publications; 2008. p. 748-9.

43.   Silverman D. Using Theories. In: K M, editor. Doing Qualitative Research. Thousand Oaks, California: Sage; 2013. p. 112.

44.   Denzin NK LinconY. The Landscape of Qualitaive Research. 2nd ed. Thousand Oaks, California: Sage; 2003.

45.   M.M. B. Wither Mixed Methods? In: M.M. B, editor. Advances in Mixed Methods Research. 1 ed. London: Sage; 2008.
46.   Cresswell J.W PCV. Designing and Conducting Mixed Methods Research. 2nd ed. Thousand Oaks, Califormia: Sage 2011.
47.   Hammersley M. Troubles with Triangulation. In: M.M. B, editor. Advances in Mixed MEthods Research. 1st ed. London: Sage 2008.
48.   BerbmanM.M. Wither Mixed Methods. In: M.M B, editor. Advances in Mixed Methods Research. London: Sage; 2008. p. 3.
49.   Bryman A. Mixed Methods Research: combining quantitative and qualitative research. Social Research Methods. 4th ed. Oxford: Oxford University Press; 2012.
50.   Cresswell J.W PCV, Garrett A. Methodological issues in conducting mixed methods research designs. In: M.M. B, editor. Advances in Mixed Methods Research. 1st ed. London: Sage; 2008.
51.   Flemming K. The synthesis of qualitative research and evidence-based nursing. Evidence Based Nursing. 2007;10:68-71.
52.   Dixon-woods MF, R. qualitative research in systematic reviews has established a place for itself. BMJ. 2001;323:1.
53.   Popay J, Rogers A, Williams G. Rationale and Standards for the Systematic Review of Qualitative Literature in Health Services Research. Qualitative Health Research. 1998;8(3):341-51.
54.   Kelly.M S, C. Integrative approaches to qualitative and quantitative evidence (forward). Integrated approaches to qualitative and quantitative evidence: Health Development Agency; 2004. p. 35.
55.   Thomas J HA. Methods for the thematic synthesis of qualitative research in systematic reviews. BMC Med Res Methodol. 2008;8(45).
56.   Centre for Research Dissemination. Systematic Reviews: CRD's guidance for undertaking reviews in health care. University of York: Centre for Research Dissemination; 2009. 277 p.
57.   Dixon-Woods. M AS, Young.B, Jones.D, Sutton. A. HDA integrative approaches to qualitative and quantitative evidence.: Health Development Agency; 2004.
58.   Barnett-Page.E TJ. Methods for the synthesis of qualitative research: a critical review. BMC Medical Research Methodology. 2009;9:1.
59.   Hannes K. Chapter 4: Critical appraisal of qualitative research in Supplementary Guidance for Inclusion of Qualitative Research in Cochrane Systematic Reviews of Interventions. Version 1: Cochrane Collaboration Qualitative Methods Group; 2011.
60.   Cutcliffe J.R MHP. Establishing the credibility of qualitative research findings: the plot thickens. Journal of Advanced Nursing. 1999;30(2):374-80.
61.   Gubba E.G LYS. Effective evaluation : improving the usefulness of evaluation results through responsive and naturalistic approaches. London: Jossey-Bass 1981.
62.   Noyes J LS. Supplemental Guidance on Selecting a Method of Qualitative Evidence Synthesis, and Integrating Qualitative Evidence with Cochrane Intervention Reviews. In: Noyes J BA, Hannes K, Harden A, Harris J, Lewin S, Lockwood C (editors), editor. Supplementary Guidance for Inclusion of Qualitative Research in Cochrane Systematic Reviews of Interventions Version 1 (updated August 2011). 1st ed: Cochrane Collaboration Qualitative Methods Group; 2011.
63.   O'Conner.S.J. Context is everything: the role of auto-ethnography, reflexivity and self-critique in establishing the credibility of qualitative research. Eur J Cancer Care. 2011;20:421-3.
64.   Gubba E.G LYS. Fourth Generation Evaluation. California: Sage Publications; 1989.
65.   McBrien.B. Evidence-based care: enhancing the rigour of a qualitative study. British Journal of Nursing. 2008;17(2):286 -9.
66.   Barbour RS. Introducing Qualitative Research. A student guide to the craft of doing Qualitative research. London: Sage Publications; 2008.
67.   Silverman S. Doing Qualitative Research 3rd ed. London: Sage Publications; 2009.

68.   Charmaz K. Constructing Grounded Theory: a practical guide through qualitative analysis. London: Sage Publications; 2006.

69.   Spencer L Ritchie J, O' Connor W. In: Ritchie J LJ, editor. Qualitative Research Practice A guide for Social Science Students and Researchers. 1st ed. London: Sage Publishing; 2003. p. 199-218.

70.   Silverman D. Doing Qualitative Research. 4th ed. London: Sage Publications; 2013.

71.   Murphy E DR, Greatbatch D, Parker S,Watson P. Qualitative research methods in health technology assessment: a review of the literature. Health Technol Assessment. 1998;2(16).

72.   Barbour RS. The newfound credibility of qualitative research? Tales of Technical Essentialism and co-option. Qual Health Res. 2003;13(7):1019-27.

73.   Schulz KF AD, Moher D, for the CONSORT Group. CONSORT 2010 Statement: updated guidelines for reporting parallel group randomised controlled trials. BMC Medicine. 2010;8.

74.   Tong A FK, McInnes E, Oliver S, Craig J. Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ. BMC Medical Research Methodology. 2012;12(181):8.

75.   Dixon-Woods M, Booth A, Sutton AJ. Synthesizing qualitative research: a review of published reports. Qualitative Research. 2007;7(3):375-422.

76.   Atkins S, Lewin S, Smith H, Engel M, Fretheim A, Volmink J. Conducting a meta-ethnography of qualitative literature: lessons learnt. BMC Med Res Methodol. 2008;8:21.

77.   Dixon-Woods M SA, Shaw R, Miller T, Smith J, Young B. et al. Appraising qualitative research for inclusion in systematic reviews: a quantitative and qualitative comparison of three methods. Journal of Health Services Research & Policy. 2007;12(1):42-7.

78.   Cochrane Collaboration. Resources and guidance [website]. Available from: ph.cochrane.org/resources-and-guidance.

79.   Seers K. What is a qualitative synthesis? Evid Based Nurs. 2012;15(4):101.

80.   Dixon-Woods M, Cavers D, Agarwal S, Annandale E, Arthur A, Harvey J, et al. Conducting a critical interpretive synthesis of the literature on access to healthcare by vulnerable groups. BMC Med Res Methodol. 2006;6:35.

81.   Centre for Research Dissemination. Systematic Reviews: CRD's guidence for undertaking reviews in healthcare. 2009.

82.   Flemming K. Synthesis of quantitative and qualitative research: an example using Critical Interpretive Synthesis. J Adv Nurs. 2010;66(1):201-17.

83.   Campbell R PP, Morgan M, Daker-White G, Britten N, Pill R, et al. Evaluating meta-ethnography: systematic analysis and synthesis of qualitative research. Health Technol Assess. 2011;15(43):1-159.

84.   Noblit GW Hare R. Meta-ethnography: synthesizing qualitative studies. London: Sage publications; 1988.

85.   Tesch R. Qualitative research:analysis types and software tools. Lewes: Farmer Press; 1990.

86.   Strauss A, Corbin J. Basics of Qualitative Research. 2nd ed. Thousand Oaks, California: Sage Publications; 1998.

87.   Richie J LJ. Qualitative Research Practice. 1st ed. London: Sage publications; 2003.

88.   Gale N. K HG, Cameron E, Rashid S, Redwood S. Using the framework method for the analysis of qualitative data in multi-disciplinary health research. BMC MEdical Research Methodology. 2013;13(117).

89.   Huberman M.A MMB. The Qualitatitive Researchers Companion. Thousand Oaks, California: Sage Publications; 2002.

90.   Flemming K, Adamson J, Atkin K. Improving the effectiveness of interventions in palliative care: the potential role of qualitative research in enhancing evidence from randomized controlled trials. Palliat Med. 2008;22(2):123-31.

91.   National Centre for Palliative Care. Palliative Care Explained 2015 [Available from: http://www.ncpc.org.uk/palliative-care-explained.

92.   Flemming K, Briggs M. Electronic searching to locate qualitative research: evaluation of three strategies. J Adv Nurs. 2007;57(1):95-100.

93.   Habermann B. Day-to-Day Demands of Parkinson's Disease. Western Journal of Nursing Research. 1996;18(4):397-413.

94.   Habermann B. Continuity challenges of Parkinson's disease in middle life. J Neurosci Nurs. 1999;31(4):200-7.

95.   Habermann BDLL. Caring for family with Alzheimer's disease and Parkinson's disease: needs, challnages and satisfactions. JOURNAL OF GERONTOLOGICAL NURSING. 2005:49-54.

96.   Birgersson AM, Edberg AK. Being in the light or in the shade: persons with Parkinson's disease and their partners' experience of support. Int J Nurs Stud. 2004;41(6):621-30.

97.   Fleming V, Tolson D, Schartau E. Changing perceptions of womanhood: living with Parkinson's Disease. Int J Nurs Stud. 2004;41(5):515-24.

98.   Hermanns M. Culturally competent care for Parkinson disease. Nurs Clin North Am. 2011;46(2):171-80.

99.   Phillips L.J. Dropping the Bomb: The Experience of Being Diagnosed with Parkinson's Disease. Geriatric Nursing. 2006;27(6):362-9.

100.   Abendroth M, Lutz BJ, Young ME. Family caregivers' decision process to institutionalize persons with Parkinson's disease: a grounded theory study. Int J Nurs Stud. 2012;49(4):445-54.

101.   Wressle E, Engstrand C, Granérus A-K. Living with Parkinson's disease: Elderly patients? and relatives? perspective on daily living. Australian Occupational Therapy Journal. 2007;54(2):131-9.

102.   van der Eijk M, Faber MJ, Al Shamma S, Munneke M, Bloem BR. Moving towards patient-centered healthcare for patients with Parkinson's disease. Parkinsonism Relat Disord. 2011;17(5):360-4.

103.   Pretzer-Aboff I GE, Resnick B. Parkinson's Disease: Barriers and Facilitators to Optimizing Function. Rehabil Nurs. 2009;34(2):55-63.

104.   Barretto M SBS, Lalkaka J, Wadia P, Devare S. An Analysis of the Needs of Homebound Patients with Parkinson's Disease and Their Caregivers in the Indian Subcontinent - a Qualitative Study. Movement Disorders. 2010;25(Suppl. 3):S710.

105.   Habermann BMW, McLennon S, Chen H, Rafail L, Wojcieszek J. Couples Living with Parkinson's Disease: Needs and Concerns at Advanced to End Stage. Movement Disorders. 2010;25(suppl.3):S694.

106.   deLau LM VD, Marinus J, van Hilten JJ. Survival in Parkinson's disease. Relation with motor and non-motor features. Parkinsonism Relat Disord. 2014;20(6):613-6.

107.   Aarsland D AK, Larsen JP, Lolk A, Kragh-Sorensen P. Prevalence and characteristics of dementia in Parkinson's disease: an 8 year prospective trial. Arch Neurol. 2003;60:387-92.

108.   Richfield E Adams D, Jones E, Johnson M, Campbell C. A palliative care service for Parkinson's disease: Patient characteristics and service interventions [abstract]. Movement Disorders. 2012;27(Suppl. 1):551.

109.   Almack K, Cox K, Moghaddam N, Pollock K, Seymour J. After you: conversations between patients and healthcare professionals in planning for end of life care. BMC Palliat Care. 2012;11:15.

110.   Parker PA BW, de Moor C, Lenzi R, Kudelka AP, Cohen L. Breaking bad news about cancer: patients' preferences for communication. J Clin Oncol. 2001;19(7):2049-56.

111.   Thomas S, MacMahon D. Parkinson's disease, palliative care and older people. Nurs Older People. 2004;16(1):22-7.

112.   Hoehn MM, Yahr M. Parkinsonism: onset, progression and mortality. Neurology. 1967;17:427-42.

113.   Pesonen.H-M RA, Isola.A. Ethical aspects of researching subjective experiences in early stage dementia. Nursing Ethics. 2011;18(5):651-61.

114.   Hely MA, Morris JG, Reid WG, Trafficante R. Sydney Multicenter Study of Parkinson's disease: non-L-dopa-responsive problems dominate at 15 years. Mov Disord. 2005;20(2):190-9.

115.   Hely M.A MJGL, Traficante R, Reid W.G.J, O'Sullivan D.J, et al. The Sydney multicentre study of Parkinson's disease: progression and mortality at 10 years. J Neurol Neurosurg Psychiatry 1999;67:300–307. 1999;67:300-7.

116.   Hemmesch A.R. The detrimental effects of atypical nonverbal behavior on older adults' first impressions of individuals with Parkinson's disease. Psychol Aging. 2014;29(3):521-7.

117.   Hemmesch A.R T-DL, Zebrowitz L.A. The Influence of Facial Masking and Sex on Older Adults' Impressions of Individuals with Parkinson's Disease. Psychol Aging. 2009;24(3):542-9.

118.   Marras C LA. Parkinson's disease subtypes: lost in translation? J Neurol Neurosurg Psychiatry. 2013;84(4):409-15.

119.   Poewe W. The natural history of Parkinson's disease. J Neurol. 2006;253 Suppl 7:VII2-6.

120.   Schrag A, Quinn N. Dyskinesias and motor fluctuations in Parkinson's disease: a community based study. Brain. 2000;123(11):2297-305.

121.   Schapira AH OC. Drug selection and timing of initiation of treatment in early Parkinson's disease. Ann Neurol. 2008;64(Suppl.2):S47-55.

122.   Gray R IN, Rick C, Patel S, Gray A et. al. Long-term effectiveness of dopamine agonists and monoamine oxidase B inhibitors compared with levodopa as initial treatment for Parkinson's disease (PD MED): a large, open-label, pragmatic randomised trial. Lancet. 2014;384:1196-205.

123.   Waller A, Girgis A, Currow D, Lecathelinais C, Palliative Care Research Program t. Development of the palliative care needs assessment tool (PC-NAT) for use by multi-disciplinary health professionals. Palliat Med. 2008;22(8):956-64.

124.   Saleem TZ, Higginson IJ, Chaudhuri KR, Martin A, Burman R, Leigh PN. Symptom prevalence, severity and palliative care needs assessment using the Palliative Outcome Scale: a cross-sectional study of patients with Parkinson's disease and related neurological conditions. Palliat Med. 2013;27(8):722-31.

125.   Higginson IJ, Gao W, Saleem TZ, Chaudhuri KR, Burman R, McCrone P, et al. Symptoms and quality of life in late stage Parkinson syndromes: a longitudinal community study of predictive factors. PLoS One. 2012;7(11):e46327.

126.   Miyasaki JM, Long J, Mancini D, Moro E, Fox SH, Lang AE, et al. Palliative care for advanced Parkinson disease: an interdisciplinary clinic and new scale, the ESAS-PD. Parkinsonism Relat Disord. 2012;18 Suppl 3:S6-9.

127.   Kempster PA, O'Sullivan SS, Holton JL, Revesz T, Lees AJ. Relationships between age and late progression of Parkinson's disease: a clinico-pathological study. Brain. 2010;133(Pt 6):1755-62.

128.   Evans JR, Mason SL, Williams-Gray CH, Foltynie T, Brayne C, Robbins TW, et al. The natural history of treated Parkinson's disease in an incident, community based cohort. J Neurol Neurosurg Psychiatry. 2011;82(10):1112-8.

129.   Carter J SBJ, Archbold PG, Inoue I, Lannon M, et.al. Living With a Person Who Has Parkinson's Disease: The Spouse's Perspective by Stage of Disease. Movement Disorders. 1998;13(1).

130.   Schrag A, Hovris A, Morley D, Quinn N, Jahanshahi M. Caregiver-burden in parkinson's disease is closely associated with psychiatric symptoms, falls, and disability. Parkinsonism Relat Disord. 2006;12(1):35-41.

131.   McDowell I. Measuring health: a guide to rating scales abd questionnaires. 3rd ed. Oxford: Oxford University Press; 2006.

132.   Streiner DL NG. Validity. Health Measurement Scales: A practical guide to their development and use. 4th ed. Oxford: Oxford University Press; 2008.

133.   Landy FJ. Stamp collecting versus science. American Psychologist. 1986;41:1183-92.

134.   Bland JM. Measuring Health and Disease: The validity of measurement methods: University of York; 2012 [Lecture notes]. Available from: http://www-users.york.ac.uk/~mb55/msc/clinimet/week8/validity.pdf.

135.   Peto V JC, Fitzpatrick R, Greenhall R. The development and validation of a short measure of functioning and well being for individuals with Parkinson's disease. Quality of life research. 1995;4(3):241-8.

136.   Fitzpatrick R DCBM, Jones DR. Evaluating patient-based outcome measures for use in clinical trials. Health Technol Assessment 1998;2(14):25.

137.   Cronbach L.J MPE. Construct validity in psychological tests. Psychological bulletin. 1955;52(4):281-302.

138.   Meyer P. Understanding Measurement: Reliability. Oxford: Oxford University Press; 2010.

139.   Abell N SD, Kamata, A. Reliability. Developing and Validating Rapid Assessment Instruments. Oxford: Oxford University Press; 2009.

140.   Streiner DL NG. Reliability. Health Measurement Scales: A practical guide to their development and use. Oxford: Oxford University Press; 2008.

141.   Campbell D.T FDW. Convergent and discriminant validation by the multitrait - mutlimethod matrix. Psychological bulletin. 1959;56(81-105).

142.   Movement Disorders Society. International Parkinson and Movement Disorders Society; [Available from: http://www.movementdisorders.org/MDS/Education/Rating-Scales/MDS-Recommended-Rating-Scales.htm.

143.   Martinez-Martin PG-NA, Morlan Cracia L, Balseiro Gomez J, Martinez-Sarries J. et.al. Unified Parkinso's disease Rating Scale Characteristics and Structure. Movement Disorders. 1994;9(1):76-83.

144.   Movement Disorder Society Task Force on Rating Scales for Parkinson's Disease. The Unified Parkinson's Disease Rating Scale (UPDRS): Status and Recommendations. Movement Disorders. 2003;18(7):738-50.

145.   Goetz CG, Fahn S, Martinez-Martin P, Poewe W, Sampaio C, Stebbins GT, et al. Movement Disorder Society-sponsored revision of the Unified Parkinson's Disease Rating Scale (MDS-UPDRS): Process, format, and clinimetric testing plan. Mov Disord. 2007;22(1):41-7.

146.   Goetz CG, Tilley BC, Shaftman SR, Stebbins GT, Fahn S, Martinez-Martin P, et al. Movement Disorder Society-sponsored revision of the Unified Parkinson's Disease Rating Scale (MDS-UPDRS): scale presentation and clinimetric testing results. Mov Disord. 2008;23(15):2129-70.

147.   Jenkinson CPV, Fitspatrick R, Greenhall R, Hyman N. Se lf-reported Functioning and Well-being in Patients with Parkinson's Disease: Comparison o f the Short-form Health Survey (SF-36) and the Parkinson's DiseaseQuestionnaire (PDQ-39). Age Ageing. 1995;24:505-9.

148.   Martinez-Martin P, Jeukens-Visser M, Lyons KE, Rodriguez-Blazquez C, Selai C, Siderowf A, et al. Health-related quality-of-life scales in Parkinson's disease: critique and recommendations. Mov Disord. 2011;26(13):2371-80.

149.   Marinus JVM, Martinez-Martin P, van Hilten J.J, Stiggelboult A.M. A short psychosocial questionnaire for patients with Parkinson's disease: the SCOPA-PS. Journal of clinical Epidemiology. 2003;56:61-7.

150.   Carter JH, Lyons KS, Stewart BJ, Archbold PG, Scobee R. Does age make a difference in caregiver strain? Comparison of young versus older caregivers in early-stage Parkinson's disease. Mov Disord. 2010;25(6):724-30.

151.   Carter JH, Stewart BJ, Lyons KS, Archbold PG. Do motor and nonmotor symptoms in PD patients predict caregiver strain and depression? Mov Disord. 2008;23(9):1211-6.

152.   Peters M, Fitzpatrick R, Doll H, Playford D, Jenkinson C. Does self-reported well-being of patients with Parkinson's disease influence caregiver strain and quality of life? Parkinsonism Relat Disord. 2011;17(5):348-52.

153.   Martinez-Martin P, Forjaz MJ, Frades-Payo B, Rusinol AB, Fernandez-Garcia JM, Benito-Leon J, et al. Caregiver burden in Parkinson's disease. Mov Disord. 2007;22(7):924-31; quiz 1060.
154.   Kelly DH, McGinley JL, Huxham FE, Menz HB, Watts JJ, Iansek R, et al. Health-related quality of life and strain in caregivers of Australians with Parkinson's disease: an observational study. BMC Neurol. 2012;12:57.
155.   Zarit S.H RKE, Bach-Paterson J. Relatives of the Impaired Elderly: Correlates of Feelings of Burden. The Gerentologist. 1980;20(6):649-55.
156.   Higginson IJ, Gao W, Jackson D, Murray J, Harding R. Short-form Zarit Caregiver Burden Interviews were valid in advanced conditions. J Clin Epidemiol. 2010;63(5):535-42.
157.   Robinson BC. Validation of a caregiver strain index. Journal of Gerontology. 1983;38:344-8.
158.   Thornton M TSS. Analysis of the Reliability of the Modified Caregiver Strain Index. Journal of Gerontology. 2003;58B(2):S127-32.
159.   Tanji H, Anderson KE, Gruber-Baldini AL, Fishman PS, Reich SG, Weiner WJ, et al. Mutuality of the marital relationship in Parkinson's disease. Mov Disord. 2008;23(13):1843-9.
160.   Aarsland D BK, Larsen J.P, Tysnes O.B, Alves G. Cognitive impairment in incident, untreated Parkinson's disease. Neurology. 2009;72:1121-6.
161.   Gallagher DA, Goetz CG, Stebbins G, Lees AJ, Schrag A. Validation of the MDS-UPDRS Part I for nonmotor symptoms in Parkinson's disease. Mov Disord. 2012;27(1):79-83.
162.   Chaudhuri KR, Martinez-Martin P, Brown RG, Sethi K, Stocchi F, Odin P, et al. The metric properties of a novel non-motor symptoms scale for Parkinson's disease: Results from an international pilot study. Mov Disord. 2007;22(13):1901-11.
163.   Martinez-Martin P, Rodriguez-Blazquez C, Alvarez-Sanchez M, Arakaki T, Bergareche-Yarza A, Chade A, et al. Expanded and independent validation of the Movement Disorder Society-Unified Parkinson's Disease Rating Scale (MDS-UPDRS). J Neurol. 2013;260(1):228-36.
164.   O'Leary N MN, O'Loughlin C, Tiernan E, McDonald K. A comparative study of the palliative care needs of heart failure and cancer patients. European Journal of Heart Failure. 2009;11(4):406-12.
165.   Ramirez A A-HJ, Richards M. ABC of palliative care: the caregivers. Br Med J 1998;316(7126):208-11.
166.   Sharpe L BP, Smith C, McConnell D, Clarke S. The relationship between available support, unmet needs and caregiver burden in patients with advanced cancer and their carers. Psycho-Oncology. 2005;14:102-14.
167.   Fleiss JL. Measuring nominal scale agreement among many raters. Psychological Bulletin. 1971;76:378-82.
168.   Cohen J. A coefficient of agreement for nominal scales. Educational and Psychological Measurement. 1960;20:37-46.
169.   M. B. Clinical Measurement. An introduction to medical statistics. 4th ed. Oxford: Oxford University Press; 2015.
170.   J.M B. Personal communication. 2014.
171.   Cohen J. Weighted kappa: nominal scale agreement with provision for scaled disagreement or partial credit. Psychological Bulletin. 1968;70:213-20.
172.   Bland M. Clinical measurement. An introduction to medical statistics. 4th ed. Oxford: Oxford University Press; 2015. p. 310.
173.   Coolican H. Research Methods and Statistics in Psychology. 5 ed. London: Hodder Education; 2009.
174.   Davis A Bremner G. The experimental method in psychology. In: Breakwell G HS, Fife-Schaw C, Smith J, editor. Research Methods in Psychology. 3 ed. London: Sage publications; 2006.
175.   Aitini E, Aleotti P. Breaking bad news in oncology: like a walk in the twighlight. Annals of Oncology. 2006;17(3):359-60.

176.	Tuck KK Brod L, Nutt J, Fromme EK. Preferences of patients with Parkinson's disease for communication about advanced care planning. Am J Hosp Palliat Care. 2015;32(1):68-77.
177.	Hellqvist C BC. Support supplied by Parkinson's nurse specialist to Parkinson's disease patients and their spouses. Applied Nursing Research. 2015;28(2):86-91.
178.	Knipe MD, Wickremaratchi MM, Wyatt-Haines E, Morris HR, Ben-Shlomo Y. Quality of life in young- compared with late-onset Parkinson's disease. Mov Disord. 2011;26(11):2011-8.
179.	Den Oudsten BL, Lucas-Carrasco R, Green AM, Whoqol-Dis G. Perceptions of persons with Parkinson's disease, family and professionals on quality of life: an international focus group study. Disabil Rehabil. 2011;33(25-26):2490-508.
180.	Martinez-Martin P, Arroyo S, Rojo-Abuin JM, Rodriguez-Blazquez C, Frades B, de Pedro Cuesta J, et al. Burden, perceived health status, and mood among caregivers of Parkinson's disease patients. Mov Disord. 2008;23(12):1673-80.
181.	Lee MA, Prentice WM, Hildreth AJ, Walker RW. Measuring symptom load in Idiopathic Parkinson's disease. Parkinsonism Relat Disord. 2007;13(5):284-9.
182.	Walker RCD, Dewhurst F, et.el. Palliative care in people with idiopathic Parkinson's disease who die in hospital. BMJ supportive and Palliative care. 2014;4:64-7.
183.	Sleeman KE HY, Verne J, Glickman M, Silber E, et.al. Place of death, and its relation with underlying cause of death, in Parkinson's disease, motor neuron disease, and multiple sclerosis: a population based study. Palliat Med. 2013;27(9):840-6.
184.	Fallon M, Foley P. Rising to the challenge of palliative care for non-malignant disease. Palliat Med. 2012;26(2):99-100.
185.	Hinnell C, Hurt CS, Landau S, Brown RG, Samuel M, Group P-PS. Nonmotor versus motor symptoms: how much do they matter to health status in Parkinson's disease? Mov Disord. 2012;27(2):236-41.
186.	Bloem BR MM. Revolutionising management of chronic disease: the ParkinsonNet approach. BMJ. 2014;348:g1838.
187.	Burn D. UK Parkinson's Excellence Network: empowering service improvement across the UK. Neurodegenerative Disease Management. 2015;5(3):173-6.
188.	British Geriatric Society. Fit forFrailty. London: British Geriatrics Society, 2014 June 2014. Report No.
189.	Stuck AE Siu AL, Wieland D, Adams J, Rubenstein LZ. Comprehensive geriatric assessment: a meta-analysis of all trials. Lancet. 1993;342:1032-6.
190.	Ellis G LP. Comprehensive Geriatric Assessment for older hospital patients. Br Med Bull. 2005;71:45-59.
191.	Donaldson L SG. Donaldson's Essential Public Health. 3 ed. Oxford: Radcliffe Publishing; 2009.

Appendix 1 NAT:PD-c

NEEDS ASSESSMENT TOOL : PROGRESSIVE DISEASE CANCER (NAT: PD-C)

COMPLETE ALL SECTIONS

PATIENT NAME: _____

DATE: _____ DIAGNOSIS: _____

PATIENT/ADDRESS LABEL

SECTION 1: PRIORITY REFERRAL FOR FURTHER ASSESSMENT

	Yes	No	If yellow boxes are ticked, consider assessment by SPCS
1. Does the patient have a caregiver readily available if required?			
2. Has the patient or caregiver requested a referral to a specialist palliative care service (SPCS)?			
3. Do you require assistance in managing the care of this patient and/or family?			

SECTION 2: PATIENT WELLBEING (Refer to the back page for assistance)

	Level of Concern			Action Taken		
	None	Some/ Potential	Significant	Directly managed	Managed by other care team member	Referral required
1. Is the patient experiencing unresolved physical symptoms (including problems with pain, sleeping, appetite, nausea, bowel, breathing or fatigue)?						
2. Does the patient have problems with daily living activities?						
3. Does the patient have psychological symptoms that are interfering with wellbeing or relationships?						
4. Does the patient have concerns about spiritual or existential issues?						
5. Does the patient have financial or legal concerns that are causing distress or require assistance?						
6. From the health delivery point of view, are there health beliefs, cultural or social factors involving the patient or family that are making care more complex?						

7. Does the patient require information about (tick any options that are relevant):
☐ The prognosis ☐ The cancer ☐ Treatment options ☐ Financial/legal issues ☐ Medical/health/support services ☐ Social/emotional issues

COMMENTS: _____

SECTION 3: ABILITY OF CAREGIVER OR FAMILY TO CARE FOR PATIENT (Refer to the back page for assistance)

Who provided this information? (please tick one) ☐ Patient ☐ Caregiver ☐ Both	Level of Concern			Action Taken		
	None	Some/ Potential	Significant	Directly managed	Managed by other care team member	Referral required
1. Is the caregiver or family distressed about the patient's physical symptoms?						
2. Is the caregiver or family having difficulty providing physical care?						
3. Is the caregiver or family having difficulty coping?						
4. Does the caregiver or family have financial or legal concerns that are causing distress or require assistance?						
5. Is the family currently experiencing problems that are interfering with their functioning or inter-personal relationships, or is there a history of such problems?						

6. Does the caregiver or family require information about (tick any options that are relevant):
☐ The prognosis ☐ The cancer ☐ Treatment options ☐ Financial/legal issues ☐ Medical/health/support services ☐ Social/emotional issues

COMMENTS: _____

SECTION 4: CAREGIVER WELLBEING (Refer to the back page for assistance)

Who provided this information? (please tick one) ☐ Patient ☐ Caregiver ☐ Both	Level of Concern			Action Taken		
	None	Some/ Potential	Significant	Directly managed	Managed by other care team member	Referral required
1. Is the caregiver or family experiencing physical, practical, spiritual, existential or psychological problems that are interfering with their wellbeing or functioning?						
2. Is the caregiver or family experiencing grief over the impending or recent death of the patient that is interfering with their wellbeing or functioning?						

COMMENTS: _____

IF REFERRAL REQUIRED FOR FURTHER ASSESSMENT OR CARE, PLEASE COMPLETE THIS SECTION

1. Referral to: (Name) _____
2. Referral to: (Specialty) ☐ General practitioner ☐ Social worker ☐ Psychologist ☐ Specialist palliative care service
   ☐ Medical oncologist ☐ Radiation oncologist ☐ Haematologist ☐ Other _____

3. Priority of assessment needed: ☐ Urgent (within 24 hours) ☐ Semi-Urgent (2-7 days) ☐ Non-Urgent (next available)
4. Discussed the referral with the client. ☐ Yes ☐ No
5. Client consented to the referral. ☐ Yes ☐ No
6. Referral from: Name: _____ Position: _____ Signature: _____

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ISSUES TO CONSIDER WHEN RATING THE LEVEL OF CONCERN

PATIENT WELLBEING

Physical symptoms
- Does the patient present with unresolved physical symptoms such as drowsiness, fatigue, dyspnoea, vomiting/nausea, pain, constipation, sleep problems, diarrhoea, or loss appetite?

Activities of daily living
- Is the patient having difficulty with toileting, showering, bathing, or food preparation?
- Is there a caregiver to assist the patient?

Psychological
- Is the patient experiencing sustained lowering of mood, tearfulness, guilt or irritability, loss of pleasure or interest in usual activities?
- Is the patient experiencing feelings of apprehension, tension, anger, fearfulness or nervousness, hopelessness or a sense of isolation?
- Is the patient requesting a hastened death?

Spiritual/Existential
- Is the patient feeling isolated or hopeless?
- Does the patient feel that life has no meaning or that his/her life has been wasted?
- Does the patient require assistance in finding appropriate spiritual resources or services?

Financial/Legal
- Are there financial concerns relating to loss of income or costs of treatment, travel expenses, or equipment?
- Is the family socio-economically disadvantaged?
- Are there conflicting opinions between patient and family relating to legal issues such as end-of-life care options and advance care plans?
- Is the patient or family aware of the various financial schemes available and do they need assistance in accessing these?

Health Beliefs, Social and Cultural
- Does the patient or family have beliefs or attitudes that make health care provision difficult?
- Are there any language difficulties? Does the patient or family require a translator?
- Is the family preventing information about prognosis from being disclosed to the patient?
- Does the information have to be passed on to a particular member of the family or cultural group?
- Is the patient or family feeling socially isolated?
- Does the family live more than 50km from the primary service provider?
- Is the patient of Aboriginal or Torres Strait Islander descent?
- Is the patient over 75 years of age? (NB: older patients are under-represented in SPCSs.)

Information
- Does the patient want more information about the course and prognosis of the disease and treatment options?
- Is the patient aware of the various care services available to assist them and do they need assistance in accessing these? (eg financial and legal assistance, psychological services, support groups, pastoral care.)

ABILITY OF CAREGIVER OR FAMILY TO CARE FOR PATIENT

Physical symptoms
- Are the patient's physical symptoms causing the caregiver and family distress?

Providing physical care
- Is the caregiver having difficulty coping with activities of daily living, medical regimes or practical issues such as equipment and transport?

Psychological
- Is the caregiver having difficulty coping with the patient's psychological symptoms?
- Is the caregiver requesting a hastened death for the patient?

Family and Relationships
- Is there any communication breakdown or conflict between patient and family over prognosis, treatment options or care giving roles?
- Is the patient particularly concerned about the impact of the illness on the caregiver or family?

Information
- Does the caregiver or family want more information about the course and prognosis of the disease and treatment?
- Is the caregiver or family aware of the care services available to assist them and do they need assistance in accessing these? (eg respite, financial and legal services, psychological services, support groups, pastoral care.)

CAREGIVER WELLBEING

Physical and psychosocial
- Is the caregiver experiencing physical symptoms eg fatigue, physical strain, blood pressure/heart problems, stress related illness, or sleep disturbances?
- Is the caregiver feeling depressed, hopeless, fearful, nervous, tense, angry, irritable or critical of others, or overwhelmed?
- Does the caregiver have spiritual/existential issues that are of concern?

Bereavement Grief (pre and post death)
- Is the caregiver or family experiencing intrusive images, severe pangs of emotion, denial of implications of loss to self and neglect of necessary adaptive activities at home or work?

Appendix 2 Analytical Framework

Tertiary code	Secondary code	Primary Code
Diagnosis	**Identifying symptoms**	
		Symptom appraisal (Pt)
		Symptom appraisal (others)
		Appraisal delay
		Presentation: misdiagnosis
		Last to notice (patient)
	Receiving a diagnosis	
		Diagnosis: reaction
		Misdiagnosis
		Diagnosis: information needs
		Diagnosis: impact on carer
		Informing others (diagnosis)
		ER: diagnosis
Emotional response	**ER: diagnosis**	
		Injustice
		Relief at diagnosis
		Denial of diagnosis
		Impact of diagnosis
		Hope (cure)
	ER: Parkinson's	
		Accounting for symptoms
		Brave face
		Biographical disruption
		New roles
		Definition of self
		Self-validation / justification
		Rationalisation
		Emotional response to symptoms
		Emotional response to disease
		Blame: patient
		Thinking too much
		Response to disease (psychiatric)
	Fear	
		Fear: symptoms
		Fear: diagnosis (carer)
		Fear: financial
		Fear: health care
		Fear for future
		Fear of unknown
		Blame: carer
		Humour
		Carer anger
		Changed personality
		Less strong character
		Patient as prism
		Carer frustration
		Stubborn (pt)
Care and Carer		Carer pride

	Carer: Individual	
		Carer bereavement
		Biographical disruption (carer)
		Effect on family
		Effect on relationship
		Family breakdown
		Carer as guardian
	Carer: Vigilant protector	Carer as expert
		Carer vigilance
		Carer as therapist
		Carer as protector
		Carer: responsible
		Carer as guide
		Loyalty tension
		Approach to cognitive decline (pos)
		Approach to cognitive decline (neg)
		Impact of gender
		Physical impact of care
	The care process	Accepting care
		Maintaining independence
		Care tension
		Dealing with decision making
		Protecting family
		Early days
		Health beliefs; prognosis
	HB: disease	Health beliefs: medication
		Health beliefs: natural hx
		Contrast with cancer
Health beliefs		
		Medication avoidance
		Fear of medication
	HB: medication	Medication tension (Pos)
		Medication tension (Neg)
		Medication: adverse events
		Medication
		Future:limbo
		Anticipating decline
		Approach to planning (pos)
Viewing the Future	**Planning**	Approach to planning (neg)
		Approach to planning (hindsight)
		Future planning: triggers
		Future planning: timing
		Care planning
		Planning: past experience

Appendix 2 Analytical Framework (cont. page 3)

	Impact of disease	Motor fluctuations
		Physical limits
		Impact of symptoms: hallucinations
		Watershed moment
	Information	Information needs
		Info needs: Natural Hx
		Misinformation
		Information sources
		Missing information
		Information tension
		Information dissonance
		Don't want to know (future)
		Face it when it comes
		Attitude to info (pos)
		Attitude to info (neg)
		Impact of information
Being supported	**Health care services**	
		Experience: HCP's
		Experience: Service
		Experience: service - MDT
		Experience diagnosis
		Experience: medication
		Expert monitoring
	Support	
		Support: professionals
		Support: charity
		Accessing support
		Attitude to support groups
		Downward comparison
		Coping
Other		Approach to social situations
		Stigma

Appendix 3 Steps in framework analysis

Step 1: Familiarisation

A stage common to all modes of qualitative analysis, this involves gaining an intimate knowledge of the raw data, through reading and re-reading the primary source material, usually transcripts. Researchers are encouraged to make notes and begin to note themes and, to consider *a priori* knowledge or themes brought to the project. This is a key difference to Grounded Theory, where pre-held beliefs are suspended, so themes emerge purely from the data.

Step 2: Indexing

Indexing is a process analogous to coding, during which sections of data are identified under one or more codes to facilitate subsequent analysis. The approach to indexing is less prescriptive than the "line by line" coding advocated by Charmaz and allows, but does not require the use of participant language – so called "source coding".

Step 3: Charting

The next step in organising data ahead of detailed analysis, involves the creation of charts, which map participants or groups against a thematic framework. The framework may have emerged entirely from the data (inductive), or include some pre-specified categories (deductive). In addition to ease of data manipulation, this allows subsequent themes to be traced directly back to the primary source material.

Step 4: Mapping and interpretation

The final analytical phase, where data and emergent themes can be compared within or between groups, allows patterns and divergent cases to be identified and scrutinised. The formation of charts in stage 3 increases the transparency of this phase and is one of the main attractions of framework in AHR, allowing non-practitioners some insight into the analytic process as opposed to the potently closed process by which themes "emerge" in other qualitative disciplines.

Appendix 4 Study Eligibility Form

Study Title:

Study Identity Number:

	Yes	No	Unclear
Is the study design Qualitative?			
Does the study include patients **OR** carers with PD?			
Does the study examine palliative / supportive care needs?			
If the study includes other diseases (ie PSP/MSA) can the participants with PD be distinguished?			

Outcome (circle):   **Include**   **Exclude**   **Unclear**

Appendix 5 SLR Data Extraction tool

<u>SLR Data Extraction Tool</u>

Study Title:

Study identity number:

Setting:

Design: Interview   FG   Other:

Research Aim:

Sample:

Patients: Male   Female   Ratio: Age range:

Carers: Male   Female   Ratio: Age range:

Disease stage Early   Late   Mixed   Bereaved   Not clear

Methodology used:
Quality assessment:
General comments:

NAT Construct	Identified Theme from Primary Study	Associated Quotes
Unresolved physical symptoms		
Problems with ADL's		
Psychological symptoms		
Spiritual / existential issues		
Financial / legal issues		
Carer / family distress		
Bereavement issues		
Requirement for information		
Other		

Appendix 6 Example RTA grid paper 2

Primary codes	Quote	Initial Concepts
Subjugation of carer health needs	"I took ill, it was probably exhaustion. . . the doctor put him in for two weeks' respite while I was in hospital"	**Subjugation of carer needs (Health / Psycho-social)**
Psycho-social impact on carer – loss of "self, loss of control, "helplessness		
Care tension – recognise need for help but reluctant to relinquish care to another person		**Care Tension**
Surprise at rate of decline / *lack of preparation for death and advanced stages.*	'I knew he was deteriorating but I didn't expect him to die too soon.'	**Role of Faith**
Role of spiritual support	'I think that [having a faith] is what carried me through.'	
Post bereavement sense of abandonment Need for bereavement support	'You know, really in a way you're grieving before they even die, whenever they have changed so much. Whenever my dad had changed so much, you are really grieving away five years before it of what they have lost. You maybe need help to come to terms that they have changed.' (FC5)	**Loss of life roles (1 – Spouse to carer, 2- Carer to bereaved)**
Loss of purpose (carer) post bereavement	'I was depressed around the time and still am. . . I find I miss them so much. Really they are part of your life for so long.'	**Poor Knowledge / Unprepared**
lack of preparation for death and advanced stages Some unaware PD incurable		
Poor knowledge of available services "*lack of signposting*"		
Consultations with specialist focused on medication – neglect of psycho-social.	'The neurologist saw him every six months and agreed the tablets; they didn't have a lot of time.' (FC10)	**Biomedical focus of consultations**
Poor communication between professionals	'. . . it was frustrating, very frustrating because you were the liaison with the health people, with the GP and you were at them to constantly to go back and say this is not working	**Dissatisfaction with clinicians**
Specialist palliative care rarely mentioned	'It's only for cancer [The Hospice], isn't it?'	
Barriers to palliative care: -Carers difficulty discussing end of life issues -Lack of knowledge -Lack of referral by professionals		**Information Tension**

Appendix 7 Example grid RTA 1 (Initial concepts in to RTA constructs)

RTA constructs	Initial concepts
Subjugation of Carer needs	Need for support (Formal and Informal, Spiritual)
	Occurrence of Trigger events
	Loss of "life role"
Care Tension	Care as "duty"
	Guilt Re: accepting help
	Benefits of care
Financial Hardship	Loss of Job
	Cost of care
Poor Knowledge	Disease specific knowledge
	Care Specific knowledge
	Service Specific Knowledge
	Information Tension
Clinical dissatisfaction	Biomedical focus of consultations
	Doctor as prescriber
	Poor communication between HCP's
	Negative experience of diagnostic process

Appendix 8 – summary of full text study selection

Study number	Study Title	Outcome Final
1	An analysis of the needs of homebound patients with Parkinson's disease and their caregivers in the Indian subcontinent - A qualitative study	**Exclude** Unable to contact authors for full data set
3	Are supportive services meeting the needs of Australians with neurodegenerative conditions and their families.	**Exclude** Data for participants with PD could not be extracted
5	Caregiver-burden in Parkinson's disease is closely associated with psychiatric symptoms, falls, and disability	**Exclude** Study design not qualitative
9	Couples living with Parkinson's disease: Needs and concerns at advanced to end stage	**Exclude** conference abstract only – author contacted but unable to give more detail
12	Effects of PD on Quality of Life of patients' spouses: a qualitative survey	**Exclude** Article in French
14	How general practice can help improve care of people with neurological conditions: a qualitative study.	**Exclude** Data for participants with PD could not be extracted
18	Older Swedish women's experiences of living with symptoms related to Parkinson's disease	**Exclude** Does not examine palliative or supportive care need
19	Palliative care and support for people with neurodegenerative conditions and their carers	**Exclude** Study design not qualitative
22	Perceptions of persons with PD, family and professionals on quality of life: an international focus group study.	**Exclude** Does not examine palliative or supportive care need
23	Symptom prevalence and severity of non-motor symptoms in people affected by advanced Parkinson's disease and related disorders. Can palliative care help?	**Exclude** Study design not qualitative
24	The effectiveness of palliative care in the later stages of MND/ALS	**Exclude** Data for participants with PD could not be extracted
25	The symptom experience of patients with Parkinson's disease	**Exclude** Study design not qualitative

Appendix 9 – Summary of included studies (page 1 of 2)

Study title	Country	Recruitment setting	Participants	Disease stage	Methodology	Method
An exploration in to the palliative and end of life experiences of carers of people with Parkinson's disease (14)	UK (N. Ireland)	Community	**Carers**	Bereaved	Content Analysis	Interviews
Being in the light or being in the shade: persons with PD and their partners' experience of support (96)	Sweden	Secondary care	**Patients and Carers**	Not clear	Content analysis	Interviews
Caring for Family with Alzheimer's Disease and Parkinson's Disease(95)	USA	Primary care and PD support groups	**Carers**	Not clear	Content analysis	Interviews
Changing perceptions of womanhood: living with Parkinson's Disease (97)	UK	Primary and Secondary care	**Patients (female only)**	Mixed (all young – pre menopause)	Unclear	Descriptive case studies
Continuity challenges of PD in middle life (94)	USA		**Patients**	Early	Paradigm cases	Interviews
Culturally competent care for people with Parkinson's disease (98)	USA	PD support groups	**Patients**		Ethnography	Interviews
Dropping the bomb: the experience of being diagnosed with PD (99)	USA	PD support groups	**Patients**	Mixed	Content analysis	
Family caregiver's decision process to institutionalize persons with PD: a grounded theory study (100)	USA	Tertiary care	**Carers**	Mixed	Grounded Theory	Interviews
Living and coping with PD: Perceptions of informal carers (13)	UK (N. Ireland)		**Carers**	Not clear	Content Analysis	Interviews

Living with Parkinson's disease: Elderly patients' relatives' perspectives on daily living (101)	Sweden	Secondary care	**Patients and Carers**	Mixed	Phenomenology	Interviews
Moving towards patient centred healthcare for patients with PD (102)	Holland	Community and secondary care	**Patients and carers**	Early	Grounded theory (constant comparison)	Focus groups
Palliative stage Parkinson's disease: patient and family experiences of health-care services (12)	Canada	Tertiary care	**Patients and Carers**	Late	Phenomenology	Interviews
Parkinson's Disease: Barriers and Facilitators to Optimizing Function (103)	USA	Community	**Patients and carers**	Not clear	Grounded theory	Interviews
Would people with Parkinson's disease benefit from palliative care? (9)	Australia	PD support groups	**Patients and carers**	Mixed		Interviews
Day to Day demands of Parkinson's Disease (93)	USA	Secondary care and support groups	**Patients**	Early	Phenomenology	Interviews

Appendix 10 – Study approvals: Qualitative

NHS
Health Research Authority

NRES Committee Yorkshire & The Humber - Bradford
Yorkshire & Humber REC Office
Millside
Mill Pond Lane
Meanwood
Leeds
LS6 4RA

Telephone: 0113 30 50128
Facsimile: 0113 85 56191

09 August 2012

Dr Edward W Richfield
Research Fellowship
Hull York Medical School
Hertford Building/Cottingham campus
Hull York Medical School
Cottingham Road, Hull
HU6 7RX

Dear Dr Richfield

Study title:	**Assessing Palliative Care needs in Parkinson's Disease: Developing a needs assessment tool**
REC reference:	**12/YH/0332**

Thank you for your letter of 07 August 2012, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

Conditions of the favourable opinion

The favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission or approval must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission ("R&D approval") should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements.

Guidance on applying for NHS permission for research is available in the Integrated Research Application System or at http://www.rdforum.nhs.uk.

Where a NHS organisation's role in the study is limited to identifying and referring potential participants to research sites ("participant identification centre"), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of approvals from host organisations

1. **Submit the invitation letter and consent forms on headed paper.**

2. **Ensure that statement 1 in the consent forms refer to the most recent version of the information sheet i.e. version 1.1 dated 07.08.2012**

You must notify the REC in writing once all conditions have been met (except for site approvals from host organisations) and provide copies of any revised documentation with updated version numbers.

The REC will acknowledge receipt and provide a final list of the approved documentation for the study, which can be made available to host organisations to facilitate their permission for the study.   Failure to provide the final versions to the REC may cause delay in obtaining permissions.

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Evidence of insurance or indemnity		14 October 2011
Investigator CV	1	11 June 2012
Letter from Sponsor		31 May 2012
Letter of invitation to participant	1.1	07 August 2012
Other: Letter to GP		11 June 2012
Other: Interview Schedule	1	11 June 2012
Other: Focus Group- Topic Guide	1	11 June 2012
Participant Consent Form: Patient	1.1	07 August 2012
Participant Consent Form: Carer	1.1	07 August 2012
Participant Information Sheet: Interview	1.1	07 August 2012
Participant Information Sheet: Focus group	1.1	07 August 2012
Protocol	1	11 June 2012
REC application		12 June 2012
Response to Request for Further Information		07 August 2012

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

<u>Reporting requirements</u>

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

<u>Feedback</u>

You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.
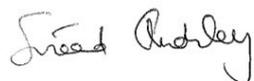
Further information is available at National Research Ethics Service website > After Review

12/YH/0332	Please quote this number on all correspondence

With the Committee's best wishes for the success of this project

Yours sincerely

[signature]

Dr Ian Woollands
Chair

Email: sinead.audsley@nhs.net

Enclosures: "After ethical review – guidance for researchers"

Copy to: *Professor John Hay, University of Hull*
               Mr James Illingworth, Hull And East Yorkshire Hospitals NHS Trust

Appendix 11 NAT: Parkinson's disease

NEEDS ASSESSMENT TOOL: PARKINSON'S DISEASE

Please complete all sections

Guidance for completion on reverse

Date:

Name:

(Or attach address label)

Section 1a: Red Flags – If present, be alert for unmet palliative care need:	Y	N	
Red flag symptoms? (Persistent hallucinations / ≥2 Falls / Aspiration / Hoehn and Yahr 3 -see guidance for details)			If yellow boxes ticked **consider** need for in depth palliative assessment +/- involvement of SPCS
Admission to residential or nursing care?			
Failure to attend clinic today?			
Section 1b: Priority referral for further assessment:	Y	N	
No carer?			
Patient or carer request referral to SPCS?			
You require assistance of SPCS?			

Comments………

Section 2: PATIENT WELLBEING ("Does the patient have……")	Level of Concern			Action Taken		
Who provided this information? Patient ☐ Carer ☐ Both☐	None	Some/ Potential	Significant	Directly Managed	Refer to other team member	Refer to SPCS
Unresolved physical symptoms? (Motor / Non-motor)						
Unresolved psychological or neuropsychiatric symptoms?						
Problems with daily living activities?						
Spiritual or existential concerns?						
Financial or legal concerns?						
Health beliefs, cultural or social factors making care delivery complex?						
Information needs: Prognosis ☐ Diagnosis ☐ Treatment options ☐ Financial/legal issues ☐ Support services ☐ Social/emotional issues ☐						

Comments………

Section 3: ABILITY OF CARER OR FAMILY TO CARE FOR PATIENT ("Is the Carer / Family…….")	Level of Concern			Action Taken		
Who provided this information? Patient ☐ Carer ☐ Both☐	None	Some/ Potential	Significant	Directly Managed	Refer to other team member	Refer to SPC
Distressed about the patient's symptoms? (Motor / Non-motor / neuro-psychiatric)						
Having difficulty providing physical care?						
Having difficulty coping?						
Concerned about financial or legal issues?						
Experiencing problems that are interfering with inter-personal relationships or functioning?						
Information needs: Prognosis ☐ The diagnosis ☐ Treatment options ☐ Financial/legal issues ☐ Support services ☐ Social/emotional issues ☐						

Comments………

Section 4: CARER/FAMILY WELLBEING "Carer or family experiencing……."	Level of Concern			Action Taken		
	None	Some/ Potential	Significant	Directly Managed	Refer to other team member	Refer to SPC
Problems that are interfering with their wellbeing or functioning?						
Grief over the impending or recent death of the patient?						

Comments………

IF FURTHER ASSESSMENT REQUIRED, PLEASE COMPLETE THIS SECTION:

Referral to: Name…………………………………………………….

Speciality: Neurology ☐   Elderly care☐   PD Nurse Specialist☐   OT ☐   PT☐   SLT ☐   Social Services ☐   SPCS☐   Other ☐

Client aware of referral: Y☐ N☐   **Client agrees to referral:** Y ☐ N☐

Referral from: Name………………………………   Position…………………………   Signature……………………………

ISSUES TO CONSIDER WHEN RATING LEVEL OF CONCERN

RED FLAGS – If present consider further assessment by own team +/- SPCS if required

Visual hallucinations –"yes" if hallucinations are formed and persistent. Do not include if associated with inter-current illness or medication change.

Recurrent falls –"yes" if multiple (>2) falls have occurred. Single, isolated falls, even in the context of injury, should not be included here.

Pneumonia / choking – May indicate aspiration and should trigger further palliative review in addition to SLT where appropriate.

Hoehn and Yahr stage 3 (Bilateral disease, mild to moderate disability and impaired postural reflexes) is associated with reduced quality of life.

24 hour care – admission to either residential or nursing care should trigger exploration of further supportive and palliative care needs.

Failure to attend clinic – may indicate physical or social difficulties, change in circumstance or acute admission preventing attendance.

PATIENT WELLBEING

Physical symptoms

Motor: Difficulty using legs (Freezing, Falls, Shuffling, Slowness) or difficulty using arms (freezing, slowness, dyskinesia, motor fluctuations)

NMS: Fatigue, drowsiness, Pain, Constipation, Poor sleep, Urinary urgency, frequency, or incontinence, swallowing difficulty, SOB, Drooling, Spasms.

Activities of daily living

• Is the patient having difficulty with toileting, showering, bathing, or food preparation?

• Do they require more information to maximise their daily function –see below

Neuro-psychiatric / Psychological - "Does the patient have…."

• Thinking or memory problems, which interfere with wellbeing / relationships?

• Hallucination or behavioural issues which require assistance or evidence of psychosis?

• Sustained lowering of mood, tearfulness or guilt? Loss of pleasure in usual activities? Feelings of anxiety, apprehension, anger or fearfulness?

• Is the patient struggling with the implications of, or emotional response to the diagnosis?

• Is the patient requesting a hastened death?

Spiritual/Existential – "Is the patient…."

• Feeling isolated or hopeless?

• Feeling that life has no meaning or that his/her life has been wasted?

• Having difficulty thinking about the future?

• Requiring assistance in finding appropriate spiritual resources or services?

Financial/Legal concerns

• Consider loss of income, costs of treatment, travel expenses, equipment, or future care needs (such as residential care)?

• Is the family socio-economically disadvantaged?

• Is the patient or family aware of the various financial schemes available and do they need assistance in accessing these (e.g. social worker)?

• Are there conflicting opinions between patient and family relating to legal issues such as end-of-life care options and advance care plans?

Health Beliefs, Social and Cultural – "Does the patient or family…"

• Have beliefs or attitudes that make health care provision difficult – for example believing that palliative / hospice care is not available to them?

• Have communication difficulties – consider language and disease related issues (hypophonia / freezing of speech)?

• Feel socially isolated? If so are they avoiding peer support groups due to concerns such as "downward comparison"?

• Need information passed on to a particular member of the family or cultural group?

• Want information about prognosis to be withheld from the patient, or are they reluctant to discuss prognosis? If so, has this been explored?

• Have logistical difficulties accessing services (distance, transport, cost)?

Information – "Is the patient aware of/that…."

• Available services and do they need assistance accessing these? (e.g. financial /legal assistance, psychological services, support groups, pastoral care.)

• Advance Care Planning (ACP) and have their views / attitudes towards it been explored?

• PD is progressive, incurable and shortens life?

• Does the patient want more information about the course and prognosis of the disease and treatment options?

ABILITY OF CARER OR FAMILY TO CARE FOR PATIENT

Physical symptoms

• Are the patient's physical symptoms causing the carer and/or family distress?

Providing physical care

• Is the carer having difficulty coping with activities of daily living, medical regimes or practical issues such as equipment and transport?

• Have they received all the practical information they require?

Neuro-psychiatric / Psychological – "Is the carer / family…"

• Having difficulty coping with the patient's memory problems, hallucinations or behavioural issues?

• Having difficulty coping with the patient's psychological symptoms (esp. anxiety and depression)?

• Requesting a hastened death for the patient?

Family and Relationships

• Is there any communication breakdown or conflict between the patient and family over prognosis, treatment options or care giving roles?

• Is the patient particularly concerned about the impact of the illness on the carer or family?

• Is the disease having an adverse effect on the relationship between patient and carer? (May wish to consider impact of physical, psychological and personal cares, sexual dysfunction and change to previous roles within relationship.)

Information – "Do/are the carer or family…"

• Require more information about the course and prognosis of the disease and treatment?

• Aware of available services/ need assistance accessing these? (e.g. Financial/legal psychological services, support groups, pastoral care.)

• Are the information needs of the patient and family congruous?

CARER AND FAMILY WELLBEING - "Do the carer or family…"

Physical and psychosocial

• Experience physical strain, ill health, fatigue, disturbed sleep? Is there evidence of anxiety, depression or feelings of hopelessness?

• Have spiritual/existential issues that are of concern?

• Currently feel that caring has a net positive or negative affect for them personally and their relationship with the patient?

Grief (pre and post death)

• Experience intrusive images, severe emotion, denial of implications of loss to self and neglect of necessary adaptive activities at home or work?

• Know of the progressive nature of PD? Has future care planning been considered? (If patient not capacitous this may include best interest decisions)

• Have access to support services (Such as PD Nurse Specialist, SPC, Local support groups, post bereavement support)?

Appendix 12 – Study approvals: Quantitative

NHS
Health Research Authority
NRES Committee Yorkshire & The Humber - Bradford

Yorkshire & Humber REC Office
Millside
Mill Pond Lane
Meanwood
Leeds
LS6 4RA

Telephone: 0191 4283545

21 March 2013

Dr Edward W Richfield
Research Fellowship
Hull York Medical School
Hertford Building/Cottingham campus
Hull York Medical School
Cottingham Road, Hull
HU6 7RX

Dear Dr Richfield

Study title:	**Assessing palliative care needs in Parkinson's disease: developing a needs assessment tool - Clinimetric testing of the NAT-Parkinson's disease.**
REC reference:	13/YH/0006
IRAS project ID:	114750

Thank you for your letter of 18th March 2013, responding to the Committee's request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the NRES website, together with your contact details, unless you expressly withhold permission to do so. Publication will be no earlier than three months from the date of this favourable opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to withhold permission to publish, please contact the Co-ordinator Hayley Jeffries, hayley.jeffries@nhs.net.

Confirmation of ethical opinion
On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Mental Capacity Act 2005
I confirm that the committee has approved this research project for the purposes of the Mental Capacity Act 2005. The committee is satisfied that the requirements of section 31 of the Act will be

A Research Ethics Committee established by the Health Research Authority

met in relation to research carried out as part of this project on, or in relation to, a person who lacks capacity to consent to taking part in the project.

Ethical review of research sites
NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

Document	Version	Date
Covering Letter		18 March 2013
Covering Letter		18 March 2013
Evidence of insurance or indemnity	Certs 1-3	06 December 2012
GP/Consultant Information Sheets	1.0	24 October 2012
Investigator CV		30 November 2012
Letter from Sponsor		31 May 2012
Other: Summary CV - M Johnson		30 November 2012
Other: Feasibility assessment for proxy completion of NAT-Parkinson's disease	1.1	06 November 2012
Other: Letter to GP	1.1	20 February 2013
Other: NAT: PD-C		
Participant Consent Form: Carer	1.2	20 February 2013
Participant Consent Form: Patient	1.2	20 February 2013
Participant Consent Form: Consultee	1.0	20 February 2013
Participant Consent Form: Feasibility	1.1	20 February 2013
Participant Information Sheet: Feasibility	1.1	31 October 2012
Participant Information Sheet: Carer	1.2	20 February 2013
Participant Information Sheet: Patient	1.3	20 February 2013
Participant Information Sheet: Consultee	1.2	20 February 2013
Participant Information Sheet	Patient	16 March 2013
Participant Information Sheet	1.4 - Patient	16 March 2013
Protocol	1.2	20 February 2013
Questionnaire: PDQ-39		
Questionnaire: Caregiver Strain Index		
Questionnaire: MDS-UPDRS parts 1 and 2		
Questionnaire: SCOPA-PS		
REC application		04 March 2013
Response to Request for Further Information		
Response to Request for Further Information		

Statement of compliance
The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review
<u>Reporting requirements</u>

The attached document *"After ethical review – guidance for researchers"* gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The NRES website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

<u>Feedback</u>
You are invited to give your view of the service that you have received from the National Research Ethics Service and the application procedure. If you wish to make your views known please use the feedback form available on the website.

Further information is available at National Research Ethics Service website > After Review

13/YH/0006	Please quote this number on all correspondence

We are pleased to welcome researchers and R & D staff at our NRES committee members' training days – see details at http://www.hra.nhs.uk/hra-training/

With the Committee's best wishes for the success of this project.

Yours sincerely

pp

Dr Ian Woollands
<u>Chair</u>

Email:hayley.jeffries@nhs.net

Enclosures: "After ethical review – guidance for
                              researchers" *[SL-AR2]*

Copy to: Professor John Hay, Cottingham Road, HU6 7RX

                              Mr Damon Foster, York Foundation Trust R&D Unit

Appendix 13 Summary and synthesis of thesis findings

	What are the palliative and supportive care needs associated with PD?	What were the clinimetric properties of the adapted tool?	Overall synthesis
SLR and synthesis	**Information tension**: dynamic state created by desire for information, to address present / future needs and maintain hope. Exists within and between individuals (i.e. patient and carer). **Care tension**: dynamic care state, responds to patient / carer needs, abilities, and perceptions of acceptability. **Negotiation**: resolution of tensions within and between individuals may be explicit or implicit and impacts the unmet needs of people with PD. **Clinician**: importance of patient centred approach. Inaction is not neutral.	Not appropriate	
Qualitative study	**Diagnosis**: associated needs Re: information / patient reaction. Target for improved practice. **Response to PD**: affects ability to assimilate and hence later needs. Fear prominent in carers. **Care:** i) vigilant protector, ii) care dynamic – *asking for help* and *taking over* vs *pushing*. **Information and planning**: crucial to assess and meet needs, wide implications, hard if discordant patient/carer needs. **Health beliefs**: role in medication avoidance and perceived role of clinical team. **Support:** downward comparison a barrier. Disease milestones: frame disease experience	Not appropriate	
Adaptation and Content validation	Identification of individuals at high risk of unmet palliative care need via "red flags" Broad agreement can be reached by a range of clinicians, regarding the nature of unmet care need and the qualities required in a screening tool.	Confirmed face and content validity of NAT:Parkinson's disease, amongst a range of clinical raters from diverse backgrounds.	
Clinimetric (construct)	*Study not designed to answer this question (sequential mixed methods).* Useful to observe the spread of unmet need across the spectrum of domains covered by the NAT:Parkinson's disease.	Fair to moderate levels of construct validity Evidence for construct validity of some areas less well demonstrated, result of broad constructs and balance between practical considerations (usability) and clinimetric accuracy.	
Clinimetric (reliability)	*Study not designed to answer this question (sequential mixed methods).* Useful to observe that independent raters can identify the same unmet palliative care needs in the same patient. This strengthens the idea that palliative care needs exist in PD, which is not always accepted.	Fair to moderate levels of inter-rater reliability Tool can be applied by variety of raters, with minimal training, provided a good knowledge of PD.	
Summary	**1**- Confirmed that palliative care needs exist in PD and are present from diagnosis. Potential to affect all domains of life, both patient and carer and to fluctuate over time. **2**- Qualitative framework for understanding **3**- "Milestones" – opportunity to screen / discuss **4**- Information tension / Care tension – key **5**- Require responsive services and clinicians **6**- Support where discordant needs exist **7**-Needs can be identified by multiple independent raters using a screening tool.	Successful adaptation of NAT for Parkinson's disease Fair to moderate clinimetric properties Fit for purpose as a means of identification and triage of unmet palliative care need – never intended as a comprehensive quantitative assessment of need. Suitable for everyday use by range of clinicians Minimal training required	**1**- Removed conjecture Re: palliative care need in PD **2**- Established presence of need from diagnosis. **3**- Suggesting key disease milestones as a time for evaluation. **4**- Successful adaptation of the NAT:Parkinson's disease, with support for clinimetric properties. **5**- Combining phase 1 (qualitative) and phase 2 (clinimetric) gives insight to clinical application: i) The need for repeated assessment during PD journey, ii) Potential triggers for re-assessment (milestones), iii) Requirement for clinicians to improve communication esp. diagnosis, iv) Need to create responsive services.

Abbreviations Used in this thesis

AHR	Applied Health Research
CGA	Comprehensive Geriatric Assessment
LoA	Lines of Argument
NAT	Needs Assessment Tool
NAT:PD-c	Needs Assessment Tool: Progressive Disease – cancer
NAT:HF	Needs Assessment Tool: Heart Failure
PD	Parkinson's disease
RTA	Reciprocal translational analysis
SD	Standard Deviation
SPC	Specialist Palliative Care