

**Submitted in partial fulfilment of the Lancaster University Doctorate in Clinical
Psychology**

December 2020

Doctoral Thesis

The experience of living with a neurodegenerative condition

Gina Wieringa

Doctorate in Clinical Psychology

Division of Health Research

Lancaster University

All correspondence should be sent to:

Gina Wieringa

Doctorate in Clinical Psychology

Furness Building

Lancaster University

Lancaster

LA1 4YG

Email: g.wieringa@lancaster.ac.uk

Word Count			
	Main Text	Appendices (inc. Tables, figures, references)	Total
Thesis Abstract	241	n/a	241
Literature Review	7997	11992	19,989
Research Paper	7968	5512	13,480
Critical Appraisal	3974	1159	5133
Ethics Section	5469	4823	10292
Total	25,649	23,486	49,135

Thesis Abstract

This thesis outlines three pieces of exploration relating to the experience of living with a neurodegenerative condition: a meta synthesis of the literature on how individuals adjust to a diagnosis of Parkinson's disease (PD); an interpretive phenomenological analysis of the experience of 10 individuals living in the pre-manifest stage of Huntington's disease (HD) and a critical analysis incorporating my reflections on the research.

In the first section, the meta-synthesis is described, which reviewed 20 papers exploring how an individual adjusts to a diagnosis of PD. Three main themes emerged relating to the adjustment process: 'maintaining coherent sense of self', 'feeling in control' and 'holding a positive mindset'. The findings provide insight into how an individual manages the challenges faced by their condition and the importance of maintaining a positive mindset. The review also discussed clinical implications, including ways to empower individuals with PD to self-manage their condition.

The second section outlines the empirical research, in which 10 individuals living in the pre-manifest stage of HD were interviewed. Three major themes emerged: 'feeling limited by time'- "trigger for a countdown", 'the perception of stalling time'- "I have no intention of becoming symptomatic" and 'making the most of time'- "I could be hit by a bus tomorrow".

In the final section, the critical analysis considers the similarities and differences between the two papers and the strengths and limitations of each piece of research. The researcher motivations are also discussed, and suggestions are made for future research.

Declaration

This thesis outlines research undertaken between October 2019 and November 2020 as part requirement of the Lancaster University Doctorate in Clinical Psychology. The work documented here is my own except where due reference has been made in the text. This thesis has not been submitted for an award of a higher degree elsewhere.

Signature: *gina wieringa*

Print name: Gina Wieringa

Date: 18 December 2020

Acknowledgements

I would like to thank the participants who took part in the empirical study for being so open in sharing their experiences with me. I would also like to thank Dr Fiona Eccles and Dr Maria Dale for the supervision they provided throughout the doctoral program and for all the comments on my drafts, which helped me to formulate my thoughts and express them on paper.

I would like to thank my parents, my brother and my twin sister for their unconditional support throughout the whole of my doctorate, and for giving me the opportunity to get onto the doctorate.

Finally, I want to thank my grandparents, particularly my grandma, whose resilience in the face of having multiple sclerosis inspired me to do this doctoral thesis and taught me much about living well despite adversity.

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**Section 1: Adjusting to Living with Parkinson's Disease; a meta-ethnography of qualitative
research**

Lancaster University

Doctorate in Clinical Psychology

2018 Intake

Word count:

7997 words (excluding title page, references, figures and appendices)

Prepared in accordance with Instructions for Authors for '**Psychology & Health**'

All correspondence should be sent to:

Gina Wieringa

Doctorate in Clinical Psychology

Furness Building

Lancaster University

Lancaster

LA1 4YG

Email: g.wieringa@lancaster.ac.uk

Abstract

Parkinson's disease is a progressive, neurodegenerative condition, which causes a range of physical difficulties including tremor, rigidity, slowness of movement and postural instability. Psychological difficulties are also common. Adjustment is thus required to manage the effects of the condition and this review presents a synthesis of research in this area. A systematic search of three databases (MEDLINE, CINAHL and PsycINFO) was carried out. After exclusion based on eligibility criteria, 20 articles were included when analysing the data. Three main themes are proposed relating to the process of adjustment: 'maintaining a coherent sense of self', 'feeling in control' and 'holding a positive mindset'. Although many of the studies described challenges of living with PD, the results are dominated by the determination of individuals to self-manage their condition and maintain positive wellbeing. The results highlight the need to empower patients to self-manage their illness, mitigating the effects of Parkinson's disease and supporting future wellbeing.

Keywords: Adjustment; Meta-ethnography; Parkinson's disease; Psychological support; Wellbeing

Introduction

PD is a debilitating, progressive condition normally diagnosed after the age of 50 although onset at a younger age is also possible. In 2016, 6.1 million individuals had Parkinson's disease globally, compared with 2.5 million in 1990, reflective of increasing numbers of older people, longer disease duration and environmental factors (Dorsey et al., 2016). The symptoms of PD are wide-ranging, but individuals typically start with motor difficulties including tremor, rigidity, bradykinesia and loss of postural reflexes (Montel, 2009). Other physical symptoms commonly experienced include excessive saliva and dribbling, urinary urgency and constipation (Khoo et al., 2013). These symptoms frequently cause difficulties for an individual to carry out daily activities and may contribute to activity limitations (Smith & Shaw, 2017). Furthermore, psychological difficulties are also common. The most commonly experienced psychological difficulty in PD is depression; although prevalence rates vary between 2 and 67%, the average prevalence is about 35% (Reijnders et al., 2008). Anxiety is also frequently experienced and substantial anxiety is believed to occur in up to 40% of individuals with PD (Walsh & Bennett, 2001). Anxiety and depression co-occur quite often and there is evidence to suggest that anxiety may cause a significant deterioration in physical symptoms, which may in turn exacerbate anxiety (Schneider et al., 2008). Estimates of the prevalence of psychotic symptoms in individuals with PD vary from 15 to 52% in those who receive treatment (Schneider et al., 2008). In the early stages, psychosis in PD typically occurs with a high level of insight, and psychological support can be sufficient in managing symptoms. However, psychotic symptoms are typically recurrent and likely to worsen over time, resulting in significant anxiety and increasing the likelihood of social and functional impairment (Wint et al., 2004). Additionally, apathy occurs in approximately 40% of individuals with PD (Starkstein et al., 2009) and can

occur independently of depression and cognitive impairment (Kirsch-Darrow et al., 2006). Finally, impulse control disorders (ICDs) are estimated to occur in around 13% of individuals and are associated with greater functional impairment in activities of daily living compared with those with PD with no ICDs, despite similar symptom severity (Voon et al., 2011).

Given the extent of the psychological difficulties of PD, and ample research which documents the relationship between psychological difficulties and excess disability, worse quality of life, poorer outcomes and increased caregiver burden (Weitraub & Burn, 2011), it is essential to better understand the factors which might facilitate an individual's adjustment to PD. Despite growing literature in the area, there is still little consensus on what constitutes psychological adjustment, especially with regard to individuals living with a chronic illness. Typically, literature has framed the concept as both a *state* and a *process* (Hammond & Winthrop, 2018). Adjustment as a state can be considered the outcome of an individual adapting to their circumstances. Optimal adjustment is when the person manages to balance the difficulties of their condition with a good quality of life (Sharpe & Curran, 2006), resulting in good physical, cognitive and emotional functioning (Hammond & Winthrop, 2018). The state of adjustment is normally defined according to objective measurement such as low negative affect and maintaining function, but this may not necessarily reflect subjective considerations of an individual (Stanton et al., 2007).

An alternative conceptualisation is to recognise that adjustment is likely to involve a process of adaptation, which is iterative and dynamic, the end goal of which might be to reach the state of living well with PD. Defining adjustment as a process enables one to consider the idiosyncratic nature of how an individual adapts to a chronic illness, including how this process may not be captured by functional outcomes (Moss-Morris, 2013). Considering adjustment as a

process acknowledges how adjustment is likely to be condition specific and that the adaptive tasks required in adjusting to a condition will vary according to the specific illness challenges. It also recognises that the process of adjustment will differ according to specific time points in the individual's journey with their disease. For instance, there is evidence to suggest that physical aspects of the disease, such as severity of motor symptoms, may be linked to psychological difficulties such as anxiety and depression (Sagna et al., 2014). Physical aspects of the disease alongside psychological aspects such as anxiety and stress also decrease an individual's ability to engage in social activity, increasing their social isolation and contributing to depression (Hartley et al., 2014). How an individual is able to adjust to their condition is clearly integrated with both the physical and psychological aspects of the disease process, and a better understanding of this relationship is needed. As such, adjustment in this review was based on the Moss-Morris (2013) model of adjustment. This model defines adjustment as the process by which an individual is able to return to equilibrium in the face of specific illness triggers (such as when one receives their diagnosis or in the case of physical deterioration) or maintain equilibrium in the face of day to day illness demands (Moss-Morris, 2013). Equilibrium involves factors such as good illness management, positive affect and less illness interference on an individual's roles and relationships (Moss-Morris, 2013). Adjustment in this sense is a dynamic process, as factors which may be successful for adjustment at one stage of disease severity may no longer be successful with advancing deterioration. This is especially relevant in PD, given in the latter stages motor control and active problem-solving strategies may be rendered impossible by worsening of symptoms.

Consequently, the present review is focused on individual adjustment processes, with the research question 'how do individuals adjust to living with Parkinson's disease?' In addressing

this question, the current review brings together the qualitative literature on adjustment to PD by conducting a meta-synthesis of relevant papers. The need to promote an individual's adjustment to their condition is highlighted by studies which outline the consequences of poor adjustment to PD, such a loss of independence and self-esteem, social withdrawal, increasing frustration and perceived stigma (Caap-Ahlgren, 2002). Individuals living with PD still have a need for preventative interventions which promote an individual's wellbeing and mitigate further deterioration in health (Grady & Gough, 2014), and a better understanding of adjustment in PD can help to inform such interventions. Although a diversity of psychological interventions have been adopted with people with PD and have evidenced preliminary positive findings of their effectiveness (Zarotti et al., 2020), there is a need to focus on factors which support people's adjustment to the condition, rather than just focusing on the reduction of symptoms.

Methods

A meta-ethnography was chosen because it is an approach which not only synthesises information but also seeks to broaden conceptual knowledge, as was the aim of the present review (Booth et al., 2016). Meta-ethnography was developed by Noblit and Hare following their perceived failure of existing approaches to synthesis to preserve the context of the individual studies (Campbell et al., 2011) and their recognition of the importance of this context in shaping the findings of a study. The approach of meta-ethnography is therefore informed by an interpretive rather than aggregative intent. There are three possible types of relationships that guide translation and subsequent synthesis (Campbell et al., 2011). The first of these is reciprocal, where studies are translated into one another and overarching metaphors are revealed which provide explanatory power for the individual studies. Reciprocal translation can be guided

by an interpretive intent, where the goal is to develop novel insight from the metaphors. Alternatively, reciprocal translation may instead summarise studies in terms of what the studies express (Dixon-Woods et al., 2006). Meta-ethnography adopts the former approach to reciprocal translation, as opposed to other forms of qualitative synthesis which are more descriptive in nature. The second type of translation is refutational, where more elaborate translations are developed which uncover the relation between the main accounts of a study and competing explanations. Finally, line of argument synthesis involves uncovering the larger inference which is implicitly conveyed in the studies but is not explicitly stated i.e. identifying the full picture from the individual pieces.

Conducting the meta-synthesis followed the seven-step process detailed by Noblit and Hare (1988). These are as follows: getting started, deciding what is relevant to the initial interest, reading the studies, determining how the studies are related, translating the studies into one another, synthesising translations and expressing the synthesis

Identifying relevant papers

The first stage of the meta-synthesis was 'getting started', which involved exploring the existing literature on individuals living with PD, to determine the suitability of an article focused on adjustment processes. This indicated the need for a review focused on adjustment specifically, which could bring together the existing literature on experiences of PD more generally. The next stage involved identifying the relevant focus of the review. This was determined following scoping searches of literature relating to PD and to adjustment in chronic illness more widely. Following this, relevant papers were identified by searching three databases: MEDLINE Complete, CINAHL and PsycINFO. Following guidance from an academic librarian, the search

involved three areas: (1) qualitative approaches, (2) adjustment and (3) Parkinson's disease. Details of the free text terms and subject headings used in each database can be seen in Table 1.

[Table 1 here]

Inclusion/exclusion criteria

Inclusion criteria for papers to be selected were: (1) used a qualitative approach to data collection (i.e. an approach which aimed to understand people's beliefs, experiences or attitudes and generated non-numerical data (Pathak et al., 2013)), (2) peer-reviewed, (3) focused on the experience of the individual with PD rather than a caregiver, and (4) at least one major theme (or concept) focused on the process of adjustment. Exclusion criteria were: (1) focused on adjustment after deep brain stimulation (DBS), (2) a comparative or dyadic study where the experiences of the individual with PD could not be clearly extracted, (3) focused on adjustment of aspects not specific to the condition e.g. coping with menstruation in individuals with PD (Tolson et al., 2002) and (4) the focus was predominantly on barriers to optimal adjustment.

Search results

During the search 1761 articles were identified, of which 904 were duplicates. The remaining papers were appraised for relevance based on the title and abstract. 807 were excluded because they were unrelated to the research question or not meeting eligibility criteria. 51 papers were accessed in full, either because the abstract suggested potential relevance to the research question or because there was insufficient information to exclude them based on eligibility criteria. From these 51 papers, 31 were excluded; 17 did not meet the inclusion criteria and 14 met exclusion

criteria. This resulted in 20 articles identified as being of relevance for inclusion in the review. See Figure 1 for details of this process.

[Figure 1 here]

Study Characteristics

Studies were conducted between 2002 and 2019. 14 studies were conducted in Europe, (including the UK, Netherlands, Spain, Sweden and Estonia) and six in the USA. Participants were all people with PD or people with PD; in total the review collates data from 458 individuals, with individual study sample sizes ranging from 6 to 85. PD disease severity as categories by Hoehn & Yahr disease stages ranged from 1-5, with the majority of participants falling within 1-3 on the disease stage spectrum. 11 studies used semi-structured interviews to collect the data and analysis included thematic analysis, grounded theory, interpretative phenomenological analysis and content analysis.

Quality of the selected studies

The quality of a meta-synthesis is somewhat dependent upon the quality of the selected studies, thus assessing the quality of the studies is important. Furthermore, evaluating the methodological quality of a study is essential to determine the relevance of the findings for clinical practice (Harrison et al., 2017). However, rather than being constrained by the quality of the data in the studies, a meta-ethnography seeks to generate understanding which is not preconditioned by the nature of quality judgments of a single study (Booth et al., 2016). As such, the Critical Appraisal Skills Programme (CASP) checklist was used to support critical engagement with the studies but not used to exclude any individual studies. The CASP checklist enables researchers to assess the trustworthiness, relevance and results of published papers and is

widely used in clinical research (CASP, 2019). Assessing the quality of the papers using this checklist involved identifying the standard of methodological reporting and study robustness and focused on three key areas; validity of the research, rigour of the findings, and utility of the results. Studies were assessed based on eight appraisal questions and each question was given a score of 1-3 according to the amount of evidence provided, with a total possible score of 24. CASP scores ranged from 14-22, with the majority of papers scoring between 17-22. Higher quality articles typically gave a more thorough description of the data collection and analysis process and produced a more detailed account of their consideration of the ethical issues. Considering the final themes, each sub-theme was present in at least two of the three strongest papers and thus no subtheme depended solely on the weaker papers. How the papers were evaluated with respect to the CASP tool and the eight appraisal questions used are detailed in Table 2.

[Table 2 here]

Analysis

In a meta-ethnography, different levels of interpretation provide weight to the eventual findings, based on Schütz' concept of first and second order constructs (Schütz', 1962). First order constructs reflect the participants' interpretations to the original questions asked within a single study. Second order constructs reflect the authors' interpretations of the participants' responses. Finally, third order constructs reflect the researcher's interpretations of the original authors' interpretations. A meta-ethnographic approach rests upon these third order constructs, which allows for an interpretative account of the shared concepts in adjustment rather than merely combining themes together. Each paper was read and annotated in detail, with key concepts drawn out for each paper, reflective of Schütz' second order constructs. Key concepts

were translated into a table and the papers which contained each concept were noted; this is detailed in Table 4 of the Appendix. Consideration was also given to divergent concepts and how these might reflect either aspects of the concept which were difficult for some individuals to achieve e.g. acceptance, or how a broader definition of the concept might capture such refutations. The key concepts were then synthesised to generate three key themes, the third order constructs, which felt to capture the important aspects of individuals' experience. How the concepts in each paper contribute to aspects of each theme is detailed in Table 3. Discussing these concepts with two supervisors helped to maintain the quality of the original findings and uphold methodological rigour (Thomas & Harden, 2008).

[Table 3 here]

Reflexivity

A meta-ethnography extends beyond the original interpretations of the author, which gives rise to potential biases from the researcher's own position. Nevertheless, despite its interpretative roots, a meta-ethnography must always be grounded in the data reported by the primary studies (Dixon-Woods et al., 2005). Since researcher preconceptions will always influence how data are gathered, interpreted, and presented (Tufford & Newman, 2012), the primary researcher regularly consulted with two supervisors. This enabled the researcher's subjectivity to be explored for its meaning and relevance and helped to minimise the potential for bias (Elliott et al., 2012).

Findings

In synthesising the 20 papers, three main themes relating to how individuals adjust to living with PD were constructed: (1) maintaining a coherent sense of self, (2) feeling in control and (3) holding a positive mindset. These three themes are described in detail below.

Maintaining a coherent sense of self

Maintaining a coherent sense of self felt to be important for individuals to adjust to the challenges of PD and to accommodate changes where necessary. There appeared a temporal dimension to this process. The early stages of the illness involved an individual's initial recognition of the chronicity of their illness and what it meant for their identity. Individual responses were often aimed at preserving aspects of their pre-illness self, with the support of friends and family being crucial in this, in supporting them to do the activities they had always done and in encouraging them to retain valued roles. In later stages of the illness, given ongoing deterioration in physical and mental ability, it was often not possible to maintain many aspects of their pre-illness identity. Here, acceptance provided the context for reshaping one's identity to incorporate the illness, in order to allow for positive self-evaluation and maintaining a coherent sense of self in the face of progressive deterioration.

Support from friends and family.

The support from friends and family was important in helping an individual to retain a valued social identity and maintain a coherent sense of self, finding the aspects of themselves which may initially have been lost following physical deterioration: “....with the help of caring and supportive family members, a few special friends, doctors and my husband I have been back to moving and finding my groove” (Gardenhire et al., 2019, p1786).

Despite some deterioration in their physical ability, family members enabled an individual to continue valued activity, reinforcing the coherence of their sense of self rather than someone whose identity had changed as a result of the diagnosis (Navarta-Sanchez et al., 2017; Kang & Ellis-Hill, 2015). Furthermore, by helping individuals with practical aspects of the condition such as daily planning or cooking, partners helped individuals with PD to maintain independence in the activities which mattered most (Gardenhire et al., 2019; Uebelacker et al., 2014; Lutz et al., 2018). Individuals were determined to maintain their usual activities as far as possible even when this conflicted doctors' advice about expected behaviours, only adjusting their behaviour when their habitual activities were no longer working for them (Lubi et al., 2019). Although negotiating the role in the family was often necessary (Habermann, 1996), focusing on the continuity of these relationships allowed an individual to maintain who they wanted to be (Sperens et al., 2018; Williams & Keady, 2008; Habermann, 1996).

Support groups were one forum where an individual gained friendship which encouraged a coherent sense of self. Coherence was provided by opportunities which allowed individuals to reinforce and confirm the experiences of themselves which mattered, such as through participation in enriching activity and offering reciprocal support (Sperens et al., 2018; Berard & Smith, 2019; Gardenhire et al., 2019):

In helping others find theirs, I have created a purpose for myself and the amazing thing is I found that I began to enjoy life more! It was not just a service for others; it became a pathway to regain my lost dignity and optimism, and sense of value as a person (Gardenhire et al., 2019 p1788).

Despite disruption to their sense of self as a result of the illness, individuals were able to re-establish coherence by providing support to others and maintaining purpose and valued social activity.

Acceptance.

Where an individual's pre-illness self-image was no longer congruent with PD related changes, acceptance of the diagnosis and illness limitations was required to adjust to their condition. This allowed individuals to maintain a coherent sense of self despite alterations in their identity. The diagnosis for many was perceived as an emotional force which changed their identity and challenged fundamental aspects of themselves (Vann-Ward et al., 2017). Individuals faced a major challenge of moving from being overwhelmed to acknowledging the illness as their own (Habermann, 1996). In order to accept the disease, individuals described going through many different emotions from initial denial, through to sadness and anger, before eventually allowing themselves to be hopeful for their new future (Hellqvist et al., 2018; Nazzal & Khalil, 2017; Habermann, 1996). The process culminated in a shift in how they thought about their diagnosis, from having to adjust to living with a diagnosis they did not want, to a situation in which they were able hold an optimistic outlook and feel a sense of mastery in living with PD (Gardenhire et al., 2019; Lutz et al., 2018).

For many individuals, acceptance involved modifying their expectations of what they were able to do, incorporating their disease related limitations into an identity which accommodated illness changes. The metaphor 'like a bird with a broken wing' highlights how participants accepted the things they were no longer able to do in order to adapt to the newfound circumstances (Smith & Shaw, 2017). There was an ongoing tension between preserving their

current sense of self and releasing aspects of the former self (Stanley-Hermanns & Engebretson, 2010; Lutz et al., 2018) in order to adjust to a self which integrated 'the broken wing'. Although aspects of their self might change, participants who had adjusted were able to build on past relationships and events to maintain coherence in their sense of self, integrating the PD into an altered understanding of the self (Williams & Keady; Vann-Ward, 2017). This acceptance facilitated proactive coping, such as through using mobility devices and making necessary adaptations to their activities, thus enabling continued social engagement alongside the ability to optimise care (Smith & Shaw, 2017; Lubi, 2019; Vann-Ward et al., 2017). By continuing to adapt their activities to allow for sustained wellbeing and activity participation, individuals were able to redefine themselves in relation to their impairments and activity limitations (Eriksson & Ahlgren, 2013; Vann-Ward et al., 2017; Lutz et al., 2018).

Support groups for individuals with PD also helped individuals to accept a sense of self which incorporated the illness and offered a forum for individuals to normalise their situation and feel validated in their illness experiences (Charlton & Barrow, 2002; Smith & Shaw, 2017; Stanley-Hermanns & Engebretson, 2010; Lutz et al., 2018). Having the opportunity to talk to others in a similar situation helped individuals to reduce disease related distress and to value an identity which incorporated their PD diagnosis (Uebelacker et al., 2014; Charlton & Barrow, 2002; Hellqvist et al., 2018).

Feeling in control

Feeling in control helped individuals to retain a sense of agency despite the illness limitations. Undertaking self-management of their condition was one way in which individuals gained back a sense of control while living with PD. In order to feel able to self-manage their

condition, information and resources were necessary to provide individuals with the required knowledge and expertise.

Self-management.

Feeling able to self-manage care was an important component of feeling in control. Learning self-management techniques helped an individual to feel they had a greater understanding of their situation and thus strengthened feelings of being control:

...and this education as a whole has helped me to take on my illness in a more powerful way. Maybe it's because I'm relatively newly diagnosed...for me...this education gave me hands-on tangible things to do for myself. That felt good for me. (Hellqvist et al., 2018, p3723).

Feeling in control and of owning responsibility for management of symptoms were stressed as important across many of the studies (Soundy et al., 2019; Smith & Shaw, 2017; den Oudsten et al., 2011; Vann-Ward et al., 2017). When primary control of PD is not possible given the unknown and fluctuating course of the disease, feelings of secondary control (i.e. the ability to control the psychological impact of circumstances) were important to maintain some sense of self-efficacy. Even previously mundane tasks such as maintaining the physical environment were described as valued tasks in helping to occupy individuals in a way which contributed to their feelings of self-efficacy: "Taking care of the house keeps me busy, I enjoy doing woodwork and there is always something to attend to" (Sperens et al, 2018 p705).

In many of the studies, individuals with PD actively monitored their health in order to tailor the management of their symptoms (Vann-Ward et al., 2017; Hellqvist et al., 2018).

Regarding medication, individuals made independent decisions about how and when to take prescribed and non-prescribed medications, often listening to their body in order to determine medication needs (Vann-Ward et al., 2017; Stanley-Hermanns & Engebretson, 2010; Williams & Keady, 2008; Habermann, 1996). Listening to the body often resulted in confrontation with the limits of formal knowledge and the acknowledgement that the person knew best in managing their condition. Medication helped individuals to stabilise their symptoms and maintain independence (Williams & Keady, 2008; Sperens et al., 2018), allowing individuals to feel 'master of [their] own fate' (Plouvier et al., 2018 p141). Despite being an external way of managing physical symptoms, taking medication allowed individuals to feel they had control over their condition and gave them agency in practical aspects such as dosage, timing and the management of side effects (Habermann, 1996).

It was often through physical activity where an individual first became motivated to be proactive at taking care of their own health (Soundy et al., 2019; Uebelacker et al., 2014; Sperens et al., 2018). Some individuals described how being motivated to exercise was something which came naturally, as physical activity was something they had always enjoyed (Soundy et al., 2019; Hellqvist et al., 2018; Lutz et al., 2018). Others described dedicating time to exercise as more of a conscious effort, deliberately chosen because of its effects on their physical and mental wellbeing (Eriksson et al., 2013; Lubi, 2019; Kang & Ellis-Hill, 2015; Habermann, 1996). Engagement in physical activity and exercise classes was seen by these individuals as a way to challenge themselves and to be the active agent in maintaining their health (Kang & Hill, 2015; Williams & Keady, 2008; Nazzal & Khalil, 2017).

Information and resources.

Information and resources were important for individuals to feel in control of their condition. Individuals reported how information about the medical aspects of PD were particularly useful in helping them to feel in control of their care (Sperens et al., 2018; Navarta-Sanchez et al., 2017; Soundy et al., 2019; Nazzal & Khalil, 2017). Unfortunately, even in later studies, individuals still reported they often received little information from their healthcare professional or received information which was delivered without due regard for the impact which disclosure might have on the individual (Plouvier et al., 2018; Shaw & Schmidt, 2017; Habermann, 1996).

Furthermore, lack of knowledge about the disease and its symptoms had forced some to conduct autonomous searches for information and try alternative solutions, to satisfy their “need to know” (Habermann, 1996 p. 404). Limitations of a service such as having insufficient resources and lack of professionals’ time were reported as making it difficult to ask the right questions, leading to concerns remaining unanswered and decreasing agency (Lubi, 2019; Nazzal & Khalil, 2017). The progressive and fluctuating course of the disease, experienced differently in every individual, could create mystery and uncertainty which may be amplified by lack of knowledge (Smith & Shaw, 2017; Habermann, 1996).

In contrast, participants who received regular contact from healthcare professionals really valued the support that was offered, describing it as sensitive to their needs and making them feel able to manage their condition (Hellqvist et al., 2018). Sufficient knowledge of- and education about- the disease could facilitate anticipation of change and could help individuals to feel more in control of their condition (Plouvier et al., 2018; Navarta-Sanchez et al., 2017; Lubi, 2019; Sperens et al., 2018). This sense of agency was important in helping individuals to feel better able to overcome the barriers of PD (Smith & Shaw, 2017). In empowering individuals to self-

manage their condition, many individuals agreed that resources from healthcare professionals helped them to feel more capable of adjusting to PD (Navarta-Sánchez et al., 2017).

Theme 3: Holding a positive mindset

Having a positive attitude enabled individuals to experience positive wellbeing despite the challenges raised by their condition. Social comparison-both upward (looking to others functioning better than oneself) and downward (reflecting on the worse situation of others) helped individuals to reflect on their ability to cope. A related concept, gratitude, helped individuals to value the things which they appreciated in their life.

Social comparison.

The ability to make both upward and downward positive social comparisons seemed to be important in helping individuals with PD to achieve a positive mindset. Self-help support groups were one environment whereby individuals had the opportunity to make social comparisons (Gardenhire et al., 2019; Charlton & Barrow, 2002; Sperens et al., 2018). Members of self-help groups were able to utilize the experience of peers to make a positive downward social comparison and reflect on their relatively good fortune (Charlton & Barrow, 2002; Hellqvist et al., 2018). Downward social comparisons were reflected in statements such as “There is always someone worse off than me” (Hellqvist et al., 2018, p3725). For many, reflecting on the worse situation of others could help to keep their own situation in perspective and facilitate emotional coping (Lutz et al., 2018). Strengthening self-image through downward social comparisons allowed individuals to adjust to their own situation and was experienced regardless of their own degree of impairment (Hellqvist et al., 2018; Eriksson & Ahlgren, 2013).

Participants were also able to make positive upward social comparisons to find inspiration and optimism from well adapted peers who had experienced PD for significantly longer duration (Charlton & Barrow, 2002; Soundy et al., 2019). By gaining inspiration from others who were coping well, broadening knowledge of PD management or heightening gratitude of their current functioning, social comparison helped individuals to maintain positivity (Soundy et al., 2019). This positivity translated into individuals perceiving their future as possible (Soundy et al., 2019; Gardenhire et al., 2019), with individuals realising that illness changes could be managed observing the positive and encouraging outcomes of peers (Soundy et al., 2019).

Gratitude.

In many of the studies, participants described how having gratitude helped them to maintain a positive mindset and facilitated their adjustment to the PD (den Oudsten et al., 2011; Lutz et al., 2018). Having gratitude for present functioning facilitated the ability to place high value on the activities participants were still able to do, which translated into individuals feeling less burdened by their activity limitations (den Oudsten et al., 2011; Stanley-Hermanns & Engebretson, 2010). In the early stages of disease progression, individuals could be grateful that their body in some aspects functioned “normally”, and thus reflect upon how they were still themselves despite the PD (Eriksson et al., 2013; Kang & Ellis-Hill, 2015; Stanley-Hermanns & Engebretson, 2010). Even experiences of tiredness and sweating when exercising or being active were considered positive, and were longed for, since they meant expressions of a healthy body (Eriksson et al., 2013). It also helped individuals to remain active in society, which they described as contributing

to feelings of being useful and maintaining high levels of self-worth (Kang & Ellis-Hill, 2015; Williams & Keady, 2008).

Gratitude for friends and family was also described in many individuals' accounts. For instance, kinship and sense of belonging were important sources of wellbeing and helped an individual with PD to reflect on the valued relationships they had despite the changes brought on by the diagnosis (Williams & Keady, 2008; Vann-Ward et al., 2017). As one individual reflected on how their relationship with their partner had changed since diagnosis: "I mean we sort of found out that our feelings for each other had multiplied. We've got a lot more time for each other" (Smith & Shaw 2017, p18).

Gratitude for the present seemed to be a way in which individuals were able to live life to the full despite the challenges faced living with PD. Many of the individuals described the importance of acknowledging that their future was likely to be associated with symptom deterioration and reduced life expectancy, and the need therefore to live in the present with renewed energy and determination (Charlton & Barrow, 2002; Williams & Keady, 2008; Vann-Ward et al., 2017). This renewed energy led participants to take up desired opportunities in the present and not the future, thereby leading to the sense that they were embracing the present (Smith & Shaw, 2017; Williams & Keady, 2008; Vann-Ward et al., 2017). By living for each day, their present became more manageable and they were able to be grateful for the things they could still achieve. For some individuals, gratitude and positive thinking felt to be part of their personality and a natural process, whereas for others it was more of a conscious strategy or through sense of obligation (Sperens et al., 2018). Many individuals experienced negative

reactions to the initial diagnosis of PD (Shaw & Schmidt, 2017), but gratitude helped an individual to navigate their path to finding an optimistic perspective (Gardenhire et al., 2019).

Discussion

The present review describes how, for many individuals, PD is experienced as a threat to their self-identity, thus maintaining a coherent sense of self was a key adaptive task in adjusting to their condition. This is in line with the Moss-Morris model of adjustment (2013) whereby critical events in an illness experience (such as the diagnosis or initial symptoms), alongside ongoing illness stressors, disrupt emotional equilibrium and quality of life. The goal of adjustment is to return to equilibrium, thereby reducing the impact of the illness on everyday life. However, the present review describes how maintaining a coherent sense of self did not just involve a restoration of an individual's previous sense of self, as assumed in the aforementioned model, but also involved altering their sense of self to integrate the PD. This suggests that rather than restore equilibrium or return to previous assumptions, individuals living with a progressive condition need to alter their equilibrium to accommodate illness changes. As reflected upon by Frank (1995), the onset of illness disrupts our life narrative and the old maps which once guided us, and new maps need to be developed to navigate this new territory. Williams' (1984) describes how reconstructing an identity narrative helps an individual to understand the illness in terms of their past experiences and to reaffirm their sense of self as one of purpose and meaning. For individuals living with chronic fatigue syndrome, reconstructing an identity narrative has been shown to be a powerful way in which individuals make sense of their illness and in providing opportunities to connect with other individuals with similar illness narratives (Bulow, 2004). In placing the experience of chronic illness within the framework of their life history, an

individual can find meaning to the events which have disrupted and changed their identity (Williams, 1984) and re-establish their relationship between their self, the world and their body (Bury, 1982). The ability of an individual to reconstruct a narrative which integrates their illness experience allows an individual to maintain a coherent sense of self, assumed to be key to psychological health (Reese et al., 2011). In developing a sense of self which not only incorporates but is enriched by their illness experience, individuals may be better able to adjust to their condition.

In order to alter their equilibrium to accommodate illness changes, individuals described the importance of gaining a sense of control over the management of their condition. Having a sense of control has consistently been linked to good psychological and physical adjustment to chronic illness (Taylor et al., 2000). Gaining control over symptoms through self-management (primary control) allowed individuals to feel more in control of their life and better able to adapt to disease related limitations (secondary control). This felt important given the uncontrollable nature of disease progression. Both increased perceptions of primary control (Pape & Kim, 2002; Krakow et al, 1999) and secondary control (McQuillen et al., 2003) have previously been associated with greater wellbeing and psychological adjustment in individuals with PD. Even if control of disease may not be possible, global life control and feelings of self-efficacy rather than control over the disease per se can also be important (Eccles & Simpson, 2011). Indeed, in one study investigating perceptions of cause and control in individuals with PD (Eccles et al., 2011), knowledge of aetiology and underlying physiological processes of PD did not contribute to a perception of control, although knowledge supporting management techniques was perceived as helpful. Whether knowledge of the disease contributes to a perception of control is likely to depend upon the individual and the meaning which potential knowledge has for them; including

whether it is congruent with their own understanding. Self-management processes can thus be understood as an ongoing negotiation between different perspectives, including own perspectives vs medical advice, present wellbeing vs future health and routines required in self-management vs being free (Auduly et al., 2009). Where individuals experience aspects of the negotiation process as complementary to one another, determining the course of self-management is unproblematic. In contrast, where individuals experience incompatibilities between advice given about self-management behaviours and individual needs or preferences, individuals may experience a kind of inner conflict, reflecting a tension between what an individual wants to do and what they experience that they should do (Bazerman et al., 1998). This highlights the importance of ensuring that information and knowledge related to self-management is tailored to an individual's understanding and experience of the disease.

In line with the Moss-Morris model (2013), cognitive factors including gratitude and social comparison were important components for individuals living with PD to hold a positive mindset despite continuing deterioration. In comparing themselves with others and experiencing gratitude for the things one appreciated in life, individuals seemed able to come to terms with their condition. For many, reaching a place of optimism involved a series of emotional stages including shock, denial and anger (Gardenhire et al., 2019), situating it as a similar process to that of bereavement. This process of adaptation may be a healthy way in which an individual grieves for the person they once were (Reynolds & Prior, 2003). In making space for and allowing oneself to experience and share negative emotional experiences with others, individuals with chronic illness may be better able to self-regulate emotions (de Ridder et al., 2008). The ability to hold a positive mindset alongside co-existence of positive and negative emotions is

likely to facilitate an optimistic outlook in the face of adversity and adjustment to the challenges imposed by their condition (Tedeschi & Calhoun, 2004).

Clinical implications

In the present study, maintaining a coherent sense of self was described as an important factor in adjusting to living with PD. Where an individual is unable to find meaning, purpose or personal efficacy, psychological interventions which are biographically informed have been shown to help people with chronic illness to retain their pre-diagnosis identities where possible but also to develop new identities which accommodate illness changes (Hubbard et al., 2010). Biographically informed interventions involve identifying the roles which an individual valued prior to their illness and supporting them to develop ways in which they can adapt these activities to continue valued social activity (Bury, 2001). When individuals are given the appropriate level of support which allows them to maintain a coherent sense of self, they may experience an enhanced sense of responsibility over their illness (Pierce et al., 2003) requiring less input in the future from medical and psychological professionals. The implications of less professional input and increased empowerment are not only financially beneficial for health services but also helps enhance individual wellbeing and quality of life. It is essential that any intervention appropriately considers the point at which an individual is at in their adjustment journey, in order to offer support which helps them to adapt to a self which has changed.

Feelings of being in control were important for individuals to adjust to living with PD, with information and resources being key to facilitating the participants' perceptions of control. Consistency in the information provided by healthcare professionals is important to minimise distress related to confusion about their symptoms and to heighten individual's self-efficacy in their ability to manage their condition. Several of the studies highlighted incidences where

individuals had felt information had been delivered insensitively or without due regard to an individual's preference in the timing or setting for the delivery. Individuals described being informed of their diagnosis in non-private settings or in an abrupt and impersonal manner (Shaw & Schmidt, 2017). This may reflect the complexity of providing the right information at the right time (Eijk et al., 2015) or reflect clinician's guilt about their inability to provide a medical treatment or cure, thus affecting their consideration of appropriate manner and timing of delivery (Schrag et al., 2018). Nonetheless it is concerning, given the importance of individuals feeling empowered through professional support to self-manage their care. Where professionals deliver recommendations without recourse to an individual's practical understandings or idiosyncratic needs, individuals may feel forced into adopting habits they are not ready to adopt and available solutions can be unsettling (Lubi et al., 2019). Instead, an integrated approach including the individual with PD in any decision making is likely to help individuals to feel more like a partner and is considered the best way to manage PD (Post et al., 2011). This integrated approach would ensure that the provision of information is tailored to the needs of the individual so that they feel able to make good decisions about their current and future care (Shaw & Schmidt, 2017). Early intervention incorporating psychoeducation about the management of symptoms is key to heightening feelings of control and facilitating adjustment to PD.

Participants across the studies spoke of the need to not dwell on their future deterioration but instead to embrace their present with renewed purpose, such that mindfulness-based cognitive therapy (MBCT) may be an effective intervention. MBCT typically takes places over 8 weeks and teaches participants to become more aware of their present thoughts and feelings. It also encourages a more detached relationship with these thoughts and feelings, rather than viewing them as accurate representations of the self or reality (Batink et al., 2013). In developing

a detached perspective to difficult thoughts and feelings which may arise from living with PD, an individual may be less likely to experience the difficult thoughts as a reflection of their inability to cope and more able to fully engage with their present. Preliminary evidence suggests the experience of mindfulness can help individuals to access inner resources and reaffirm their existing coping ability, enabling them to confront previously avoided social situations and to have more confidence in their ability to self-manage situations which they may have previously avoided (Fitzpatrick et al., 2010). Facilitating this self-management and encouraging individuals to fully engage with their present experience may provide a protective mindset against PD related challenges and an acceptance of the feelings or emotions of having PD. In encouraging individuals to focus on their present and enhancing existing coping ability, MBCT may help individuals to adjust to their condition.

An important theme across individual accounts was that optimal adjustment relates to embracing positive living despite a readjusted state of health, and healthcare professionals need to support an individual to accept this readjusted state wherever possible. Interventions based on acceptance and commitment therapy (ACT: Hayes & Storsahl, 2011) may help individuals to develop acceptance to the changes which PD brings. Acceptance is a stance an individual adopts rather than a process of coping and enables an individual to pursue a life in accordance with their values, despite the limitations imposed by their illness (Chan, 2013). ACT encourages this accepting stance through techniques such as cognitive defusion, which help an individual to detach from difficult thoughts and in doing so develop more flexible ways of responding to challenging situations (Hayes & Storsahl, 2011). In developing more flexible responses, individuals are supported to lead a meaningful life despite the continual challenges which PD presents. It is likely that any ACT protocol would need to accommodate the neuropsychological

profile of PD, where deficits in attention and executive control are common, and may lead to deficits in managing the demands of complex processing tasks (Foley et al., 2019), such as defusing from difficult thoughts. Although there is little evidence for the efficacy of ACT in PD populations, there is evidence that ACT may be effective in conditions such as MS, where greater acceptance of the condition (according to ACT principles) was found to relate to better adjustment (Pakenham & Fleming (2011). Furthermore, a recent intervention utilizing a trans-diagnostic approach for individuals with chronic illness has highlighted the effectiveness of ACT principles in reducing psychological distress, reducing physical limitations, and increasing valued behaviour (Brassington et al., 2016). This effect was seen despite the individuals' evaluation of their health changes remaining the same. It is important that the effectiveness of the intervention was independent of the evaluation of health status, given that the progressive nature of PD means that an individual's health status is likely to continually deteriorate. The efficacy of ACT based interventions in other chronic illnesses highlights its potential efficacy in supporting individuals with PD to adjust to their condition.

Limitations and future research

Conducting a meta-ethnography largely ensures that the experiences of the participants' in the individual studies are preserved (Britten et al., 2002). Nevertheless, the synthesizing of multiple studies meant that some of the individual experiences may have been lost. For instance, nuances in adjustment which may have reflected differences in disease duration, extent of symptoms and disease severity of individual participants were not considered in depth. Furthermore, the majority of the studies included individuals with PD of either low or medium severity. Although some studies included participants with PD of high severity, it is likely that these participants represent those individuals who are functioning relatively well despite the

severity of their condition. Although recruiting individuals with high severity PD is challenging, reflective of both the physical and cognitive aspects of severe PD which would make research contribution more difficult, future research should aim to explore how adjustment might differ in the more advanced stages of the disease. An additional avenue for future research would be to study the experience of individuals with PD who additionally experience cognitive impairment. Although not discussed in detail in the present synthesis it is nevertheless an important aspect to consider. It may be expected that cognitive changes would limit an individual's ability to flexibly adapt their sense of self and to adopt self-management skills, with consequent implications on an individual's ability to adjust to their condition.

The characteristics of the included studies, in being all located in Western countries, means the findings are biased towards experiences of Western cultures and healthcare systems. Consequently, future research should explore the experiences of adjustment for individuals living with PD in non-Western countries. Furthermore, there is evidence to suggest that developing countries experience greater disease burden from chronic illness (Murray & Lopez, 2013), evidencing the need to understand and support individuals living with a chronic illness to adjust to and self-manage their illness.

Conclusion

Findings from this review indicate the need for individuals to maintain a stable sense of self and feel in control, in spite of illness related deterioration. Having a positive mindset was also important in helping individuals to experience positive wellbeing. Understanding how an individual adapts to living with a chronic illness is important not only to better understand the full implications of the condition but also so we are better able to offer early intervention for individuals who are finding adjustment difficult. Empowering individuals to self-manage their

condition can provide individuals and their families with the ability to cope and maximise their wellbeing.

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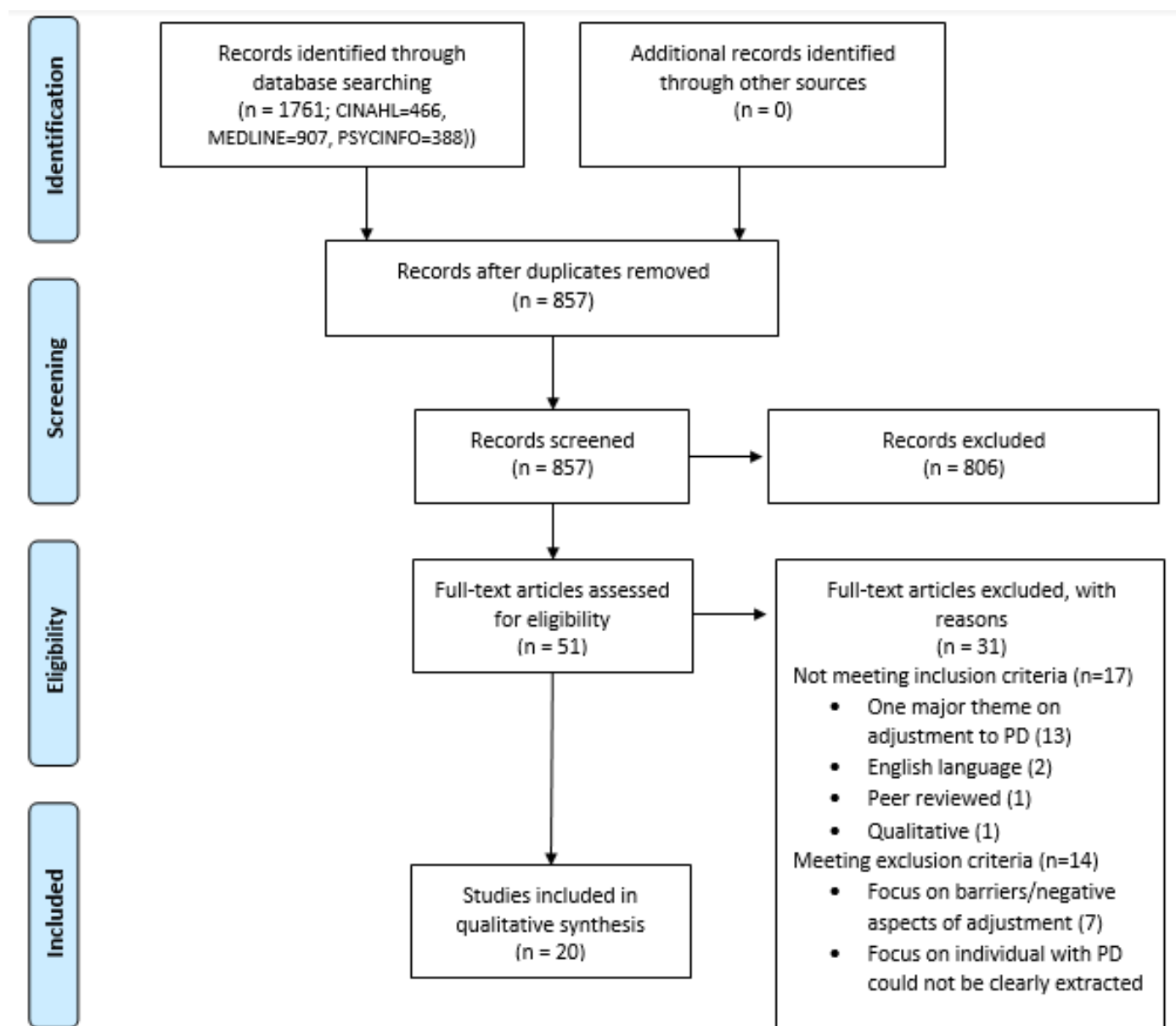
Figure 1: PRISMA flowchart identifying the process of article identification

Table 1: Table displaying the search strategy and the terms used for each database

Database	Free Text – Methodology (Title/Abstract/Keyword)	Subject Headings- Methodology	Free Text - Parkinson's disease (Title/Abstract)	Subject Headings- Parkinson's Disease	Free Text- Adjustment (Title/Abstract)	Subject Heading- Adjustment
PsycInfo (388 results)	“interview” OR “thematic analysis” OR “phenomenol*” OR “narrative” OR “focus group” OR “discourse analysis” OR “grounded theor*” OR “content analysis” OR “hermeneutic*” OR “heuristic*” OR “experience” OR “perception”	DE "Qualitative Measures" OR DE "Qualitative Methods"	"Parkinson* Disease"	DE "Parkinson's Disease"	“adjust*” OR “adapt*” OR “accept*” OR “manag*” OR "coping OR “resilience”	DE "Adjustment" OR DE "Resilience (Psychological)" OR DE "Adaptability (Personality)" OR DE "Self-Management"

MEDLINE	As above	MH "Qualitative As above	MH	"adjust*" OR	MH "Adaptation,
(908)		Research"	"Parkinson	"adapt*" OR	Psychological+" OR
			Disease"	"accept*" OR	MH "Self-
				"manag*" OR	Management/PX"
				"coping" OR	
				"resilience"	
CINAHL	As above	MH "Qualitative As above	MH	"adjust*" OR	MH "Adaptation,
(468)		Studies"	"Parkinson	"adapt*" OR	Psychological+"
			Disease"	"accept*" OR	OR MH "Self-
				"manag*" OR	Management/PF"
				"coping" OR	
				"resilience"	

Table 2: Table outlining the evaluative process using the CASP tool.

Study	Research Design*	Sampling	Data collection	Reflexivity	Ethical Issues	Data analysis	Findings	Value of research	Total (Low=<18 Medium =18-21, High= 22-24)
Soundy	2	3	3	3	3	3	3	2	High
Uebelacker	1	2	2	2	2	1	2	2	Low
Plouvier	3	2	3	3	2	3	3	2	High
Shaw	2	2	2	1	1	2	2	2	Low
Charlton	2	3	2	1	3	1	3	3	Medium
Lutz	3	3	3	1	3	3	3	3	High
Navarta-Sanchez	1	3	2	2	3	3	3	3	High
Hellqvist	1	2	2	2	1	3	3	3	Medium
Smith	2	1	2	1	1	3	3	2	Medium
Gardenhire	3	3	2	3	3	2	3	3	High
Sperens	3	2	3	1	2	3	3	2	High
Den Oudsten	1	2	2	3	3	1	2	2	Medium
Eriksson	3	2	3	2	0	3	2	2	Medium
Kang	2	1	3	3	2	3	3	2	High

Lubi	2	3	1	1	2	1	2	3	Medium
Nazzal	3	1	2	1	2	3	2	3	Medium
Vann-Ward	3	3	3	2	2	1	3	3	High
Stanley-Hermanns	3	3	2	2	3	3	3	3	High
Williams	3	2	2	1	2	1	3	3	Medium
Habermann	3	2	2	2	2	2	3	2	Medium

* CASP questions

Research Design	Was the research design appropriate to address the aims of the research?
Sampling	Was the recruitment strategy appropriate to the aims of the research?
Data collection	Was the data collected in a way that addressed the research issue?
Reflexivity	Has the relationship between researcher and participants been adequately considered?
Ethical Issues	Have ethical issues been taken into consideration?
Data analysis	Was the data analysis sufficiently rigorous?
Findings	Is there a clear statement of findings?
Value of research	How valuable is the research?

ble 3: Characteristics of the studies included for the synthesis

First author	Year	Country	Study Aim	Participants	Data collection	Data analysis
Soundy	2019	Oxford, UK	To consider the impact of providing the First Steps program on the stories of people with Parkinson's Disease (PD) and to investigate the mechanisms which may explain this impact	40 participants; 18 from the intervention group and 22 from the active control group 16 female and 24 male. Mean age 66.7 years. Avg. Mean length of PD 5.yrs.	Audio record single semi-structured interviews	Narrative analysis to consider the impact of the program. Thematic analysis to consider the established psychosocial mechanisms of impact

Uebelacker	2014	Butler hospital, Providence RI, USA	To identify and understand potential issues of importance to individuals with PDs when coping with Parkinson's Disease.	75 individuals with PD who were participating in a Movement Disorders Program. 43 male, 32 female. Aged 47-88. Length of PD 0- 18 yrs. Hoehn & Yahr disease stages ranged from 1-5	Open-ended surveys	Qualitative data analysis
Plouvier	2018	Nijmegen, The Netherlands	To explore PD individuals with PDs' coping with care changes	16 people with PD (11 male, 5 female). Aged 58-79. Hoehn & Yahr disease stages ranged from 1-4	Audio record semi- structured interviews	Inductive approach to comparative content analysis

Shaw	2017	North Yorkshire, UK	To investigate the current ethical issues in relation to recognizing and managing Parkinson disease (PD) from the individuals with PD's perspective	12 individuals with PDs living with PD who were from the medical school's Individuals with PDs as Educators program. 5 female, 7 male. Aged 51-86. Length of PD 11mths- 24yrs. Disease severity not described	Audio recorded semi- structured interviews	Thematic analysis
Charlton	2002	Bolton, UK	To investigate how Parkinson's disease had affected the lives of eight participants and explore coping methods they used	8 participants; four of the participants were members of the Parkinson's Disease Society and four were not. Aged 62-86, length	Audio recorded semi- structured interviews	Thematic analysis

of PD 3-8 yrs. Disease

severity not described

Navarta-Sanchez	2017	Navarre, Spain	To explore the meaning that coping with Parkinson's disease has for individuals with PDs and family carers and suggest the components of an intervention focused on enhancing their coping	21 participants: 9 people with PD, 7 family carers and 5 healthcare professionals. Persons with PD: 6 male, 3 female. Mean age 71 yrs. Mean length of PD 6.1yrs. Hoehn & Yahr disease stage: mean=1.5	Audio recorded focus groups (divided by participant group)	Qualitative content analysis
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Hellqvist	2018	Linköping University, Sweden	To identify and describe experiences valuable for managing daily life after participation in the NPS self-management intervention	25 persons with PD (14 male, 11 female) and 17 relatives. Length of PD <1yr-13yrs. Hoehn & Yahr disease stages ranged from 1-3	Audio recorded group discussions	Thematic analysis according to Braun and Clarke
Smith	2017	Aston, Birmingham	To investigate lived experience of Parkinson's disease (PD) aiming to investigate opportunities for wellbeing	Individuals with PDs and their partners (total 9) recruited via a Parkinson's UK support group. 4 PwD and 4 partners, 1 widowed partner. Person with PD aged 67-85, length of PD 2-21yrs. Disease	Audio recorded in-depth individual interviews in individuals with PD's home	Interpretative phenomenological analysis

severity not described.

Gardenhire	2019	Lubbock, USA	To investigate how individuals cultivate optimism despite challenges presented by PD	85 first person accounts. Demographic info not collected.	Audio record personal accounts	Grounded theory
Sperens	2018	University Hospital, Northern Sweden	To explore how people with Parkinson's disease manage the effect of the disease on everyday life	24 people with PD (12 male, 12 female). Aged 46-80. Length of PD 1- 19 yrs. Thirteen reported needing help in ADLs.	Audio recorded semi- structured interviews	Grounded Theory
Den Oudsten	2011	International centres; people with	To identify factors that persons perceive as eminently important	Participants included persons with PD, caregivers and health	Audio recorded focus groups	Thematic analysis

		PD from centres in Sweden, Norway and Holland	for QOL	professionals. 38 people with PD (15 female, 23 male). Aged 54-74. Disease severity not described.	(divided by participant group)	
Eriksson	2013	Värmland, Sweden	To explore and generate an understanding of the meaning of physical exercise in the lives of individuals with PD	11 people with PD (6 male, 5 female) participating in an exercise program. Aged 61-81. Length of PD 2- 17 yrs. Hoehn & Yahr disease stages ranged from 1-4	Audio recorded semi- structured interviews	Grounded Theory
Kang	2015	Durham, UK	To explore how people with PD live life successfully and what	8 people with PD (3 male, 5 female). Aged 57-78. Length of PD 2-	Audio recorded semi-	Thematic analysis

			contributes to this	16 yrs. Disease severity	structured	
			success	not described.	interviews	
Lubi	2019	University of Tartu, Estonia	To examine the role of information seeking in the maintenance of existing lifestyle and illness related adjustments	16 people with PD (9 male and 7 female). Aged 35-72 yrs. Disease severity not described.	Audio recorded semi-structured interviews	Thematic analysis using social practice theory
Nazzal	2017	Irbid, Jordan	To describe the daily living experiences and impact on lines of Jordanian individuals with PD	8 people with PD (4 male, 4 female). Aged 32-76 yrs. Length of PD 2-14 yrs. Hoehn & Yahr disease stages ranged from 2-3	Audio recorded semi-structured interviews	Thematic analysis

Stanley-Hermanns	2010	University of Texas, USA	To provide a rich understanding of the illness experience of individuals living with PD	14 people with PD (7 male, 7 female). Aged 38-82. Hoehn & Yahr disease stages ranged from 1-5	Field data collected from support groups, individual semi-structured interviews using grand tour questions	Thematic analysis
Williams	2008	Bangor University, UK	To understand adjustment and decision-making in day-to-day living with late-stage PD	13 people (10 men and 3 women) and their family carers. Aged 61-89. Length of PD 19-26 yrs. Disease severity: late (not defined)	Longitudinal semi-structured interviews	constructivist grounded theory and centre-stage storyline generation

Vann-Ward	2017	USA	To identify, explore, and theorize the social and psychological processes used by people with Parkinson disease	15 men and 10 women with PD (ages 40–95). Length of PD 3mths to >30 yrs. Hoehn & Yahr disease stages ranged from 3-5	In-depth interviews, nonparticipant observation, and participant photos, videos, and related document	constructivist grounded theory
Habermann	1996	University of San Francisco	To explore the experience of having Parkinson's disease in middle life	16 people (9 men and 7 women), aged 42-59. Length of PD 1-16 years. Hoehn & Yahr disease stages ranged from 1-3	Audio recorded semi structured interview	Phenomenological framework of stress and coping guided the analysis

Lutz	2017	University of Western Ontario	To deepen the current understanding of the experience of living with Parkinson's disease and its implications for occupation	6 people with PD (4 men and 2 women), aged 57-73. Length of PD 5-10 years.	Audio recorded semi structured interview using participants' photographs	Thematic analysis interpreted through narrative theory
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Appendix 1-A: Psychology and Health: Instructions for Authors

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Appendix 1-B: Constructing the synthesis through the generation of initial concepts to develop the final themes.

First author	Maintaining a coherent sense of self	Sense of agency and independence	Staying positive in daily life
Uebelacker 2014	<ul style="list-style-type: none"> Medication as important to help diminish the tremor symptoms and to allow for continued activity Support groups offer social interaction with similar others so one doesn't feel so alone Practical support allows an individual to maintain independence in others aspects of their life 	<ul style="list-style-type: none"> Physical activity providing benefits on physical symptoms and in helping an individual feel in control over managing the PD 	<ul style="list-style-type: none"> Emotional support helps with reducing anxiety for the future Relaxation techniques, tai chi and massage create a less stressful environment and contribute to relaxation.
Soundy 2019	<ul style="list-style-type: none"> Acceptance of the PD and 	<ul style="list-style-type: none"> Exercise/physical activity as a 	<ul style="list-style-type: none"> The outcome of social

being willing to identify PD
in social situations helping
an individual to continue
engaging in activities which
mattered to them.

way to look after yourself and
manage your own wellbeing

- Physical activity programs
helping individuals to become
more aware of their own
limitations and better able to
adapt accordingly
- The effort of physical activity
as contributing to the idea that
action could be taken to promote
mobility, balance and general
health
- An increase in self-
confidence and self-efficacy from
the group leading to engaging in
more activities

comparisons and taking
the messages of others on
board in order to have a
positive outlook on the
future

- Positive atmosphere of
support groups providing
a protective mindset
against the experiences of
PD symptoms
 - The ability to relate to
other individuals and to
compare coping strategies
helping individuals to feel
better about their own
situation.
-

and social interaction.

- The valuable role of information and advice in helping individuals to better understand how to manage their condition

Smith 2017	<ul style="list-style-type: none"> • The difficulties in accepting the PD causing powerful ontological challenges for the individual • The feelings of being “taken over” by the illness leading to PD dictating what one could and couldn’t do. 	<ul style="list-style-type: none"> • Finding ways to maintain involvement in activities outside the home to retain a sense of vitality • The importance of agency and taking action to overcome barriers in helping one come to terms with PD 	<ul style="list-style-type: none"> • Holding onto optimism as a way to find new ways of supporting oneself and adapting to the newfound circumstances. • Focusing on the positive aspects of the diagnosis on relationships and experiencing gratitude from those connections. • Sense of urgency leading
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to participants to take up desired opportunities in the present and not the future.

- Making the most of the situation preventing individuals from becoming overwhelmed by their disabilities and deficits.

Navarta Sanchez 2017	<ul style="list-style-type: none"> • The importance of accepting and coming to terms with the PD in order to adapt and live with the disease • Carers supporting the individual to continue 	<ul style="list-style-type: none"> • Getting activity underway as a way to improve symptoms or overcome the difficulties in activities of daily living • The importance of information in order to manage the PD and 	<ul style="list-style-type: none"> • Focusing on the positive aspects of the condition and the way the PD had helped them find new balance in their lives. • Living in the present by
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previously enjoyed activity.

reduce uncertainty

not thinking about the future; different to not ignoring the future, just not living in the future day to day.

- Being positive because it's the only thing one has to face the ups and downs.
- Negativity as a cause of bitterness, both for themselves and all those around them.

Sperens 2018

- The need to adapt occupational performance to accommodate obstacles

- Efficacious medical treatment to manage the symptoms of PD helped individuals feel able to

- Living with PD described as a conscious and demanding strive to
-

- Reducing expectations of performance-for some an unproblematic matter of fact, for others more troublesome and threatening their identity as a result
 - Managing time and energy so one could adapt according to daily functioning and activity-this helped to remain in charge of what they did each day
 - Physical exercise to keep up both mobility and wellbeing was an important way of challenging participants and helped them feel more in control.
 - Input from physiotherapists or attendance at a group were important aspects of social interaction and of motivation
 - Achieving a satisfactory life involved keeping up a good mood and having a good social life
 - Positive thinking for some as part of their personality and the way they had always acted, for others more of a conscious attitude or "act".
 - The need to stay positive and keep spirits up due to their awareness of the
-

effect of PD on their
loved ones.

- The value of certain
places and environments
to provide
peace, tranquility and
comfort. Places just to
enjoy being.

Plouvier 2018

- The importance of accepting
the inevitable disabilities and
limitations in order to search
for solutions
that minimize the impact on
their everyday life
 - Sufficient knowledge of the
disease facilitating the ability to
anticipate changes in care.
 - Not being able
to anticipate changes in care
leading to feelings of lack of
control
 - Being able to make
-

adjustments independently,

helping individuals have realistic

expectations of the post-change

situation

- Knowledge about medication

leading to feelings that one can

better manage the situation and be

“master of your own fate”

- When individuals were able to

solve problems independently,

this led to a sense of control over

changes to care and the situation

afterwards

Gardenhire 2019

- The opportunity to listen and understand others' stories and relate to their

- The confidence to speak out that you have PD and to engage in more activities and social

- The future as possible from listening to experiences of others and
-

experiences, providing an insight into how others cope. Equally, the opportunity to share their own experiences

interaction, leading to feelings of self-efficacy and feeling confidence in living well with PD

- The ability to embrace advice given and take it on board in order to adopt exercise and devote more time to it
-

taking on board positive messages in order to utilize it in their own life situation

- The program as providing a protective mindset against the experiences of symptoms and acceptance of their emotions about having PD.
-

Charlton 2002

- The inability to accept physical deterioration leading to vivid fears for the future
- Not wanting to accept the

- The fear of increasing dependence and loss of mobility causing difficulties in everyday living
- The importance of taking time out and switching activities where

- Social comparison helping the individual to recognize their relatively good fortune
 - Living one day at a time
-

- | | | |
|-------------------------------|---------------------------------|-----------------------------|
| effects of the PD leading to | necessary in order to deal with | and doing the things |
| feelings of loss of identity | tremor and immobility | which mattered to them |
| • Acceptance of the condition | | before it was too late |
| as crucial to manage the | | • Maintaining a normal life |
| illness; for members of | | as possible in spite of the |
| a self-help group, leading to | | illness |
| adaptations to their life | | • Maintaining a positive |
| whereas for non-members, | | outlook either by positive |
| acceptance as something | | thinking or by focusing on |
| which you just had to do. | | the positive aspects of the |
| | | condition. |
| | | • Dysfunctional cognitions |
| | | and bias, such |
| | | as catastrophizing, |
| | | leading to low self- |
| | | esteem, low positive |
-

affect and negative illness
perceptions

Hellqvist 2018	<ul style="list-style-type: none"> • Meeting others with PD helping individuals to find a sense of connection and to feel more comfortable in owning and sharing their experiences • Spending time with family as a way to forget about the disease and the symptoms for a while. • Accepting the disease as a way to acknowledge the present situation but to live life as well as possible 	<ul style="list-style-type: none"> • Long term relations and good access to healthcare staff helping individuals feel more confident in managing PD. • Taking ownership of the condition as important in adapting to it • Knowledge and advice about PD and medications helping to support decision making around finances and mobility aids • Having knowledge and seeking information as a way to understand and manage the 	<ul style="list-style-type: none"> • Support groups providing common ground and making participants feel less isolated • Support from others providing emotional support and promoting the feeling there was a hopeful and good future. • Thinking through situations which cause stress and anxiety and trying to make a plan to change negative thoughts
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despite the PD.

changed conditions

- Inner motivation and participating in a group helping individuals to initiate behaviour change with regard to physical activity and engaging more in self-monitoring

- Actively focused on the small things that generated happiness in daily life, such as acknowledging the good things in life and spending time with friends and family.

Shaw 2017

- The importance of disclosure to help an individual to come to terms with the condition and reduce uncertainty related to unexplained symptoms
 - PD diagnoses being given in non-private settings leading
 - Loss of independence as a major challenge for participants, yet wanting to take charge of making decisions which impacted upon increasing dependence.
 - Lack of interdisciplinary cooperating amongst professionals causing individuals
-

to perceptions of it being impersonal and undignified to feel confused and not in control with their care.

- Physical changes due to PD threatening their identity, leading to low self-esteem and worthlessness
- Medication helping an individual to restore balance with their previous self
- The need to balance the provision of information according to an individual's needs so as to equip individuals to make informed decisions but not overwhelm them in the early stages of the condition

Den Oudsten 2011

- | | | |
|---|---|--|
| <ul style="list-style-type: none"> • Taking leave of activities, they could not perform anymore in order to adapt to everyday life • The difficulties faced by individuals with PD in having to lose their job or | <ul style="list-style-type: none"> • Individuals with PD stressed that it was very important for them to be able to take care of themselves, to be independent | <ul style="list-style-type: none"> • The importance of thinking in positive ways, and putting high value on the activities one could still do • The challenges of maintaining their positive |
|---|---|--|
-

knowing that they probably
will lose it in the near future.

Most of
the participants stressed that
they would like to be useful
in some way in case they
were not able to work
anymore

- Remaining active in society
as way to maintain levels of
self-esteem
- The importance of support
groups in offering a sense of
belonging

way of thinking,
especially when the
disease progresses

Eriksson 2013

- | | | |
|---|--|---|
| <ul style="list-style-type: none"> • Individuals aimed to
continue to participate in | <ul style="list-style-type: none"> • Being able to live independently,
such managing daily activities | <ul style="list-style-type: none"> • The rational decision to
try to think positively to |
|---|--|---|
-

- | | | |
|---------------------------------|------------------------------------|-------------------------------|
| valuable areas of life, even if | like grooming, dressing, moving | try and make the best of |
| support was needed for | around and managing house and | the situation |
| managing to do so. To retain | garden were essential life goals | • Having gratitude that their |
| their identity and to be | • Confidence in own ability to | body functioned in |
| needed by significant others | attain goals and the determination | a “normal” way in order |
| was important to this | to stay active, despite disease | to maintain wellbeing. |
| • The importance of being part | intrusion, were strongly | |
| of a social context where the | connected with exercising | |
| participants share | • Acting in the best way according | |
| mutual experiences was | to their knowledge, beliefs and | |
| stressed by participants in | experiences enabled the | |
| order to generate feelings of | individual to take as much | |
| coherence | responsibility and command of | |
| | the body as possible | |
| | • Exercising meant slowing or | |
| | preventing progression of not | |
-

only symptoms of the disease, but
also the disease itself

Kang, 2015	<ul style="list-style-type: none"> • Successful living as being able to live the usual life that they had before having PD. Participants rated their level of success by comparing between their pre-illness life and current life with PD • Pretending one had not been diagnosed with PD which seemed to be a positive psychological response at the earlier stage of illness • The importance of personal effort and the ability to 	<ul style="list-style-type: none"> • Participants felt success when their symptoms seemed not to be getting worse and when they became comfortable and competent with their new adjustment to illness • The importance of independence and contributing to others' lives. This seemed to lead participants to believe in their self-worth and the dignity of their lives • An important aspect linked to feeling successful was a sense of being in control 	<ul style="list-style-type: none"> • Having peace with oneself and with life was important in order to counteract social stigma and maintain a positive outlook on life • Participants tried to make most of life by being thankful to be alive and loving self and others • Social comparison and family support seemed to create positive feelings that they were not left
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- continue previous activities
- Positive family support helped participants to manage their illness by being always beside them, being co-operative, accepting their illness and treating them as normal rather than treat them differently
- Engagement in physical activity and exercise classes as a way to challenge self and maintain health
- alone

Nazzal, 2017

- Many participants had experienced shock, denial and grief. However, all participants stated that once they accepted the diagnosis they started to feel more in line.
 - Decreased independence and difficulty in performing a wide range of areas of occupation was the biggest challenge
 - Sense of helplessness lead to feelings of hopelessness and depression
 - Feelings of comfort from faith helping individuals to feel they could overcome the challenges
-

- Family support was reported by participants as an adaptation tool to overcome challenges of the disease and to accommodate the disease process into their lives
- Information and advice helped individuals to take action to manage their condition
- Participants reported that lack of communication with the physicians was a common practice. The delay in diagnosis at one hand, and lack of knowledge about the disease and its symptoms and treatment on the other hand, had forced some to try some natural alternative and folk remedies
- Independence and taking charge of own activity was important.

Lubi 2019

- The difficulty in accepting
 - Individual information-seeking
-

- | | |
|-----------------------------------|------------------------------------|
| future deterioration making | making it possible to maintain |
| joining a individuals with PD | control over information and its |
| society difficult | inflow |
| • The need to hold onto | • Autonomous searches for other |
| previous lifestyle and | information and solutions were |
| activities related to it in order | carried out despite |
| to feel better | acknowledgement of the rules of |
| • The need in the early stages | the medical system |
| to continue existing practices | • Understanding of the benefits of |
| to prove they are “normal”. | exercise in promoting wellbeing |
| In the later stages, hiding | and the need for individual |
| symptoms in every possible | responsibility |
| way. | • Lack of information can mean |
| • Acceptance and knowing | questions are unanswered leading |
| limits supporting individuals | to decreases or abandonment in |
| to tolerate reduced | agency |
-

capabilities and continue in
interrupted activities

Stanley-Hermanns	<ul style="list-style-type: none"> • Individuals had to accept the fact that PD will never go away in order to incorporate the PD into their perception of the self • There was an ongoing tension between preserving the self and releasing aspects of the former self in order to reconstruct a self which integrated PD 	<ul style="list-style-type: none"> • The need to continually assess wellbeing and determine daily activities accordingly, allowing individuals to continue doing things which mattered • Listening to their body helped to manage and negotiate daily living 	<ul style="list-style-type: none"> • Many individuals described gratitude for their current state • Reflecting on how they were still themselves despite the PD helped to maintain a sense of integrity • Support groups helped individuals to feel less alone
Williams	<ul style="list-style-type: none"> • In order to make sense of their present, individuals had to build on past relationships, 	<ul style="list-style-type: none"> • Individuals needed to feel a sense of control over late-stage PD by maintaining daily activities, 	<ul style="list-style-type: none"> • Finding meaning helped individuals to focus on living in the present

	<p>identity and events in order to reconstruct the everyday in the light of PD</p> <ul style="list-style-type: none"> • Focusing on past events helped individuals to provide continuity between previous relationships and a shared sense of identity with partners and professionals • Biography and previous life experiences helped individuals to shape their identity in the face of PD 	<p>medication regimes and interests.</p> <ul style="list-style-type: none"> • Taking control of routines was essential and involved participants developing expertise of their symptoms • Medication helped individuals to stabilise their symptoms and maintain independence • In order to maintain a perception of independence, individuals avoided seeing other PD individuals with PDs with more advanced symptoms, fearful of their own dependency 	<p>whilst fighting the PD</p> <ul style="list-style-type: none"> • Individuals were determined not to focus too much on their fears for the future
Vann-Ward	<ul style="list-style-type: none"> • Participants were determined and creative in efforts to 	<ul style="list-style-type: none"> • Individuals took responsibility for own care; they actively monitored 	<ul style="list-style-type: none"> • Relief and gratitude that their condition was not

- | | | |
|-------------------------------|-----------------------------------|----------------------------|
| maintain their identities and | their health and showed | more serious |
| fulfill responsibilities | proactivity regarding treatment | • Having the ability to |
| • Participants redefined | due to beliefs that their efforts | relinquish negative |
| themselves as | would maintain a sense of self- | relationships and nurture |
| accommodating the | responsibility, delay symptoms, | others. New relationships |
| parkinsonian symptoms. | and improve their condition | providing companionship |
| Acknowledging losses of | • Individuals made independent | and feelings of closeness. |
| past identities was a means | decisions regarding how and | • Renewed energy and |
| of coming to terms with their | when to take prescribed and non- | motivation from |
| circumstances | prescribed medications | Parkinson's dance groups |
| • Participants examined and | • Independence of activity was | • Becoming more |
| adjusted former interests to | likewise important, such as | understanding of others |
| accommodate what they | planning how one would travel | less fortunate than |
| were no longer able to do | when driving was no longer | themselves and |
| • Participants strove to be | possible | developing greater self- |
| engaged in daily life. Most | | tolerance |
-

people became involved with
volunteer activities; this
helped them to maintain a
sense of purpose and sense
of self

Habermann	<ul style="list-style-type: none"> • The diagnosis necessitated acknowledgement and acceptance of the disease, but often this was experienced alongside feelings of anger and denial • Individuals faced a major demand of moving from being overwhelmed to acknowledging the illness as their own 	<ul style="list-style-type: none"> • Many began to view their body as uncooperative. The mind had to control the body, instruct it, and try to get it to cooperate • Participants identified a need to know about the disease. • After a few years of diagnosis, individuals confronted the limits of formal knowledge and began to acknowledge that they knew best re managing their condition 	<ul style="list-style-type: none"> • Being able to ask for support and receive support from others were key adaptive tasks in managing the demands of the illness
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- Loss of identity as a working person was a major struggle for participants
 - Changes in role and reinterpreting responsibility was a necessary task within the family
 - Practical knowledge involving timing, dosage and side effects of medication was invaluable
 - Equally important was learning how diet and activity influenced functioning
 - Ongoing monitoring helped individuals deal with the unpredictability of the disease
 - Being unable to drive and giving up working were sources of feelings of dependence
 - Participants focused on the need to accept PD as part of their reality and how they saw themselves
 - participants developed ways to facilitate their engagement in self-care tasks, connecting the importance of these tasks to
 - Social comparison with people of other diseases helped individuals feel lucky for having PD
-

Lutz

- Some strove to maintain their sense of continuity with their pre-PD self and being a person with PD
 - New leisure engagements were important for reconstructing sense of self amidst life transition
 - For many, continuing engagement in occupations acted as a means to maintain the participants' desired identities
 - Support from friends and family helped one maintain meaningful activity
- maintaining a sense of independence
 - Adaptations to physical activity helped individuals to maintain control over their identity
- Photos often represented the positive memories and appreciation whilst living with PD
 - Participants were appreciative of remaining cognitive function
-

Section 3: The experience of living in the pre-manifest stage of Huntington's disease

Lancaster University

Doctorate in Clinical Psychology

2018 Intake

Word count:

7968 words (excluding references, figures and appendices)

Prepared in accordance with Instructions for Authors for '**Psychology & Health**'

All correspondence should be sent to:

Gina Wieringa

Doctorate in Clinical Psychology

Furness Building

Lancaster University

Lancaster

LA1 4YG

Email: g.wieringa@lancaster.ac.uk

Abstract

This paper explored the experience of living in the pre-manifest stage of Huntington's disease (HD)-a hereditary, neurodegenerative disorder. Pre-manifest refers to the stage between receiving a predictive genetic test confirming an individual has the HD gene expansion, and before the person reaches the threshold for receiving a formal diagnosis of clinical (or manifest) HD. Individuals thus know they will definitely develop the disease but live with the uncertainty of when this will be. Data was gathered from ten pre-manifest individuals at least one year after the genetic test using semi-structured interviews and analysed using interpretative phenomenological analysis. Three themes were developed from the data: 'feeling limited by time', 'the perception of stalling time' and 'making the most of time', all highlighting the way in which time holds significant meaning when living in the pre-manifest stage of HD. It is proposed that psychological support could be beneficial for individuals experiencing difficulties with living in the pre-manifest stage of the disease and suggestions for promising interventions are outlined.

Keywords: Huntington's disease; chronic illness; interpretative phenomenological analysis; qualitative; wellbeing

Introduction

Huntington's disease (HD) is a progressive neurodegenerative condition which is caused by an expanded CAG trinucleotide repeat in the gene encoding the huntingtin protein. HD is inherited in an autosomal dominant pattern, meaning a child of a parent with the HD gene has a 50% chance of inheriting the disease. The age of onset differs across individuals, but signs of the condition and clinical diagnosis typically occur aged 30-50 (Carlozzi et al., 2016). The expansion number of the CAG repeat shows a very strong negative correlation with age at onset of motor signs, with a larger expansion number leading to earlier onset (Lee et al., 2012), but this is still hugely variable across individuals. A myriad of functional difficulties is experienced. These include problems with both voluntary and involuntary movements, such as difficulties with swallowing, balance and mobility as well as chorea and bradykinesia (Ho & Hocaoglu, 2011). Cognitive difficulties such as problems with concentration, memory and attention are often also experienced; communication ability is often impacted by motor speech disorders, linguistic difficulties and behavioural changes, leading to negative effects on social interaction and diminished quality of life (Nuzzi, 2018). Emotional difficulties are also common, including apathy, depression, irritability and anger, aggression, suicidal ideation, obsessional compulsive traits, anxiety and psychosis (Eddy et al., 2016; Wetzel et al., 2011; van Duijn et al., 2008). Motor and cognitive difficulties typically worsen with advancing disease progression, resulting in reduced health related quality of life (Carlozzi & Tulskey, 2013). In advanced stages, individuals are likely to require 24-hour care and become totally dependent on others for their most basic needs, a level of need which is very frequently not met by healthcare and social support services (van Walsem et al., 2015).

In order to receive a diagnosis of HD, an individual must experience the onset of motor symptoms (Reilman et al., 2014). However, difficulties across the motor, cognitive and psychiatric domains are likely to occur gradually and may be present more than a decade before the actual diagnosis (Paulsen, 2010). Furthermore, many individuals report anxiety related to potential symptom onset, often many years before showing motor difficulties (Konrad, 2003). There is no way of preventing the onset of the disease, currently no cure, and HD is generally fatal within 15-20 years of clinical diagnosis (Carlozzi et al., 2016). Predictive testing has allowed individuals who are at risk of HD (because of their family history) to find out whether they will develop the disease, prior to showing motor symptoms. Although this has allowed researchers to study the natural course of the disease and provide opportunities for early intervention, the test raises significant ethical issues regarding the potentially devastating consequences which living with the test result entails (Lilani, 2005). Additionally, given the inherited nature of the disease, many individuals will have witnessed first-hand the impact of advancing deterioration. It is often the immediate family who take on responsibility of caring (Coulson et al., 2007); thus, many pre-manifest individuals may be caring or have cared for family members. By offering the predictive test, health services have an ethical and moral responsibility to understand the impact of genetic testing and to support individuals with the long-term consequences (Andersson et al., 2016).

Although much research has been done identifying psychological consequences of a positive test result, the focus is generally on clinical symptoms such as anxiety and depression or on adverse events such as family breakdown or suicide (Mccusker & Loy, 2017). Furthermore, the focus is generally on the short-term (typically under one year since testing) psychological implications of predictive testing (Almqvist et al., 2003). These include symptoms of depression,

obsessive compulsiveness, anxiety, and psychoticism, as documented by large scale quantitative studies (Berrios et al., 2002; Duff et al., 2007; Crozier et al., 2014). Yet there is little knowledge about the in-depth experiences of these individuals from a qualitative perspective, including the longer-term experience after having tested positive. For instance, there has been little research into how a person who has tested positive to the HD gene expansion but is not yet symptomatic perceives their status as ‘embodied risk’ (Kavanagh & Broom, 1997). The uncertainty about when individuals will develop the physical symptoms (and thus receive a diagnosis) is likely to compound any psychological difficulties not least because of the difficulties inherent in deciding whether symptoms in pre-manifest individuals constitute a clinical diagnosis (McCusker & Loy, 2017).

Whilst quantitative studies have highlighted the psychological and social difficulties experienced by those in the pre-manifest stage and sought to understand how they can affect quality of life, the research is limited in its ability to explore how an individual’s meaning of their experience affects them in everyday life. Furthermore, the implications of the research tend to be on the detection of subtle subclinical psychiatric symptoms which might indicate earlier markers for the disease, rather than on the implications for psychological support (Duff et al., 2007; Berrios et al., 2002). Exploring the meaning of an individual’s experience is fundamental in order to better understand variations in psychological outcomes across individuals and how to provide effective psychological support to those experiencing difficulties. Few qualitative studies to date have focused on the lived experience of individuals in the pre-manifest stage of HD. In contrast to quantitative approaches, qualitative research is well suited to develop understanding of the everyday experience of pre-manifest individuals and substantiate the existing quantitative literature. Consequently, this study will take a qualitative approach and use interpretative

phenomenological analysis (IPA) as the methodology because of its ability to explore in detail how individuals make sense of their experience (Smith et al., 2009). Thus, this study aimed to answer the question: ‘how do individuals experience the pre-manifest stage of HD’.

Method

Design

The study design followed guidelines for IPA research (Smith et al., 2009). As a hermeneutic approach it gives precedence to the individual’s personal interpretation of and meaning given to their experience. As an idiographic approach, IPA provides a framework for thorough analysis of individual experiences (Smith et al., 2009), which felt important to capture the complexity of living with a chronic illness (Brocki & Wearden, 2006). Furthermore, IPA recognizes the important role of the researcher in making sense of an individual’s account of their experience (Smith & Osborn, 2003). This awareness felt important to the present study in order to continually reflect on the ways in which the researcher’s active involvement in the participant’s narrative might shape the research outcome (Pringle et al., 2011) and thus the importance of constructive self-reflection in the research process. IPA appreciates that it is through our experiencing that meaning is generated and enables the researcher to more fully explore the way the participant’s experience influences the meaning that they give to being human (Engward & Goldspink, 2020).

Semi-structured interviews were used to ensure the focus was on the pre-manifest stage but were also guided by participants’ responses. Interviews were conducted via telephone due to differing geographical location of participants. Arguably, telephone interviews are an effective option for qualitative data collection due to it complementing the style of semi-structured

interview schedules (Cachia & Millward, 2011) and have been used previously in IPA research (Swift & Wilson, 2001).

Expert by experience

People with either pre-manifest or manifest HD were approached for their input into the design of the study via HD voice (a patient and public involvement group which sits within the UK Huntington's Disease Association (HDA)). They were asked their opinions on the participant information sheet and the invitation letter to ensure they were understandable and inviting. The flyer was simplified and made more professional following this discussion, and the questions in the interview schedule made more accessible.

Participants

To be included in the study, participants were required to be between 18 and 65 and to have tested positive for the HD gene expansion at least one year prior to the interview. It was felt that interviewing participants at least a year following the test would encourage participants to focus on their experiences of living with their genetic status rather than those of the testing period. Participants self-reported that they had the gene expansion for HD as confirmed through genetic testing. They also confirmed that they had not been diagnosed with the disease. Participants were excluded if they had a comorbid significant illness. In choosing the eligibility criteria, the researcher attempted to ensure homogeneity of sampling with regards to "obvious social factors or other theoretical factors relevant to the study" (Smith & Larkin, 2009 p. 50). Two participants were recruited through the HD clinic at an NHS Trust and 8 were recruited through a Facebook advertisement on the Huntington's Disease Association's webpage; all provided written informed consent prior to interview commencement. Pseudonyms were used in

all tables, transcripts and subsequent analyses and any other identifying information was changed. In brief, there were 3 male and 7 female participants, mean age was 38 years (range 25-50) and mean time since their genetic test was 5 years (range 2-10). All were White British. There were 6 individuals who were married or in a relationship with one or more children, 2 married with no children and 2 single with no children. Most (8) individuals were in paid employment. The demographics of participants is shown in Table 1. As can be ascertained from the aforementioned overview, there was some variance in their experience of the pre-manifest stage, which allowed the idiographic and divergent aspects of the experience to be explored.

Recruitment and Data collection

Participants were invited to take part through letters of invitation provided by research nurses at the host Trust, or by responding to the Facebook advertisement. The researcher's contact details were included, and potential participants were asked to contact her if they were interested in participating. After answering any questions and confirming they were happy to go ahead, a mutually convenient time was arranged with each participant for the interview. Consent forms had to be completed in advance of the interview and emailed or posted back to the researcher; at the beginning of this meeting further questions were clarified and participants were asked to reaffirm consent.

Data analysis

Data analysis followed guidance from Smith et al. (2009). There were four stages to the analysis. The first stage involved multiple readings of an individual participant's transcript with comments noted around points of interest (see Appendix A). These comments were closely grounded in the raw data. The second stage involved re-reading of the transcript, with emergent

themes noted in the right-hand column which attempted to describe the participant's experiences (see Appendix A). In the third stage of the analysis, emergent themes were listed, and similar/connecting themes were grouped into superordinate themes (Appendix B). In the fourth stage, analysis was conducted across all the transcripts and superordinate themes for the entire data sample were generated by looking for concordance across participants (Appendix C). Bias was reflected upon and incorporated into the analysis using a reflective journal and regular supervision. Supervision was a space for analytic interaction, highlighting which interpretations belonged to the researcher and which belonged to the participants, strengthening the rigour and transparency of the interpretations (Engward & Goldspink, 2020). Sensitivity to context is equally important to high quality research (Yardley, 2008) and this was enhanced through regular discussion with one supervisor who works in a HD clinic. Furthermore, engaging with individuals with HD prior to data collection and once analysis was complete enhanced the commitment and rigour (Yardley, 2008).

Reflexivity is a core component within IPA, given the need to continually reflect on the meaning which we generate from the data in the analysis process (Shaw, 2010). It was important to have space within supervision to reflect upon the researcher's interpretations were filtered through the lens of the participant's own sense making, in that access to the participant's experience was only generated through the participant's own account of it (Smith et al., 2009).

Ethical approval

The study received ethical approvals from the UK Health Research Authority Tayside Research Ethics Committee (reference: 19/ES/0105) and approval to conduct the study from the Research and Development Department at the host trust.

Results

The findings indicated that the way in which individuals adjust to living in the pre-manifest stage of HD is underpinned through their representation of time. The results span past, present and future and are depicted by three main themes: ‘Feeling limited by time’, ‘The perception of stalling time’ and ‘Making the most of time’. Each of these themes and the sub-themes are described in the following discussion.

Theme 1: “It’s like a trigger for a countdown”-Feeling limited by time

The theme feeling limited by time captures participants’ reflections of the way in which receiving their genetic test results felt to limit the time they had left. This theme incorporated two main sub-themes; the countdown to symptom onset and HD dictating the present.

Countdown to symptom onset.

One recurring theme across participants’ accounts was the way in which receiving their test results felt to initiate a process of counting down to symptom onset. The acuity of the countdown period was maintained by a hypervigilance towards any possible signs which might suggest symptom onset. The overwhelming fear of possible symptom onset was described by most participants and was alluded to by Daisy:

And if you do things like you forget things or names or you put your purse in the fridge or whatever, it only takes a couple of things to happen over a period of time for you to start panicking and think ‘oh my god, is this it, is it starting?’

Many individuals were able to question their fears and think through alternative explanations given the context of their experiences. As Sharon pointed out: “you know, it could well be it's a symptom, or it could just be that you're very, very stressed at that point and actually, the kinds of things that happened to the brain when it's stress [ed] is very similar.” Despite this sense of rationality, symptom questioning was pervasive, resulting in the countdown to symptom onset being an ever-present feature in the background of the participants' lives. Paul summed this up neatly: “is it Huntington's starting? I'll always have that”

The idea of the HD status being an ever-present feature in an individual's life was echoed by others, in which HD became part of “my forever radar rather than my temporary radar” (Daisy) and “an unresolved threat just lurking in the background” (Angie) . For Steve, there was a period at which “it settle(d) into the background noise and has kind of remained fairly constant ever since”. Alongside “threat”, “radar” and “noise”, the contextualized nature of “background” becomes apparent. It would seem as though the word ‘background’ reflects the underlying and insidious backdrop of HD in the participants' lives, always reminding them of the countdown which commenced following the test result.

The countdown to symptom onset was often made more pertinent by seeing the course of parental disease. As Catherine described, seeing the way her mum experiences symptoms: “can be upsetting sometimes because it is a window into your own future almost”. This window into the future is the lens through which many individuals described seeing their own onset. It is sobering that the phrase ‘window into the future’ often describes an opportunity which makes it

possible to see or understand something clearly, in order to affect the present in positive ways. Here though, this window only intensified the reality of their own symptom onset, in which the counting down was exacerbated by family experience of the disease.

Interestingly however, for some individuals, parental onset did not intensify their own fears for their future. Instead, many were too consumed by their caring role to give any space to their own fears. As Joanna explained of symptom onset: “Honestly at the moment it doesn't normally bother me I'm more focused on my mum.”

Similarly, some were able to feel reassured by their knowledge of how disease progression is a very idiosyncratic experience and thus the way HD affected their parent could only give some indication of how it might affect them. Steve described how: “Even going to see mum it's not, I'm not thinking that's going to be me in a few years it's just ‘oh it's my mum’”

HD dictating the present.

The ways in which an individual's HD status dominated their thought processes highlights how participants often felt HD was dictating their present. As Joanna explained with regards to her decision to change careers from events marketing to teaching following the test results:

Honestly it was the Huntington's gene. Originally I trained in events, it's a very active job and you are up and down all the time running around different venues and it's not something I would be able to do long term.

The personification of the Huntington's gene depicts Joanna's sense that the gene had control over her decision making, where she felt forced to re-train because she knew she may not be able to continue in her preferred career once motor difficulties started. The negotiations individuals

made with regard to their HD status is echoed in Angie's account when speaking about navigating the dating world with HD, where she described that she had to "negotiate that with a ball and chain around your foot which is how I view Huntington's." The ball and chain metaphor depicts an image of a criminal being physically restrained and held back; although participants were not to blame for having HD, perhaps this describes the regret Angie alludes to in taking the test, because before the test everything remained possible for her.

It seemed the test results had prompted urgency about what one could realistically afford to do long term. A similar sense of urgency is reflected in Carol's comments: "I've always been sort of a straightforward person, but it made me re-evaluate a lot of things. We moved house kind of because I knew that I needed to do certain things within a certain timeframe." Here Carol described feeling "made" into re-evaluating things and making the house move due to the sense of urgency which came about from having the test. This sense of urgency is triggered by the knowledge that one has HD, such that one felt the result: "Is a trigger for a countdown if you like." (Paul)

Interestingly, the word "trigger" was used by several participants, highlighting the sense felt by participants of the results setting off something, whether that is forced decision making or emotional rollercoasters. It feels apt that the word trigger defines the mechanism by which one fires a gun; perhaps for some individuals the knowledge of the result creates a similar sort of finality to their life before the result.

Theme 2: "I have no intention of becoming symptomatic"- The perception of stalling time

The theme "perception of stalling time" encompasses the way in which individuals gained some sense of agency over the unpredictable and uncertain nature of living in the pre-

manifest stage. Individuals were able to distance themselves from HD by viewing symptom onset as a future reality and by holding onto hope for future medical advances which might delay or eradicate symptom onset.

Distancing self from HD.

The perception of stalling time reflects participants' perspectives that they were able to delay symptom onset by distancing themselves from the reality of their situation, or by situating it as very much a concern for the future. For some, distancing themselves was achieved by viewing their future as free of symptoms:

So this time in 10 years I fully intend to be part of, if not a drug trials programmes, certainly using whatever it is that's going to delay onset for as long it needs, so that's where I'm coming from when I say I've no intention of becoming symptomatic. (Steve)

Not only did Steve describe how he still expects to be asymptomatic in 10 years' time, but also that he has no intention of becoming symptomatic. He distanced himself from present concerns about symptom onset by reinforcing his belief that it will not happen for at least another 10 years.

Most participants seemed to refer to family patterns of the disease in order to maintain the belief that symptom onset was very much for the future. Hannah explained how "nobody in the family [have shown any] symptoms til 60+ so I think I'm very lucky in that sense and I think I use that as sort of a crutch like 'oh I won't be sick until I'm 60'"

Similar certainty about onset was described by Daisy, who was working on an “18-year timeframe”, Catherine, who used her CAG count to “give (her) at least 20 healthy years” and Paul, who thought “there is a good chance that (he’s) got another 5 to 10 years”. For most participants, their own predictions of symptom onset seemed to be heavily influenced by parental experience of symptom onset, providing a sense of certainty that one had a defined period prior to showing symptoms. Thus, family experience of HD helped participants to create time between the present and their own symptom onset, thereby distancing themselves from HD.

Holding onto hope.

A closely related concept which links with an individual’s perception of stalling time was their ability to hold onto hope. As explained above by Steve, not intending to be symptomatic reflected his hope that drug trials and research programmes will eradicate or delay his symptoms. The hope that research progresses before symptom onset was also described by other participants. Carol described the hope she shared with her children when telling them that she had testing positive for HD: “‘by the time mummy is 70 there will definitely be a medicine out there’....so to look at it from that point of view, so the future isn’t all dark and bleak there is hope, there is hope there.” Here Carol described how she holds onto hope that future medical trials will alleviate or stop her symptom onset, much in the same way that Steve described his hope for future drugs progressing faster than his symptom onset. Carol’s use of the word “mummy” in the opening of this statement indicates the significance of hope for her; it is though she needs to remain hopeful as part of her responsibilities as a parent to her children.

The idea that hope is almost necessary in order to manage life in the present is echoed by Catherine when responding to why hope was important: “because if you don't have hope then what do you have?” Hope was not around initially for Catherine. As she described: “there is a

massive adjustment that you go through when you are told you've got the gene and rather than think about hope and stuff you're kind of adjusting to the news that you've got this defective gene". This adjustment period is echoed by Steve, who describes that "once you find out it's quite an emotional roller coaster", Jake, who thought: "it does take a period of adjustment", Angie who had "a period of time where I was really anxious about them and upset by them" and Paul who mentioned that "initially I didn't see that there was a lot of hope and I guess I was approaching it from a terminal illness point of view." It was almost against this backdrop however which hope emerged as even more significant in the participants' lives. Paul summed up the importance echoed by many of having hope in their life:

I guess it stood a chance of almost paralysing me knowing that it was happening or would happenBut I kind of overcame that knowing that, well you could waste a lot of time which is bad for everyone around you.... I want to live my life with the view that I'm free of Huntington's.

Thus, despite the very real difficulties which HD presents to individuals and their families, hope was a very tangible force for participants in preventing them from being paralysed by the fear of HD and allowing them to live their lives.

Theme 3: "I could be hit by a bus tomorrow"-Making the most of time

Making the most of time describes an individual's acknowledgement that, despite symptom onset being a future reality, their time in the present was precious. This is reflected in the participants' determination to stay positive, and in their determination to choose the people they connected with.

Staying positive.

For most participants, the need to stay positive was significant in helping them to make the most of their present. As Carol described: “I need to put all my time and energy into positive things and positive people and doing good things and making a positive impact.” The importance which Carol attached to remaining positive is reflected in its reiteration three times within this statement. It is by staying positive that she was able to maximize her time and energy and really make the most of the time she has, implicitly conveying her caution that she might not be able to make a positive impact for as long as she might like. Similar descriptions of the need to be positive were conveyed by Daisy, who talked about being positive about her future in order to embrace her present; Jake, who felt having a positive outlook prevented problems elsewhere in his life; Joanna, who expressed determination to keep positive until the symptoms started; and Steve, who talked about making a positive out of a negative situation. It is thus through positivity that individuals seemed able to make the most of the time they had symptom free and prevent themselves from dwelling on their potential future.

It is important to acknowledge that one participant found it more difficult to hold a positive outlook on her life. For Angie, the test result “changed things in terms of the way you view your future and your life” and “is a thing that looks over your shoulder saying ‘oh no you can't do that now I'm going to close that door for you’”. Rather than having a positive outlook on her life and seeing possibilities, for Angie it felt as if all the doors were closing, limiting both her opportunities and her ability to be positive. Although she later reflected on ways she dealt with her fears for the future by finding ways to feel positive, her honesty about the realities of HD perhaps reflect the fears which others chose to avoid.

Catherine for instance avoided dwelling on future fears by focusing on the things she wanted to do with her life and reflecting on the ways it might not be HD which limited her life.

As she explained: “I could dwell on it and worry myself sick about it and not do things that I want to do and then go out and get hit by a bus...” Interestingly, the phrase “I could get hit by a bus” is echoed in several participants’ accounts. It highlights that, for many of the participants, there was comfort to be gained from knowing that it is impossible to predict what can happen in the future, regardless of having HD. An individual can either waste time worrying about something which may never transpire, or instead choose to live for today and be positive for what they currently have. In reflecting on the relative benefits of focusing on symptoms versus living their life with a positive mindset, many of the participants were able to “push it to the back of [their] mind and carry on living life” (Daisy).

Choosing valued connections

Making the most of time was also reflected in the way that individuals made conscious decisions about who to spend their time with. For many, spending time with family helped them to make the most of a symptom free present. As Sharon described: “The family is very supportive and it’s [HD] not something that’s not talked about”. For Sharon, it felt very important to talk about and share her concerns with family in order to move on from her worries and embrace her time in the present. This was echoed in others’ accounts: “But I guess I felt a lot better after I talked to Jane [partner] about my concerns in that my anxiety almost completely melted away” (Paul)

In sharing concerns with family members, Sharon felt able to reduce the power of HD from being that which was hidden and stigmatized in the past to something which could be dropped into casual conversation, similar to Carol who valued talking with her family about her HD concerns even if it wasn’t “deep and meaningful”.

Being selective with who one spent time with extended to being less tolerant of people who did not share their perspective about life. As Carol described when reflecting on some of the implications of the HD result: “And I think I’ve become less, not patient, I can’t think of the word for it, less tolerant of idiots and people that are moaning about something completely ridiculous and shallow.” By discarding negative relationships, individuals were able to focus on the positive relations in their life, and even used their HD status as an impetus for coming together as a family. Joanna summed this up nicely when describing how HD had brought the family closer: “life cannot get in the way anymore. You can’t be putting stuff off thinking I’ll do it later I’m too busy because there might not be a later.”

Discussion

The results of this study describe the experience of living in the pre-manifest stage of HD. Embodied in participants’ accounts and reflected in each of the three themes is the salience of time; its ability to take on new meaning and value once an individual is aware of their test result. The participants’ experience of knowing they will develop a chronic illness induced different relationships to time than are experienced by an otherwise healthy person (Jowsey, 2006), where expectations for the future changed, the meaning of their present altered and their past relationships to others also adjusted. For the present participants, this was reflected in their alterations of future plans, their discarding of negative past relationships and their determination to see gratitude in their present. The salience of time for individuals with chronic illness is not new and its origins can be found in the work of Bury (1982), who suggested that a chronic illness can disrupt a person’s previously accustomed experience of time and their expectations of living

within established time-frames of childhood through to old age. His work has since been applied to many chronic illnesses (e.g. Faircloth et al., 2004; Harris, 2009; Carricaburu & Pierret, 1995). Although this research differs from the experience of individuals in the present study who are not yet experiencing symptoms, Bury's account of "biographical disruption" appears to mirror the participants' accounts, in which their understanding of time and their life within it felt to be disrupted upon receiving the genetic test results. Similarly, Charmaz, in her longitudinal work on chronic illness describes how an individual's struggle to control their illness is really an effort to control time and to preserve the defining image of the self (Charmaz, 1993). The experience of chronic illness presents ambiguity for an individual over whom they are in the world, which is translated by the individual and understood through their relation to time (Ellingsen et al., 2014). Such ambiguity and unpredictability are reflected in the participants' accounts as they described their initial struggle when finding out they had tested positive. The uniquely certain entity in our lifeworld, which is time, becomes unpredictable and uncertain when knowing you will experience chronic illness in future. Thus, the participants' way of adjusting to the results was to gain back some control over their present and to exercise some agency in decision making for an uncertain future.

As illustrated in the first theme, many of the participants in the present study expressed hypervigilance towards potential signs of the disease and were constantly 'symptom watching'. They began to interpret insignificant events through a lens of knowing they had the HD gene, generating meaning about an experience they were unsure of and believing it to be a sign of disease. Interestingly, this has also been demonstrated to occur in individuals who are aware they are at risk, but later go on to test negative for the gene (Hagen, 2018). Being unable to determine the meaning of illness-related events presents a life-long challenge for individuals with a chronic

illness (Mishel) and therefore interpreting events through a framework of HD may be a strategy to manage uncertainty and to create meaning (Hagen, 2018). When individuals experience high uncertainty, they are less able to process new information, predict outcomes and adapt to their illness situation (Mishel, 1990) and consequently may experience increased burden of managing chronic illness (Johnson et al., 2006). Mishel highlighted the importance of four factors in evaluating the perception of uncertainty; ambiguity, complexity, unpredictability and inconsistency (Mishel, 1990). For individuals in the pre-manifest stage of HD, indistinct and vague signs of possible symptoms contribute to ambiguity, the lack of contingency between these signs and the illness outcome (i.e. actual onset) contributes to unpredictability and the different messages individuals received about the significance of these signs from healthcare professionals versus their own perception contribute to inconsistency, indicating a high level of illness uncertainty (Giammanco et al., 2015). Reconfiguring this uncertainty into a tangible experience such as symptom onset-from which participants could generate meaning - was a complex undertaking which at times felt to dictate the participants' lives.

As indicated in theme 2, another way that participants managed the unpredictability was to distance themselves from the reality of HD. For many individuals, denial of the situation or of approaching symptom onset appeared to be part of a process which enabled them to gain control over an uncertain future. Within Mishel's (1990) theory, denial might remove the unpredictability of the period between a positive test result and symptom onset. Interestingly, theories of coping typically propose a passive coping style of responding to a chronic condition (such as avoidance or denial of one's future) to be associated with poorer adjustment to the condition (Hesselink et al., 2004; Klein et al., 2007; Park et al., 2008). In contrast, more active coping styles (such as using positivity to manage difficult thoughts or problem-solving activities)

are believed to be adaptive and lead to better adjustment (Pucheu et al., 2004). However, such broad coping styles neglect consideration of the ways in which the effectiveness of a distinct strategy is likely to be context specific (Smith et al., 1997). Denial, for instance, can be adaptive in helping the mind defend itself from threatening information which an individual can do little about, thus allowing them to accept and cope with the life-threatening effects of their condition (Siemerink et al., 2011). By denying knowledge of future deterioration, participants in the present study were able to focus on a present which was free of symptoms rather than a future plagued by uncertainty.

An alternative way in which individuals managed the uncertainty was by holding onto hope. By holding onto hope for future medical advances which might prolong or even eliminate symptom onset, participants described how hope allowed them to live each day as if they didn't have HD. Hope is an internal response which develops over time and helps individuals to deal with difficult and unexpected circumstances (Rustøen & Moum, 1997). In holding onto hope, an individual can visualise a positive outcome to their situation and believes in their ability to overcome the challenges presented to them (Acquaye et al., 2016). For individuals diagnosed with a life-threatening illness, with limited opportunities to improve the expected outcome of their predicament, hope offers a means to gain some agency over the situation and results in meaningful outcomes for the individual (Acquave et al., 2016). As a means of maintaining a healthy coping response despite the adversity they faced and helping them to feel able to cope with uncertainty, hope was an important factor in the participants' wellbeing.

Closely related to hope and the management of uncertainty was the ability to maintain a positive outlook, discussed in theme 3, which allowed individuals to embrace their present and lead a meaningful life. Embracing the present is reflected in another study of pre-manifest

individuals who saw their quality of life as being intertwined with their present attitude and behaviour, and thus were determined to live every day to the fullest (Ready et al., 2011). In the present study, participants also expressed gratitude for the ways in which they were still able to live life to the fullest, in the absence of HD symptoms. Gratitude is typically considered to be an important factor in an individual's ability to find benefit after a traumatic experience (Peterson & Seligman, 2003) and may act as a protective factor in buffering the effects of adversity on a person's wellbeing (Tugade & Fredrickson, 2007). Gratitude has been shown to be positively related to resilience and growth and negatively related to depressive symptoms in survivors of traumatic experiences (Fredrickson et al., 2003) and, as a dispositional trait, may represent the common factor underlying resilience and post-traumatic growth (Kashdan et al., 2006). Higher levels of gratitude may enable individuals to find possible benefits from their illness experience and develop personal qualities such as individual spirituality and appreciation of life; this in turn is likely to lead to a greater acceptance of adversity and improved coping to negative events (Cordova et al., 2001). This sense of gratitude and renewed energy is replicated in other genetic illness, in which individuals experienced new levels of strength and increased fulfilment, described as "redemptive adjustment" (McAllister et al., 2007, p2656). In that study, the experience of knowing they would develop a progressive condition, although traumatic, enabled individuals to find their positive characteristics they were not aware they had, such as their strength and ability to fight the negativity (McAllister et al., 2007). For participants in the present study, this strength and energy could be focused in new directions such as contributing to the research or the work of support groups, resulting in unexpected fulfilment. Embracing the present and finding meaning is one way to find hope for the future and holding a positive outlook in life worked well for many of the participants in this regard.

Clinical implications

The participants in the study described the importance of a positive mindset and an optimistic outlook in order to accept and come to terms with an uncontrollable future. For those who find it hard to adjust to a positive genetic test result, psychological support may be beneficial in helping individuals to maintain control where possible and to support healthy cognitive appraisals towards the uncontrollable aspects of living in the pre-manifest stage. Preliminary evidence suggests that some individuals who are pre-symptomatic or in the early stages of HD value the opportunity to engage with psychological therapy in order to manage the psychological difficulties they experience (Theed et al., 2018). Techniques derived from acceptance and commitment therapy (ACT) such as cognitive defusion (seeing thoughts for what they are-just thoughts) can help individuals to manage difficult thoughts and anxiety, without such thoughts dominating their overt or covert behaviour in problematic ways (Harris, 2009). Supporting individuals to cognitively defuse may help enable the thoughts about loss of control and feelings of helplessness to recede into the background of their life, which many participants in the present study had found beneficial.

For participants in the present study, being able to embrace their present was an important part of their wellbeing. An important component within ACT is encouraging individuals to fully engage with their present experiences and clarify their values, which enabled the participants to lead a meaningful life despite the limitations of their illness (Vowles et al., 2007). ACT not only encourages individuals to step back from their thoughts but also encourages value congruent activity (Hayes et al., 2006). By fully embracing the present and committing to valued activity, individuals may be better able to embrace their pre-manifest status.

An alternative approach to supporting individuals to manage their anxieties related to symptom onset and minimise hypervigilance towards possible signs of the disease is mindfulness based cognitive therapy (MBCT). MBCT teaches individuals to detach from negative thinking habits and repetitive thought patterns by encouraging people to turn their perspective to more engaged, meaningful processing of their experience (Segal et al., 2002) and has been shown to be a promising approach for pre-manifest individuals in helping them to adopt a non-judgmental and less reactive approach (Eccles et al., 2020). Indeed, MBCT encourages individuals to allow thoughts and feelings to occur even when they generate anxiety, placing greater emphasis on trying to understand the cognitive and psychological aspects of their experience rather than being overwhelmed by it (McCay et al., 2016). MBCT can thus help individuals to manage the overwhelm of facing an uncertain and uncontrollable future, instead encouraging them to accept and create meaning from their present experience.

More generally, offering group based psychological interventions may be an important means of providing a space where individuals can experience commonality and validation in their experiences. The benefits of a group-based intervention were articulated by individuals in an MBCT feasibility study, where participants described how it was helpful to meet others in a similar situation and that just working together in a group was itself therapeutic (Eccles et al., 2020). Furthermore, there is evidence to suggest that group sessions can help individuals in the pre-manifest stage of the disease to share personal stories and discuss sensitive areas of HD, which can sometimes be difficult outside of an established setting (Stopford et al., 2019). Many of the participants in the present study valued the forum of the HDA to gain validation and support from others in a similar position, highlighting the utility of group settings.

Limitations

Several limitations of the study should be acknowledged. Participants were largely recruited from a flyer advertised on the HDA website, which may have biased the sample to individuals who were already engaged with the HD community and thus more motivated to increasing awareness and understanding of HD. As a result, their attitude toward living in the pre-manifest stage may not generalise to others in the pre-manifest stage of HD. Secondly, as participants were required to proactively express their interest to participate, it is likely that the participants felt able to talk about their experiences and thus may have been coping better mentally and physically than the general population of HD gene expansion carriers.

It was notable that the majority of the individuals in the present study were coping remarkably well in the pre-manifest stage of HD. It may be anticipated that the eligibility criteria may have falsely limited the sample to those who were coping well; studies of the psychological impact of the test results highlight that the first year after testing may be especially difficult for individuals (Almqvist et al., 2003), possibly because of the difficult period of initial adjustment. Nevertheless, it is worth considering the participant's level of insight of their situation, as lack of insight can be an issue for individuals with manifest HD (Ho et al., 2006). It is possible that executive functioning deficits may have limited participants' introspection; thus, relying solely on self-report is a limitation of the study. Future studies should therefore also obtain partner/family perspectives. Nevertheless, for the participants in the present study, denying knowledge of future deterioration appeared to be a proactive coping mechanism, helping them to accept and cope with the threat of a future they could do little about (Siemerink et al., 2011). As evidenced too by Andrews et al. (2018), lack of insight seems to be more common in individuals diagnosed with manifest HD.

The participants' experience of pre-manifest HD was varied regarding the length of their experience as pre-manifest, and their age both when receiving the test result and at interview. Considering IPA requires homogeneity regarding the research construct being explored, this divergence is a possible limitation. Future research should perhaps employ a more narrowly defined sample, where the timeframe since receiving the genetic test result is limited, say, to 2-5 years. Nevertheless, Smith and Osborn (2003) suggest that how the specificity of a sample is defined depends upon the research construct; where the researched experience is rare (as is the case for individuals living in the pre-manifest stage of HD), the experience may itself define the boundaries of the relevant sample

Future research

Given the present sample may have represented a subset of individuals with relatively high levels of optimism than the typical pre-manifest population, future studies might explore levels of optimism in people with pre-manifest HD and the relationships between levels of optimism and other related factors such as locus of control, perceived stress and life events. A quantitative study would help to explore the relationships between these factors. Pre-manifest individuals could be divided into groups based on estimated proximity to HD diagnosis, using data obtained from their CAG repeat number and parental age of onset. Furthermore, a longitudinal study would be beneficial to consider how these variables change over the period of an individual's pre-manifest journey. There is evidence to suggest that individuals less than 9 years away from diagnosis may experience higher levels of wellbeing and lower perceived stress than other pre-manifest groups (Downing et al., 2012). There may be several reasons for this, such as that these individuals have had more time to adjust to the results (Downing et al., 2012) or have less insight into their difficulties (Duff et al., 2010). Alternatively, it may be that the

longer period of time since diagnosis may have resulted in individual's developing more adaptive views of their illness and experiencing meaning from their experience, both of which have been shown to lead to increased optimism in individuals with other chronic illness (Lee et al., 2006). Given little is known about the effects of optimism on perceived stress, mood and overall wellbeing, and the relationship between levels of optimism and HD disease trajectory, this exploration would offer an important avenue for future research.

Conclusion

This study has highlighted the difficulties experienced by individuals when adjusting to the pre-manifest stage of HD. The results found that the experience of living in the pre-manifest stage of HD involves navigating the difficulties of feeling limited by the time they have left symptom free by holding onto hope for future treatments and in making the most of their symptom free present. Indeed, positivity and hope were key factors in helping individuals to manage their anxieties and dealing with ongoing uncertainty related to future deterioration. In developing our understanding of how people in the pre-manifest stage maintain their wellbeing, we may be better equipped to support those who show more difficulty in managing their experiences.

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Table 1: Participants' demographics

Participant	Gender	Age	Marital status	Children	Time since genetic test
Paul	Male	44	Married	One	3 yrs
Angie	Female	36	Single	None	4 yrs
Daisy	Female	31	Married	Two	5 yrs
Catherine	Female	38	Single	None	6 yrs
Steve	Male	50	Co-habiting	One	3 yrs
Joanna	Female	29	Married	Two	2 yrs
Carol	Female	50	Married	Two	4 yrs
Hannah	Female	25	Cohabiting	One	4 yrs
Sharon	Female	36	Married	None	8 yrs
Jake	Male	45	Married	None	10 yrs

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Appendix 2: B Extract from transcript detailing initial coding and theme identification (Sharon)

<p>"People with HD"-choosing not to associate herself with this group Obviously implies expertise</p> <p>Took a long time for mum to accept</p> <p>Awareness of previous difficulties for mum-value judgment of better-what does better mean (for Sharon vs for mum)</p>	<p>problems, and obviously people with HD who it can be difficult for them to accept, you know, the diagnosis. So, yeah, but it did take a long time for her to be able to, to accept it but she is, she has accepted it now and is in a much better place now.</p> <p>Interviewer: Yeah, I'm glad, glad to hear that and you mentioned that it can be difficult for people to accept it. Do you think that you've accepted it?</p>	<p>Difficulties in accepting the diagnosis</p> <p>Acceptance as a long process</p>
<p>Felt she would be able to accept it quicker than she did? Language implies reluctance to acknowledge-reflective of disappointment of self?</p> <p>More than 5 yrs but doesn't want to say?</p> <p>Settled in-idea that it is part of her but not necessarily so unwelcome</p> <p>"Okay"-very qualified relationship</p> <p>Engaging oneself fully in the research-perhaps as a way to gain some sense of control over the uncontrollable?</p> <p>Language conveys urgency-time feels limited?</p> <p>The need to contribute</p> <p>No repeated 4 times-highlights lack of any cure/treatment. Gravity of negativity amplified by language use</p> <p>Sense of achievement in taking part</p> <p>Hope for future involvement</p> <p>Providing a different focus for one's life</p> <p>Hope for future drugs</p>	<p>Participant: Yes, I think it took me longer than I thought it would to really for it to really be able to sink in. I mean yeah, probably easily five years, I would say, for it to really have settled in and me sort of be, be okay with it. I mean, I'm, as soon as I got pretty much, as soon as I got my result I signed up to do any form of research I possibly could. And that was the thing for me was that if I felt that I was actively participating in a disease that you know, there is no treatment and there is no there's no treatment let alone even, you know, no cure or anything. So I did it and I do, I take part usually at least one form of research each year. So I've done quite a few studies. And I would, I'm certainly hoping to take part in the big drug trials once they're at the stage where they take more participants, when they do take</p>	<p>Coming to terms with it as something which takes time</p> <p>Actively participating in the research</p> <p>HD as uncontrollable, incurable</p> <p>Sense of achievement in contributions</p> <p>Hope for future involvement</p> <p>Hope for future drugs</p>

<p>Highlighting the difficulties-reference to the fact that it's very difficult to understand when don't have HD?</p> <p>Justifying her decision</p> <p>Collective pronoun-validating her decision as shared with sister</p> <p>Test taking as a reasoned decision</p> <p>Reiterates own peace of mind-the peace which comes with certainty</p> <p>Planning for the future.</p> <p>2nd person "you know"-invites reader to understand her point of view</p> <p>The difficulties of this period</p> <p>Had talked through the implications</p> <p>Language implies inevitability-suggesting there was no other option</p> <p>Having knowledge which someone else doesn't want to know</p> <p>Mum fine about them getting tested but didn't want to know herself-a paradoxical situation</p> <p>Reiterates "fine about it/us"-keen to convey the sense that their decision resulted in right outcome?</p>	<p>Participant: I mean, it's a very difficult position, you know, I mean, we wanted to do it for ourselves. That was our reasoning behind it, we wanted to have, for our peace of mind we needed to know so that we could plan for the future, so you know.</p> <p>Yeah, I mean, there was there was no easy way around it and we did talk about that through the genetic counselling in that it unfortunately is one of those things that we did know her result without her wanting to know it. You know now when we talk about it you know she is she's fine about it and can understand why we wanted to get tested. So she was always fine about us getting tested it but she didn't want to know about herself but she didn't, didn't have a problem with us.</p> <p>Interviewer: But I guess with a positive result the two of those can't go alongside each other, can they? In terms of not wanting to know her own status but being happy for you to get find out for yourselves?</p>	<p>The implications of HD as a genetic condition</p> <p>Test taking as reasoned decision</p> <p>Test taking as a way to get peace of mind, plan for future</p> <p>No easy way around it</p> <p>Unfortunate consequences of the test</p> <p>A process to being fine about it</p> <p>Collective identity in test taking</p>
<p>Test taking as very difficult</p> <p>Difficult for whom-self or mum-or both?</p> <p>Changed focus from self to mum</p> <p>Always a challenge</p> <p>Anybody you know-normalises experiences</p>	<p>Participant: Yeah, yeah. Yeah, I mean, it was very difficult because she was struggling really with you know, mental health and it was, it's just always a challenge. I mean with anybody you know about, people who have mental health</p>	<p>Difficult period around test taking</p> <p>Challenge as nothing new</p> <p>Mental health difficulties</p>

Appendix 2-C: From initial concepts to emerging themes (Steve)

Initial concepts	Emerging themes	Supporting Quotes
Having control over when one discloses the result Test taking as a way to find out the unknown The result enabling plans to be made Knowledge as something one can do something about	The test result as providing power	It's not so much keeping it a secret it's just not letting information come out until it's ready to and in the right way It's far better to know than not to know And again she tested positive before we told my mum. I told her and we sort of came up with the plan You can do something about it or not as the case may be. But can do something about it
Reassurance provided by family history of the disease Reliance on the drug trials as a way to maintain belief in not getting symptoms Hope that one will never reach symptom onset Protecting himself from the emotional side of HD The ability to throw walls around in your mind	No intention of becoming symptomatic	So, it's been up and down but obviously it's mitigated by the fact it's late onset I am relying on the fact that there are many drugs in the pipeline that will delay it I have no intention of becoming symptomatic So I could protect myself from the emotional side of things by thinking, you know, my mum's gonna be really upset if she finds out It's having an ability to throw, throw walls around in your mind throw walls around things you don't want to think about
The need to do things you've always wanted to do Living for the moment No stressing over the small stuff Think about the positive things Gratitude and feeling fortunate	Re-evaluating what matters in life	Yes, it does focus you, it means I do try to just do things I want to do ...right you've got to go and do all your bucket list type things and do various things and live for the momenttry not to get too stressed about stuff It's had a positive impact because it's made me do things and appraise various aspects of my life I'm fortunate in that the work that I do is inside and outside so it's quite a mixture

Participating in research as way to cope Research as a selfish act-a way to get the drugs first Research as a way to help others Making a positive out of a negative	The need to contribute to the research	It's my own way of burying my head in the sand. Just ignoring it, and everyone that has got it, so I can take part in the research like I say in a slightly selfish version of if the drugs come out or if some form of delaying the onset comes about then I'm first in the queue to get it all Taking part in the research it makes you feel good because you are helping other people to find out thing trying to make a positive action now to something that is quite negative
Constantly questioning HD as constant background noise Emotional rollercoaster as being a typical response	Emotional rollercoaster of the result	So that's the biggest problem is just constantly going "yeah is that an early sign, or not?" It's that sort of constant background noise in your life ...this seems to be typical talking to people that have been through the testing process as well in that once you find out it's quite an emotional roller coaster

Appendix 2-D: Theme identification across participant transcripts¹

Individual participant themes	Refining Themes	Superordinate theme (subordinate themes in brackets)	Participants contributing to the theme
Assuming the worst, feeling one knows the result, period of adjustment after the test, ups and downs, lack of control, test impacting future planning, test changing decision to have children, doors closing after the test	The implications of test taking in limiting their life and the emotions which come with that	Feeling limited by time (countdown to symptom onset, HD dictating the present)	Paul, Angie, Daisy, Steve, Joanna, Catherine, Hannah, Sharon, Carol, Jake
Activities aimed to prolong symptom onset, family experience of the disease, dark side of HD, knowing when one will get symptoms based on family pattern, hope for future cure, believing own symptom onset will be different, taking part in the research to contribute to the HD knowledge	Using family experience of the disease to predict own onset whilst hoping for medical cures to alleviate symptoms	The perception of stalling time (distancing self from HD, holding onto hope)	Paul, Angie, Daisy, Steve, Joanna, Catherine, Hannah, Sharon, Carol, Jake
Making the most out of life, holding onto the positives, being grateful for family and friends, feeling able to cope, refocusing attention, not worrying about the little things, avoiding thoughts of the future, reflecting on the worse situation of others, not putting up with negative environments	Being positive in the present and making the most of the now and spending time with valued others	The need to embrace time (the need to stay positive, choosing valued connections)	Paul, Angie, Daisy, Steve, Joanna, Catherine, Hannah, Sharon, Carol, Jake

¹ All participants contributed to every theme

Section 3: Critical Appraisal

Lancaster University

Doctorate in Clinical Psychology

2018 Intake

Word count: 3974 (excluding title page and references)

Prepared in accordance with Instructions for Authors for '**Psychology & Health**'

All correspondence should be sent to:

Gina Wieringa

Doctorate in Clinical Psychology

Furness Building

Lancaster University

Lancaster

LA1 4YG

Email: g.wieringa@lancaster.ac.uk

Overview

This critical analysis will first consider how the findings from both the research paper and the literature review might relate to one another, and in doing so suggest themes which might be applicable to living with a chronic illness more widely. The analysis also reflects on the clinical implications of the two papers and the need for a more coherent definition of adjustment. The analysis which follows will then discuss personal reflections on conducting the research as well as considering the crucial role of reflexivity to this process. Finally, the paper will make suggestions for areas of future research.

Research Findings

Before drawing comparisons between the papers with the aim of discussing the implications for transdiagnostic themes for adjusting to living with a neurological condition, it is worth considering the validity of the findings of the individual papers. Regarding the meta-synthesis, the themes developed from the data were all present in several of the papers assessed as being of high quality on the Critical Appraisal Skills Programme (CASP) checklist, suggesting the validity of the findings of the review. Regarding the research paper, which sought to obtain the experiences of individuals in the pre-manifest HD, it is worth considering possible deficits in their levels of insight, as problems with executive function are observed in individuals with manifest HD (Ho et al., 2006). Regarding the research participants, whilst no objective determinant of their cognitive function was obtained, the participants seemed to demonstrate a high level of insight into the future challenges they faced, reflected in their planning for future deterioration. Whilst not expressing acute psychological distress, participants did reflect on the real psychological difficulties they had experienced at times; as such their avoidance of thinking about future deterioration did not reflect lack of awareness but served as a coping mechanism to help them cope with their

fears. As such, the research findings from both papers can be considered valid in considering the experiences of individuals of the phenomenology under question.

The research paper found three main themes that captured participants' experiences of adjusting to living in the pre-manifest stage of Huntington's disease (HD): (1) Feeling limited by time-"Trigger for a countdown", (2) The perception of stalling time-"I have no intention of becoming symptomatic and (3) Making the most of time-"I could be hit by a bus tomorrow". Such findings highlight how the nature of time takes on a new level of meaning for individuals when they become aware they will develop HD. Describing their experiences through the lens of time may be one way in which individuals make sense of their self as an individual with pre-manifest HD and create a coherent narrative of their illness experience (Ellingsen et al., 2014). Interestingly, the meta-synthesis-exploring how individuals adjust to living with Parkinson's disease (PD)- proposed three main themes relating to the process of adjustment: 'maintaining a coherent sense of self', 'feeling in control' and 'holding a positive mindset'. The theme 'maintaining a coherent sense of self' revealed a similar need for participants to create a narrative of their illness experience in order to maintain their sense of self. This narrative reflected how individuals with PD can integrate their illness identity into their previous sense of self through a process of accepting their limitations alongside the need to maintain past roles and responsibilities (Williams & Keardy, 2008). Although the research participants were not yet experiencing any symptoms of their condition, accounts from across the papers indicate that adjustment may be a process which begins at the point at which one experiences a threat to their sense of self.

For individuals living in the pre-manifest stage of HD, regaining control over the uncontrollable helped them to manage its unpredictable nature, in which they described clinging onto hope for future medical treatments and contributing to the research as important ways of 'stalling time'. This need to regain control over the illness trajectory was also

prevalent in the meta-synthesis, contributing to the theme 'feeling in control'. For individuals living with PD, the physical and mental deterioration prompted many to start or re-engage with physical activities, in order to provide them with a sense that they were able to prolong illness deterioration. Similarly, medication management and control over their day to day routines helped individuals to feel better able to manage their symptoms. Consequently, both papers describe the connection between the uncontrollable nature of the illness and the personal efforts and motivation of participants to regain control. Interestingly, although some participants interviewed for the empirical paper described the importance of physical activity in order to prolong symptom onset, it did not emerge as a significant theme. Perhaps the very tangible nature of physical deterioration experienced by those with PD heightens the relevance of exercise in order to maintain one's wellbeing. Gaining control over symptoms through self-management (primary control) allowed individuals to feel more in control of their life and better able to adapt to disease related limitations (secondary control). This felt important given the uncontrollable nature of disease progression. Even if control of disease may not be possible, global life control and feelings of self-efficacy rather than control over the disease per se can also be important for individuals experiencing PD symptoms (Eccles & Simpson, 2011). For individuals living in the pre-manifest stage of HD, primary control may be higher given their functioning remains intact and they are not yet experiencing symptoms, thus secondary control efforts (such as the belief that exercise can prolong symptom onset) may have less relevance. As long as an individual's circumstances are able to promote personal control efforts, there is likely to be a fit between an individual's perception of control and what they are actually able to influence, such that secondary control efforts are less important (Reich & Zautra, 1990).

Despite the challenges imposed by a progressive, neurodegenerative condition, participants across both papers were able to reflect on the positive aspects of their

experiences. Indeed, such experiences were deemed integral to the adjustment process. It was this ability to focus on the positive aspects of the knowledge and embrace their present which helped the research participants to maintain a positive mindset and look optimistically towards one's future. Many of the individuals living with PD also described how acknowledging their future deterioration allowed them to focus on their present with positivity and determination. By focusing on the small things which generated happiness in their daily lives and being grateful for one's circumstances, individuals were able to reduce the burden of PD. A positive mindset and sense of gratitude were also reinforced in both papers through social comparison. It seems that irrespective of their own degree of impairment, an individual can strengthen their self-image by finding someone else to compare with who they perceive as worse off than themselves (Eriksson & Ahlgren, 2013). In a review outlining the process of adjustment to chronic illness, Sharpe and Curran (2006) highlight how benefit-finding in chronic illness is an adaptive strategy that decreases the incongruence between situational and global meaning. Congruence between an individual and their environment enables an individual to satisfy their needs and motivations and thus contributes to positive adjustment (Lawton, 1985).

Of equal relevance is the way in which the papers provide novelty in describing condition-specific aspects of individuals' experiences. Regarding the research paper, individuals described the emotional rollercoaster of the period after receiving the test results. Although the meta-synthesis described the emotional challenges of receiving a diagnosis, the complexity of this experience was not described in detail. In contrast, individuals living in the pre-manifest stage of HD described the seemingly precarious nature of the peaks and troughs, where fear and hope could be described as simultaneous experiences. It was this ability to hold onto hope despite the "constant background threat" of HD which allowed individuals to adjust to their situation. The meta-synthesis also described how some individuals perceived

PD as a threat to their identity, with physical and cognitive deterioration threatening fundamental aspects of who they wanted to be. However, in the research paper, this threat was transformed into something of an unrelenting nature; whilst most individuals with PD acknowledged the threat as a transitory concept, for individuals living in the pre-manifest stage of HD this threat was always lurking in the background of their life. It was the fear of the unknown which seemed to heighten the intimidation of the threat; for individuals living with PD, diagnostic support and first-hand experience of symptoms allowed them to accommodate and manage how the threat operated within their lives.

Clinical Implications

Both papers indicate the psychological processes which seem to be beneficial to support someone either diagnosed with, or with the knowledge of, a progressive neurodegenerative illness. This is important considering that, in conditions such as PD, a greater understanding of the role of psychological difficulties in presenting distress has led to an increasing number of psychologically informed interventions for people with PD (Zarotti, Eccles et al., 2020). This has important relevance for the research paper, given the current research on psychological interventions is extremely limited in people affected by HD (Zarotti, Dale et al., 2020). The empirical research develops our understanding of the important factors which facilitate an individual's adjustment to the condition, and in doing so provide a focus for the application of psychological approaches.

The meta-synthesis identified the importance of peer support groups in offering a space for sharing and validating of one's experiences, a concept echoed by individuals in the research paper in helping individuals to feel less alone. Providing a safe forum to share experiences and managing emotive group dynamics is an area of expertise which lends itself well to the role of clinical psychologist (Brown et al., 2002). The benefits of group-based

intervention was identified in the systematic review, and also in a study of individuals in the pre-manifest stage of HD, where participants described how it was helpful to meet others in a similar situation and that just working together in a group was itself therapeutic (Eccles et al., 2020). Furthermore, there is evidence to suggest that group sessions can help to provide a safe space for self-expression (Stopford et al., 2020). Such group-based interventions may involve the principles of acceptance and commitment therapy (ACT), which encourages individuals to embrace valued living despite the challenges they face (Harris, 2009). ACT has shown promising evidence of its effectiveness for people with neurological conditions (Graham et al., 2014; McLeod, 2015; Whiting et al., 2019). Considering the literature on adjusting to PD highlighted the importance of social comparison in benefit finding and positive thinking, introducing ACT principles through a peer-support format appears fitting. Any group-based intervention needs to accommodate the neuropsychological profile of the individual group members, as an individual with cognitive difficulties may find abstract psychological concepts or ACT metaphors more challenging, or experience difficulties with inhibiting thoughts and thus struggle with cognitive defusion (Hill et al., 2017). Furthermore, difficulties in skills which underlie social cognition have been observed in conditions such as PD (Dodel et al., 2010), which could preclude therapeutic work which requires social interaction. Nevertheless, more recent studies have suggested that difficulties in social skills observed in some individuals may reflect deficits in executive function rather than social cognition per se, and impaired social cognitive performance can be eliminated by reducing the processing demands of the task (Foley et al., 2019). This highlights the need to adapt to tailor delivery of the approach to the person's idiosyncratic neuropsychological context (Wilson et al., 2009). This might involve using more written handouts for complex concepts, using more visual aids, explaining metaphors using more concrete examples and repeating key messages (Hill et al., 2017).

Reflexivity

An assumption of IPA is that researchers interpret data through the lens of their own psycho-social context and prior understandings (Shaw, 2004); IPA thus involves a dialogue between the participant's understanding of their experience and the researcher's interpretation of that understanding (Shaw et al., 2014). Reflexivity is thus a core component within IPA, given the need to continually reflect on the meaning which we generate from the data in the analysis process (Shaw, 2010). The active involvement of the researcher in the generation of the participant's narrative has been criticised as potentially limited the validity of the research results (Pringle et al., 2011). Consequently, it was important to follow guidelines outlined by Yardley (2008) which help to improve the validity of qualitative research. I implemented a number of processes to improve the validity of this qualitative research. Of note, the supervisory process enabled me to explore the theme identification and content of the interviews to ensure credibility of the interpretation. Regular discussion with my field supervisor, who works in a HD clinic, also contributed to the sensitivity to context. With regards to the meta-synthesis, regular reflection using a reflective diary enabled me to appreciate my own preconceptions of what adjustment might mean when considering the relevance of papers and in the identification of papers. Consequently I was able to appreciate my own influence on both the way I which I collected data and the weight of interpretation given to participant accounts (Finlay, 2002).

Personal reflections

Recruitment of participants

Recruiting participants was initially focused on inviting people through a clinical psychology service, as it was hoped this would provide accounts of individual's who may provide insight into their experience of HD alongside an appreciation of what psychology can

offer to the adjustment process. It also meant that participants were already known to one of the supervisors, thus facilitating eligibility checks and enabling recruitment to be undertaken by research nurses who were already known to the individual. However, following initial difficulties with recruitment, and considering the additional burden it placed on the research nurses (who also maintained a clinical role), it felt appropriate to extend recruitment to the Huntington's Disease Association (HDA), which was identified as the second recruitment route within the ethics application. In retrospect, it may have been helpful to recruit through one channel only, as a key aspect of IPA studies is the homogeneity of the sample (Smith et al., 2009), which produces an in depth analysis of participants' accounts rather than a representative sample (Touroni and Coyle, 2002). I think recruiting through a community organisation rather than solely through NHS services ensured I also captured the experience of those not receiving the support required to adjust to their condition and this lack of healthcare support is something which many individuals with HD experience (Dawson et al., 2004).

Eligibility criteria for participants were that they needed to have tested positive for the HD gene one year or more prior to interview and to have no significant comorbid physical condition. This felt important because living with a comorbid physical health condition is likely to present significant additional challenges to living with HD pre-manifest and it would be difficult to separate out those experiences from the experiences which were related to having the HD gene. Nevertheless, future research should focus on the experience of those with pre-manifest HD and a comorbid physical health condition; given such a population are likely to be in even greater need of psychological support, a better understanding of their experiences is warranted. It was notable that the majority of the individuals in the present study were coping remarkably well in the pre-manifest stage of HD and themes of positive coping were predominantly not reflected in my field supervisor's experience of those

accessing psychological input. It may be anticipated that the eligibility criteria may have falsely limited the sample to those who were coping well, given that studies of the psychological impact of the test results highlight that the first year after testing may be especially difficult for individuals (Almqvist et al., 2003). The experience of individuals in the pre-manifest stage of HD is unlikely to be one of linear adjustment and difficulties are to be anticipated when living with the knowledge of future deterioration. It has been suggested that individuals around 9-15 years away from HD diagnosis may experience increased stress due to possible signs of the disease becoming more evident (Downing, et al., 2012). Most of the participants in the present study were likely further away from diagnosis yet had experienced some time to adjust to the initial test results which in turn, may have contributed to them describing their adaptive experiences.

Data analysis

IPA was chosen as my method for data analysis given its ability to offer an idiographic lens on each of the participant's experience. The way in which emphasis was placed on the uniqueness of each individual and the personal meaning ascribed to their experience (Smith et al., 2009) felt important to me in order to truly validate each person's account. In contrast, an alternative approach such as grounded theory or thematic analysis focuses on the emerging patterns within the data or superordinate theories in its analysis, rather than the way in which an experience is embodied and given meaning by the individual's relationship to their experience (Starks et al., 2016). IPA enables the patterns in the data to generate salience or meaning for the researcher, but crucially, an individual's voice in those patterns are not lost.

An alternative approach to hearing the voice of the participants is narrative analysis. Narrative analysis is an approach where the participants' stories are the raw data (Bleakley,

2005) and focuses on the context of the narrator, including their culture, history, identity and lifestyle (Lieblich et al., 1998). An attempt is made to understand what participants do with narratives in the process of storytelling and how they position themselves in the storytelling process (De, Fina & Georgakopoulou, 2015). The narrative analyst also recognises that telling stories is fundamental to the process of identity construction and seeks to explore how this identity construction takes place in a situated social context. It helps us to understand the different levels involved in stories and the way it functions as a means for the individual to share and disseminate knowledge. As an approach however, the focus is on studying a small number of individuals, collecting their stories and reporting their experiences and then chronologically ordering the meaning of those experiences (Creswell et al., 2007). The aim is to restory the participants' narratives by collecting their story, analysing the key elements and then writing them within a chronological sequence (Ollerenshaw & Creswell, 2000). Whilst time did emerge through the process of data analysis, the meaning of time in relation to the individuals' experiences did not account for a chronological order but reflected the participant's concurrent differing understandings of time as situated in their present day experience. As an approach it does not typically deal with self-contained stories and does not treat an individual's story as a holistic account, instead focusing on the meaning of an individual's story as it unfolds in a societal context (Esin et al. 2014). As the aim of the present research was to gain a better understanding of the experience of the pre-manifest stage as obtained by individual accounts, rather than focusing on the context of an individual's narrative and how it had shaped their experience, IPA was deemed more appropriate for the present research.

One criticism of the IPA approach, namely that it incorporates researcher bias (Kacen & Chaitin, 2006). Nevertheless, IPA openly acknowledges this criticism, being explicit about the assumptions of researcher involvement in interpretation of the data and highlighting the

importance of reflexivity as a means of quality control (Berger, 2013). Smith and Osborn (2003) acknowledge that the researcher often influences the direction of a participant's account and must be mindful both of how much directing of the interview is acceptable but also that different directions often enlighten the exploration. An essential component within IPA research is the recognition that the researcher's relationship to the participant's account is dependent upon and shaped by their subjective perspective and that this subjective perspective also occurs within their specific context (Pietkiewicz & Smith, 2014). This researcher interpretation of the participant's interpretation of their experience, a double hermeneutic, is made transparent within the research process. A fundamental part of the IPA process is continual reflexivity, ongoing engagement with personal context and immersivity with the data, which ensure the results are grounded in the data. IPA understands that the experience of an individual cannot be separated from the individual's context and their relationships (Chapman & Smith, 2002); for the individuals in the present study, living with pre-manifest was a familial and social experience, thus IPA appeared to be the most appropriate approach.

Future research

The sample in the empirical paper were all White British, despite recruiting from an NHS Trust with a diverse ethnic population and a social media channel which was not limited to geographical location. Although this lent itself well to the requirements of homogeneity within IPA studies, it raises the questions as to whether services for individuals with HD/pre-manifest HD are accessible to ethnic minority groups. Furthermore, the meta-synthesis included studies which were all conducted in European countries with western populations, suggesting research on adjustment processes more widely has been limited to Eurocentric populations. An inclusion criterion for the papers in the meta-synthesis was that the paper had to be written in English, further limiting the research to those conducted in Western countries.

This limits the generalisability of the study and means we have little understanding of adjustment processes to neurological disease within other cultures. It may be anticipated that adjustment is a culturally dependent construct, given many aspects of the adjustment process was intertwined with an individual's social context and the meaning of illness within their society (Stanton et al., 2007). Future research should therefore seek to explore how individuals from other cultures adjust to the experience of a neurological disease.

It is also noteworthy that the levels of optimism in the research participants', whilst appearing to contribute to successful adjustment for the studied individuals, could be considered a form of denial. Their ambitious hopes for treatment which would prevent their own symptom onset was not matched by the reality of the medical advances; so too, their convictions that their own symptom onset was a distant reality because of their parent's disease onset belied the reality that parental age of onset is only one predictor of the age of onset of the offspring. Both beliefs could be considered unrealistic levels of optimism which helped them to avoid or deny the reality of their likely future. Researching the longitudinal outcomes of unabated optimism to better understand whether there is an 'optimal' level of optimism is important in order to help psychological professionals to tailor their interventions. Regarding PD, there is evidence to suggest that the relationship between optimism and outcome may be non-linear (Hurt et al., 2014), where individuals with very high levels of optimism have been observed to use less adaptive coping strategies and do not experience higher levels of psychological wellbeing than those displaying medium levels of optimism (deRidder et al., 2000). It is suggested that high levels of optimism may prevent people from feeling the need to self-manage their condition or take action to minimise health related deterioration (deRidder et al., 2000), much in the same way the denial shown by participants in the present study may have prevented them from engaging in efforts to improve their wellbeing. Nevertheless, optimism has been shown to be beneficial for

individuals with PD in helping to protect against the effect of negative illness perceptions (Hurt et al., 2014), highlighting that some level of optimism is important. Future research needs to better understand the relationship between levels of optimism and clinical outcome for those experiencing other neurological conditions such as HD. Educating health professionals about how to empower individuals living with a progressive condition through realistic levels of hope, whilst broaching sensitive conversations about the very real prognosis, may better equip individuals with the tools needed to self-manage or indeed to adjust to the realities of their condition.

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Welcome to the Integrated Research Application System**IRAS Project Filter**

The integrated dataset required for your project will be created from the answers you give to the following questions. The system will generate only those questions and sections which (a) apply to your study type and (b) are required by the bodies reviewing your study. Please ensure you answer all the questions before proceeding with your applications.

Please complete the questions in order. If you change the response to a question, please select 'Save' and review all the questions as your change may have affected subsequent questions.

Please enter a short title for this project (maximum 70 characters)

Living in the premanifest stage of Huntington's disease.

1. Is your project research?

☒ Yes ☐ No

2. Select one category from the list below:

- ☐ Clinical trial of an investigational medicinal product
- ☐ Clinical investigation or other study of a medical device
- ☐ Combined trial of an investigational medicinal product and an investigational medical device
- ☐ Other clinical trial to study a novel intervention or randomised clinical trial to compare interventions in clinical practice
- ☐ Basic science study involving procedures with human participants
- ☐ Study administering questionnaires/interviews for quantitative analysis, or using mixed quantitative/qualitative methodology
- ☒ Study involving qualitative methods only
- ☐ Study limited to working with human tissue samples (or other human biological samples) and data (specific project only)
- ☐ Study limited to working with data (specific project only)
- ☐ Research tissue bank
- ☐ Research database

If your work does not fit any of these categories, select the option below:

☐ Other study

2a. Please answer the following question(s):

- a) Does the study involve the use of any ionising radiation? ☐ Yes ☒ No
- b) Will you be taking new human tissue samples (or other human biological samples)? ☐ Yes ☒ No
- c) Will you be using existing human tissue samples (or other human biological samples)? ☐ Yes ☒ No

3. In which countries of the UK will the research sites be located? *(Tick all that apply)*

- ☒ England
- ☐ Scotland

- ☐ Wales
☐ Northern Ireland

3a. In which country of the UK will the lead NHS R&D office be located:

- ☒ England
☐ Scotland
☐ Wales
☐ Northern Ireland
☐ This study does not involve the NHS

4. Which applications do you require?

- ☒ IRAS Form
☐ Confidentiality Advisory Group (CAG)
☐ Her Majesty's Prison and Probation Service (HMPPS)

**Most research projects require review by a REC within the UK Health Departments' Research Ethics Service.
Is your study exempt from REC review?**

- ☐ Yes ☒ No

5. Will any research sites in this study be NHS organisations?

- ☒ Yes ☐ No

5a. Are all the research costs and infrastructure costs (funding for the support and facilities needed to carry out research e.g. NHS Support costs) for this study provided by a NIHR Biomedical Research Centre, NIHR Collaboration for Leadership in Health Research and Care (CLAH C), NIHR Patient Safety Translational Research Centre or Medtech and In Vitro Diagnostic Cooperative in all study sites?

Please see information button for further details.

- ☐ Yes ☒ No

Please see information button for further details.

5b. Do you wish to make an application for the study to be considered for NIHR Clinical Research Network (CRN) Support and inclusion in the NIHR Clinical Research Network Portfolio?

Please see information button for further details.

- ☐ Yes ☒ No

The NIHR Clinical Research Network provides researchers with the practical support they need to make clinical studies happen in the NHS e.g. by providing access to the people and facilities needed to carry out research "on the ground".

If you select yes to this question, you must complete a NIHR Clinical Research Network (CRN) Portfolio Application Form (PAF) immediately after completing this project filter question and before submitting other applications. Failing to complete the PAF ahead of other applications e.g. HRA Approval, may mean that you will be unable to access NIHR CRN Support for your study.

6. Do you plan to include any participants who are children?

☐ Yes ☒ No

7. Do you plan at any stage of the project to undertake intrusive research involving adults lacking capacity to consent for themselves?

☐ Yes ☒ No

Answer Yes if you plan to recruit living participants aged 16 or over who lack capacity, or to retain them in the study following loss of capacity. Intrusive research means any research with the living requiring consent in law. This includes use of identifiable tissue samples or personal information, except where application is being made to the Confidentiality Advisory Group to set aside the common law duty of confidentiality in England and Wales. Please consult the guidance notes for further information on the legal frameworks for research involving adults lacking capacity in the UK.

8. Do you plan to include any participants who are prisoners or young offenders in the custody of HM Prison Service or who are offenders supervised by the probation service in England or Wales?

☐ Yes ☒ No

9. Is the study or any part of it being undertaken as an educational project?

☒ Yes ☐ No

Please describe briefly the involvement of the student(s):

The student will be undertaking the research as part of a required piece of assessed work in order to obtain a doctorate in clinical psychology. The student will be the primary researcher supervised by a member of the university and an NHS psychologist.

9a. Is the project being undertaken in part fulfilment of a PhD or other doctorate?

☒ Yes ☐ No

10. Will this research be financially supported by the United States Department of Health and Human Services or any of its divisions, agencies or programs?

☐ Yes ☒ No

11. Will identifiable patient data be accessed outside the care team without prior consent at any stage of the project (including identification of potential participants)?

☐ Yes ☒ No

Integrated Research Application System

Application Form for Research involving qualitative methods only

The Chief Investigator should complete this form. Guidance on the questions is available wherever you see this symbol displayed. We recommend reading the guidance first. The complete guidance and a glossary are available by selecting [Help](#).

Please define any terms or acronyms that might not be familiar to lay reviewers of the application.

Short title and version number: (maximum 70 characters - this will be inserted as header on all forms)
Living in the premanifest stage of Huntington's disease.

PART A: Core study information

1. ADMINISTRATIVE DETAILS

A1. Full title of the research:

Exploring individuals' experiences of living in the premanifest stage of Huntington's disease.

A2-1. Educational projects

Name and contact details of student(s):

Student 1

	Title	Forename/Initials	Surname
	Miss	Gina	Wieringa
Address	Lancaster University		
	Lancaster		
Post Code	LA14YT		
E-mail	g.wieringa@lancaster.ac.uk		
Telephone	07985185645		
Fax			

Give details of the educational course or degree for which this research is being undertaken:

Name and level of course/ degree:
Doctorate in clinical psychology

Name of educational establishment:
Lancaster University

Name and contact details of academic supervisor(s):

Academic supervisor 1

	Title	Forename/Initials	Surname
	Dr	Fiona	Eccles
Address	Lancaster University		

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	Lancaster
Post Code	LA14YT
E-mail	f.eccles@lancaster.ac.uk
Telephone	44 (0)1524 592807
Fax	

Please state which academic supervisor(s) has responsibility for which student(s):

Please click "Save now" before completing this table. This will ensure that all of the student and academic supervisor details are shown correctly.

Student(s)	Academic supervisor(s)
Student 1 Miss Gina Wieringa	<input checked="" type="checkbox"/> Dr Fiona Eccles

A copy of a current CV for the student and the academic supervisor (maximum 2 pages of A4) must be submitted with the application.

A2-2. Who will act as Chief Investigator for this study?

- ☒ Student
☐ Academic supervisor
☐ Other

A3-1. Chief Investigator:

	Title	Forename/Initials	Surname
	Miss	Gina	Wieringa
Post	Trainee Clinical Psychologist		
Qualifications	BSc Psychology		
ORCID ID			
Employer	Lancaster University		
Work Address	Doctorate in Clinical Psychology		
	Lancaster University		
	Lancaster		
Post Code	LA1 4YT		
Work E-mail	g.wieringa@lancaster.ac.uk		
* Personal E-mail	g.wieringa@lancaster.ac.uk		
Work Telephone			
* Personal Telephone/Mobile	07985185645		
Fax			

** This information is optional. It will not be placed in the public domain or disclosed to any other third party without prior consent.*

A copy of a current CV (maximum 2 pages of A4) for the Chief Investigator must be submitted with the application.

A4. Who is the contact on behalf of the sponsor for all correspondence relating to applications for this project? This contact will receive copies of all correspondence from REC and HRA/R&D reviewers that is sent to the CI.

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	Title Forename/Initials Surname
	Ms Becky Gordon
Address	Head of Research Quality and Policy
	Research Services
	Lancaster University
Post Code	LA1 4YT
E-mail	sponsorship@lancaster.ac.uk
Telephone	(0)1524 592981
Fax	

A5-1. Research reference numbers. *Please give any relevant references for your study:*

Applicant's/organisation's own reference number, e.g. R & D (if available): N/A

Sponsor's/protocol number: N/A

Protocol Version: 0.1

Protocol Date: 25/03/2019

Funder's reference number (enter the reference number or state not applicable): N/A

Project website: N/A

Additional reference number(s):

Ref.Number	Description	Reference Number
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Registration of research studies is encouraged wherever possible. You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you have registered your study please give details in the "Additional reference number(s)" section.

A5-2. Is this application linked to a previous study or another current application?

☐ Yes ☒ No

Please give brief details and reference numbers.

2. OVERVIEW OF THE RESEARCH

To provide all the information required by review bodies and research information systems, we ask a number of specific questions. This section invites you to give an overview using language comprehensible to lay reviewers and members of the public. Please read the guidance notes for advice on this section.

A6-1. Summary of the study. *Please provide a brief summary of the research (maximum 300 words) using language easily understood by lay reviewers and members of the public. Where the research is reviewed by a REC within the UK Health Departments' Research Ethics Service, this summary will be published on the Health Research Authority (HRA) website following the ethical review. Please refer to the question specific guidance for this question.*

Huntington's Disease (HD) is an inherited neurodegenerative disorder which causes significant impairment in movement and thinking. Depression and other mood changes have been reported in individuals who have tested positive to the genetic test for HD but prior to showing motor symptoms (Martinez-Horta et al., 2014), which are necessary to receive a formal diagnosis. This suggests that living at risk of HD is itself very challenging emotionally and this study aims to explore the experience of these individuals. Interviews will be used to explore the process by which individuals adjust to the positive test result and how the individual's identity changes to include being a gene-positive carrier. The study will explore both positive and negative coping mechanisms which individuals describe in order to get a better understanding of how they come to terms with finding out they will develop HD. The study will also

discuss the important clinical implications which arise from getting a better understanding of the process of adjustment; this includes a) long-term support for individuals who test positive to carrying the HD gene mutation but have not yet received a diagnosis and b) sharing important findings about coping strategies which may help individuals to show more positive adjustment responses.

A6-2. Summary of main issues. Please summarise the main ethical, legal, or management issues arising from your study and say how you have addressed them.

Not all studies raise significant issues. Some studies may have straightforward ethical or other issues that can be identified and managed routinely. Others may present significant issues requiring further consideration by a REC, R&D office or other review body (as appropriate to the issue). Studies that present a minimal risk to participants may raise complex organisational or legal issues. You should try to consider all the types of issues that the different reviewers may need to consider.

Ethical issues

1. Confidentiality. The research team will not have access to participants' personal details without their consent. The nature of how we obtain personal details of the participant will depend upon the recruitment method:

If recruitment is via one of the NHS Trusts, participants will be initially approached by a member of the clinical team involved in their care and will have to explicitly provide their details to members of the research team, or give their verbal consent to the clinical team for their details to be passed to myself (Gina).

If enough participants cannot be recruited via the NHS Trusts, then participants will be recruited via the HDA website. Participants will have to approach myself in order to provide their details and give their verbal consent for their details to be used by the research team

2. Risk of harm to participants. The interview is not expected to cause participants significant distress. However, participants who participate will be fully briefed beforehand about any possible adverse effects of the interview. Should any of the participants disclose they are at risk of harming themselves or others then a member of the participant's clinical team will be informed. For participants recruited via the HD, the appropriate authority will be informed. Where possible the participant will be informed if this is necessary.

3. Risk of harm to researchers. In the event that researchers become distressed by the emotive subject of the research, there are support networks surrounding the research team which can be utilised for confidential emotional support. The students will be able to receive support from their supervisors. If conducting any home visits to conduct the interview, the researchers will follow Lancaster University's guidance regarding lone working.

4. Data storage. All electronic data will be encrypted and stored on the secure University Network for the duration of the project. Such storage location may include secure cloud storage (e.g. Box) which is accessible via the university and is deemed to meet the university's requirements for secure storage. Again, this data will be encrypted via password protection. Paper data (e.g. consent forms) will be scanned in and destroyed.

3. PURPOSE AND DESIGN OF THE RESEARCH

A7. Select the appropriate methodology description for this research. Please tick all that apply:

- ☐ Case series/ case note review
- ☐ Case control
- ☐ Cohort observation
- ☐ Controlled trial without randomisation
- ☐ Cross-sectional study
- ☐ Database analysis
- ☐ Epidemiology
- ☐ Feasibility/ pilot study
- ☐ Laboratory study
- ☐ Metanalysis

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- ☒ Qualitative research
- ☒ Questionnaire, interview or observation study
- ☐ Randomised controlled trial
- ☐ Other (please specify)

A10. What is the principal research question/objective? *Please put this in language comprehensible to a lay person.*

How do individuals with pre-manifest Huntington's disease adjust to living with knowing they will develop the disease?

A11. What are the secondary research questions/objectives if applicable? *Please put this in language comprehensible to a lay person.*

How do individuals living with pre-manifest Huntington's disease cope?

A12. What is the scientific justification for the research? *Please put this in language comprehensible to a lay person.*

Huntington's disease (HD) is an inherited disorder which often starts with affective disturbances such as depression and culminates in significant cognitive deficits and motor abnormalities (Du, Pang, yc & Hannan, 2013). Predictive testing has allowed individuals who are at risk of HD to find out whether they will develop the disease, prior to showing any motor symptoms. Although this has allowed researchers to study the natural course of the disease and provide opportunities for early intervention, the test raises significant ethical issues regarding the potentially devastating consequences which living with the knowledge of the test result entails (Lilani, 2005). By offering the predictive test, the NHS has an ethical and moral responsibility to understand the impact of genetic testing and to support individuals with the long-term consequences (Andersson, Petersén, Graff & Edberg, 2016).

Although much research has been done identifying psychological consequences of a positive test result, the focus is generally on clinical symptoms such as anxiety and depression, the implications for a person's quality of life or on adverse events such as family breakdown or suicide (Codori & Brandt, 1994). There has been little research into how an individual who has tested positive to the HD gene but is not yet symptomatic adjusts to living in the knowledge they will develop HD.

Additionally, the nature of adjustment to living in the pre-manifest stage of HD needs clarification because this process is likely to be idiosyncratic, depending on a multitude of different factors. The perceived prognosis of the condition (likely shaped by an individual's experiences), the rapidity of any health decline and the duration of the asymptomatic period will all likely influence the adaptive nature of one's adjustment to the illness (Stanton, Revenson & Tennen, 2007). Within the literature on chronic illness, there is a lack of consistency as to how adjustment is defined and no definitive way of measuring it as a construct (Moss-Morris, 2013). Yet the diagnosis of a chronic condition presents an individual with a significant challenge to their usual ways of coping (Dekker & deGroot, 2018) and thus psychosocial adjustment is key in order to maintain their quality of life. Although not clinically diagnosed until the individual shows unequivocal signs of involuntary movement (Sturrock & Leavitt, 2010), living in the pre-manifest stage of HD is likely to raise similar challenges to that presented by diagnosis of a chronic condition, especially given that many non-motor symptoms develop years before the onset of motor symptoms. Understanding the nature of this adjustment is key to supporting individuals to maintain a good quality of life (Hammond & Winthrop, 2018).

Furthermore, there has been little research to how adjustment processes change according to unknown and unpredictable elements of chronic conditions, such as when symptom onset is uncertain, as is the case for individuals living with pre manifest HD. Yet this uncertainty is likely to be important to consider in understanding adjustment, given that uncertainty in illness has been associated with increased psychological distress, decreased quality of life, and difficulty coping (Johnson Wright, Afari, & Zautra, 2009). It is thus important to explore how these feelings of uncertainty fluctuate over the time since receiving the genetic screening results, in order to better understand the relationship between adjustment and uncertainty in chronic conditions where disease progression may not be known or already determined.

Regarding HD, research has already indicated that individuals who have tested positive for the HD gene expansion are significantly more likely to experience depression and irritability than those without the gene expansion, even prior to experiencing disease related symptoms (Julien et al., 2007). Given that the period of adjusting to living with the gene-expansion is likely to raise significant challenges for individuals in coming to terms with their gene-positive status, a better understanding of the psychological difficulties they face is necessitated. It is hoped that this

understanding will contribute to knowledge of the needs of this group and could help inform the types of support that are needed.

A13. Please summarise your design and methodology. *It should be clear exactly what will happen to the research participant, how many times and in what order. Please complete this section in language comprehensible to the lay person. Do not simply reproduce or refer to the protocol. Further guidance is available in the guidance notes.*

Design

This study will collect qualitative data using Interpretative phenomenological analysis (IPA)

Data collection

The primary aim of the study is to collect data on the experiences of individuals living in the pre-manifest stage of Huntington's disease. Semi-structured interviews will therefore be conducted to gain participants' experiences and their thoughts and opinions on these experiences. This data will be analysed using IPA which has its focus on how individuals make sense of their experiences and thus appears best suited to this line of research.

Recruitment

Participants will be recruited via three routes:

In the first instance, participants will be recruited through a multidisciplinary team at Leicestershire Partnership NHS Trust which works with families affected by HD and includes a clinical psychologist (the field supervisor). Participants will be identified via research nurses working at the Leicestershire Trust who will identify people who meet the inclusion criteria and will invite them to participate (either in person or via post).

In the second instance, participants will be recruited from Northamptonshire (an area which receives consultancy from the Leicestershire Service but where patients don't receive ongoing HD psychological support via the same method). Participants will be identified via the advisory service for Northamptonshire which is commissioned by Leicestershire Trust

If enough participants cannot be recruited via the first two methods, participants will also be recruited via the HDA website. In this instance, participants will be identified responding directly to myself (Gina) via an advert published on the HDA website.

Once I have a potential participant's contact details via one of the methods outlined above, I will then contact the potential participant to explain more about the study and to answer any questions. If the participant wishes to proceed with study participation, then a convenient time will be arranged to conduct the interview. This will be via phone or Skype (or another similar system) unless there are extenuating circumstances making this difficult, in which case I will arrange to conduct the interview in person.

Participants

All participants will be between 18 and 65 and will have to have tested positive for the HD gene expansion at least one year prior to the interview. This decision was taken because it was hoped that they will have had some time to live with the sure knowledge of their genetic status. Participants will have to be well enough to tolerate a one hour interview which may cause some distress.

Participants will be asked to talk about their experiences of adjusting to living in the pre-manifest stage of Huntington's Disease. Questions will be asked based on their responses. Participants will not be required to talk about anything they don't wish. An individual can choose to stop the interview at any time or move on from a question at any point.

Service user involvement

Experts by experience have been involved in the study design; members of HD voice (a patient and public involvement group which sits within the Huntington's Disease Association (HDA)) were asked to comment on the proposed interview schedule with the aim that the experiences of people living in the pre-manifest stage will be accurately captured from the interview. The letter of invitation, the consent form, the flyer for the HDA and the information sheet were also reviewed to ensure it was understandable and inviting. On the back of this feedback, I simplified the questions in the interview schedule as feedback indicated this might be too complex for some participants, given that very early symptoms of HD can make cognitive processing more difficult. I also made the documents a lot more personal, as I felt it was important to acknowledge how much I appreciated participants' input. Furthermore, I clarified the support available for anyone who experiences distress.

A14-1. In which aspects of the research process have you actively involved, or will you involve, patients, service users, and/or their carers, or members of the public?

- ☒ Design of the research
- ☐ Management of the research
- ☐ Undertaking the research
- ☐ Analysis of results
- ☐ Dissemination of findings
- ☐ None of the above

Give details of involvement, or if none please justify the absence of involvement.

People with the HD gene expansion were approached for their input in the study conduct through HD voice (a patient and public involvement group which sits within the HDA).

The research findings will be disseminated to participants directly and published on the HDA website with permission. It will be presented at suitable conferences (e.g. European Huntington's Disease Network Plenary Meeting) and Special Interest Groups. The research will also be published in peer-reviewed academic journals.

4. RISKS AND ETHICAL ISSUES

RESEARCH PARTICIPANTS

A15. What is the sample group or cohort to be studied in this research?

Select all that apply:

- ☐ Blood
- ☐ Cancer
- ☐ Cardiovascular
- ☐ Congenital Disorders
- ☒ Dementias and Neurodegenerative Diseases
- ☐ Diabetes
- ☐ Ear
- ☐ Eye
- ☐ Generic Health Relevance
- ☐ Infection
- ☐ Inflammatory and Immune System
- ☐ Injuries and Accidents
- ☐ Mental Health
- ☐ Metabolic and Endocrine
- ☐ Musculoskeletal
- ☐ Neurological
- ☐ Oral and Gastrointestinal
- ☐ Paediatrics
- ☐ Renal and Urogenital
- ☐ Reproductive Health and Childbirth
- ☐ Respiratory
- ☐ Skin
- ☐ Stroke

Gender: Male and female participants

Lower age limit: 18 Years

Upper age limit: 65 Years

A17-1. Please list the principal inclusion criteria (list the most important, max 5000 characters).

Participants will need to be between 18 and 65. They will need to have tested positive for the HD gene expansion at least one year prior to the interview. Participants will have to be well enough to tolerate a one hour interview which may cause some distress.

Participants will be taken on a first come first served basis. Participants will be recruited primarily through Leicestershire Partnership NHS Trust, and opened out to the HDA if we do not have enough participants recruited.

A17-2. Please list the principal exclusion criteria (list the most important, max 5000 characters).

Participants will be excluded if they have any significant physical illness and if they have already received a formal diagnosis of HD. Participants may also be excluded if sufficient responses have already been gathered but this will be made clear when the participant initially expresses an interest to participate.

RESEARCH PROCEDURES, RISKS AND BENEFITS

A18. Give details of all non-clinical intervention(s) or procedure(s) that will be received by participants as part of the research protocol. These include seeking consent, interviews, non-clinical observations and use of questionnaires.

Please complete the columns for each intervention/procedure as follows:

1. Total number of interventions/procedures to be received by each participant as part of the research protocol.
2. If this intervention/procedure would be routinely given to participants as part of their care outside the research, how many of the total would be routine?
3. Average time taken per intervention/procedure (minutes, hours or days)
4. Details of who will conduct the intervention/procedure, and where it will take place.

Intervention or procedure	1	2	3	4
Conversation with research nurse or responding to H A advert	1	0	10	research nurse or myself (Gina)
Initial conversation with potential participant	1	0	20	Gina
Consent forms completed (remotely)	1	0	15	Participant
Consent forms completed (in person)	1	0	15	Gina
Interview participant	1	0	45-60	Gina via skype or phone (or at a venue convenient to the participant if Skype/phone is not possible)

A21. How long do you expect each participant to be in the study in total?

Participants will be involved in the study for approximately 3-4 months (though the actual interview lasts around one hour)

Months 1-4: Recruitment and Interviews

A22. What are the potential risks and burdens for research participants and how will you minimise them?

For all studies, describe any potential adverse effects, pain, discomfort, distress, intrusion, inconvenience or changes

to lifestyle. Only describe risks or burdens that could occur as a result of participation in the research. Say what steps would be taken to minimise risks and burdens as far as possible.

The interview is not anticipated to cause any significant distress. However, some of the experiences of the individual may evoke some distressing reactions and/or discuss difficult content.

If the interview is being conducted in person, the interview will be stopped as soon as any distress is identified by myself (Gina). The participant will be offered a break and, if a family member has attended the interview with them, whether they wish to request their support (presuming the family member is not already in the interview room). After any required break, they will be asked if they wish to stop the interview, or to continue the questions but move on from the distressing topic. Participants will also be made aware that they can rearrange the interview for another date if they wish to do so.

If the interview is being conducted by phone or by Skype, distress may be more difficult to become immediately aware of, as subtle changes in body language are not always clear. However, the same principles will apply to interviews conducted via Skype and to those in person. However, there will need to be more confidence from myself that any distress that has been identified has been minimised appropriately as there will be no opportunity for face-to-face debriefing after the interview has ended. I will ensure before finishing the interview that I check on the wellbeing of the participant at the end.

For both in person and Skype interviews, if any of the research team becomes concerned that a participant poses a risk of serious harm (to themselves or others) they will inform a member of the person's clinical team or their GP/A&E if deemed appropriate.

Where an individual has been recruited via the HDA, any risk identified will be passed onto my supervisor and the rest of the research team who will decide if any intervention is necessary

Participants can withdraw from the study at any time without giving a reason. They can choose to stop the interviews at any time. They can choose to withdraw their data that they have already given at any time up to the point at which it has been analysed. We will make every effort to withdraw data if a participant requests this, but will let them know if this is not possible (due to the data already having been analysed)

A23. Will interviews/ questionnaires or group discussions include topics that might be sensitive, embarrassing or upsetting, or is it possible that criminal or other disclosures requiring action could occur during the study?

☒ Yes ☐ No

If Yes, please give details of procedures in place to deal with these issues:

I (Gina) will be conducting the study (ie performing the interviews) and am a trainee clinical psychologists who is able to assess for signs of distress and act accordingly. If an individual appears distressed at any point in the interview, I will stop the interview, check on the wellbeing of the participant and offer a break. The interview will only be resumed if the participant is willing and feels able to do so.

A24. What is the potential for benefit to research participants?

None

A26. What are the potential risks for the researchers themselves? (if any)

When conducting interviews and particularly when lone working the researchers will adhere to Lancaster University's guidance on fieldwork. I (Gina) will leave the details of the interview (e.g., participant, date, time, location) in a sealed envelope/password protected document to a peer, which they could open if they couldn't reach me. I will telephone or text this trainee when the interview is concluded. If this telephone call/text does not take place, attempts will be made to contact myself. If contact cannot be made, the appropriate authorities will be informed. It is also possible that I will use an electronic lone working system connected to an electronic database.

If the student experience distress during the study they will be able to speak to their supervisors and they will receive regular supervision throughout the study.

RECRUITMENT AND INFORMED CONSENT

In this section we ask you to describe the recruitment procedures for the study. Please give separate details for different study groups where appropriate.

A27-1. How will potential participants, records or samples be identified? Who will carry this out and what resources will be used? *For example, identification may involve a disease register, computerised search of social care or GP records, or review of medical records. Indicate whether this will be done by the direct care team or by researchers acting under arrangements with the responsible care organisation(s).*

Participants will be recruited via three routes:

In the first instance, participants will be recruited through a multidisciplinary team at Leicestershire Partnership NHS Trust which works with families affected by HD and includes a clinical psychologist (the field supervisor). Participants will be identified via research nurses working at the Leicestershire Trust who will identify people who meet the inclusion criteria and will provide them with a participant information sheet for them to gain further information about the study (either provided in person or via post). If they are interested, the research nurses will invite them to participate.

In the second instance, participants will be recruited from Northamptonshire (an area which receives consultancy from the Leicestershire Service but where patients don't receive ongoing HD psychological support via the same method. Participants will be identified via the advisory service for Northamptonshire which is commissioned by Leicestershire Trust.

If enough participants cannot be recruited via the first two methods, participants will also be recruited via the HDA website. In this instance, participants will be identified responding directly to myself (Gina) via an advert published on the HDA website.

A27-2. Will the identification of potential participants involve reviewing or screening the identifiable personal information of patients, service users or any other person?

☒ Yes ☐ No

Please give details below:

Where recruitment is via research nurses working for the NHS, the clinical care team will use patients' clinical data to decide if they meet the inclusion criteria for the research. The research team will not have access to identifiable information without the permission of the potential participant themselves

Where recruitment is via the HDA, no screening of identifiable patient information will be used.

A27-3. Describe what measures will be taken to ensure there is no breach of any duty of confidentiality owed to patients, service users or any other person in the process of identifying potential participants. *Indicate what steps have been or will be taken to inform patients and service users of the potential use of their records for this purpose. Describe the arrangements to ensure that the wishes of patients and service users regarding access to their records are respected. Please consult the guidance notes on this topic.*

None anticipated (as only the clinical team has access to patient's record)

A27-4. Will researchers or individuals other than the direct care team have access to identifiable personal information of any potential participants?

☒ Yes ☐ No

A27-5. Has prior consent been obtained or will it be obtained for access to identifiable personal information?

☒ Yes ☐ No

If Yes, please give details below.

Consent will be sought by the clinical care team where recruitment is via Leicestershire NHS Trust in order to for me to identify potential participants (only basic contact information such as their name and telephone number/email address will be obtained). This consent will be verbal consent.

Where recruitment is via the HDA, potential participants will make contact with myself first, and provide their contact details to myself first.

A28. Will any participants be recruited by publicity through posters, leaflets, adverts or websites?

☒ Yes ☐ No

If Yes, please give details of how and where publicity will be conducted, and enclose copy of all advertising material (with version numbers and dates).

If enough participants cannot be recruited via NHS trusts, participants will be recruited via an advert posted on the HDA website.

A29. How and by whom will potential participants first be approached?

Participants recruited through both Leicestershire and Northamptonshire NHS Trusts will first be approached by research nurses working for the Leicestershire Trust or via an invitation letter sent in the post.

Participants recruited through the HDA will first be approached by advertising the study on the HDA website.

If participants at the NHS trust meet inclusion criteria they will be given a pack of information or letter of invitation and asked to contact Gina directly. They can also indicate to their clinical that they wish for further details.

For participants responding to the HDA advert, they will be able to contact Gina directly.

A30-1. Will you obtain informed consent from or on behalf of research participants?

☒ Yes ☐ No

If you will be obtaining consent from adult participants, please give details of who will take consent and how it will be done, with details of any steps to provide information (a written information sheet, videos, or interactive material). Arrangements for adults unable to consent for themselves should be described separately in Part B Section 6, and for children in Part B Section 7.

If you plan to seek informed consent from vulnerable groups, say how you will ensure that consent is voluntary and fully informed.

Participants will have had access to a written information sheet before deciding to opt in.

Where consent is taken in person e.g. with the NHS recruitment route Gina will meet with the participant to take informed written consent (on a pre-prepared consent form) or the consent form will be sent in advance and posted back to me (Gina).

Where participants are recruited via the H A (i.e. remote consent), consent forms will be sent to the person's address to be returned to myself.

If you are not obtaining consent, please explain why not.

Please enclose a copy of the information sheet(s) and consent form(s).

A30-2. Will you record informed consent (or advice from consultees) in writing?

☒ Yes ☐ No

A31. How long will you allow potential participants to decide whether or not to take part?

Participants will have several days from receiving the information sheet to decide whether to take part in the interview. Participants can stop the study at any point (by just choosing not to attend/participate in the interview).

A33-1. What arrangements have been made for persons who might not adequately understand verbal explanations or written information given in English, or who have special communication needs? (e.g. translation, use of interpreters)

Participants will need to understand verbal explanations in English in order to participate in the intervention so they will be excluded if this is not possible. If someone was not able to understand written information (or not able, for instance, to see written information) then the participant information sheet would be read to them (or provided in large font, if this was helpful). Consent would be taken verbally and this would be audio recorded.

A35. What steps would you take if a participant, who has given informed consent, loses capacity to consent during the study? Tick one option only.

- ☐ The participant and all identifiable data or tissue collected would be withdrawn from the study. Data or tissue which is not identifiable to the research team may be retained.
- ☒ The participant would be withdrawn from the study. Identifiable data or tissue already collected with consent would be retained and used in the study. No further data or tissue would be collected or any other research procedures carried out on or in relation to the participant.
- ☐ The participant would continue to be included in the study.
- ☐ Not applicable – informed consent will not be sought from any participants in this research.
- ☐ Not applicable – it is not practicable for the research team to monitor capacity and continued capacity will be assumed.

Further details:

If you plan to retain and make further use of identifiable data/tissue following loss of capacity, you should inform participants about this when seeking their consent initially.

CONFIDENTIALITY

In this section, personal data means any data relating to a participant who could potentially be identified. It includes pseudonymised data capable of being linked to a participant through a unique code number.

Storage and use of personal data during the study

A36. Will you be undertaking any of the following activities at any stage (including in the identification of potential participants)? (Tick as appropriate)

- ☐ Access to medical records by those outside the direct healthcare team
- ☐ Access to social care records by those outside the direct social care team
- ☒ Electronic transfer by magnetic or optical media, email or computer networks
- ☐ Sharing of personal data with other organisations
- ☐ Export of personal data outside the EEA
- ☒ Use of personal addresses, postcodes, faxes, emails or telephone numbers
- ☒ Publication of direct quotations from respondents
- ☐ Publication of data that might allow identification of individuals
- ☒ Use of audio/visual recording devices
- ☒ Storage of personal data on any of the following:
- ☒ Manual files (includes paper or film)
 - ☒ NHS computers
 - ☐ Social Care Service computers
 - ☐ Home or other personal computers

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- ☒ University computers
- ☐ Private company computers
- ☐ Laptop computers

Further details:

A37. Please describe the physical security arrangements for storage of personal data during the study?

Personal data on paper will be made electronic and destroyed immediately afterwards.
Electronic personal data will be kept securely on the university secure network or on a secure cloud (e.g. Box) accessible through the university and which has similar security credentials to the university network

The data recorders that the university uses do not encrypt so all data will be kept securely until it is uploaded onto the university system

A38. How will you ensure the confidentiality of personal data? Please provide a general statement of the policy and procedures for ensuring confidentiality, e.g. anonymisation or pseudonymisation of data.

All personal data will be kept confidential. Qualitative data will be anonymised as far as possible for publication. Names and identifiable information will not be used in any published quotes. Basic demographic information may be included alongside their quote but every effort will be made to anonymise participant identifiable data.

A40. Who will have access to participants' personal data during the study? Where access is by individuals outside the direct care team, please justify and say whether consent will be sought.

All members of the research team will have access to participants' contact details in order to communicate with them during the study. This will only take place when a participant has consented to be part of the study.

Storage and use of data after the end of the study
A41. Where will the data generated by the study be analysed and by whom?

Data will be analysed by myself (Gina) at Lancaster University under supervision from Fiona Eccles and Maria Dale and will be shared with the research team members who work at Leicestershire Partnership NHS Foundation Trust

A42. Who will have control of and act as the custodian for the data generated by the study?

	Title Forename/Initials Surname
	Dr Fiona Eccles
Post	Lecturer
Qualifications	MPhys, DPhil, Grad Dip Psych, DClinPsy, Certificate in Academic Practice
Work Address	Doctorate in Clinical Psychology Lancaster University Lancaster
Post Code	LA1 4YT
Work Email	f.eccles@lancaster.ac.uk
Work Telephone	01524592807
Fax	

A43. How long will personal data be stored or accessed after the study has ended?

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- ☐ Less than 3 months
- ☐ 3 – 6 months
- ☐ 6 – 12 months
- ☐ 12 months – 3 years
- ☒ Over 3 years

If longer than 12 months, please justify:

Consent forms and transcripts will be stored for 10 years or 10 years from publication, whichever is longer. These will be anonymised and kept separately. Audio recordings will be kept until thesis examination at the viva and then destroyed.

All other personal data (e.g. to send participants a consent form/copy of the results) will be destroyed within 6 months of the study completion.

A44. For how long will you store research data generated by the study?

Years: 10

Months: 0

A45. Please give details of the long term arrangements for storage of research data after the study has ended. Say where data will be stored, who will have access and the arrangements to ensure security.

Paper data (e.g. consent forms) will be scanned in electronically and deleted immediately after doing so. The electronic consent forms and transcripts will be stored securely on the Lancaster University network (or Box) by the research coordinator of the doctorate in clinical psychology programme under the direction of the academic supervisor (Fiona Eccles) for 10 years and then deleted.

INCENTIVES AND PAYMENTS**A46. Will research participants receive any payments, reimbursement of expenses or any other benefits or incentives for taking part in this research?**

- ☐ Yes ☒ No

A47. Will individual researchers receive any personal payment over and above normal salary, or any other benefits or incentives, for taking part in this research?

- ☐ Yes ☒ No

A48. Does the Chief Investigator or any other investigator/collaborator have any direct personal involvement (e.g. financial, share holding, personal relationship etc.) in the organisations sponsoring or funding the research that may give rise to a possible conflict of interest?

- ☐ Yes ☒ No

NOTIFICATION OF OTHER PROFESSIONALS**A49-1. Will you inform the participants' General Practitioners (and/or any other health or care professional responsible for their care) that they are taking part in the study?**

- ☐ Yes ☒ No

If Yes, please enclose a copy of the information sheet/letter for the GP/health professional with a version number and date.

PUBLICATION AND DISSEMINATION

A50-1. Will the research be registered on a public database?

☐ Yes ☒ No

Please give details, or justify if not registering the research.
I am not aware a suitable database exists.

Registration of research studies is encouraged wherever possible.

You may be able to register your study through your NHS organisation or a register run by a medical research charity, or publish your protocol through an open access publisher. If you are aware of a suitable register or other method of publication, please give details. If not, you may indicate that no suitable register exists. Please ensure that you have entered registry reference number(s) in question A5-1.

A51. How do you intend to report and disseminate the results of the study?

ick as appropriate:

- ☒ Peer reviewed scientific journals
- ☒ Internal report
- ☒ Conference presentation
- ☒ Publication on website
- ☐ Other publication
- ☐ Submission to regulatory authorities
- ☐ Access to raw data and right to publish freely by all investigators in study or by Independent Steering Committee on behalf of all investigators
- ☐ No plans to report or disseminate the results
- ☐ Other (please specify)

A52. If you will be using identifiable personal data, how will you ensure that anonymity will be maintained when publishing the results?

Care will be taken when publishing any demographic data to try to ensure anonymity is maintained

A53. Will you inform participants of the results?

☒ Yes ☐ No

Please give details of how you will inform participants or justify if not doing so.

The research findings will be disseminated to participants directly should they express an interest in being contacted after the study with the results. The research findings will also be published on the HDA website with permission of the HDA administrators.

5. Scientific and Statistical Review

A54-1. How has the scientific quality of the research been assessed? *Tick as appropriate:*

- ☐ Independent external review
- ☐ Review within a company
- ☐ Review within a multi-centre research group

- ☒ Review within the Chief Investigator's institution or host organisation
- ☒ Review within the research team
- ☒ Review by educational supervisor
- ☐ Other

Justify and describe the review process and outcome. If the review has been undertaken but not seen by the researcher, give details of the body which has undertaken the review:

For all studies except non-doctoral student research, please enclose a copy of any available scientific critique reports, together with any related correspondence.

For non-doctoral student research, please enclose a copy of the assessment from your educational supervisor/ institution.

A59. What is the sample size for the research? *How many participants/samples/data records do you plan to study in total? If there is more than one group, please give further details below.*

Total UK sample size: 12

Total international sample size (including UK): 12

Total in European Economic Area: 12

Further details:

We aim to recruit between 8-12 participants

A60. How was the sample size decided upon? *If a formal sample size calculation was used, indicate how this was done, giving sufficient information to justify and reproduce the calculation.*

The study will aim to have around eight to twelve participants, in keeping with Smith et al. (2009), who suggest four to ten participants for professional doctorates using IPA. This number permits an in depth focus of each person's experience (in line with IPA's idiographic nature)

A62. Please describe the methods of analysis (statistical or other appropriate methods, e.g. for qualitative research) by which the data will be evaluated to meet the study objectives.

Analysis will be conducted using guidelines from IPA theory (Smith et al., 2009). Interviews will be transcribed and then analysed line by line, making note of all emerging themes and any apparent contradictions. Emergent themes will be collated and put into overarching categories. Continual re-reading will be used to ensure individual accounts are reflected in the themes and that the main themes are consistent with most of the data. Bias will be reflected upon and incorporated into the study through the use of a reflective journal and discussed during supervision.

6. MANAGEMENT OF THE RESEARCH

A63. Other key investigators/collaborators. *Please include all grant co-applicants, protocol co-authors and other key members of the Chief Investigator's team, including non-doctoral student researchers.*

	Title	Forename/Initials	Surname
	Dr	Maria	Dale
Post			
Qualifications	DClinPsy		
Employer	Leicestershire Partnership NHS Trust		
Work Address	Mill Lodge Narborough	Leicestershire	
Post Code	LE19 4SL		

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Telephone	(0) 116 295 3649
Fax	
Mobile	(0) 116 295 3649
Work Email	Maria.Dale@leicspart.nhs.uk

A64. Details of research sponsor(s)**A64-1. Sponsor****Lead Sponsor**Status: ☐ NHS or HSC care organisation

Commercial status:

☒ Academic☐ Pharmaceutical industry☐ Medical device industry☐ Local Authority☐ Other social care provider (including voluntary sector or private organisation)☐ Other*If Other, please specify:***Contact person**

Name of organisation Lancaster University

Given name Becky

Family name Gordon

Address Research Services

Town/city Lancaster University

Post code LA1 4YT

Country UNITED KING OM

Telephone 01524 592981

Fax

E-mail sponsorship@lancaster.ac.uk

A65. Has external funding for the research been secured?*Please tick at least one check box.*☐ Funding secured from one or more funders☐ External funding application to one or more funders in progress☒ No application for external funding will be made

What type of research project is this?

☒ Standalone project

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- ☐ Project that is part of a programme grant
☐ Project that is part of a Centre grant
☐ Project that is part of a fellowship/ personal award/ research training award
☐ Other

Other – please state:

A66. Has responsibility for any specific research activities or procedures been delegated to a subcontractor (other than a co-sponsor listed in A64-1) ? Please give details of subcontractors if applicable.

- ☐ Yes ☒ No

A67. Has this or a similar application been previously rejected by a Research Ethics Committee in the UK or another country?

- ☐ Yes ☒ No

Please provide a copy of the unfavourable opinion letter(s). You should explain in your answer to question A6-2 how the reasons for the unfavourable opinion have been addressed in this application.

A68-1. Give details of the lead NHS R&D contact for this research:

	Title Forename/Initials Surname
	Dr David Clarke
Organisation	
Address	Leicestershire Partnership NHS Trust Research & Development Unit Swithland House 352 Londonoad Leicester
Post Code	LE2 2PL
Work Email	david.clarke@leicspart.nhs.uk
Telephone	0116 295 7641
Fax	
Mobile	0116 295 7641

Details can be obtained from the NHS R&D Forum website: <http://www.rdforum.nhs.uk>

A69-1. How long do you expect the study to last in the UK?

Planned start date: 01/09/2019

Planned end date: 01/12/2020

Total duration:

Years: 1 Months: 3 Days: 1

A71-1. Is this study?

- ☒ Single centre
☐ Multicentre

A71-2. Where will the research take place? (Tick as appropriate)

- ☒ England
☐ Scotland
☐ Wales
☐ Northern Ireland
☐ Other countries in European Economic Area

Total UK sites in study 2

Does this trial involve countries outside the EU?

- ☐ Yes ☒ No

A72. Which organisations in the UK will host the research? Please indicate the type of organisation by ticking the box and give approximate numbers if known:

- | | |
|---|---|
| <input checked="" type="checkbox"/> NHS organisations in England | 1 |
| <input type="checkbox"/> NHS organisations in Wales | |
| <input type="checkbox"/> NHS organisations in Scotland | |
| <input type="checkbox"/> HSC organisations in Northern Ireland | |
| <input type="checkbox"/> GP practices in England | |
| <input type="checkbox"/> GP practices in Wales | |
| <input type="checkbox"/> GP practices in Scotland | |
| <input type="checkbox"/> GP practices in Northern Ireland | |
| <input type="checkbox"/> Joint health and social care agencies (eg community mental health teams) | |
| <input type="checkbox"/> Local authorities | |
| <input type="checkbox"/> Phase 1 trial units | |
| <input type="checkbox"/> Prison establishments | |
| <input type="checkbox"/> Probation areas | |
| <input type="checkbox"/> Independent (private or voluntary sector) organisations | |
| <input checked="" type="checkbox"/> Educational establishments | 1 |
| <input type="checkbox"/> Independent research units | |
| <input type="checkbox"/> Other (give details) | |

Total UK sites in study: 2

A73-1. Will potential participants be identified through any organisations other than the research sites listed above?

- ☒ Yes ☐ No

A73-2. If yes, will any of these organisations be NHS organisations?

- ☐ Yes ☒ No

If yes, details should be given in Part C.

A74. What arrangements are in place for monitoring and auditing the conduct of the research?

The research team will be in regular contact during the study to ensure it is progressing as planned. The trainee clinical psychologist (Gina) involved in the study will receive regular supervision from her supervisors.

A76. Insurance/ indemnity to meet potential legal liabilities

Note: in this question to NHS indemnity schemes include equivalent schemes provided by Health and Social Care (HSC) in Northern Ireland

A76-1. What arrangements will be made for insurance and/or indemnity to meet the potential legal liability of the sponsor(s) for harm to participants arising from the management of the research? Please tick box(es) as applicable.

Note: Where a NHS organisation has agreed to act as sponsor or co-sponsor, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For all other sponsors, please describe the arrangements and provide evidence.

- ☒ NHS indemnity scheme will apply (NHS sponsors only)
- ☒ Other insurance or indemnity arrangements will apply (give details below)

Lancaster University legal liability cover will apply

Please enclose a copy of relevant documents.

A76-2. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of the sponsor(s) or employer(s) for harm to participants arising from the design of the research? Please tick box(es) as applicable.

Note: Where researchers with substantive NHS employment contracts have designed the research, indemnity is provided through NHS schemes. Indicate if this applies (there is no need to provide documentary evidence). For other protocol authors (e.g. company employees, university members), please describe the arrangements and provide evidence.

- ☐ NHS indemnity scheme will apply (protocol authors with NHS contracts only)
- ☒ Other insurance or indemnity arrangements will apply (give details below)

Lancaster University legal liability cover will apply

Please enclose a copy of relevant documents.

A76-3. What arrangements will be made for insurance and/ or indemnity to meet the potential legal liability of investigators/collaborators arising from harm to participants in the conduct of the research?

Note: Where the participants are NHS patients, indemnity is provided through the NHS schemes or through professional indemnity. Indicate if this applies to the whole study (there is no need to provide documentary evidence). Where non-NHS sites are to be included in the research, including private practices, please describe the arrangements which will be made at these sites and provide evidence.

- ☒ NHS indemnity scheme or professional indemnity will apply (participants recruited at NHS sites only)
- ☒ Research includes non-NHS sites (give details of insurance/ indemnity arrangements for these sites below)

Lancaster University legal liability cover will apply

Please enclose a copy of relevant documents.

A78. Could the research lead to the development of a new product/process or the generation of intellectual property?

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☐ Yes
 ☒ No
 ☐ Not sure

PART C: Overview of research sites

Please enter details of the host organisations (Local Authority, NHS or other) in the UK that will be responsible for the research sites. *For further information please refer to guidance.*

Investigator identifier	Research site	Investigator Name
IN1	<input checked="" type="radio"/> NHS/HSC Site <input type="radio"/> Non-NHS/HSC Site	Forename Maria Middle name Family name Dale Email Maria.Dale@leicspart.nhs.uk Qualification (MD...) DClinPsy Country UNI ED KINGDOM
	Organisation name Leicestershire Partnership NHS Trust Address Mill Lodge Narborough Leicestershire Post Code LE19 4SL Country GB	

**Project Title**

Living in the pre-manifest stage of Huntington's disease

IRAS ID: 264354 Version 0.1

Created : 25/03/19

Applicants**Principal Investigator**

- Gina Wieringa

Trainee Clinical Psychologist, Lancaster University, Lancaster, LA1 4YT, UK T: +44 (0)7852 516 566 Email: g.wieringa@lancaster.ac.uk

Co-investigators

- Dr Maria Dale (field supervisor)

Clinical Psychologist

Huntington's disease Service, Leicestershire Partnership NHS Trust, Mill Lodge, Narborough, Leicestershire, LE19 4SL T: +44 (0) 116 295 3649 email:

Maria.Dale@leicspart.nhs.uk

- Dr Fiona Eccles (academic supervisor)

Lecturer in health research

Lancaster University, Lancaster, LA1 4YT: +44 (0)1524 592807 Email: f.eccles@lancaster.ac.uk

Introduction

Huntington's disease (HD) is an inherited disorder which often starts with affective disturbances such as depression and culminates in significant cognitive deficits and motor abnormalities (Du, Pang, Tyc & Hannan, 2013). Predictive testing has allowed individuals who are at risk of HD to find out whether they will develop the disease, prior to showing any motor symptoms. Although this has allowed researchers to study the natural course of the disease and provide opportunities for early intervention, the test raises significant ethical issues regarding the potentially devastating consequences which living with the knowledge of the test result entails (Lilani, 2005). By offering the predictive test, the NHS has an ethical and moral responsibility to understand the impact of genetic testing and to support individuals with the long-term consequences (Andersson, Petersén, Graff & Edberg, 2016).

Although much research has been done identifying psychological consequences of a positive test result, the focus is generally on clinical symptoms such as anxiety and depression, the implications for a person's quality of life or on adverse events such as family breakdown or suicide (Codori & Brandt, 1994). There has been little research into how a person who has tested positive to the HD gene but is not yet symptomatic perceives their status as 'embodied risk' (Kavanagh & Broom, 1997). This precarious status-known as "premanifest HD" is a prominent part of an individual's experience with HD and of the way in which they adjust to this new identity, yet attention has not been given to this important time period in the lives of those affected. It is clear that more research is needed.

Additionally, the nature of adaptive adjustment to living in the pre-manifest stage of HD needs clarification because this process is likely to be idiosyncratic, depending on a multitude of different factors. Within the literature on chronic illness, there is a lack of consistency as to how adjustment is defined and no definitive way of measuring it as a construct (Moss-Morris, 2013). Yet the diagnosis of a chronic condition presents an individual with a significant challenge to their usual ways of coping (Dekker & deGroot, 2018) and thus psychosocial adjustment is key in order to maintain their quality of life. Although not clinically diagnosed until

the individual shows unequivocal signs of involuntary movement (Sturrock & Leavitt, 2010), living in the pre-manifest stage of HD is likely to raise similar challenges to that presented by diagnosis of a chronic condition, especially given that many non-motor symptoms develop years before the onset of motor symptoms. Understanding the nature of this adjustment is key to supporting individuals to maintain a good quality of life (Hammond & Winthrope, 2018).

Furthermore, there has been little research into how adjustment processes change according to unknown and unpredictable elements of chronic conditions, such as when symptom onset is uncertain, as is the case for individuals living with pre manifest HD. Yet this uncertainty is likely to be important to consider in understanding adjustment, given that uncertainty in illness has been associated with increased psychological distress, decreased quality of life, and difficulty coping (Johnson Wright, Afari, & Zautra, 2009). It is thus important to explore how these feelings of uncertainty fluctuate over the time since receiving the genetic screening results, in order to better understand the relationship between adjustment and uncertainty in chronic conditions where disease progression may not be known or already determined.

Regarding HD, research has already indicated that individuals who have tested positive for the HD gene expansion are significantly more likely to experience depression and irritability than those without the gene expansion, even prior to experiencing disease related symptoms (Julien et al., 2007). Given that the period of adjusting to living with the gene-expansion is likely to raise significant challenges for individuals in coming to terms with their gene-positive status, a better understanding of the psychological difficulties they face is necessitated. It is hoped that this understanding will contribute to knowledge of the needs of this group and could help inform the types of support that are needed.

Interpretative phenomenological analysis (IPA), with its focus on how individuals make sense of their experiences, appears best suited to this line of research (Smith, Flowers & Larkin, 2009). IPA also lends itself well to the multiple

perspectives which give rise to shared experiences (Seamark, Blake, Seamark & Halpin, 2004). The current study will utilise IPA in order to explore the adjustment experiences of individuals living with HD, an approach which has been shown to be useful in exploring adjustment to other neurodegenerative conditions (Ali & Bokharey, 2015) and [Obj] for individuals with long term health conditions (e.g. Hogg, Garratt, Shaw & Tagney, 2007).

Method

Participants

To be included in the study, participants will need to be aged between 18 and 65 (i.e. be of typical working age in the UK). They will need to have tested positive for the HD gene expansion at least one year prior to the interview. This decision was taken because it was hoped that they will have had some time to live with the sure knowledge of their genetic status. Additionally, previous research has suggested that symptoms of depression and anxiety are most common in carriers in the first two months post-test, but that levels of depression return to baseline level one year after the test (Huggins, Bloch & Wiggin, 1992). It was felt that interviewing participants at least a year following the test would encourage participants to focus on their experiences of living with their genetic status rather than those of the testing period. Participants will have to be well enough to tolerate a one hour interview. Participants with additional significant physical health conditions will be excluded from the study.

The study will aim to have around 8-12 participants in keeping with Smith et al. (2009), who suggest four to ten participants for professional doctorates using IPA. This number permits an in depth focus of each person's experience (in line with IPA's idiographic nature) but also allows comparisons across participants' narratives to highlight similarities and differences.

Participants will be made aware of the commitment involved in study participation, but as participation is entirely voluntary, a participant may choose to discontinue the study at any time without giving a reason, up until the point at which data analysis has commenced. The participant is not required to give a reason for discontinuing the study, any reason given will be documented. Once recordings have been transcribed and analysis has begun withdrawing data will not be possible.

Design

The study will collect qualitative data consisting of participants' responses. The primary aim of the study is to collect data on the experience of participants in adjusting to living with pre-manifest Huntington's disease. Semi-structured interviews will therefore be conducted to explore their experiences.

Experts by experience have been involved in the study design; members of HD voice (a patient and public involvement group which sits within the Huntington's Disease Association (HDA)) were asked to comment on the proposed interview schedule with the aim that the experiences of people living in the pre-manifest stage will be accurately captured from the interview. The letter of invitation, the consent form, the flyer for the HDA and the information sheet were also reviewed to ensure it was understandable and inviting. On the back of this feedback, I simplified the questions in the interview schedule as feedback indicated this might be too complex for some participants, given that very early symptoms of HD can make cognitive processing more difficult. I also made the documents a lot more personal, as I felt it was important to acknowledge how much I appreciated participants' input. Furthermore, I clarified the support available for anyone who experiences distress.

The methodology for the analysis will be interpretative phenomenological analysis (IPA), as this is in keeping with constructivist epistemology which is not guided by prior hypotheses. Choosing the constructivist epistemology of IPA serves two main aims:

1. Firstly, there is little previous research studying adjusting to the pre-manifest stage of Huntington's Disease and thus there is a paucity of prior hypotheses to guide the line of questioning from a theoretical perspective
2. Secondly, adjustment to living with a chronic condition is likely to be a very idiosyncratic process, which cannot be indicated prior to in depth data collection and analysis. Hypothesising about adjustment processes within this participant group would likely misrepresent the convergence and divergence of participant experiences, key aspects which IPA focuses on.

Furthermore, as phenomenology is a philosophical approach to the study of the experience of others (Boland, Levack, Hudson & Bell, 2012), IPA fits well with a study exploring how individuals make sense of their experience (Smith & Osborn, 2003) and has a clear focus on psychological processes. The flexible nature of the interview schedule in IPA and the broad, open ended questioning allows the participant to choose the aspects of their experience which feels most pertinent to them, and thus the participant's narrative is what shapes the researcher's understanding of the phenomenon (Smith et al., 2009).

At the same time, the researcher is actively involved in the process, and my interpretations of what the participant discusses will be important part of trying to make sense of how the participant has come to understand their adjustment process. IPA can therefore be considered a double hermeneutic, as it involves two separate layers of interpretation (Smith & Osborn, 2007).

Experts by experience were approached for their input in the study conduct; the principal investigator (myself) approached HD voice (a patient and public involvement group which sits within the HDA). They were invited to express their opinions on the suggested questions provided in the interview schedule as well as provide any more general feedback on the study conduct.

Procedure

Recruitment

Participants will be recruited via three routes:

1. In the first instance, participants will be recruited through a multidisciplinary team at Leicestershire Partnership NHS Trust which works with families affected by HD and includes a clinical psychologist (the field supervisor). Participants will be identified via research nurses working at the Leicestershire Trust who will identify people who meet the inclusion criteria and will invite them to participate (either in person or via post). Any interested patients will be asked to contact me directly (using the contact details on the information sheet) or they can express interest to their clinician/nurse and then the nurse will pass on their details to myself.
2. In the second instance, participants will be recruited from Northamptonshire (an area which receives consultancy from the Leicestershire Service but where patients don't receive ongoing HD psychological support via the same method.) Participants will be identified via the advisory service for Northamptonshire which is commissioned by Leicestershire Trust. Interested participants will contact myself via the methods above.
3. If enough participants cannot be recruited via the first two methods, participants will also be recruited via the Huntington's Disease Association (HDA) website. In this instance, participants will be identified by responding directly to myself via an advert published on the HDA website.

Consent

The participant will provide consent prior to the interview.

If the interview is being conducted in person, then written consent will be taken at the start of the interview. Interviews will only be conducted in person however if conducting it via Skype or phone is difficult for the participant.

If the interview is conducted via telephone or skype (as it is anticipated will be the case with most or all participants) then the participant will complete a consent form in advance and return it to myself via email or post prior to the interview date.

Participants will have several days from receiving the information sheet to decide whether to take part in the interview. Participants can stop the study at any point (by just choosing not to take part in the interview).

Data collection

Semi-structured interviews will be used to ensure they are guided by participants' responses, whilst also ensuring responses are framed around the process of adjustment. Semi-structured interviews are frequently used in IPA research (Smith et al., 2009). Interviews will be conducted either in person, by phone or via Skype dependent upon individual circumstances, but it is anticipated most will be via phone or Skype (or other similar online platforms). Interviews will be audio recorded.

Analysis

After each interview the data will be transcribed verbatim and made anonymous. Analysis will be conducted using guidelines from IPA theory (Smith et al., 2009). Interviews will be transcribed and then analysed line by line, making note of all emerging themes and any apparent contradictions. Emergent themes will be collated and put into overarching categories. Continual re-reading will be used to ensure individual accounts are reflected in the themes and that the main themes are consistent with most of the data. Bias will be reflected upon and incorporated into the study through the use of a reflective journal and supervision.

Direct quotes will be used in the writing up of the findings but these will be anonymised. Basic demographic information may be included alongside their quote but every effort will be made to anonymise participant identifiable data.

Dissemination

The research findings will be disseminated to participants directly and published on the HDA website with permission. It will be submitted as my thesis and presented at the thesis presentation day at Lancaster University. It will be presented at suitable conferences (e.g. European Huntington's Disease Network

Plenary Meeting) and Special Interest Groups. It is also intended that the research will be published in peer-reviewed academic journals.

Practical issues

The recruitment of participants will be taking place in a location and Trust which is different from that of the person conducting the interviews. Where Skype (or another video calling facility) or phone is amenable to participants, this will be used to conduct the interviews to minimise research costs.

Ethical concerns

Risk to participants

The interview is not anticipated to cause any significant distress. However, some of the experiences of the individual may evoke some distressing reactions and/or discuss difficult content.

If the interview is being conducted in person, the interview will be stopped as soon as any distress is identified by the researcher. The participant will be offered a break and, if a family member has attended the interview with them, whether they wish to request their support (presuming the family member is not already in the interview room). After any required break, they will be asked if they wish to stop the interview, or to continue the questions but move on from the distressing topic. Participants will also be made aware that they can rearrange the interview for another date if they wish to do so.

If the interview is being conducted by Skype, distress may be more difficult to become immediately aware of, as subtle changes in body language are not always clear. However, the same principles will apply to interviews conducted via Skype and to those in person. However, there will need to be more confidence from the researcher that any distress that has been identified has been minimised appropriately as there will be no opportunity for face-to-face debriefing after the

interview has ended. The researcher will therefore check on the wellbeing of the participant at the end of the interview.

For both in person and Skype interviews, if any of the research team becomes concerned that a participant poses a risk of serious harm (to themselves or others) they will inform a member of the person's clinical team (if recruited from the NHS) who will assess if any intervention is required and seek guidance where necessary. If the person is recruited via the HDA website then I will ensure that appropriate action is taken (e.g. directing the person to their GP or to A&E as appropriate).

Risk to researchers

Where (if) interviews are conducted in person and particularly when lone working the researchers will adhere to Lancaster University's guidance on fieldwork. The interviewer will leave the details of the interview (e.g., participant, date, time, location) with another trainee clinical psychologist in a password protected document who will be able to access the details if they were not able to reach me. They will telephone another member of the team when the interview is concluded. If this telephone call does not take place, attempts will be made to contact the interviewer. If contact cannot be made, the appropriate authorities will be informed. The details of the interview will be destroyed as soon as the interview has taken place. It is also possible that the researcher will use a commercial electronic lone working system connected to a call centre, when details of the visit will be held briefly on the company database.

Confidentiality and anonymity

The personal information that participants provide will be kept confidential. The data collected for this study will be stored securely and only the researchers conducting this study will have access to the raw data.

- Audio recordings from the interviews will be kept until thesis examination and then securely destroyed
- Lancaster University will keep copies of the interview transcriptions and the consent forms electronically for 10 years after the study has finished or 10 years from publication, whichever is longer. At the end of this time, they will be securely destroyed. Research supervisor Fiona Eccles will be responsible for overseeing the data during this time.
- Files held on the computer will be encrypted (meaning no one other than the researchers can access them) and the computer itself will be password protected
- The typed transcript interviews will be made anonymous by removing any identifying information. Anonymised direct quotations may be used in the report or in publications of the study.
- Personal data collected by the researcher will be confidential and will be kept separately interview responses.

Approximate Timetable for study completion

July 2019: Ethics applications

Sept-Dec 2019: Ethics approval

Jan-Jun 2020: Recruitment and Data collection

Jul-2020-Dec 2020: Data analysis and write up

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East of Scotland Research Ethics Service (EoSRES)



Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

Tayside medical Science Centre
Residency Block Level 3
George Pirie Way
Ninewells Hospital and Medical School
Dundee D D1 9SY

Miss Gina Wieringa
Trainee Clinical Psychologist
Lancaster University
Doctorate in Clinical Psychology
Lancaster University
Lancaster
LA1 4YT

Date: 2 October 2019
Your Ref:
Our Ref: LR/AG19/E S/0105
Enquiries to: Arlene Grubb
Direct Line: 01382 383848
Email: eosres.tayside@nhs.net

Dear Miss Wieringa

Study title: Exploring individuals' experiences of living in the premanifest stage of Huntington's disease
REC reference: 19/ES/0105
Protocol number: N/A
IRAS project ID: 264354

Thank you for your letter of 30 September 2019, 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.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or NHS management permission (in Scotland) should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).

Guidance on applying for HRA and HCRW Approval (England and Wales)/ NHS permission for research is available in the Integrated Research Application System.

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 management permissions from host organisations

Registration of Clinical Trials

It is a condition of the REC favourable opinion that **all clinical trials are registered** on a publicly accessible database. For this purpose, 'clinical trials' are defined as the first four project categories in IRAS project filter question 2. Registration is a legal requirement for clinical trials of investigational medicinal products (CTIMPs), except for phase I trials in healthy volunteers (these must still register as a condition of the REC favourable opinion).

Registration should take place as early as possible and within six weeks of recruiting the first research participant at the latest. Failure to register is a breach of these approval conditions, unless a deferral has been agreed by or on behalf of the Research Ethics Committee (see here for more information on requesting a deferral: <https://www.hra.nhs.uk/planning-and-improving-research/research-planning/research-registration-research-project-identifiers/>

As set out in the UK Policy Framework, research sponsors are responsible for making information about research publicly available before it starts e.g. by registering the research project on a publicly accessible register. Further guidance on registration is available at: <https://www.hra.nhs.uk/planning-and-improving-research/research-planning/transparency-responsibilities/>

You should notify the REC of the registration details. We will audit these as part of the annual progress reporting process.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a site (as applicable).

After ethical review: Reporting requirements

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, including early termination of the study
- Final report

The latest guidance on these topics can be found at <https://www.hra.nhs.uk/approvals-amendments/managing-your-approval/>.

Ethical review of research sites

The favourable opinion applies to all NHS/HSC sites listed in the application subject to confirmation of Capacity and Capability (in England, Northern Ireland and Wales) or management permission (in Scotland) 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
Confirmation of any other Regulatory Approvals (e.g. CAG) and all correspondence [REC response]		
Copies of advertisement materials for research participants [Participant Flyer REC reviewed-highlighted]	0.2	13 September 2019
Copies of advertisement materials for research participants [Participant Flyer REC reviewed-clean]	0.2	13 September 2019
Covering letter on headed paper [Study Approval]		17 July 2019
Evidence of Sponsor insurance or indemnity (non-NHS Sponsors only) [Sponsor Insurance]		18 July 2019
Interview schedules or topic guides for participants [Interview Schedule]	0.1	25 March 2019
IRAS Application Form [IRAS_Form_30092019]		30 September 2019
IRAS Checklist XML [Checklist_30092019]		30 September 2019
Letter from sponsor [Sponsors hip letter]		08 August 2019
Letters of invitation to participant [Letter of invitation]	0.1	25 March 2019
Participant consent form [Participant Consent Form REC reviewed-highlighted]	0.2	13 September 2019
Participant consent form [Participant Consent Form REC reviewed-clean]	0.2	13 September 2019
Participant information sheet (PIS) [Participant Information Sheet Highlighted]	0.2	13 September 2019
Participant information sheet (PIS) [Participant Information Sheet Clean]	0.2	13 September 2019
Research protocol or project proposal [Research Protocol]	0.1	25 March 2019
Response to Request for Further Information		
Summary CV for Chief Investigator (CI) [CV for Chief Investigator]		
Summary CV for student [CV for student]		
Summary CV for supervisor (student research) [Supervisor CV]		14 June 2019

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.

User Feedback

The Health Research Authority is continually striving to provide a high-quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website: <http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/>

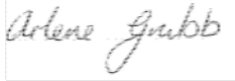
HRA Learning

We are pleased to welcome researchers and research staff to our HRA Learning Events and online learning opportunities— see details at: <https://www.hra.nhs.uk/planning-and-improving-research/learning/>

19/ES/0105**Please quote this number on all correspondence**

With the Committee's best wishes for the success of this project.

Yours sincerely



Mrs Samantha Downie
Chair

Email: eosres.tayside@nhs.net

Enclosures: "After ethical review – guidance for
researchers" [\[SL-AR2\]](#)

Copy to Becky Gordon, Lancaster University

IRAS ID: 264354 Version 0.1

Created: 25/03/19

Semi-structured interview schedule

This interview schedule indicates topic areas to be discussed during the interview with example questions. The exact questions will depend on participants' responses and the focus of each interview will be partly guided by what the individual being interviewed deems to be important. It may be that several questions in each section are not covered, given that for some people the processing of questions may take considerably longer.

Introduction

Introduce myself. Revisit participant information sheet and purpose of interview (To explore the experience of individuals living with the Huntington's disease (HD) gene expansion to gain a better understanding of their experiences)

Remind them of information on consent form and limits of confidentiality ("All information you provide will be kept confidential unless you disclose information regarding risk to yourself or others. In such an event, you will be informed of the action that would be necessary in order to ensure your safety and that of others").

Collect basic info about age, gender, ethnic group, partnership status, living status (who they live with) and employment status

Reflecting on initial genetic screening results

In this section of the interview, I will get information about how long it has been since the genetic test and who else in the family knows they have the HD gene expansion. This section will also include their initial reactions and those of family members, and their expectations for the future. This will not be the focus of the interview but will provide context to their adjustment response.

Example questions:

1. How long ago did you initially receive your screening results?

2. Did you have difficulties in accepting the test results, if so for how long?

Reflecting on experiences adjusting to living in the pre-manifest stage

In this section I will ask about the participant's experiences of having to adjust to living in the pre-manifest stage

Example questions:

1. How do you feel now knowing you will develop HD?
2. How has living with the HD gene affected to your ability to achieve your life goals?
3. How has living with the HD gene changed any future planning?
4. Has there been any positive impact to living in the pre-manifest stage?
5. How have you coped with the negative impacts of living in the pre-manifest stage?
6. Have you shared knowledge that you will develop HD with others?
7. Are you involved in the HD community? If so, what support have you had from them?
8. If you could go back to finding out about your genetic results, is there anything that you think you would have done differently?
9. Is there anything you find helpful in supporting your wellbeing?
10. Tell me about others' reactions to your positive test result?

Reflecting on the situation now

I will ask participants about the ongoing impact of the adjustment process

Example questions:

1. What are your thoughts about where you are at now in relation to and symptom onset?
2. Has your relationship with HD changed since the initial test results? If so, what do you think has led to this change?
3. What does living in the pre-manifest stage mean to you?
4. Do you think things will change in future with how you view where you are at now in relation to HD?
5. What kind of feelings and thoughts do you have for the future?
6. Is there anything else you would like to share about your experiences which you think are relevant?

Conclusion

In this part of the interview the participant will be thanked for taking part. I will ensure the participant has not been distressed by the interview and if necessary will direct the participant to sources of support on the participant information sheet.



Living in the pre-manifest stage of Huntington's disease

IRAS ID: 264354 Version 0.1

Created: 25/03/19

I am writing to you to see if you would be interested in taking part in the above study. The study is being conducted as part of my doctoral qualification in clinical psychology at Lancaster University. I am supervised by Dr Maria Dale, a clinical psychologist working at Leicestershire Partnership NHS Trust and Dr Fiona Eccles, a lecturer at Lancaster University.

I am really keen to better understand the process an individual goes through in order to cope with living with the HD gene, in the period prior to receiving a diagnosis. I hope that this understanding will help others in a similar position to yourself to navigate what I imagine can be a difficult time period. There are no right or wrong answers to this and it is important to gain a variety of viewpoints because everyone will have different experiences.

Before you decide whether or not you would like to participate in this study, it is important for me that you feel as though you understand what will be involved in this project should you decide to participate and have the opportunity to ask any further questions. I would appreciate if you could take time to read the enclosed information sheet carefully and think about whether or not you would like to take part.

If you have any questions about the study then please do contact me. My email address is g.wieringa@lancaster.ac.uk or telephone 07852 516 566 and I will be happy to discuss with you any questions you may have. Alternatively, you can contact one of your clinical team at Leicestershire Partnership NHS Trust or Northamptonshire Healthcare NHS Foundation Trust, depending on where you receive your care.

Thank you very much for reading this letter,

Gina Wieringa (Lead investigator of the study)

Dr Maria Dale (field supervisor)

Dr Fiona Eccles (academic supervisor)



IRAS ID: 264354 Version 0.1

Created : 25/03/19

Participant Information Sheet

Living in the pre-manifest stage of Huntington's disease

My name is Gina Wieringa and I am conducting this research as a student in the clinical psychology doctoral programme at Lancaster University, Lancaster. I really appreciate you taking the time to read this information sheet and your interest in my study.

What is the purpose of this study?

- To explore the experience of individuals living with the Huntington's disease (HD) gene expansion to gain a better understanding of their experiences
- To help inform long-term support for such individuals
- To share important findings about coping strategies which may help others

Why have I been approached?

You have been approached because the study requires information from people who have received testing results which indicate they have the gene expansion for HD. Your contact details have been obtained via your clinical care team at Leicestershire Partnership NHS Trust who felt you may be interested in participating, or because you have responded to the flyer on the HDA website.

Do I have to take part?

No. It's completely up to you to decide whether or not you take part and any care you currently receive (in addition to any future care) will not be affected by your decision. Unfortunately you will not be eligible to participate if you are actively psychotic due to the difficulty in managing psychotic episodes within an interview situation

What will I be asked to do if I take part?

If you decide to participate, I will arrange with you a convenient time to conduct an interview, either by Skype or by telephone. If Skype/phone is not possible for you then I can arrange to meet you in person. This interview should last approximately one hour. During the interview I will ask you some questions about your experiences related to adjusting to living with the HD gene. You will not be required to talk about anything you do not wish to speak about.

Example Questions

1. How do you feel now knowing you will develop HD?
2. Does it affect your life on a day to day basis?
3. Has it had implications on planning for the years ahead (e.g. living arrangements, family support etc.)

Will my data be identifiable?

In order to enable the research to take place, I will need to audio record the interview. Only I will be involved in the audio transcription. All information you provide will be kept confidential unless you disclose information regarding risk to yourself or others. In such an event, you will be informed of the action that would be necessary in order to ensure your safety and that of others.

- This research will be written up as part of my doctoral training and will also be submitted for publication in a journal article. This research may also be presented at conferences
- Audio recordings and consent forms will be destroyed and/or deleted once the project thesis has been examined (December 2020)
- Lancaster University will keep copies of the interview transcriptions and the consent forms electronically for 10 years after the study has finished or 10 years from publication, whichever is longer. At the end of this time, they will be securely destroyed.
- The files on the computer will be encrypted (that is no-one other than the researchers will be able to access them) and the computer itself password protected.
- The typed version of your interview will be made anonymous by removing any identifying information including your name. Anonymised direct quotations from your interview may be used in the reports or publications from the study, so your name will not be attached to them.
- All your personal data will be confidential and will be kept separately from your interview responses. The exception to this is if it is thought that there is a risk of harm to yourself or others, in which case I will need to share this information with my research supervisor.
- Research data will be discussed with my supervisor in order to ensure accuracy and reliability of any themes generated.
- You can choose to withdraw your data that you have already given at any time, up to the point at which it has been analysed, at which point it will not be possible.

Data protection

Lancaster University will be the data controller for any personal information collected as part of this study. Under the GDPR you have certain rights when personal data is collected about you:

- You have the right to access any personal data held about you, to object to the processing of your personal information, to rectify personal data if it is inaccurate, the right to have data about you erased and, depending on the circumstances, the right to data portability.
- Please be aware that many of these rights are not absolute and only apply in certain circumstances.

If you would like to know more about your rights in relation to your personal data, please speak to the researcher on your particular study. For further information about how Lancaster University processes personal data for research purposes and your data rights please visit our webpage: www.lancaster.ac.uk/research/data-protection

What will happen to the results?

The results will be summarised and reported and may be submitted for publication in an academic or professional journal and presented at conferences. I will also give you a copy of the results if you would like one.

Are there any risks?

There are no risks anticipated with participating in this study. However, if you experience any distress following participation you are encouraged to inform me so that I can support you as best as possible. I will endeavour to minimise any distress you are experiencing and will stop the interview if necessary. It may be that I feel it is important to inform your HD care team (if you have one). Alternatively, you will be encouraged to see your GP if you feel this would be helpful. You may also contact the HDA on 0151 331 5444 or email info@hda.org.uk

Are there any benefits to taking part?

Although you may find participating interesting, there are no direct benefits in taking part. It is hoped that gaining a better understanding of how individuals cope in this stage will help others in a similar situation to yourself.

Who has reviewed the project?

This study has been reviewed and approved by the Health Research Authority Research Ethics Committee and the Trust has also given approval for this study to be conducted. The East of Scotland Research Ethics Service REC 2, which has responsibility for scrutinising all proposals for medical research on humans, has examined the proposal and has raised no objections from the point of view of research ethics. It is a requirement that your records in this research, together with any relevant medical records, be made available for scrutiny by monitors from << name of sponsor company (if appropriate) >> and NHS <<insert name of Health Board/Trust>>, whose role is to check that research is properly conducted and the interests of those taking part are adequately protected

Where can I obtain further information about the study if I need it?

If you have any questions about the study or think you might like to take part, please feel free to contact me on g.wieringa@lancaster.ac.uk or telephone 07852 516 566. You can also post details to: Gina Wieringa, Doctorate in Clinical Psychology

Furness College,
Lancaster University,
Bailrigg, Lancaster,
LA1 4YG

You can also contact:

Dr Maria Dale (field supervisor) on Maria.Dale@leicspart.nhs.uk or

Dr Fiona Eccles (academic supervisor) on f.eccles@lancaster.ac.uk or 01524 592807

Complaints

If you wish to make a complaint or raise concerns about any aspect of this study and do not want to speak to the researcher, you can contact:

Ian Smith
Research Director
Lancaster University,
Tel: +44 (0)1524 592981
Email: i.smith@lancaster.ac.uk

If you wish to speak to someone outside of the Doctorate Programme, you may also contact:

Professor Roger Pickup Tel: +44 (0)1524 593746
Associate Dean for Research Email: r.pickup@lancaster.ac.uk
Faculty of Health and Medicine
(Division of Biomedical and Life Sciences)
Lancaster University
Lancaster
LA1 4YG

Thank you for taking the time to read this information sheet.



Living in the pre-manifest stage of Huntington's disease

- Are you willing to take part in an interview of ~1 hour to help with my doctoral thesis (due to be completed by December 2020), which aims to better understand how people comes to terms with HD?

Participants (8 -12 required)

- Need to have tested positive for the HD gene expansion at least one year ago, but not yet showing physical symptoms
- Unfortunately, you will not be eligible to participate if have any other significant health difficulties as this may confound the results

I would really value hearing about any experiences you would be willing to share.

For further information (with no obligation to take part) please contact Gina Wieringa on g.wieringa@lancaster.ac.uk

Participant Consent Form

IRAS ID: 264354 Version 0.2

Project Title: Living in the pre-manifest stage of Huntington's Disease (HD)

Name of researcher: Gina Wieringa

We are asking if you would like to take part in a research project exploring how individual's experience the pre-manifest stage of Huntington's Disease.

Before you consent to participating in the study we ask that you read the participant information sheet and sign at the bottom if you agree with all the statements. If you have any questions or queries before signing the consent form please speak to the principal investigator, Gina Wieringa

1. I confirm that I have read the information sheet and fully understand what is expected of me within this study
2. I confirm that I have had the opportunity to ask any questions and to have them answered.
3. I understand that my interview will be audio recorded and then made into an anonymised written transcript.
4. I understand that audio recordings will be kept until the research project has been examined.
5. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.
6. I understand that once my data have been anonymised and incorporated into themes it might not be possible for it to be withdrawn, though every attempt will be made to extract my data, up to the point of publication.
7. I understand that the information from my interview will be pooled with other participants' responses, anonymised and may be published.
8. I consent to information and quotations from my interview being used in reports, conferences and training events. Any information and quotations used will be anonymised
9. I understand that the researcher will discuss data with their supervisor as needed.

10. I understand that any information I give will remain confidential and anonymous unless it is thought that there is a risk of harm to myself or others, in which case the principal investigator will need to share this information with their research supervisor.
11. I consent to Lancaster University keeping written transcriptions of the interview for 10 years after the study has finished
12. I consent to take part in the above study

Name of Participant_____ **Signature**_____ **Date** _____

Name of Researcher _____ **Signature** _____ **Date** _____

**A University Teaching Trust**

Research & Development
Swithland House
352 London Road
Leicester
LE2 2PL

4th October 2019

Tel: 0116 295 3435
www.leicspart.nhs.uk

Private and Confidential

Miss Gina Wieringa
Trainee Clinical Psychologist
Lancaster University
Lancaster
LA1 4YT

Dear Gina

RE: Letter of Access for Research (Doctorate in Clinical Psychology Student: IRAS 264354) – Exploring individuals' experiences of living in the pre-manifest stage of Huntington's disease.

This letter confirms your right of access to conduct research as a supervised medical student at Leicestershire Partnership NHS Trust for the limited purpose and on the terms and conditions set out below. This right of access commences on **1st October 2019** and ends on **31st December 2020** unless terminated earlier in accordance with the clauses below. This letter of access may be renewed should the need for access be required beyond the lifetime of this document, and subject to further review.

You have a right of access to conduct such research as confirmed in writing in the "confirmation of capacity and capability" letter for research from this NHS organisation, and as specified in the study protocol and OID. Please note that at all times you are responsible to the Local Collaborator for the research project at this site, who is responsible for your conduct, and for delegating appropriate duties.

It is important to understand that receipt of a signed Letter of access from the Trust is only valid if:

- a. You return a signed copy to the R&D Office, and this receipt is acknowledged (and a copy of the LoA is added to the local site file with the local PI and coordinators notified).**

The information supplied about your role in research at Leicestershire Partnership NHS Trust has been reviewed and you do not require an honorary research contract with this NHS organisation. We are satisfied that such pre-engagement checks as we consider necessary have been carried out.

You are considered to be a legal visitor to Leicestershire Partnership NHS Trust premises. You are not entitled to any form of payment or access to other benefits provided by this NHS organisation to employees and this letter does not give rise to any other relationship between you and this NHS organisation, in particular that of an employee.

While undertaking research through Leicestershire Partnership NHS Trust, you will remain accountable to your academic institution, the University of Lancaster but you are required to follow the reasonable instructions of **Dr Dave Clarke (Operational Lead, R&D)** and to liaise with **Dr Maria Dale and staff delegated by her** in terms of your particular duties within the project or those given delegated responsibility on her/his behalf in relation to the terms of this right of access.

Where any third party claim is made, whether or not legal proceedings are issued, arising out of or in connection with your right of access, you are required to co-operate fully with any investigation by this NHS organisation in connection with any such claim and to give all such assistance as may reasonably be required regarding the conduct of any legal proceedings.

You must act in accordance with Leicestershire Partnership NHS Trust policies and procedures, which are available to you upon request, and the requirements of the UK Policy Framework for Research in Health & Social Care.

You are required to co-operate with Leicestershire Partnership NHS Trust in discharging its duties under the Health and Safety at Work etc Act 1974 and other health and safety legislation and to take reasonable care for the health and safety of yourself and others while on Leicestershire Partnership NHS Trust premises. You must observe the same standards of care and propriety in dealing with patients, staff, visitors, equipment and premises as is expected of any other contract holder and you must act appropriately, responsibly and professionally at all times.

You are required to ensure that all information regarding patients or staff remains secure and *strictly confidential* at all times. You must ensure that you understand and comply with all of the requirements of the NHS Confidentiality Code of Practice which is referenced at (<http://www.dh.gov.uk/assetRoot/04/06/92/54/04069254.pdf>) and the Data Protection Act 1998. Furthermore you should be aware that under the Act, unauthorised disclosure of information is an offence and such disclosures may lead to prosecution.

You should ensure that, where you are issued with an identity or security card, a bleep number, email or library account, keys or protective clothing, these are returned upon termination of this arrangement. Please also ensure that while on the premises you wear your ID badge at all times, or are able to prove your identity if challenged. Please note that this NHS organisation accepts no responsibility for damage to or loss of personal property.

We may terminate your right to attend at any time either by giving seven days' written notice to you or immediately without any notice if you are in breach of any of the terms or conditions described in this letter or if you commit any act that we reasonably consider to amount to serious misconduct or to be disruptive and/or prejudicial to the interests and/or business of this NHS organisation or if you are convicted of any criminal offence. Your substantive employer is responsible for your conduct

during this research project and may in the circumstances described above instigate disciplinary action against you.

Leicestershire Partnership NHS Trust will not indemnify you against any liability incurred as a result of any breach of confidentiality or breach of the Data Protection Act 1998. Any breach of the Data Protection Act 1998 may result in legal action against you and/or your substantive employer.

If your current role or involvement in research changes, or any of the information provided in your Research Passport changes, you must inform your employer through their normal procedures. You must also inform your nominated manager in this NHS organisation.

Please sign and return 1 copy of the attached Letter of Access to the address above and forward one signed copy to your HR Department for their files.

Your Letter of Access may be checked at any time during the lifetime of the research, please ensure that you have your copy with you at all times. The information it contains must therefore remain up to date and accurate.

In the absence of a returned fully signed copy of the contract you will NOT be allowed onto Trust premises to conduct any research. As described above, your role in the study must be listed in the Delegation Log and this approved and counter-signed by the local Principal Investigator.

Yours sincerely



Dr Dave Clarke
Research & Development Operational Lead

CC – HR

LETTER OF ACCESS BETWEEN

NHS Organisation	Leicestershire Partnership NHS Trust
	AND
Name	Gina Wieringa
Employer OR Place of Study	University of Lancaster
Report to: (Local Collaborator)	Dr Maria Dale

PERIOD OF AGREEMENT


From:	1 st October 2019	To:	31 st December 2010
OR			
Fixed-term Contract for:	Years:	Months:	Effective Date:

STUDY

Study Title:	Exploring individuals' experiences of living in the pre-manifest stage of Huntington's disease.
Study Location(s):	Mill Lodge (HD Service and Referral/Advice Network)
Primary Activity:	Identification and contact with participants

SIGNATURES

Researcher:	
Name:	Gina Wieringa
Date:	

On behalf of the NHS Organisation:	
Name/Role:	Dr Dave Clarke (Operational Lead – Research & Development)
Date:	4 th October 2019

For Office Use Only.

	Date	Signed
Date Signed Copy received in R&D Office		
Confirmation Individual added to delegation log and signed-off by Principal Investigator:		
Date Added to local Site File		



Miss Gina Wieringa
 Trainee Clinical Psychologist
 Lancaster University
 Doctorate in Clinical Psychology
 Lancaster University
 Lancaster
 LA1 4YT

Email: hra.approval@nhs.net
HCRW.approvals@wales.nhs.uk

03 October 2019

Dear Miss Wieringa

**HRA and Health and Care
 Research Wales (HCRW)
 Approval Letter**

Study title:	Exploring individuals' experiences of living in the premanifest stage of Huntington's disease.
IRAS project ID:	264354
Protocol number:	N/A
REC reference:	19/ES/0105
Sponsor	Lancaster University

I am pleased to confirm that [HRA and Health and Care Research Wales \(HCRW\) Approval](#) has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications received. You should not expect to receive anything further relating to this application.

Please now work with participating NHS organisations to confirm capacity and capability, in line with the instructions provided in the "Information to support study set up" section towards the end of this letter.

How should I work with participating NHS/HSC organisations in Northern Ireland and Scotland?

HRA and HCRW Approval does not apply to NHS/HSC organisations within Northern Ireland and Scotland.

If you indicated in your IRAS form that you do have participating organisations in either of these devolved administrations, the final document set and the study wide governance report (including this letter) have been sent to the coordinating centre of each participating nation. The relevant national coordinating function/s will contact you as appropriate.

Please see [IRAS Help](#) for information on working with NHS/HSC organisations in Northern Ireland and Scotland.

How should I work with participating non-NHS organisations?

HRA and HCRW Approval does not apply to non-NHS organisations. You should work with your non-NHS organisations to [obtain local agreement](#) in accordance with their procedures.

What are my notification responsibilities during the study?

The standard conditions document “[After Ethical Review – guidance for sponsors and investigators](#)”, issued with your REC favourable opinion, gives detailed guidance on reporting expectations for studies, including:

- Registration of research
- Notifying amendments
- Notifying the end of the study

The [HRA website](#) also provides guidance on these topics, and is updated in the light of changes in reporting expectations or procedures.

Who should I contact for further information?

Please do not hesitate to contact me for assistance with this application. My contact details are below.

Your IRAS project ID is **264354**. Please quote this on all correspondence.

Yours sincerely,

Natalie Wilson
Approvals Specialist

Email: hra.approval@nhs.net

Copy to: Becky Gordon, Lancaster University, Sponsor contact

List of Documents

The final document set assessed and approved by HRA and HCRW Approval is listed below.

<i>Document</i>	<i>Version</i>	<i>Date</i>
Confirmation of any other Regulatory Approvals (e.g. CAG) and all correspondence [REC response]		
Copies of advertisement materials for research participants [Participant Flyer REC reviewed-highlighted]	0.2	13 September 2019
Copies of advertisement materials for research participants [Participant Flyer REC reviewed-clean]	0.2	13 September 2019
Covering letter on headed paper [Study Approval]		17 July 2019
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Sponsor Insurance]		18 July 2019
Interview schedules or topic guides for participants [Interview Schedule]	0.1	25 March 2019
IRAS Application Form [IRAS_Form_30092019]		30 September 2019
IRAS Checklist XML [Checklist_30092019]		30 September 2019
IRAS Checklist XML [Checklist_30092019]		30 September 2019
Letter from sponsor [Sponsorship letter]		08 August 2019
Letters of invitation to participant [Letter of invitation]	0.1	25 March 2019
Organisation Information Document [Organisation Information Document]	0.1	25 March 2019
Participant consent form [Participant Consent Form REC reviewed-highlighted]	0.2	13 September 2019
Participant consent form [Participant Consent Form REC reviewed-clean]	0.2	13 September 2019
Participant information sheet (PIS) [Participant Information Sheet Highlighted]	0.2	13 September 2019
Participant information sheet (PIS) [Participant Information Sheet Clean]	0.2	13 September 2019
Research protocol or project proposal [Research Protocol]	0.1	25 March 2019
Response to Request for Further Information		
Schedule of Events or SoECAT	1	05 September 2019
Summary CV for Chief Investigator (CI) [CV for Chief Investigator]		
Summary CV for student [CV for student]		
Summary CV for supervisor (student research) [Supervisor CV]		14 June 2019

Information to support study set up

The below provides all parties with information to support the arranging and confirming of capacity and capability with participating NHS organisations in England and Wales. This is intended to be an accurate reflection of the study at the time of issue of this letter.

Types of participating NHS organisation	Expectations related to confirmation of capacity and capability	Agreement to be used	Funding arrangements	Oversight expectations	HR Good Practice Resource Pack expectations
Research activities and procedures as per the protocol and other study documents will take place at participating NHS organisations.	Research activities should not commence at participating NHS organisations in England or Wales prior to their formal confirmation of capacity and capability to deliver the study.	An Organisation Information Document has been submitted and the sponsor is not requesting and does not expect any other site agreement to be used.	Sponsor is providing envelopes and stamps to participating NHS organisations (please see Organisation Information Document).	A Local Collaborator (LC) is expected at participating NHS organisations.	No Honorary Research Contracts, Letters of Access or pre-engagement checks are expected for local staff employed by the participating NHS organisations. Where arrangements are not already in place, research staff not employed by the NHS host organisation undertaking any of the research activities listed in the research application would be expected to obtain a Letter of Access based on standard DBS checks and occupational health clearance.

Other information to aid study set-up and delivery

This details any other information that may be helpful to sponsors and participating NHS organisations in England and Wales in study set-up.

The applicant has indicated that they do not intend to apply for inclusion on the NIHR CRN Portfolio.



**Leicestershire Partnership
NHS Trust**

A University Teaching & Research Active Trust

Research & Development

Swithland House
352 London Road
Leicester
LE2 2PL

Direct dial: 0116 295 7641

Email: dave.clarke@leicspart.nhs.uk

Tel: 0116 295 3534

Web: www.leicspart.nhs.uk

DC/FE/GW/MD/NEUR0850/264354

4th October 2019

Miss Gina Wieringa
Trainee Clinical Psychologist
Lancaster University
Lancaster
LA1 4YT

Dear Gina

RE: CONFIRMATION OF CAPACITY & CAPABILITY:

Exploring individuals' experiences of living in the pre-manifest stage of Huntington's disease.

Study Codes:			
Trust Reference:	NEUR0850	IRAS (REC) Reference:	264354
EDGE ID:	126866	Portfolio ID:	N/A
Study Sponsor:	University of Lancaster (Becky Gordon)		

I am writing in respect of your application to conduct research involving Leicestershire Partnership NHS Trust (the "Trust"). This study has now been validated and reviewed according to local ORCA processes. Therefore the Trust can **confirm that we have capacity to support this research** on the condition that the Trust suffers no unforeseen costs as a result of this study being undertaken. A study record has been created on the Trust's Research Database and/or LPMS. All research studies taking place are now subject to monitoring in respect of confirmation timelines, recruitment to time and target and so on. The key monitoring target is a 70-day timeline from receipt of a Local Information pack at site, which incorporates the 30-day timeline, within which the first patient or participant should be recruited.

Confirmation of Capacity & Capability STATUS					
Confirmed in FULL		<input checked="" type="checkbox"/>	Confirmed in PRINCIPLE		<input type="checkbox"/>
			Not Confirmed		<input type="checkbox"/>
TIMELINES				Milestones	
Date of Favourable Ethical Review (Time A)	3 rd October 2019	Date Site Invited ¹ (Time B ¹)	13 th September 2019	(A-B ¹)	N/A
		Date Site Selected ² (Time B ²)	18 th September 2019	(B ¹ -B ²)	N/A
Date of Initial HRA Assessment	N/A	Date Sponsor Confirmation (Time B ³)	3 rd October 2019	(B ² -B ³)	14 Days
Date of Final HRA Approval ³	3 rd October 2019	Date Site Confirmed (CCC) (Time C)	4 th October 2019	(B ² -C)	1 Day
Target Date: First Patient/Participant Visit (FPFV) (B ² + 70)			27 th November 2019		

The conduct of your study (including examination of the site file) at this site may be subject to audit for protocol adherence and other monitoring. This approval is subject to the accuracy of the following information:

¹ Date of receipt of e-mail including approved study protocol

² Receipt of minimum document set (VRA) from Sponsor

³ Where required; studies may not begin if final HRA Approval not secured.

Study Summary			
Chief Investigator (Supervisor):	<i>Miss Gina Wieringa (Dr Fiona Eccles)</i>		
Principal Investigator (Local):	<i>Dr Maria Dale</i>	CRN Delivery Co-ordinator(s)	<i>N/A</i>
Indemnity Provider:	<i>NHS And University of Lancaster</i>	Start Date (Local)	<i>7th October 2019</i>
NIHR Portfolio:	<i>No</i>	End Date (Local)	<i>1st December 2020</i>
Student Project	<i>Yes (Doctorate in Clinical Psychology)</i>	Target Recruitment	<i>12</i>
Funding Source:	<i>N/A</i>	Amount:	
Local NHS Support Costs:	<i>N/A</i>		
PID Qualifying Study:	<i>No</i>		
Sponsor Reference:			

Please note that all research with an NHS element is subject to the provisions of the UK Policy Framework for Health & Social Care (November 2017). If you are unfamiliar with the standards contained in this document, or the LPT policies that reinforce them, you can obtain advice from the R&D Office or your Sponsor. You must stay in touch with the R&D Office during the course of the research project, particularly if/ when:

- There is a change of Principal Investigator;
- To fulfil requirements for performance reporting;
- The project finishes (please complete a summary report form);
- Amendments are made, whether minor or substantial;
- Serious Adverse Events occur (adhere to local and Sponsor SOPs).


This is necessary to ensure that your indemnity cover is and remains valid. Should any issues arise that inhibit study delivery it is essential that you contact the R&D Office immediately. If patients or staff members are involved in an incident, you should also contact the Clinical Risk Manager and report as per Trust Policy.

Provision against NHS Costs: The Trust reserves the right to invoice the study team, in the unlikely event of any unexpected costs arising from this study, including, but not limited to:

- Staff Time attending interviews.
- Travel and administrative costs

I hope the project goes well, and if you need any help or assistance during its course, please do not hesitate to contact the Office.

Kind regards



Dr Dave Clarke
Operational Lead (R&D): *for Trust R&D Office*

Copies to:

Becky Gordon (Sponsor)