Doctoral Thesis

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

A qualitative exploration of individual and couple's experiences of movement disorders

Doctorate in Clinical Psychology

Lancaster University

Louise Glover

2019 intake

October 2022

Word Count

	Main Text	Tables, Figures, References and Appendices	Total
Abstract	300	-	300
Literature Review	7,916	10,779	18,695
Empirical Paper	7,492	11,385	18,877
Critical Appraisal	3,956	817	4,773
Ethics Section	5,353	6,610	11,963
Total	**25,017**	**29,591**	**54,608**

Thesis Abstract

The thesis titled ‘A qualitative exploration of individual and couple’s experiences of movement disorders’ explores experiences relevant to two common movement disorders; Parkinson's and cervical dystonia (CD).

Section one presents a meta-ethnography, synthesising 19 qualitative studies relevant to the couple’s experience of Parkinson’s. The analysis of which produced three themes, which were: 1) Disruption of the couple’s roles and responsibilities and attempting to maintain the relationship; 2) Challenges to communication and closeness which can lead to increased resilience; and 3) Increased cognitive and behavioural symptoms leading to grief, burden and isolation. The findings highlight the challenges that couples experience and the individual and relational resources that support them to cope. The relevant strengths, weaknesses, and implications for clinical practice and future research are discussed.

Section two presents a qualitative interpretative phenomenological analysis (IPA) study exploring the experience of diagnosis in CD. Six participants were interviewed, and their data analysed. The analysis produced three themes: 1) Losing control over one’s body and attempts to regain control: ‘You don't know what's going on and your whole life is kinda falling apart’; 2) Feeling powerless in response to the health system: ‘I had this change and I couldn't do anything about it, and nobody seemed to have any kind of solution’; 3) Mixed feelings upon diagnosis: ‘It's kinda like a relief, but scary at the same time’. The findings provide further insight and interpretation of the experience of diagnosis and highlight how this can be a significant and distressing experience. Clinical implications and suggestions for further study are discussed.

Section three presents a critical appraisal where the author has discussed broader research issues and reflections on the reports contained in sections one and two. Considered, are the

implications for the findings presented, their strengths, limitations, and suggestions for future research.

Declaration

This thesis contains research undertaken for the Doctorate in Clinical Psychology at the Division for Health Research, Lancaster University. The work presented here is the author's own, except where due reference is made. The work has not been submitted for the award of a higher degree elsewhere.

Name: Louise Glover

Signature:

Signature removed for the published version.

Date: 21st October 2022

Acknowledgements

I would firstly like to thank the six people who participated in this research; thank you for your trust and for taking the time to speak with me. I hope the report contained in section two is a respectful interpretation of your experiences.

I would like to thank my supervisors, Dr Fiona Eccles, Dr Clare Dixon and Dr Christopher Kobylecki. Thank you for sharing your wisdom and expertise with me, without which, this project would not have been possible. I have valued the opportunity to work with you throughout this research. It has been an opportunity to gain insight into the world of research, which has left me keen to continue to contribute to the psychological evidence base throughout my career as a clinical psychologist.

I would like to give a special thank you to my husband, Joe, who has supported me throughout this journey to completing the thesis project. You have played a huge role in keeping me sane. Thank you for the many 'surprise' cups of tea that you brought to me whilst I was focused on the computer screen, not noticing that you had come and gone. You never expected any thanks as you were just being you, but I will forever be grateful. Finally, thank you to my daughter, Molly, as being a positive role model for you has been a great motivation to keep going through challenging times.

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Section One: Systematic Literature Review

Parkinson's and the couple relationship: A qualitative meta-synthesis

Word Count (excluding references, tables and appendices): 7,896

Louise Glover

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

All correspondence should be sent to:
Louise Glover
Health Innovation Campus
Lancaster University
Sir John Fisher Drive
Lancaster,
LA1 4AT

Email: l.harris7@lancaster.ac.uk

Prepared for: Aging and Mental Health

Abstract

Objective***:*** The aim of this meta-synthesis is to explore the current qualitative literature on the experience of Parkinson's and the couple's relationship including the perspectives of both people with Parkinson's and their caregiving spouse or partner.

Rationale: There is a shift toward a relational focus within the broader field of health research. Several relevant qualitative studies were available that considered aspects of the experience of Parkinson's and the couple's relationship. Previous reviews have not considered the qualitative literature in-depth and from the perspective of both people with Parkinson's and partners.

Methods: Noblit and Hare's (1988) meta-ethnography approach was applied; 19 studies were included in the review following a systematic search of four electronic databases.

Findings: Analysis produced three themes: 1) Disruption of the couple's roles and responsibilities and attempting to maintain the relationship; 2) Challenges to communication and closeness which can lead to increased resilience, and 3) Increased cognitive and behavioural symptoms leading to grief, burden and isolation. The themes are discussed with supporting extracts from the 19 included studies.

Conclusion: The findings highlight the challenges that couples experience and the individual and relational resources that support coping. The present review suggests that services may work more effectively with people with Parkinson's and their partners rather than approaching them as separate individuals. Support should be individually tailored to each couple as the impact on the couple may change in response to individual and contextual factors. This review adds further evidence to the case for relationally focused multidisciplinary team input at all stages of Parkinson's disease.

Keywords: Parkinson's; couple relationship; dyad; experiences; qualitative research, systematic review.

Background

Parkinson's is the second most common neurodegenerative disorder, currently estimated to affect six million people worldwide (Dorsey et al., 2018). Although younger onset is possible, Parkinson's typically develops over 50, its prevalence increases with age, and it is more common among males than females (Pringsheim et al., 2014).

Parkinson's is characterised by three main physical symptoms; involuntary shaking of parts of the body, slowness in movement and stiff and inflexible muscles (DeMaagd & Philip, 2015). A range of other physical and psychological difficulties may also present, including anxiety, depression, loss of sense of smell, problems with balance, and problems with sleep (Kobylecki, 2020). People with Parkinson's may also experience cognitive impairment, hallucinations, delusions, behaviour change, and dementia (Kobylecki, 2020). Parkinson's has an idiosyncratic trajectory. Therefore, disease stage and progression are commonly measured in clinical practice with the use of four stages, which include diagnosis, maintenance, complex, and palliative stages (Kobylecki, 2020), with a recognition that time spent within each phase may vary significantly. Treatments, including pharmacological and surgical treatments, are available to help manage the presenting symptoms and improve mobility. However, treatments may become less effective over time and can cause unwanted side effects, including dyskinesia, and impulsive and compulsive behaviours, such as hypersexuality (Weintraub et al., 2010). People with Parkinson's require increasing levels of care over time, a role often taken up by close relatives, many of whom are either a spouse or partner (DeMaagd & Philip, 2015). Much of the research on Parkinson's has focused on the needs of caregivers or people with Parkinson's individually. Findings of which have highlighted the importance of supporting caregivers with the many challenges related to

caregiving and the impact of people with Parkinson's increasing dependence on their sense of autonomy and emotional well-being (Chen et al., 2021; Greenwell et al., 2015; Henry et al., 2020; Mosley et al., 2017; Perepezko et al., 2019; Theed et al., 2017; Vescovelli et al., 2018).

Due to the progressive nature of Parkinson's and increasing need for care over time, much of which is provided by a spouse or partner, the development of Parkinson's can have a great impact upon the couple relationship. This is because individuals do not exist in isolation; instead, when one person experiences illness, it has wide-reaching effects on affected individuals, those close to them, and the relationships between them (Rosland et al., 2012). Impact on the relationship can be exacerbated or mitigated by several factors, such as the severity of motor symptoms, psychological symptoms, reciprocity, and intimacy (Bertschi et al., 2021; Perepezko et al., 2019). A positive dyadic relationship, however, is said to mediate the effect of the burden associated with caregiving (Bertschi et al., 2021; Goldsworthy & Knowles, 2008; Mosley et al., 2017). Furthermore, higher rates of 'mutuality' are beneficial - a term used to encompass features that signify relationship quality including love and affection, shared pleasurable activities, shared values, and reciprocity (Archbold, 1992; Archbold et al., 1990). Increased mutuality between spouses and people with Parkinson's was found to be associated with better mental health of both partners, reduced caregiver burden and improved quality of life for people with Parkinson's and their significant other (Karlstedt et al., 2017; Tanji et al., 2008; Wielinski et al., 2010). Therefore, the couple relationship has an important role in supporting the wellbeing and coping ability of both people with Parkinson's and their partners.

Several theoretical models have proposed explanations of dyadic coping within long term chronic illness. Dyadic coping is a term used to describe the interpersonal mechanisms between individuals with a health condition and their close relatives who provide care during illness (Bodenmann, 1997). The models include the developmental-contextual model (DCM;

Berg & Upchurch, 2007; Checton et al., 2015), the cognitive transactional model (CTM; Badr & Acitelli, 2017) and the dyadic-regulation connectivity model (DRCM; Karademas, 2022). The DCM and CTM highlight the relationship between illness and dyadic coping, suggesting that illness characteristics such as disease stage, or knowledge about disease, influence the potential for adaptive dyadic coping. They also suggest, in a reciprocal manner, that effective dyadic coping influences more positive appraisals of illness and well-being and leads to improved health outcomes. The CTM highlights that both intra- and interpersonal coping; for example, the ability of one individual to communicate their needs to their partner, influences the couple's ability to share the illness experience and thus achieve dyadic coping. Whilst the DCM highlights dynamic and fluctuating factors influencing coping, such as financial strain, gender, age, life stage, relationship quality and illness symptoms and stage. Most recently, the DRCM (Karademas, 2022), proposed that three corresponding 'hubs' influence dyadic coping. The first hub is 'The representations hub', which involves appraisals of illness, coping, relationship quality, and emotional reactions. The second hub is 'the coping hub', which encompasses the ongoing interactions between partners, coping strategies, congruence between intra and interpersonal coping, and the broader context of social support and environment that facilitates couples coping. The third hub is 'the outcomes hub', which includes the stage and impact of illness, patient and partner adaptation to illness, and the impact of illness on their relationship. The DRCM aimed to recognise the complexity and fluidity of couples' experiences, suggesting that difficulties within any of the hubs can impact the entire system.

The relevant theoretical models highlight the complexity of couple's experiences of illness, and highlight the dynamic and reciprocal processes that exist between individuals that affect coping. However, despite the increasing focus on the couple's relationship in Parkinson's, gaps in our understanding remain. A recent scoping review by Weitkamp et al.

(2021) provided a helpful overview of pertinent issues relevant to dyadic coping in chronic illnesses, including Parkinson's. The review considered a small number of studies with differing methodologies (i.e., quantitative and qualitative), lacked in-depth insights, and did not include the experience of the caregiving partner. Yet, several qualitative studies exist whose findings are relevant to Parkinson's and the couple's relationship that were not included in the aforementioned scoping review, that include the views of people with Parkinson's and their partners or spouses both together and individually.

Qualitative research can provide an in-depth insight into the lived experience of couples experiencing Parkinson's, shedding light on the pertinent interpersonal mechanisms which are highly valuable to stimulate further research and influence clinical care. Since there is a number of existing qualitative research studies available, a review of the qualitative literature would be worthwhile. Meta-ethnography (Noblit & Hare, 1988), is a method of meta-synthesis aiming to synthesise qualitative research findings on a related topic to provide new and developed understandings. This method results in a rich and broad understanding of the experience in question, more so than can be gained by the articles individually.

Therefore, the current review aims to synthesise the available qualitative literature related to the experience of Parkinson's and the couple's relationship, including the perspective of both people with Parkinson's and their caregiving spouse or partner. The research question that informed the review was 'What is the impact of Parkinson's on the couple's relationship, and conversely, what is the impact of the couple's relationship on the experience of Parkinson's?'. The review findings will stimulate further research initiatives and provide an enhanced understanding of the impact of Parkinson's on the couple's relationship to guide clinical practice with particular attention paid to the role of clinical psychologists within the care pathway.

Method

Design

The approach of meta-ethnography was employed during the synthesis. Meta-ethnography is an interpretive and inductive approach to qualitative synthesis developed by Noblit and Hare (1988) and commonly used in qualitative health research. Meta-ethnography seeks to provide a novel and interpretive account of the topic in question by drawing on all relevant literature available. The current synthesis was influenced by Noblit and Hare's (1988) original works as well as more recent guidance for increasing the quality and rigour of meta-ethnography methods and reporting (France et al., 2019; Sattar et al., 2021). The PRISMA guidelines (Page et al., 2021) guided the reporting of the synthesis.

Search Strategy

Prior to conducting a systematic search, the research aims were organised into four concepts: (1) Parkinson's (2) couples/partners OR (3) relationship satisfaction/quality, and (4) qualitative research methods. After completing an initial scoping search and consultation with an academic librarian, the search strategy contained in Tables 1 and 2 was produced which includes both subject heading search terms and free text search terms.

[Tables 1 & 2 about here]

Studies were included if they met the following inclusion criteria: (1) were available in English; (2) were published in a peer-reviewed journal; (3) employed qualitative data collection and analysis methods; (4) Parkinson’s was the primary diagnosis or the main focus; (5) findings were presented thematically or narratively, and the use of quotes provided evidence; and (6) results addressed to some degree the interplay between Parkinson’s and the couple relationship and involved people with Parkinson’s and their partners or spouses either together or separately. Where a small number of participants were not a spouse or partner, e.g., an adult child acting as a care partner, papers were included, but only findings that

related to the couple's relationship were included (Lawson et al., 2018; Roger & Medved, 2010; Thomson et al., 2020). Studies were excluded if they had experiences of people with a separate diagnosis where it was not clear which findings related to which diagnosis.

Using the search strategy outlined in Tables 1 and 2, relevant qualitative research papers were identified by searching four academic databases: MEDLINE (searchable years 1977-, 'English' and 'peer reviewed' selected), PsycINFO (searchable years 1989-, 'peer reviewed' selected), CINAHL (searchable years 1994-, 'peer reviewed' and 'English language' selected), and Academic Search Ultimate (searchable years 1997-, 'peer reviewed' selected). The search was conducted in November 2021 and repeated in March 2022, which enabled the inclusion of one further paper (Constant et al., 2022).

The database search returned 3,267 papers (PsycINFO = 739, MEDLINE = 1,066, CINAHL = 400, Academic Search Complete = 1,062). Duplicates were removed (n = 1,464), and the remaining papers were reviewed by title and abstract. The full text of the papers deemed suitable (n = 31) were read, and the inclusion and exclusion criteria were applied. In total, 19 papers were identified that met the criteria for inclusion in the current review. Then the 19 studies' reference sections were searched for any other relevant papers, and no further papers were identified. This process is illustrated within the PRISMA flow diagram in Figure 1.

[Figure 1 - PRISMA flow diagram, about here]

Study Characteristics

A summary of the study characteristics is contained in Table 3. The papers were published between 2000 and 2022. Studies took place in several countries including France (1), Denmark (n=1), Sweden (n= 1), New Zealand (n= 1), Australia (n= 1), Mexico (n=1), Canada (n=2), the USA (n= 4), and the UK (n= 7). Sample sizes varied (range=7-44). All studies used semi-structured interviews as a data collection method. Two studies used

supplementary data collection methods, such as reflective diaries, creative writing, surveys, and focus groups, to support the collection of sensitive data related to violence or sexual intimacy (Fleming et al., 2004; Sánchez-Guzmán et al., 2022). Various approaches of qualitative analysis were used in the studies. Seven studies used thematic analysis (Haahr et al., 2013; Lawson et al., 2018; McKeown et al., 2020; Thomson et al., 2020; Vatter et al., 2018; Whitehead, 2010; Wootton et al., 2019). Six studies used phenomenological approaches, such as interpretative phenomenological analysis (Constant et al., 2022; Habermann, 2000; Hodgson et al., 2004; Mach et al., 2019; Smith & Shaw, 2017; Williamson et al., 2008). Two studies used grounded theory (Martin, 2016; Roger & Medved, 2010). One study described their analysis methods as inductive and comparative (Sánchez-Guzmán et al., 2022). One study used a content analysis approach (Birgersson & Edberg, 2004). Finally, two studies gave no explicit detail of their analysis methods (Deutsch et al., 2021; Fleming et al., 2004).

A summary of the participants characteristics is contained in Table 4. The characteristics of participants included in the studies varied. Eleven studies included people with Parkinson's and their spousal heterosexual or heterosexual romantic partner/cohabiting partners (Birgersson & Edberg, 2004; Constant et al., 2022; Hodgson et al., 2004; Lawson et al., 2018; Martin, 2016; Roger & Medved, 2010; Smith & Shaw, 2017; Sánchez-Guzmán et al., 2022; Thomson et al., 2020; Whitehead, 2010; Wootton et al., 2019). Seven studies included the spouse or partner of people with Parkinson's only (Deutsch et al., 2021; Haahr et al., 2013; Habermann, 2000; Mach et al., 2019; McKeown et al., 2020; Vatter et al., 2018; Williamson et al., 2008). One study included people with Parkinson's only (Fleming et al., 2004). Relationship duration also varied within and across studies, between 18 months-64 years. One study included four people with Parkinson's who had previously been in a relationship that had ended (Fleming et al., 2004).

The studies included 137 people with Parkinson's and 191 partners. Of the total people with Parkinson's included in the studies, 83 were male, and 54 were female. Of the partners, 147 were female, and 44 were male. Nine studies included information about participants' ethnicity. Of these studies, most were from majority ethnic (white, western) backgrounds (Haahr et al., 2013; Habermann, 2000; Hodgson et al., 2004; Mach et al., 2019; Martin, 2016; Vatter et al., 2018; Whitehead, 2010; Williamson et al., 2008; Wootton et al., 2019). Nine studies included detail about the participants' employment status suggesting half of people with Parkinson's and 60 per cent of partners were employed (Fleming et al., 2004; Haahr et al., 2013; Habermann, 2000; Hodgson et al., 2004; Lawson et al., 2018; Mach et al., 2019; Sánchez-Guzmán et al., 2022; Thomson et al., 2020; Wootton et al., 2019).

Information about the duration of Parkinson's was included in 15 papers and varied from one to 30 years (Birgersson & Edberg, 2004; Constant et al., 2022; Deutsch et al., 2021; Fleming et al., 2004; Habermann, 2000; Hodgson et al., 2004; Mach et al., 2019; Martin, 2016; McKeown et al., 2020; Smith & Shaw, 2017; Sánchez-Guzmán et al., 2022; Thomson et al., 2020; Whitehead, 2010; Williamson et al., 2008; Wootton et al., 2019). Details about the severity or stage of Parkinson's were provided by 11 studies (Constant et al., 2022; Habermann, 2000; Hodgson et al., 2004; Lawson et al., 2018; Martin, 2016; McKeown et al., 2020; Thomson et al., 2020; Vatter et al., 2018; Whitehead, 2010; Williamson et al., 2008; Wootton et al., 2019). The methods used to measure Parkinson's stage varied across studies, including reference to specific dimensions of Parkinson's, such as receiving deep brain stimulation, dopaminergic medications (Thomson et al., 2020; Whitehead, 2010), displaying impulsive and compulsive behaviours (McKeown et al., 2020), and experiences of Parkinson's delusions and psychosis (Deutsch et al., 2021; Williamson et al., 2008). Four studies used the Hoehn & Yahr Scale (Constant et al., 2022; Habermann, 2000; Hodgson et

al., 2004; Martin, 2016). The Hoehn & Yahr Scale is a tool which was introduced to describe Parkinson's stage and severity with seven stages that represent increasing severity of physical disability (Goetz et al., 2004). One study aimed to overcome the lack of disease stage homogeneity across other studies by including participants who were classified as advanced stage only according to the Hoehn & Yahr Scale (Constant et al., 2022). Where studies were not concerned with a specific Parkinson's experience, the homogeneity of disease stage and severity varied within studies; for example, one study included participants with and without cognitive impairment and people with Parkinson's dementia (Lawson et al., 2018).

[Table 3 & 4 about here]

Quality Appraisal

The results of the quality appraisal process are contained in Table 5. The papers were appraised using the Critical Appraisal Skills Programme for qualitative studies (CASP, 2018). The tool comprises ten items that aim to prompt and guide the researcher in assessing the quality of research studies and a helpful guide to support this process for novice researchers (Noyes et al., 2018). Papers were not excluded on the basis of quality appraisal, as they may have contained valuable findings despite differences in methodological rigour and reporting (Atkins et al., 2008). However, this process highlighted methodological issues that were important to be aware of when including the study's findings in the synthesis and their weighted contribution to the results. Quality rating scores were applied using a system introduced by Duggleby et al. (2010), whereby a score of 1-3 was given for each quality appraisal item on the CASP, which resulted in a total score out of 24. The scores provided a quick and accessible method to compare quality ratings across papers. Three papers were randomly selected for quality appraisal by an independent researcher due to the often-subjective nature of quality ratings. This process revealed an agreement in quality rating across the three papers.

[Table 5 about here]

Analysis and Synthesis

The seven-step approach to meta-ethnography proposed by Noblit and Hare (1988) informed the analysis. Themes were developed in an iterative process whilst repeatedly returning to the original texts to build an in-depth understanding of the studies' content and meaning. Firstly, all 19 papers were read and re-read, and key themes and metaphors were identified and compared across papers. The quality appraisal processes also aided familiarity. Next, all relevant second-order data was extracted along with relevant participant quotations. Annotations accounted for emerging themes that supported comparison across studies to identify similarities and divergences. Several key concepts were identified through this process, which were then grouped to represent broader interpretations. A line of argument synthesis developed, which allowed the bringing together of various aspects of the Parkinson's experience to provide a new and dynamic narrative of the Parkinson's experience from the couple's point of view. Appendix 1-A offers an example of the data extraction process; Appendix 1-B depicts the theme development process.

Researcher's position

Within the present report, the researcher recognises their position as a trainee clinical psychologist and completion of this project as partial fulfilment of the doctoral training programme in clinical psychology. Therefore, the researcher has an interest in the psychological and emotional processing of experiences, issues of which will have stood out to the researcher over others. To reduce the introduction of bias due to the researchers interests and experiences, the researcher engaged in reflective supervision throughout the review process, and the extraction of data and analysis were assessed by experienced researchers at incremental stages.

Findings

Three themes were identified that captured the experience of Parkinson's and the couple's relationship. These were: 1) Disruption of the couple's roles and responsibilities and attempting to maintain the relationship; 2) Challenges to communication and closeness which can lead to increased resilience; and 3) Increased cognitive and behavioural symptoms leading to grief, burden, and isolation. Below, the themes are discussed alongside supporting extracts from the 19 included studies.

Theme 1: Disruption of the couple's roles and responsibilities and attempting to maintain the relationship

A Parkinson's diagnosis and changes in abilities to carry out daily activities resulted in changes in the distribution of domestic roles and responsibilities between couple members. This change resulted in relationship uncertainty and dissatisfaction for both people with Parkinson's and their partners. The extent to which couples were impacted negatively by this change seemed to be influenced by several factors, such as values held within the pre-existing relationship, disease severity and the context in which couples resided. Couples described the use of individual and relationally focused methods of coping.

Due to people with Parkinson's decreased physical and cognitive ability, partners were required to take on an increasing number of household tasks. Partners experienced a constant sense of responsibility: 'I've had to take on all the responsibility, money, power of attorney, I have to do the maintenance.' (Partner) (Vatter et al., 2018, p. 608). The reallocation of domestic responsibilities represented a loss of role for people with Parkinson's (Constant et al., 2022; Smith & Shaw, 2017). People with Parkinson's who could no longer work experienced frustration at spending more time at home, feeling that they should go out to work to contribute to their family and household (Hodgson et al., 2004). Changes to the roles and responsibilities of people with Parkinson's burgeoned a sense of redundancy,

reduced confidence and dissatisfaction, the strain of which caused them to fear that their partners would leave them (Fleming et al., 2004; Martin, 2016).

> I used to, you know, I didn't wield a stick but you know I couldn't, I could never do that, but I felt as though I had a position in the family, but now I don't, I feel downgraded a bit, whether that's paranoia setting in or not I don't know but I just feel a lesser person ... I feel as though she's the boss now, really and it's quite rightly is too because she's got me to put up with, so there you are. (Person with Parkinson's) (Lawson et al., 2018, p. 5).

Partners' experienced a further role change due to their loved one's need for care and support. However, the degree of change experienced differed between individuals. Some partners developed an in-depth knowledge of their loved one's body and symptoms, whilst others viewed themselves as supporters, with their crucial role being to encourage independence and autonomy (Haarh et al., 2013; Habermann, 2000; Vatter et al., 2018). These differences seemed related to the nature of the relationship prior to Parkinson's; for example, when couples had previously valued their independence, these values remained. In these cases, difficulties arose when the need for care outweighed the ability to retain independence. For example, when care needs were higher, some partners began to view their role as a caregiver rather than identifying as a partner (Thomson et al., 2020; Vatter et al., 2018). Partners recognised a need to stay physically close to their loved ones: 'I must think about being close by. If I go out, I bring the mobile phone.' (Partner) (Haarh et al., 2013). For some, increased closeness represented a loss of independence and freedom.

The extent couples were affected by changes in their roles and responsibilities seemed to be influenced by external factors. For example, people with Parkinson's who were not experiencing cognitive problems were less likely to report a change in how they saw themselves and their relationship with their partner (Constant et al., 2022; Lawson et al.,

2018). This suggested that factors such as the severity of symptoms influenced the relationship quality. Parkinson's dominated the lives of some couples, causing confusion and distress with the degenerative and fluctuating nature of the condition and subsequent fluctuating need for care (Roger & Medved, 2010; Smith & Shaw, 2017). Furthermore, stress increased in the context of external factors such as financial strain, which resulted in some partners needing to continue working whilst also balancing the needs of their loved ones (Martin, 2016).

Many couples described methods of coping with their change in roles and responsibilities. For example, finding meaning and purpose in their new roles as caregivers or cared for, or opportunities to share their learnings with others, for example, partners or people with Parkinson's sharing their experiences with others in a support group setting (Habermann, 2000; Lawson et al., 2018). Couples described a wish to preserve their identities and autonomy by continuing with pre-diagnosis activities, which they felt, helped their relationship to thrive (Constant et al., 2022; Habermann, 2000; Hodgson et al., 2004; Mach et al., 2019; Roger & Medved, 2010; Smith & Shaw, 2017). Couples stressed the importance of finding a balance between people with Parkinson's needs and the support provided so as not to disempower. Nevertheless, partners struggled to hold back and watch their loved one's struggle (Constant et al., 2022; Habermann, 2000; Hodgson et al., 2004; Mach et al., 2019; Roger & Medved, 2010; Smith & Shaw, 2017).

Theme 2: Challenges to communication and closeness which can lead to increased resilience

Parkinson's introduced challenges to couples' lives, disrupting how they communicated and achieved closeness. Symptom-related changes affected couples' verbal and emotional communication and their ability to participate in activities they enjoyed together pre-diagnosis, affecting their sense of familiarity and togetherness. However, the

pre-existing relationship was a source of strength and resilience, and couples who managed to overcome such challenges experienced a strengthening of their relationship.

When symptom-related difficulties with verbal expressivity were experienced, couples' communication reduced, which led to a sense of distance between them (Birgersson & Edberg, 2004; Mach et al., 2019; Vatter et al., 2018; Whitehead, 2010; Williamson et al., 2008; Wootton et al., 2019). Facial masking, whereby the facial muscles are affected, causing slowed facial expressivity or a blank expression, disrupts non-verbal communication, affecting couples' ability to communicate emotionally. This lack of emotional reciprocity led to feelings of rejection, loss and disconnection for partners who worried their loved one was no longer satisfied with their relationship (Wootton et al., 2019). 'If that information's not there, you fill it in, and everything you fill in no facial expression with is boredom, tiredness, anger. The really negative emotions.' (Partner) (Wootton et al., 2019, p. 2520).

The degenerative and fluctuating nature of Parkinson's symptoms restricted the time and freedom couples felt they had, causing difficulty with planning and participating in activities they previously enjoyed. These changes impacted couples' sense of togetherness and led to feelings of estrangement from one another (Birgersson & Edberg, 2004; Haarh et al., 2013; Habermann, 2000; Lawson et al., 2018; Martin, 2016).

> Well, I guess maybe we're less close because we can't do everything together. We always did everything together, and now we can't. (Partner) (Martin, 2016, p. 234)

Social activities were reduced, and many couples reported spending less time with friends due to a concern about stigma towards Parkinson's symptoms (Haarh et al., 2013; Habermann, 2000; Martin, 2016). This reduced socialisation led couples to become increasingly isolated, which was a further source of relational dissatisfaction. Subsequently, partners expressed grief for the lives they had previously led together (Birgersson & Edberg, 2004; Habermann, 2000).

> And I think it's just since he has gotten more advanced, probably in the last two years or so. We still get together with our close friends which fills the need, but I've noticed a decline socially, a smaller circle. (Partner) (Habermann, 2000, p. 1412).

Furthermore, symptoms and medication disrupted couples' sexual intimacy, which for some was expected and viewed as a normal part of ageing (Fleming et al., 2004; Haarh et al., 2013; Habermann, 2000; Martin, 2016; Vatter et al., 2018). However, many couples, particularly those with younger onset Parkinson's, found that reduced sexual intimacy strained their relationship (Martin, 2016; Vatter et al., 2018; Wootton et al., 2019).

> Well, I think it's just that, the changes that are happening [from Parkinson's] are stressful on our relationship sometimes. He has all these health problems from Parkinson's [so] he can't sleep in bed. We are on different floors. He sleeps in a recliner. I sleep in the bedroom. It's just kind of like, sometimes, it's like we are roommates. (Partner) (Martin, 2016, p. 235).

Couples valued support and information from healthcare professionals, which helped them to develop an understanding of Parkinson's signs and symptoms and develop skills which helped to maintain a positive relationship, such as alternative communication strategies and challenging negative thoughts and feelings which arose (Thomson et al., 2020; Wootton et al., 2019). However, throughout the challenges that Parkinson's presented, the pre-existing couple relationship was a source of strength and resilience to cope: 'When you've been married as long as we have, it takes a lot to shake things up'. (Partner) (Martin, 2016, p. 232). This statement suggests that the length and strength of a relationship may be influential on a couple's ability to cope with difficulties associated with Parkinson's. Furthermore, pre-existing communication styles, problem-solving ability and evidence of overcoming previous challenges were also thought to influence couples' coping (Haarh et al., 2013; Martin, 2016; Roger & Medved, 2010; Sánchez-Guzmán et al., 2022; Thomson et al., 2020). When couples

were able to overcome challenges, they experienced a strengthening of their relationship with enhanced communication, understanding, appreciation, and a sense of resilience to cope with future challenges (Habermann, 2000; Roger & Medved, 2010; Wootton et al., 2019).

Theme 3: Increased impairment leading to grief, burden and isolation

Periods of enhanced need or complexity due to cognitive and behavioural changes, which increased the partner's sense of burden, were a common theme among papers. Due to this, the partners' experiences are highlighted more so than the perspective of people with Parkinson's throughout this theme. However, this experience was relevant to the nature of the couple's relationship; for example, emotional distance within the couple's relationship at this stage was common. Nevertheless, partners continued to draw on the strength of their pre-existing relationship to continue caring for their loved ones.

Partners experienced loss and grief for their loved ones as their impairments grew. Furthermore, people with Parkinson's had been their partner's source of emotional support during times of challenge, which due to increased impairment, they were often no longer able to provide (Deutsch et al., 2021; Williamson et al., 2008). Partners also contended with the increasing weight of responsibility to care for their loved ones and to make decisions alone. As such, partners no longer viewed themselves as part of a mutually beneficial relationship (Deutsch et al., 2021; Lawson et al., 2018; Thomson et al., 2020; Vatter et al., 2018; Williamson et al., 2008; Wootton et al., 2019).

> This man, this personality change he's gone through, it's crazy. It's not—he's not the man I married. He's definitely not the man I married. He's changed so much. If that's just part and parcel of Parkinson's, I guess? (Partner) (Thomson et al., 2020, p. 2220).

Furthermore, behaviour changes such as jealous delusions were a significant challenge. Partners blamed themselves for these difficulties and spent time ruminating over their behaviours: 'I had to be really careful that I didn't trigger something that would upset

him because he would become very upset.' (Partner) (Deutsch et al., 2021, p. 4). This led partners to experience a sense of exhaustion and a subsequent lack of energy to consider any actions that may help to maintain their relationship (Deutsch et al., 2021; Mach et al., 2019; McKeown et al., 2020). There was also evidence of a common perception that people with Parkinson's lacked the care and interest to maintain the relationship at this stage which seemed to highlight a lack of understanding or capacity to empathise regarding the impact of Parkinson's on their loved ones physical and cognitive abilities. Yet, this further fuelled partner's sense of emotional distance (Constant et al., 2022),

Some partners described a wish to continue to protect their loved ones when impulsive and compulsive behaviours or delusional beliefs were present by explaining and justifying behaviours and symptoms or concealing these issues. Such coping methods meant that partners could not access the support they needed from family, friends, and healthcare professionals (Haarh et al., 2013; Deutsch et al., 2021; McKeown et al., 2020). Hesitancy to share openly and, therefore access support seemed to be connected to a wish to protect their loved ones but also due to shame or embarrassment regarding people with Parkinson's symptoms and behaviours.

> I couldn't tell anybody about the violence and aggression or the pornography – my daughter would have been shocked and that is not the way I want him to be remembered. (Partner) McKeown et al., 2020, p. 4628)

Overall, coping as a couple was markedly more challenging when cognitive and behavioural changes were dominant. However, partners employed several strategies to manage, such as externalisation of unwanted behaviours by focusing on them as part of the disease rather than their loved one's character and the purposeful recall of more positive memories of their relationship (Deutsch et al., 2021; Mach et al., 2019; McKeown et al., 2020; Thomson et al., 2020; Vatter et al., 2018). Such strategies evidenced the source of

strength that was the couple's relationship, which partners could continue to draw on to support them in their caring role. At this stage, partners stressed the need to access external support such as respite care and enhanced support from friends and family (Deutsch et al., 2021; Lawson et al., 2018; Thomson et al., 2020). However, this needed to be considered carefully as, for some couples, separation further fuelled issues such as jealous delusions (Deutsch et al., 2021). Those who experienced shame and embarrassment valued being asked directly about behaviour change by health professionals or opportunities to access anonymous sources of support such as online support groups (McKeown et al., 2020).

Discussion

This review aims to synthesise the available qualitative evidence about the experiences of Parkinson's and the couple's relationship. It was hoped that this would provide a deeper understanding and new insights into the impact of Parkinson's on the couple and how being a couple influences the management of Parkinson's. Three main themes were identified, which outlined the challenges couples faced and their inter and intrapersonal coping methods. The themes expressed the impact of Parkinson's on couples' roles and responsibilities, how couples communicate and achieve closeness, and the impact of increased impairment and behavioural changes related to Parkinson's. Here the findings will be discussed with reference to the existing literature to consider how they fit with or extend current understandings.

The first theme discussed the impact that Parkinson's has on the roles and responsibilities of people with Parkinson's and their partners. In response to a diagnosis of Parkinson's and their affected partner's reduced physical and cognitive abilities, unaffected partners took on increasing roles and responsibilities. For some couples, new roles and responsibilities resulted in distress and relational dissatisfaction. Where possible, retaining a sense of autonomy for both partners and people with Parkinson's was helpful. The nature of

the pre-existing relationship and values held by the couple influenced coping, for example, whether they previously held views that regarded independence as a priority or lived a comparatively more intertwined life. Such values appeared to influenced interpretations about needing to provide or receive care from one's partner. The current evidence base reflects this diversity of coping styles. On the one hand, encouraging independence and autonomy in people with Parkinson's is suggested to support the maintenance of functional ability, well-being, and quality of life (Soundy et al., 2014; Zhao et al., 2021). On the other hand, research on 'we-ness' suggests that couples who view illness as a collective experience have improved health and well-being (Rohrbaugh, 2021).

The findings presented within this review indicate that dyadic coping may differ across the trajectory of Parkinson's, as its fluctuating nature and external stressors influence change in roles and responsibilities. These findings fit most closely with the DCM, whereby many contextual factors are believed to control couples' coping at any given time (Berg & Upchurch, 2007). However, couples described both intra and interpersonal dimensions of their dyadic coping. For example, retaining normalcy within a relationship was a relational endeavour that preserved individual well-being. Equally, individual coping methods seemed to support their relational bond, suggesting that both intra- and inter-personal coping mechanisms support both couple members with their role adjustments. This finding is in line with the DRCM, the most recent model proposed for couples coping with illness, which considers dyadic coping as a resource that fits alongside individual coping (Karademas, 2022).

The second theme related to the challenges couples experience, such as difficulties communicating and reduced ability to engage in activities. Such challenges impacted couples' sense of closeness and caused uncertainty, dissatisfaction, and strained relationships. Firstly, difficulties with communication led to a dichotomy between people with Parkinson's and

partners. The CTM (Badr & Acitelli, 2017) highlights the necessity for couples to be able to communicate their needs when attempting to cope with a stressor. This communication allows couples to view a stressor, such as illness, as a shared experience and thus support their coping. The findings presented were also reminiscent of research focused on social identity (Soundy et al., 2014). This research suggests that the sense of oneself as a member of a social group is a key factor influencing the well-being of people with Parkinson's.

The present review extends these suggestions that social identity as a couple is affected when Parkinson's symptoms and concerns about stigma, prohibit couples' usual functioning. A further finding within this theme was that many couples found their relationship strengthened when they overcame challenges. This finding is reminiscent of the idea of 'benefit finding' in chronic illness, which suggests that experiencing illness may lead to benefits such as personal growth or improved relationships (Pakenham, 2010). In MS, benefit finding has been shown to positively influence adjustment in people with the health condition and their caregivers (Pakenham & Cox, 2009).

The third theme highlighted partners' experiences related to the impact of increased impairment, psychological and cognitive changes, and changes in personality and behaviour. These experiences made people with Parkinson's appear unlike themselves, which was associated with feelings of distress, loss, and grief for partners and a loss of their mutually beneficial relationship. In Parkinson's, cognitive and psychological changes are suggested to have the most significant impact on couples' estimations of mutuality (Archbold, 1992; Archbold et al., 1990; Karlstedt et al., 2017; Karlstedt et al., 2020). Whereas higher rates of mutuality between spouses and people with Parkinson's were found to be associated with the improved mental health of both partners, reduced caregiver burden and improved quality of life for both spouses and people with Parkinson's (Karlstedt et al., 2017; Tanji et al., 2008; Wielinski et al., 2010). Therefore, it was not surprising to find that at this stage, partners

experienced distress. However, partners experienced difficulties accessing the support they needed to cope, and it seemed to be interpersonal dynamics or relational issues which were central inhibitors of partners' help-seeking behaviours. Therefore, this review provides a relational viewpoint to previous understandings, which suggest social support alongside individual personalities were factors influencing partners' ability to cope (Greenwell et al., 2015; Lyons et al., 2009).

Clinical implications

The present review suggests that services may work more effectively with people with Parkinson's and their partners rather than approaching them as separate individuals. The couple relationship is vital and allows health professionals to draw on the resources this contains whilst providing further avenues for clinical interventions. A relational focus also provides an opportunity to forecast where difficulties may present by drawing on available information about the relationship history and common problems encountered, thus allowing for preventative care. To date, UK professional guidance for care and supportive interventions for both people with Parkinson's and carers, has focused on individual rather than dyadic needs (National Institute for Health and Care Excellence, 2017; Simpson et al., 2021). Furthermore, partners receive a 'carers assessment', which amplifies the suggestion of a power imbalance within couples who may be actively striving against this dynamic. Critiques of the term 'carer' suggest that this term may inhibit support-seeking due to the negative connotation of burden that it may present to some (Molyneaux et al., 2011). Hence, there is a clear need to acknowledge the need for a dyadic focus within professional care guidelines.

Parkinson's requires adjustments on behalf of people with Parkinson's and their partners regarding their roles and identities throughout the Parkinson's trajectory. Therefore, dyadic needs should be considered throughout the care pathway. However, there is currently

little evidence for effective psychological interventions to support couples living with Parkinson's. Preliminary suggestions have included support with problem-solving and facilitating role adjustment, as well as therapies such as narrative therapy, solution-focused therapy, and emotion-focused therapy (Beaudet & Ducharme, 2013; Haahr et al., 2020; Spencer & Haub, 2018). The evidence for psychological interventions for couples living with neurodegenerative conditions and other chronic illnesses is promising for the efficacy of behavioural interventions, emotionally focused couple therapy, narrative therapy, solution-focused therapy, and couple's psychotherapy (Ghedin et al., 2017; Martire & Helgeson, 2017; Spencer & Haub, 2018; Vincent, 2019).

The duration of Parkinson's experienced by the couples represented in this review varied greatly, and some couples lived with Parkinson's for many years. The findings presented here suggest that there were benefits for couples to empower people with Parkinson's to maintain autonomy and independence in order to maintain individual well-being and relational satisfaction. However, the pre-existing relationship and the values held by the couple influenced how they coped and what was deemed 'normal' to them. Acceptance and commitment therapy (ACT; Hayes & Smith, 2005) may be a helpful approach to support these processes both when working with couples or individuals. ACT is a talking therapy, derived from CBT, that focuses on helping patients to focus on connecting with and behaving in line with their values and supports the application of mindfulness and acceptance skills to difficult and uncontrollable experiences (Hayes & Smith, 2005). ACT has been helpfully applied to help couples repair and rebuild their relationships and has become more widely used within health psychology (Graham et al., 2016; Hosseinpanahi et al., 2020).

The present review provides a first-person account of the many challenges experienced by couples when cognitive and behavioural symptoms are present. For example, couples described the distance they felt in their relationships due to the impact of Parkinson's

symptoms. Such issues are essential to consider from a clinical point of view, particularly as the review identified the value of clinical interventions which supported couples to reconnect and understand Parkinson's. Therefore, interventions, such as supporting communication and providing education about Parkinson's, ensure that couples can be prepared for the challenges and increase their resilience (Gamarel & Revenson, 2015). Furthermore, research suggests that couple interventions may be more effective than individual interventions in couples living with chronic illnesses (Berry et al., 2017). Further understanding issues faced by couples will allow professionals to draw on relational resources to enhance coping and empower couples to live well with Parkinson's.

The current review highlights that the nature of support should be tailored to each couple as the impact on the couple may change in response to many factors, making this work complex and multifaceted. Clinical supervision can support health professionals in reflecting on contextual issues and improving mental health outcomes (Snowdon et al., 2017). Clinical psychologists are trained to recognise and work with complex, systemic and contextual issues which impact well-being and to provide clinical supervision to support this practice in other health professionals (Duits et al., 2020). As such, the role of clinical psychologists is vital in providing couples' interventions and service-wide shifts toward relationally focused Parkinson's care.

Limitations

Whilst this review aims to provide a comprehensive, in-depth review of the qualitative literature relating to couples' experiences of Parkinson's, it includes several limitations. Three times the number of males represented people with Parkinson's compared to females. This gender imbalance is in line with estimations regarding Parkinson's prevalence (Wooten et al., 2004). However, it means the findings presented may be weighted towards the female partner experience and male people with Parkinson's experience. It is

important to consider this gender bias within the reported findings as females with Parkinson's are more likely to experience poorer quality of life compared to males (Crispino et al., 2020). Furthermore, a lack of diversity across participants in ethnic background and sexual identity of the couples represented means any findings or conclusions that are transferred to other groups should be done sensitively and with this in mind.

Although the papers included scored well on quality ratings, issues such as homogeneity were not assessed. Recruitment of a homogenous sample ensures that participants' experiences are relevant to the research question (Denny & Weckesser, 2022). There was a lack of homogeneity in the participant samples of several studies with a variation in Parkinson's duration and severity. Experiences differ throughout the disease stage; without clear distinction, it is difficult to determine the findings that relate to each. An awareness of this was held during the reporting of the synthesis, and findings were not applied prescriptively to distinct disease stages. Finally, this review is concerned with couples' experiences of Parkinson's, yet for many couples, the impact of Parkinson's is so great that the relationship breaks down (Fleming et al., 2004). Due to the nature of the review, only one individual's perspective was provided whose relationship had broken down, so the most distressing experiences may not be represented within the current review.

Future research

Further areas for research include couples' experiences of Parkinson's in diverse populations, for example, in minority populations, such as different ethnic groups or sexual identities. Future research should focus on the experience of couples whereby partners are all male or all female which would further our understanding of the first-hand experience of couples living with Parkinson's. Furthermore, quantitative exploration might consider whether gender distribution affects the impact on couples as suggested in other chronic health conditions (DeLongis, & Revenson, 2010). A qualitative exploration of the couple's

experiences with further consideration of sample heterogeneity would be advantageous. As suggested, this exploration of couples' experiences at specific points of the disease trajectory, such as diagnosis experience. Alternatively, during the 'complex stage' according to the widely accepted four-stage model of Parkinson's, which includes cognitive and behavioural symptoms (Kobylecki, 2020). Areas for quantitative research may explore the relationships between individual factors, such as benefit-finding on couples coping with Parkinson's. Longitudinal research would be helpful to see the impact of changes over time on the couple's relationship as the disease progresses. Finally, research to consider the efficacy of specific therapies (e.g., ACT) would provide evidence for the provision and funding for couples' interventions or interventions with a relational focus within Parkinson's services.

Conclusion

This review adds further evidence to the case for relationally focused multidisciplinary care in Parkinson’s and explores the relevance of the couple’s relationship to many dimensions of the Parkinson’s experience. This review highlights the source of strength and resource for coping that the couple relationship represents that deserves to be preserved. Supporting couple hood has the potential to enable couples experiencing Parkinson’s to live well for longer. Further research is needed to understand relational dimensions at discreet junctures within Parkinson’s and to provide evidence for effective interventions to support couples.

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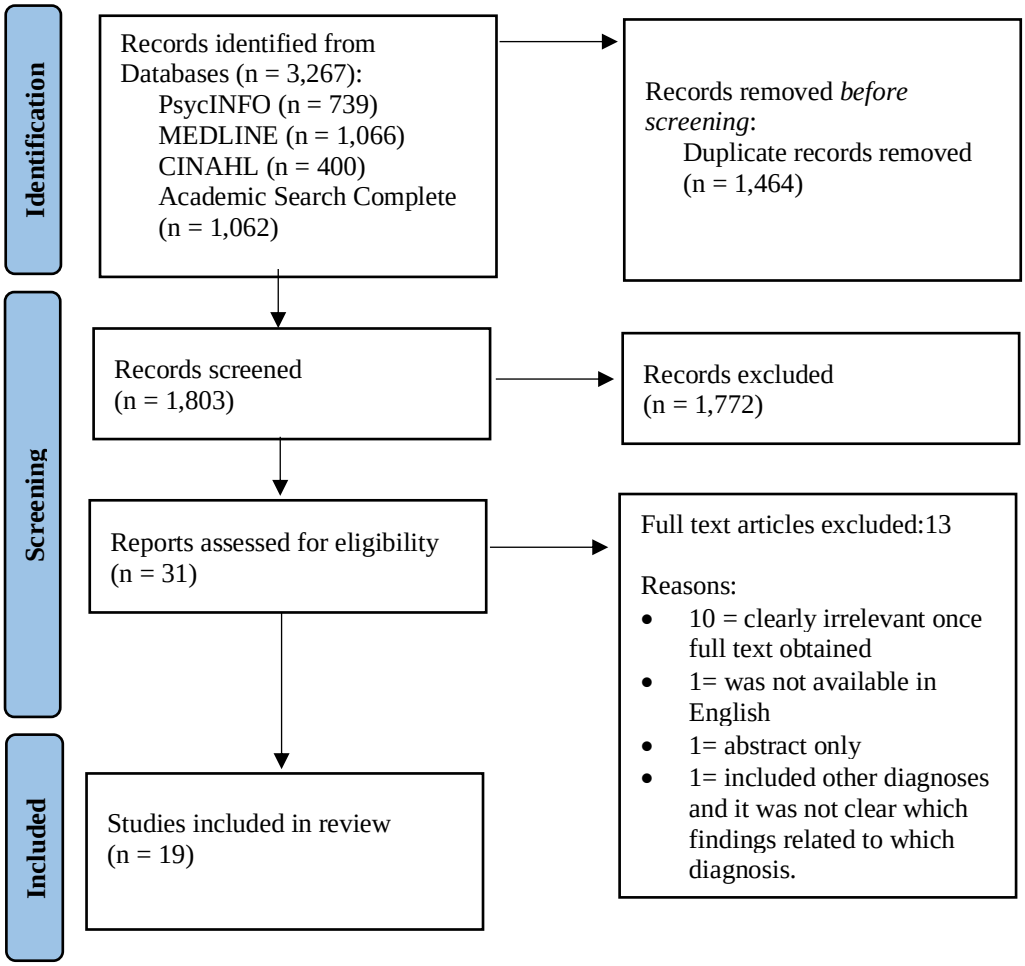
Tables and Figures

Figure 1

PRISMA flow-diagram depicting the process of literature searching and selection.



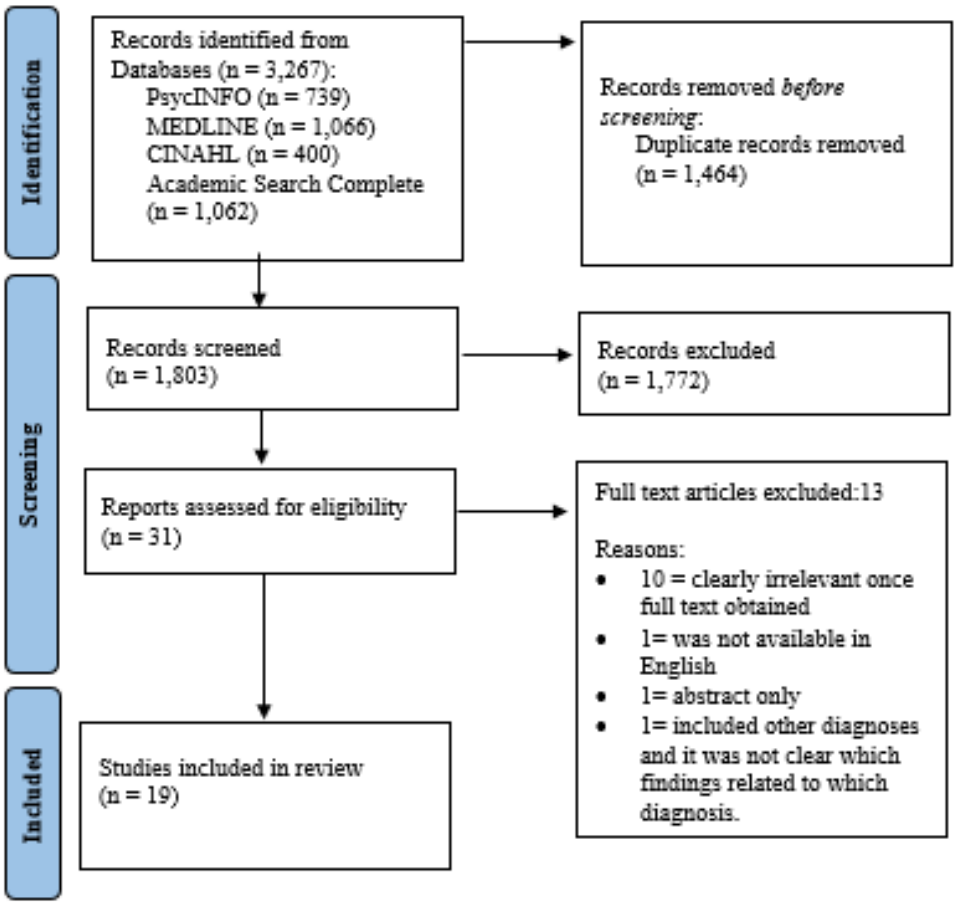


Figure 1 demonstrates the process undertaken when identifying relevant papers for inclusion in the review.

Table 1

Database subject headings and thesaurus terms

	Concept	Database*	Subject terms (+ indicates terms exploded)
Sample	Parkinson's	MEDLINE MESH headings	MH "Parkinson Disease" DE "Parkinson's Disease"
		PsycINFO Thesaurus terms	(MH "Parkinson Disease")
		CINAHL	
Phenomenon (1)	Couples/partners	MEDLINE MESH headings	(MH "Interpersonal Relations+") OR (MH "Family") OR (MH "Family Relations+")
		PsycINFO Thesaurus terms	DE "Interpersonal Relationships" OR DE "Same-Sex Couples" OR DE "Cohabitation" OR DE "Dyad*" OR DE "Family" OR DE "Romance" OR DE "Spouses" OR DE "Husbands" OR DE "Wives" OR DE "Couples" OR DE "Significant Others" OR DE "Partner" OR DE "Marital Satisfaction"
		CINAHL MESH headings	(MH "Interpersonal Relations+")
Phenomenon (2)	Relationship satisfaction/quality	MEDLINE MESH headings	MH "Personal Satisfaction"
		PsycINFO Thesaurus terms	DE "Marital Satisfaction" OR DE "Marriage and Partner Measures" OR DE "Relationship Quality" OR DE "Role Satisfaction" OR DE "Intimacy" OR DE "Male Female Relations" OR DE "Interpersonal Relationships" OR DE "Relationship Satisfaction"
		CINAHL MESH headings	(MH "Interpersonal Relations") OR (MH "Intimacy")
Research design/methods	Qualitative research methods	MEDLINE MESH headings	(MH "Qualitative Research+")
		PsycINFO Thesaurus terms	(DE "Group Discussion" OR DE "Case Report" OR DE "Focus Group Interview" OR DE "Participant Observation" OR DE "Focus Group" OR DE "Grounded Theory" OR DE "Interpretative Phenomenological Analysis" OR DE "Narrative Analysis" OR DE "Semi-Structured Interview" OR DE "Thematic

CINAHL	Analysis" OR DE "Mixed Methods Research" OR DE "Phenomenology" OR DE "Qualitative Measures" OR DE "Qualitative Methods") (MH "Qualitative Studies")

**Note.* Function not available on the Academic Search Ultimate database.

Note. free text terms were the same across all databases (see, Table 2).

Table 2

Free text search terms

	Concepts	Search Terms
Sample	Parkinson's	Parkinson* OR (parkinson* N3 diseas*) AND
Phenomenon (1)	Couples/partners	wive* OR wife* OR husband* OR couple* OR dyad* OR "significant other" OR spous* OR partner* OR carer* OR famil* OR
Phenomenon (2)	Relationship satisfaction/quality	(life OR relationship* OR interpersonal OR inter-personal) N5 (impact OR satisfaction OR quality OR wellbeing OR well-being OR well being) AND
Research design/methods	Qualitative research methods	experience* OR qualitative OR interview* OR grounded theory OR phenomenolog* OR narrative OR thematic analysis

Table 3

Study Characteristics

Paper	Research Question	Methodology	Findings (Themes)
Martin (2016)	To understand couples' experiences and the effects of Parkinson's on a relationship. "What relational issues do PWP (people with Parkinson's) and partners report experiencing as a result of the disease?"	*Data collection:* One to one semi structured interviews *Qualitative Analysis:* iterative approach using constant comparative techniques borrowed from grounded theory	(1) Experiencing changes in overall relational closeness, (2) shifting relational roles, (3) experiencing changes in sexual intimacy, (4) facing financial stress, (5) engaging in fewer leisure and social activities together, (6) experiencing uncertainty about their relationship
Haahr et al. (2012)	To explore the lived experience of being a spouse to a person living with advanced Parkinson's disease, before and during the first year of deep brain stimulation.	*Data collection:* Longitudinal interviews. 2–4 weeks prior to DBS and again 6 weeks, 6 months, and 1 year following DBS Qualitative Analysis: Hermeneutic approach, thematic analysis	(1) Solidarity – the base for joined responsibility and concern, (2) living in partnership, (3) a sense of freedom embracing life, (4) the challenge of changes and constraint.
Vatter et al. (2018)	To explore the changes in long-term intimate relationships in Parkinson's-related dementia, as perceived by spouses providing care to their partners.	*Data collection:* semi structured Interviews *Qualitative Analysis:* inductive thematic analysis	(1) Altered relationship, (2) care partner challenges, (3) acceptance and adjustment
Birgersson a & Edberg (2004)	To describe persons with Parkinson's disease and their partners' experience of support received.	*Data collection:* one-to-one interview Qualitative Analysis: Content analysis	(1) Being in the light, (2) being in the shade.
Williamson et al. (2008)	To explore the experiences of individuals living with a partner with Parkinson's	*Data collection:* one-to-one interview	(1) Uncertainty and the search for understanding, (2) adapting to symptoms over time, (3) the contribution of psychosis to

	disease and psychotic symptoms.	*Qualitative Analysis:* interpretative phenomenological analysis	changing identities, (4) the use of social comparison as a coping strategy.
Fleming et al. (2004)	To develop understandings of the experiences and adjustments made by women with PD in relation to womanhood.	*Data collection:* individual or small group interviews, reflective diaries or creative writing. Qualitative Analysis: No detail	(1) Intrapersonal health, (2) interpersonal health, (3) extra personal health
Lawson et al. (2018)	To explore the subjective impact of cognitive impairment on people with PD and their carers.	*Data collection:* one to one interview Qualitative Analysis: Thematic analysis	(1) threats to identity and role, (2) predeath grief and feelings of loss in carers, (3) success and challenges to coping in people with PD, (4) and problem-focused coping and finding meaning in caring.
Thomson et al. (2020)	To examine the significance and meaning of DBS-related changes in personality and self for patients and caregivers	Data collection: Interviews Qualitative Analysis: Thematic analysis	(1) Impact of illness on personality and self, (2) Awareness and beliefs about DBS-related personality change, (3) Hopes and fears, (4) Restoration of the "old self.", (5) Lived experiences of personality change, (6) Clinical management of personality changes.
Sánchez-Guzmán et al. (2020)	To study the intimate partner violence among PP and their caregivers. More specifically, we explored whether the presence of violence was related to the type of couple relationship before the disease onset.	*Data collection:* Survey, interviews, focus groups *Qualitative Analysis:* Data analysed inductively and comparatively	(1) Disease and history of violence, (2) disease as a buffer of violence, and (3) the burden of disease as an inductor of violence
Smith & Shaw (2017)	To understand family members' lived experience of PD and their opportunities for well-being.	Data collection: Interviews *Qualitative Analysis:* interpretative	(1) It's more than just an illness, (2) like a bird with a broken wing, (3) being together with PD, (4) carpe diem.

		phenomenological analysis	
Roger & Medved (2010)	To discuss how individuals living with Parkinson's disease and their main family supports perceive communications with each other, with a focus on their roles related to care.	Data collection: Interviews Qualitative Analysis: Grounded Theory	(1) The first moment of change after diagnosis, (2) managing identity together: a part of ongoing daily experience, (3) assisting others as part of managing identity together,
Deutsch et al. (2021)	To understand the lived experience of spouse carers of PWP and jealous delusional phenomena.	Data collection: Interviews Qualitative Analysis: No detail	(1) Managing incredulity: trying to make sense of delusion content; (2) Hypervigilance: constant alertness to bizarre and threatening discourse and behaviour; (3) Defensive strategizing: anticipating delusions and potential consequences; (4) Concealing and exposing: ambivalence about disclosing the effect of delusions yet wanting support.
Haberman n (2000)	To broaden the discourse on the spousal experience of PD. The aim was to explore the challenges faced by middle aged spouses and the coping strategies used by these spouses.	Data collection: Interviews *Qualitative Analysis:* interpretative phenomenological analysis	(1) Challenges experienced, (2) coping strategies.
McKeown et al. (2020)	To understand the experiences of carers who were confronted by the development of impulsive and compulsive behaviours.	Data collection: Interviews Qualitative Analysis: Thematic analysis	(1) realisation—developing awareness of ICB symptoms and their causes; (2) reacting—confronting and attempts to manage ICBs; (3) reaching out—help-seeking and selective disclosure; (4) reframing—shifting perspectives on ICBs over time; and (5) resignation—impact on relationships and facing the future.

Whitehead (2010)	To explore the perspectives of individuals with Parkinson's disease and their spouses on their experiences of living with communication difficulties as a consequence of the disease.	Data collection: Interviews Qualitative Analysis: Thematic analysis	(1) individuals' perceived change in self, misperception by others, and coping strategies; and (2) spouses' experiences, including communication changes in partner, the impact on the spouse, other people's reactions, and speech and language therapy.
Mach et al. (2019)	To explore the impact of communication disorders on family members of people with Parkinson's disease (PD) through the lens of third-party disability.	Data collection: Interviews *Qualitative Analysis:* Phenomenological approach	(1) What? (2) It brings up things that have changed.
Wootton et al. (2019)	To provide an initial overview of patient & spousal experiences of living with an acquired nonverbal expressive impairment in Parkinson's disease.	*Data collection:* Semi structured one to one interview *Qualitative Analysis:* Thematic content analysis	(1) Misidentification of masking as negative affect, (2) poor symptom recognition, (3) unmet health resource needs.
Hodgson et al. (2004)	To conduct a qualitative investigation into the impact of PD on the couple relationship.	*Data collection:* interviews (couples together) *Qualitative Analysis:* Phenomenological data analysis	(1) Relationship and disease history, (2) impact on the couple relationship, (3) impact on self and others, (4) connecting with resources, (5) strategies for survival.
Constant et al. (2022)	To explore the experience of couples with advanced PD	*Data collection:* Individual interviews *Qualitative analysis:* Dyadic interpretative phenomenological analysis	(1) A closeness that separates, (2) the adversity is not unbearable, but going it alone would be, (3) be prepared for anything, and facing an uncertain future.

Table 4

Participant characteristics

Paper	Participants	Relationship detail and duration	Parkinson's duration and stage
Martin (2016)	Sample size: 44 *Who:* 21 PWP, 23 partners Age PWP: 41-89 *Sex PWP:* 13 males, 8 females Age partners: 38-84 *Sex partners:* 15 females, 8 males *Ethnicity:* 42 White, 2 African American. Employment status: No detail Setting: USA	*Relationship status:* All participants were married, except for one dyad who identified as cohabiting, unmarried partners Relationship duration: 3 to 64 years	*Parkinson's duration:* less than 1 year to 26 years *Parkinson's stage:* Hoehn and Yahr's (1967) scale: 8 PWP Stage 1; 3 PWP Stage 2; 5 PWP Stage 3; 5 PWP Stage 4
Haahr et al. (2013)	Sample size: 9 *Who:* Spouses of PWP *Age:* 27-61 *Sex:* 3 males, 6 females *Ethnicity: "*participants were all the same cultural background" *Employment status:* Three spouses were working at the time of DBS, six were retired. Setting: Denmark	Relationship status: Married Relationship duration: No detail	Parkinson's duration: No detail Parkinson's stage: No detail
Vatter et al. (2018)	Sample size: 12 *Who:* Spouses of PWP *Age:* 63-78 *Sex:* All female *Ethnicity:* White British Employment status: No detail *Setting:* UK (United Kingdom)	*Relationship status:* all but one interviewee was married, and all couples lived together Relationship duration: No detail provided	Parkinson's duration: No detail *Parkinson's stage:* Four of the participants' partners had PD-MCI, five had PDD and three had DLB
Birgersona & Edberg (2004)	Sample size: 12 *Who:* 6 couples Age PWP: 61-82 *Sex PWP:* 2 males, 4 females Age partners: 66-76 *Sex partners:* 4 males, 2 females	Relationship status/duration: all married Relationship duration: 35-50 years	Parkinson's duration: 3-30 years Parkinson's stage: No detail

	Ethnicity: No detail Employment status: No detail Setting: Sweden		
Williamson et al. (2008)	Sample size: 10 *Who:* Wives of PWP *Age:* 63-79 *Sex:* female *Ethnicity:* White, British Employment status: No detail Setting: UK	Relationship status: Married Relationship duration: 16-58 years	Parkinson's duration: 3-24 years *Parkinson's stage:* Experiencing psychotic symptoms
Fleming et al. (2004)	Sample size: 19 Who: PWP *Age:* 34-56 *Sex:* Females Ethnicity: No detail *Employment status:* 16 employed; 3 retired Setting: UK	*Relationship status:* 14 stable heterosexual relationships, 4 separated. *Relationship duration:* 14 in stable relationships, 4 separated	Parkinson's duration: 18 months-27 years Parkinson's stage: No detail
Lawson et al. (2018)	Sample size: 36 *Who:* 18 PWP, 18 carers Age PWP: 62-87 *Sex PWP:* 14 males, 4 females Age partners: 52-77 *Sex partners:* 15 females, 3 males Ethnicity: No detail *Employment status:* Of PWP – 17 were retired, one was semi-retired. Of partners – 6 were still working, 12 were retired. Setting: UK	*Relationship status:* mostly spousal dyads with one adult child/parent dyad, one friend dyad. Relationship duration: No detail	Parkinson's duration: No detail *Parkinson's stage:* 6 Parkinson's without cognitive impairment; 6 Parkinson's and mild cognitive impairment, 6 Parkinson's dementia
Thomson et al. (2020)	Sample size: 22 *Who:* PWP & care partner Age PWP: 45-73 *Sex PWP:* 7 males, 4 females Age partners: 51-69 *Sex partners:* 2 males, 9 females Ethnicity: No detail *Employment status:* PWP - Six were employed and five	*Relationship status:* spouses (n = 9), parents (n = 1), and children (n = 1) Relationship duration: 29 to 51 years	Parkinson's duration: 3 to 12 years Parkinson's stage: Receiving DBS

	had retired. Partners – seven working 4 retired *Setting:* Australia		
Sánchez-Guzmán et al. (2022)	Sample size: 80 *Who: 40* PWP and 40 spouses' *Age PWP:* No detail *Sex PWP:* 3 female, 17 male Age partners: No detail *Sex partners:* 17 females, 3 males Ethnicity: No detail *Employment status:* PWP 19 retired, 21 employed, Partners – 12 retired, 28 working. Setting: Mexico	Relationship status: Marital Relationship duration: No detail	Parkinson's duration: 4-20 years Parkinson's stage: No detail
Smith & Shaw (2017)	Sample size: 9 *Who:* 4 PWP, 5 partners Age PWP: 67-75 *Sex PWP:* 2 males, 2 females Age partners: 67-74 *Sex partners:* 2 males, 3 females Ethnicity: No detail Employment status: No detail Setting: UK	Relationship status: No detail Relationship duration: No detail	Parkinson's duration: 2-21 Parkinson's stage: No detail
Roger & Medved (2010)	Sample size: 6 *Who:* 4 PWP, 4 care partners *Age:* 40-80 *Sex:* No detail provided Ethnicity: No detail Employment status: No detail Setting: Canada	*Relationship status:* three spousal couples and one male/female sibling pair Relationship duration: No detail	Parkinson's duration: No detail Parkinson's stage: No detail
Deutsch et al. (2021)	Sample size: 12 *Who:* Spouses of PWP *Age:* 66-80 *Sex:* 4 males, 8 females Ethnicity: No detail Employment status: No detail Setting: Canada	Relationship status: No detail Relationship duration: No detail	Parkinson's duration: 3–24 years Parkinson's stage: No detail

Habermann (2000)	Sample size: 8 *Who:* Spouses of PWP *Age:* 54-58 *Sex:* 3 males, 5 females Ethnicity: Caucasian *Employment status:* PWP – five employed, three not working; Partners - seven employed, one not working Setting: USA	Relationship status: Married Relationship duration: 4 months- 30 years	Parkinson's duration: 1-16 years *Parkinson's* stage: two were stage 1; one was stage 2; and five were stage 3
McKeown et al. (2020)	Sample size: 13 *Who:* Spouses of PWP *Age:* 52–65 *Sex:* 12 females, 1 male Ethnicity: No detail Employment status: No detail Setting: UK	Relationship status: Married Relationship duration: No detail	Parkinson's duration: 5–12 years *Parkinson's stage:* Experiencing impulsive and compulsive behaviours
Whitehead (2010)	Sample size: 7 *Who:* 4 PWP's, 3 spouses of PWP Age PWP: m=64.29 *Sex PWP:* 1 female, 3 males Age partners: mean 61.03 *Sex partners:* 3 females, 1 male Ethnicity: White Employment status: No detail Setting: UK	Relationship status: Married Relationship duration: No detail	Parkinson's duration: over 5 years *Parkinson's stage*: all were receiving dopaminergic medication for PD
Mach et al. (2019)	Sample size: 9 *Who:* Carers of PWP *Age:* 64-81 years *Sex:* 8 females, 1 male Ethnicity: White *Employment status:* 5 retired; 4 working. Setting: USA	*Relationship status:* 7 spouses, 1 long-term domestic partners, 1 adult child carer Relationship duration: No detail	Parkinson's duration: 5-16 years Parkinson's stage: No detail
Wootton et al. (2019)	Sample size: 18 *Who:* 9 PWP, 9 romantic partners of PWP *Age PWP:* No detail provided *Sex PWP: 8* females, 1 male Age partners: No detail *Sex PWP:* 1 male, 8 females	Relationship status: Most married Relationship duration: 4-45 years	Parkinson's duration: Less than 10 years *Parkinson's* stage: Most were independent in activities of daily living

	Ethnicity: little variation in the ethnicity of the sample cohort, with all participants reporting themselves of New Zealand or Australian European descent *Employment status:* PWP - Three were employed; six not working, Partners – no detail *Setting:* New Zealand		
Hodgson et al. (2004)	Sample size: 20 *Who:* 10 PWP's, 10 partners *Age PWP:* 46-79 year (m=61.8) *Sex PWP:* 4 female, 6 males *Age partners:* 52-79 (m=62.4) *Sex partners:* 6 females, 4 males *Ethnicity:* All Caucasian *Employment status:* 40% of couples were retired. 60% partner still working. Setting: USA	*Relationship status:* 9 spouses, 1 cohabiting couple *Relationship duration:* lived together for 8.5-58 years	PD duration: 2-20 years *PD stage:* Hoen & Yahr – two late stage 2, six stage 3, two stage 4.
Constant et al. (2022)	Sample size: 30 *Who:* 15 PWP, 15 partners *Age PWP:* m=65.27, r= 55-79 *Sex PWP:* 4 female, 11 males *Age partners:* m= 65.27, r=54-71 *Sex partners:* 11 females, 4 males Ethnicity: No detail Employment status: No detail Setting: France	Relationship status: No detail Relationship duration: 20-56 years	*PD duration:* m=12.6 years r = 6-22 years *PD stage*: advanced stage according to assessment via Hoen & Yahr

Table 5

Quality Appraisal Ratings

	Paper					
CASP Question	Martin (2016)	Haahr et al. (2013)	Vatter et al (2018)	Birgerssona & Edberg (2004)	Williamson et al. (2008)	Fleming et al. (2004)
Was there a clear statement of the aims of the research?	Yes	Yes	Yes	Yes	Yes	Yes
Is a qualitative methodology appropriate?	Yes	Yes	Yes	Yes	Yes	Yes
Was the research design appropriate to address the aims of the research?	2	3	2	3	3	3
Was the recruitment strategy appropriate to the aims of the research?	3	3	2	3	3	3
Was the data collected in a way that addressed the research issue?	3	3	2	3	3	3
Has the relationship between researcher and participants been adequately considered?	2	2	3	1	2	2
Have ethical issues been taken into consideration?	3	2	2	2	3	3
Was the data analysis sufficiently rigorous?	3	3	3	3	3	3
Is there a clear statement of findings?	3	3	3	3	3	3
How valuable is the research?	3	2	2	3	3	3
Total score (/24)	22	22	19	21	23	23

	Paper					
CASP Question	Lawson et al. (2018)	Thomson et al. (2020)	Sánchez-Guzmán et al. (2022)	Smith & Shaw (2017)	Roger & Medved (2010)	Deutsch et al. (2021)
Was there a clear statement of the aims of the research?	Yes	Yes	Yes	Yes	Yes	Yes
Is a qualitative methodology appropriate?	Yes	Yes	No	Yes	Yes	Yes
Was the research design appropriate to address the aims of the research?	3	3	1	3	3	3
Was the recruitment strategy appropriate to the aims of the research?	3	3	3	2	3	3
Was the data collected in a way that addressed the research issue?	3	3	1	3	3	3
Has the relationship between researcher and participants been adequately considered?	3	2	1	2	2	3
Have ethical issues been taken into consideration?	3	2	3	2	3	3
Was the data analysis sufficiently rigorous?	3	3	3	3	3	3
Is there a clear statement of findings?	3	3	3	3	3	3
How valuable is the research?	3	3	3	3	3	3
Total score (/24)	24	22	18	21	23	24

	Paper						
CASP Question	Habermann (2000)	McKeown et al. (2020)	Whitehead (2010)	Mach et al. (2019)	Wooton et al. (2019)	Hodgson et al. (2004)	Constant et al. (2022)
Was there a clear statement of the aims of the research?	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Is a qualitative methodology appropriate?	Yes	Yes	Yes	Yes	Yes	Yes	Yes
Was the research design appropriate to address the aims of the research?	3	3	3	3	3	3	3
Was the recruitment strategy appropriate to the aims of the research?	2	3	3	3	3	3	3
Was the data collected in a way that addressed the research issue?	3	3	3	3	3	3	3
Has the relationship between researcher and participants been adequately considered?	2	2	2	3	2	3	3
Have ethical issues been taken into consideration?	3	3	3	3	3	3	3
Was the data analysis sufficiently rigorous?	3	2	3	3	3	3	3
Is there a clear statement of findings?	3	3	3	3	3	3	3
How valuable is the research?	2	3	3	3	3	3	3
Total score (/24)	21	22	23	24	23	24	24

Appendices

Appendix 1-A: Example of data extraction (Analysis 1: Martin, 2016)

Participant quotes (First order data)	Primary author interpretations (Second order data)	Key theme/concept
"No, [our closeness] hasn't changed. We've known each other for a long time. We've been married 44 years. We dated in high school and actually go back to [name omitted] nursery school. Our relationship is pretty solid."	those who said there has been no change, many participants cited their long and stable history with their partner as the reason that PD has not affected their relational closeness P.233	PD did not affect the relationship
"I would say [since my diagnosis] it's gotten even better. He's just been great with everything. It doesn't affect our sex life in any way. He does the dishes, he cooks, he cleans."	coping with the challenges of the disease has made them feel closer to their partner P.223	PD increases the sense of closeness in relationships
"I'm more involved with her daily activities. . . . Yeah, there's more collusion there. We work together and do stuff now. . . . It's spooky sometimes: She'll finish my sentences before I even start one. We've just never been happier. It's a lot more work on my part, and she knows it."	Support provision from the partner to the PWP. P.233 a shift in how much attention is focused on one another. P.234	
"It kind of brings you closer together because she's helping me out a lot."		
"I think we are probably closer. It's a protective instinct in me. So to make sure he has good food and this and that."		
"Well, I guess maybe we're less close because we can't do everything together. We always did everything together, and now we can't."	not being able to participate in leisure activities together p234	PD can result in diminished closeness
Well, I think it's just that, the changes that are happening [from PD] are stressful on our relationship sometimes. He has all these health problems from Parkinson's [so] he can't sleep in bed. We are on different floors. . . . He sleeps in a recliner. I sleep in the bedroom. It's just kind of like, sometimes, it's like we are roommates.	Managing PD in a dyadic context can lead to shifts in relational roles. p235 sleeping in the same bed becomes impractical for couples because of the symptoms of PD p235	Changes in relational role/identity
"One thing that is difficult is sex. Because he can't turn. He can't roll over. We get in bed [but] if you want to turn over, you can't hardly get over to that side. Physically, it's very difficult."	A shift, at times, from a romantic partnership to a roommate-type relationship p.235	

	Move to a caregiver–patient dyad. P.235 Erectile dysfunction (through PD and meds). P237 changes in sexual intimacy p237	
"At this point, I don't consider myself a caretaker, no. I think he does as much for me as I do for him." They need to rephrase all that. It made me feel bad because I feel like I'm putting such a burden on my husband. [We watched the video together] and it makes you feel bad because I don't want him to be a caretaker. I want him to help me when I need it, but—When he asks me to do something for him, I do it. Is that not being a caretaker? I want to know where they get that [label of "caregiver"] from, and why they have to use that.	Labelling the partner as a caregiver was inaccurate p236 both members of the couple give care to, and receive care from, one another. P236 The caregiver label may go beyond seeming inaccurate or misleading to inducing hurt feelings p236. The term caregiver does not seem to have negative implications because it reinforces a relational identity that they do not object to. P236	Interactions with health care professionals
The biggest reason she works is for the insurance. Otherwise, I'd be uninsurable, and with all this Obama stuff, it's scary for us right now. So she works to keep that— the prescriptions and all that stuff. Financially, right now, our lives are stressful.	Financial implications of PD as the biggest source of stress on their relationship. P238 Primary reason that at least one member of the couple continues to have a full-time job rather than retiring. P238	Challenges - financial
"Yeah, [my wife and I] like to travel and stuff, and that's kind of been put to the wayside. I can't drive distances or anything like that anymore." It's changed travel. See, some days you have a really good day, and some days you don't have a really good day, and you can't predict which day it's going to be. You might take the train to [a nearby city] and have a great time, or you might get up there and can barely walk. "We used to go to the mall and walk around, and he can't do that any longer. It's just very limited. There's just not much you can do, really."	"although they used to enjoy traveling as a couple, it has become increasingly difficult to do so because of the manifestations of PD" p.239 "tremor makes it difficult to drive" p.239 "traveling hard to plan" p.239 "difficulty with walking as a symptom of PD that can be especially limiting in terms of	Challenges – Practical challenges

	being able to continue the activities they once enjoyed as a couple" p.239	
We always played cards. . . . And [our friends] play games all the time. I mean, it's people's houses, you play whatever. He can't hold the cards. . . . So then you're not going to play cards. That makes it hard, because then your circle of friends—you can't do much in the evening because that's what people do. . . . Usually the routine is you go out to eat and you come back to somebody's house and you play cards. Now you can't do that.	"PD can also restrict other joint leisure activities that couples previously enjoyed on a regular basis" p239 "going out to eat"P.239 "Socialising jointly with friends". P.240 "a consequence of PD is the loss of valued social activities, which can, in turn, isolate them from others in their social network."	Challenges – Reduced leisure and social activities
She has a little problem thinking that [because] she's got Parkinson's, I don't want to be around her. I tell her, "Bullshit. If I didn't want to be around you— when I come home every night—if I get off work at 5:00, I'm home by 5:05. I could always say, 'Oh, I'm stressed out. I need to go have a beer.' I don't. I come home." "I can't do this anymore if you don't change what you're doing." . . . He started gambling a few years back, and started lying about things to hide things. And what it was, was that the medication he was on was heightening the gambling, the OCD	making the PWP feel less secure p234 Uncertainty about their relationship for either the PWP, the partner, or both. P.240 PWP... uncertain about whether the partner would stay in the relationship after the diagnosis. P.240	Challenges – uncertainty about relationships

Appendix 1-B: Development of themes

Key themes/concepts	Conceptual cluster	Broad themes (3rd order constructs)	Relevant papers
Reassignment of household duties, role change, partner to caregiver, relational impact of changes to roles and responsibilities, values influence perceptions of togetherness, financial difficulties increase strain	Shifts in relational roles.	Disruption of couple roles and responsibilities and attempting to maintain the relationship	Martin, 2016 Vatter et al., 2018 Fleming et al., 2004 Hodgson et al., 2004 Constant et al., 2022 Birgerssona & Edberg, 2004
Positive impact of diagnosis and changes to roles and responsibilities, role change unquestioned by partners, disease experience was shared, an evolution of the relationship	Contextual issues impacting adjustment to role change.		Haarh et al., 2013 Habermann, 2000 Deutsch et al., 2021 Lawson et al., 2018
Partner dissatisfaction in relationship due to role change, togetherness threatened autonomy for PWP, dislike of caregiver label, resistance to role change, stress increased with financial strain			Smith & Shaw, 2017 Roger & Medved, 2010 Mach et al., 2019
Importance of retaining personal identity. Establishing a balance between needs and support provided supported the relationship, challenges are manageable as long as sense of autonomy and enjoyable activity is retained for PWP. Making sense of experiences supports interpersonal relationships.	Maintaining the relationship		
Tension and frustration due to difficulties with verbal and emotional communication strained relationships, PD can result in diminished closeness, frustration, tension, sense of criticism and rejection, loneliness. Change to relationship from change in social activities, leisure activities, and intimacy.	Communication and closeness	Challenges to communication and closeness: increased resilience	Birgerssona & Edberg, 2004 Whitehead, 2010 Mach et al., 2019 Wootton et al., 2019 Martin, 2016 Habermann, 2000 Hodgson et al., 2004

Uncertainty and distance in the relationship.			Vatter et al., 2018 Fleming et al., 2004
Support or lack of support can affect emotional connection and sense of togetherness. The role of the pre-existing relationship PD increases the sense of closeness in relationships, improved communication, increased resilience	Strengthening of the relationship		Smith & Shaw, 2017 Roger & Medved, 2010 Sánchez-Guzmán et al., 2022 Lawson et al., 2018
Increased burden has a relational impact, breakdown of trust, emotional distancing, deflated resignation, lack of awareness from PWP to maintain or repair relationship, partners lack energy to maintain the relationship.	Increased burden	Increased cognitive and behavioural symptoms leading to grief, burden and isolation	Thomson et al., 2020 Vatter et al., 2018 Deutsch et al., 2021 McKeown et al., 2020
Partners had lost their source of social support, loss, grief, isolation	Loss of mutual relationship		Mach et al., 2019 Constant et al., 2022
Wish to protect partner and relationship results in lack of support, difficulties accessing respite due to need to separate. Continuing to provide love and care despite feeling alone, recalling past relationship, externalization of the problem away from PWP.	Relational methods of coping.		Hodgson et al., 2004 Lawson et al., 2018

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Contents

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Version 3.1

Date of original release: 5 December 2014	Date of current version's release: 28 September 2021

Updated to include new models for

1. FORCE11-compliant software reference entry (with version number)
2. FORCE11-compliant software reference entry (without version number)

Table of Contents

Section Two: Empirical Paper

The experience of being diagnosed with cervical dystonia: A phenomenological interpretative analysis study

Word Count (excluding references, tables and appendices): 7,233

Louise Glover

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

All correspondence should be sent to:
Louise Glover
Health Innovation Campus
Lancaster University
Sir John Fisher Drive
Lancaster,
LA1 4AT

Email : l.harris7@lancaster.ac.uk

Prepared for: Psychology and Health

Abstract

Purpose: Difficulties are commonly experienced in the journey to receiving a diagnosis of cervical dystonia. The lived experience of the difficulties experienced by people diagnosed with cervical dystonia are not well understood. This study aims to explore the experience of seeking and receiving a diagnosis in CD.

Method: This qualitative study uses interpretative phenomenological analysis (IPA) methodology. Participants were recruited from a specialist movement disorder clinic in the UK. Semi-structured interviews were conducted with a sample of six individuals diagnosed with cervical dystonia.

Results: Analysis produced three themes: 1) Losing control over one's body and attempts to regain control: 'You don't know what's going on and your whole life is kinda falling apart'; 2) Feeling powerless in response to the health system: 'I had this change and I couldn't do anything about it, and nobody seemed to have any kind of solution'; 3) Mixed feelings upon diagnosis: 'It's kinda like a relief, but scary at the same time'.

Conclusion: The findings highlight how the pre-diagnosis phase can be a significant and distressing experience for people with cervical dystonia. This study suggests that understanding the experiences of those who have come to be diagnosed with cervical dystonia may support health professionals in effectively navigating their interactions with patients during the diagnosis and treatment phases. Implications for clinical practice and further research are discussed.

Keywords: Cervical dystonia; dystonia; diagnosis; experiences; qualitative research; IPA.

Introduction

Cervical dystonia, also known as spasmodic torticollis, is the most common type of dystonia (Defazio, 2010; Albanese et al., 2013). Dystonia is the term given to a group of neurological movement disorders characterised by uncontrollable muscle spasms which can, for some, be painful. It is the 3rd most common movement disorder diagnosis after Parkinson's and essential tremor (Steeves et al., 2012). Dystonia affects approximately 16.43 per 100,000 people worldwide; however, due to difficulties with diagnosis, the actual figure is may be far greater (Defazio, 2010; Steeves et al., 2012). Some people develop dystonia following an illness or injury but for others the cause of dystonia is unclear with both genetic and environmental factors suggested (Defazio, 2010).

Symptoms of cervical dystonia commonly include tightening of the neck and head muscles, leading to twisting of the head and jerking movements (Albanese et al., 2013). Cervical dystonia is more prevalent in females than males, and onset can occur at any age but is most common between the ages of 30 and 60 (LaHue et al., 2020; LeDoux et al., 2016; Norris et al., 2016).

CD is associated with several difficulties, each impacting considerably upon the quality of life of those affected, such as pain, physical impairment, anxiety, depression, experiences and perceptions of stigma and sleep disturbance (Tomic et al., 2016). Psychological difficulties are particularly prominent in CD as between 30 and 40 per cent of those affected experience clinically significant levels of depression and anxiety, and 32 per cent report suicidal behaviour (Medina Escobar et al., 2021a; Medina Escobar et al., 2021b; Worthley & Simonyan, 2021).

There are several available treatments which can improve symptoms which include oral medications, botulinum toxin injections, deep brain stimulation (DBS), and selective peripheral denervation surgery (Adam & Jankovic, 2007; Blahak et al., 2021; Chou et al.,

2011; Girach et al., 2019; Mordin et al., 2014). Benefits from physiotherapeutic interventions and psychosocial interventions, such as behavioural and relaxation interventions, include improved physical symptoms, increased treatment acceptance, reduced anxiety, reduction in perceived stigma and improved quality of life (Bernstein et al., 2016;; Jinnah & Factor, 2015; Useros-Olmo et al., 2020).

However, CD is a highly heterogenous and relatively rare condition that relies on clinical assessment for diagnosis due to a lack of diagnostic tests and biomarkers (Bertram & Williams, 2016; Supnet et al., 2020). As a consequence, many face difficulties in obtaining a diagnosis, commonly experiencing multiple assessments, incorrect diagnoses (90 per cent) and delays between one and seven or more years (Bertram & Williams, 2016; Jinnah & Factor, 2015; Jog et al., 2011; LaHue et al., 2020; Macerollo et al., 2015; Steeves et al., 2012 Tiderington et al., 2013). Difficulties in obtaining a diagnosis mean many people with the condition live and cope with its painful and limiting symptoms without adequate treatment for a considerable time.

Research suggests that adverse diagnosis experiences, such as delays or incorrect diagnosis, can contribute to difficulties in adjusting to illness. Adjustment is the process of adapting to and making sense of a condition to gain control and live well (Lazarus & Folkman, 1984; Taylor & Aspinwall, 1996). The Self-Regulation Model (SRM; Leventhal et al., 1984) suggests that maintaining or gaining a new sense of self-identity is a central agent to successful adjustment. Research in multiple sclerosis and Parkinson's highlights that receiving a diagnosis can influence one’s sense of self, which may impact adjustment, according to the SRM (Leventhal et al., 1984; Strickland et al., 2017; Warren et al., 2016). In rare cancers, where the diagnosis is often delayed and involves multiple referrals or visits to a GP, patients are more likely to lack trust in health professionals (Larsen et al., 2011; Mendonca et al., 2016). Trust in healthcare professionals is associated with fewer symptoms

and constructive health behaviours, such as treatment adherence (Birkhäuer et al., 2017). Furthermore, research in motor neuron disease suggests that a problematic pre-diagnostic phase increases uncertainty which may lead to anxiety and depression, which in turn can negatively impact adjustment to illness (Remm et al., 2019; de Ridder et al., 2008). Overall, it appears that adjustment to chronic illness depends on a complex interplay of bio-psycho-social factors and highlights the need to understand the nuanced challenges faced in different conditions to consider support needs (Moss-Morris, 2013; Walker et al., 2004).

In dystonia, more generally, quantitative research has highlighted difficulties with a diagnostic delay, such as unnecessary healthcare costs for inappropriate testing and treatment, unpleasant side effects of unnecessary treatments, worry, anxiety, impact on emotional wellbeing, relationship problems and negative impact on work and recreational activities (Bertram & Williams, 2016; Camargos & Cardoso, 2016; Jinnah & Factor, 2015; LaHue et al., 2020). While highlighting these issues is essential, this research is limited in being able to explore the complex individual impact of such experiences, which is better addressed through qualitative research. One previous qualitative study aimed to understand how people experience living with dystonia (Morgan et al., 2019). A theme emerged about the experience of diagnosis representing a source of extreme distress. It was apparent that none of the participants involved felt they were able to access formal support due to the barriers that emerged during their journey to diagnosis, such as fearing that their physical symptoms might be dismissed as related to this psychological distress (Morgan et al., 2019). However, this study did not aim to understand the experience of diagnosisand , it includes individuals with different types of dystonia and variances in the time since diagnosis, which affected the strength of findings. Therefore, further targeted research is required to adequately understand the first-hand experience of the journey to diagnosis in dystonia in order to better understand the issues that are encountered and to elucidate understandings of how individuals may be

better supported during and following this experience. As CD is the most common diagnosis within the umbrella of dystonia (Albanese et al., 2013), further research is warranted to understand the experiences of diagnosis in CD specifically, thus supporting sample homogeneity.

Therefore, the present study seeks to understand the experience of diagnosis in CD. It uses the qualitative method of interpretative phenomenological analysis (IPA; Smith et al., 2009). IPA is concerned with uncovering the personal meaning of lived experiences and the emotional and psychological processes involved. The findings will deepen insight into this experience and, in doing so, guide future research and clinical practice with particular attention to the role of clinical psychologists. The research question underpinning this study is: ‘What is the experience of diagnosis in CD?'

Method

Design

IPA was selected as the framework to explore the diagnosis experience in cervical dystonia. IPA seeks to gain an in-depth understanding of specific experiences via interviews with fewer participants than other approaches, such as thematic analysis (TA, Braun & Clarke, 2021). IPA aims to explore the in-depth psychological processes and meaning-making related to experiences and to uphold the richness of individual accounts rather than obtaining a broader view of the phenomenon in question, as may be expected in TA (Braun & Clarke, 2021; Smith & Osborn, 2008; Smith et al., 2009). IPA's epistemology is rooted in hermeneutics and phenomenology (Smith et al., 2009). As such, IPA is a helpful approach when applied to healthcare as it is an approach that enables the development of sensitive and reflective interpretative accounts, which can enrich understanding and, importantly, highlight support needs (Peat et al., 2019).

Furthermore, the double hermeneutic process, central to IPA, ensures that interpretations relate to the data whilst recognising the contribution and influence of the researcher's own experiences (Smith & Osborn, 2008; Smith et al., 2009). As such, IPA is a rigorous qualitative method that provides a robust framework to guide the research process during sampling, data collection and analysis, which can be helpfully applied to under-researched areas such as dystonia (Peat et al., 2019).

With the aim to uphold and highlight unique and individual stories, IPA seeks to include a small number (e.g., 6-10) of homogenous individuals for whom the research question is salient and significant (Murray & Wild, 2020; Smith et al., 2009). However, Murray and Wilde (2020) highlight that the critical nature of homogeneity is in the experience in question. Hence, it is possible to include individuals who differ demographically, for example, by gender or age. The participants in this study were approached with this in mind, owing to the relatively small population in question. The inclusion and exclusion criteria were applied to achieve an appropriately homogenous group.

Semi-structured interviews were the most appropriate choice for this study using an interview topic guide (see Appendix 2-A) which was informed by existing theory and issues raised through quantitative and qualitative research into dystonia and related conditions (Bertram & Williams, 2016; Camargos & Cardoso, 2016; Jinnah & Factor, 2015; LaHue et al., 2020 Larsen et al., 2011; Mendonca et al., 2016; Morgan, et al., 2019; Remm et al., 2019; Strickland et al., 2017; Warren et al., 2016). Research materials, including the interview topic guide, were shared with an expert by experience to improve accessibility.

Participants

Participants were required to have a confirmed diagnosis of idiopathic adult-onset isolated CD (Albanese et al., 2013). The diagnosis must have been received within the previous three years for twofold reasons: 1) to obtain a homogenous sample of participants

and 2) to ensure accurate recall of events (Moscovitch et al., 2016). Furthermore, the diagnosis must have been received more than six months prior to participation to ensure some level of adjustment had taken place in order to limit distress. Participants were required to be between age 35 and 75. The age range was decided upon with reference to the literature about common age of onset in CD (Norris et al., 2016), combined with observations of patients presenting in clinical practice. This limit on age range meant that the participants were likely to be at an adult stage of their life with work and/or social commitments when first experiencing symptoms and during the help-seeking phases. Participants were required to speak English fluently due to a lack of resources to fund interpreters. Finally, participants were excluded from the study if they had any additional physical or mental health difficulties that may have significantly altered their diagnosis experience. For example, another movement disorder diagnosis, cognitive impairment, or a significant level of psychological distress.

Seven people who met the study inclusion criteria agreed to be contacted by the researcher. Six people, two male and four females, took part in the study with an age range of 46-73. Participants were all diagnosed within three years of taking part. However, time since having first noticed symptoms varied between one and 20 years. Participants' characteristics are presented in Table 1. Participant names provided are pseudonyms to protect anonymity.

[Table 1 about here]

Recruitment

Recruitment was conducted in collaboration with an NHS specialist centre for neurosciences, allowing for a sample with a confirmed diagnosis. In the first instance, potential participants were identified and approached by their clinician from a specialist movement disorder clinic who provided the study information pack. Either consent was

provided for the researcher to contact the potential participants, or potential participants contacted the researcher directly.

Ethics

Ethical approval for this study was received from the NHS Health Research Authority following review from the South Central Oxford-C Research Ethics Committee (REC reference: 21/SC/0065). Following ethical approval, further approval was obtained from the relevant NHS trust where recruitment took place. Each participant provided informed verbal recorded consent, which was recorded prior to the interview. All participants were informed in writing and verbally about their rights to withdraw. Participants received details of relevant sources of support and information. Participation was anonymous, and all names and identifiers were changed or removed.

Data collection and analysis

Semi-structured interviews lasting between 60-90 minutes took place remotely. The participants chose the interview mode; five took place by telephone and one by video call. The researcher recorded interviews via a digital recording device and transcribed them verbatim. Analysis followed the steps advised by both Murray and Wild (2020) and Smith et al. (2009), with each transcript analysed before moving on to the next to aid the process of setting aside one's pre-conceptions as far as possible. Firstly, the researcher listened to the interview recording repeatedly to aid familiarity, and initial codes were applied. Initial codes were refined before being printed so they could be physically moved into related clusters. Grouping the refined themes was an iterative process which led to more explicit understandings and sensemaking. Then, interpretative summaries were written for each theme, and individual titles provided before allocating supportive quotes derived from the original transcript. This process was repeated for each transcript individually before the process of comparing and contrasting the refined themes, and interpretative summaries from

across participants began. This was a further iterative process whereby clustered themes were deconstructed and re-constructed in light of new information from each participant's analysis. Finally, this led to merging analysis across transcripts to produce new interpretations and a coherent narrative whilst highlighting and retaining the unique perspectives gathered.

IPA's double hermeneutic perspective accepts and embraces the inevitable interaction between the researcher's experiences and the interpretations made throughout the analysis (Smith & Osborn, 2008; Smith et al., 2009). However, this must be balanced with the assurance of sufficient quality. Thus, researchers must seek to avoid bias by ensuring awareness of one's pre-conceptions and attempting to put them aside during interviews and analysis (Murray & Wild, 2020). The researcher kept a reflective journal throughout the interview and analysis process to aid awareness of the influence of bias on their interpretations. The researcher engaged in reflective supervision throughout the research process, and experienced researchers provided feedback on the interview, initial coding, and grouping of themes and interpretations. In order to ensure that the research was of sufficient quality, principles outlined by Yardley (2000) were applied, which were: 1) sensitivity to context, 2) commitment and rigour, 3) transparency and coherence, and 4) impact and importance. The participants' voices are present throughout the findings to illustrate that the interpretations are rooted in the original data. An analysis audit trail is available in Appendix 2-B, 2-C & 2-D.

Personal Statement

The researcher had no prior personal experience of dystonia, although they held some familiarity with the evidence base surrounding this issue. To aid understanding and ensure that the research was relevant to clinical and personal experience, the researcher worked closely with an expert by experience and a professional expert from the clinical field. The researcher held a dual role of researcher and clinician as the research was conducted as part

of the clinical psychology doctorate training programme. Helpfully, the dual role of clinician and researcher allowed for comfortable exploration of emotive topics. However, psychological and emotional content likely stood out to the researcher over and above other issues upon viewing and interpreting data.

Findings

The analysis produced three themes: 1) Losing control over one's body and attempts to regain control: 'You don't know what's going on and your whole life is kind of falling apart'; 2) Feeling powerless in response to the health system: 'I had this change and I couldn't do anything about it, and nobody seemed to have any kind of solution'; 3) Mixed feelings upon diagnosis: 'It's kinda like a relief, but scary at the same time'. Each theme will now be explored alongside supportive quotes from each participant.

Theme 1: Losing control over one's body and attempts to regain control: 'You don't know what's going on and your whole life is kind of falling apart'

This theme explores participants' experiences of dystonia symptoms throughout their journey to diagnosis as they experienced a loss of control of their bodies. The symptoms experienced led to difficulties in different areas of life. In the absence of explanation or treatment, participants took matters into their own hands to try to reduce or cope with their symptoms. However, such efforts to control or hide what was happening to their bodies led some to develop further difficulties.

All participants spoke of a process of first noticing symptoms and experiences of trying to make sense of what was causing them. Nigel described noticing a sudden change in his body which caused him to panic. Similarly, Jane and Diana's initial experiences of involuntary head movements were hard to ignore; Diana's head movements were painful, and Jane experienced an unusual head tremor and the sensation that her head was too heavy for her shoulders. Diana proclaimed: 'it (my head) would just turn to the side, and it wouldn't

move’; highlighting her frustration with the lack of control over her body. Sara and Andrea experienced a more insidious onset whereby they initially experienced mild symptoms they could ignore at first, such as neck and shoulder pain. However, these mild symptoms gradually developed into noticeable alarming symptoms, such as the head moving involuntarily, which understandably increased their concerns. David suggested that it was not him but his wife who noticed that his head began to move to the side while using his phone. At first, this was subtle before gradually increasing in frequency and intensity.

Seemingly trying to gain control over what was happening, all participants described a process of trying to make sense of what was causing their bodies to behave in this strange way. Jane and Andrea both suspected that their symptoms were caused by an emerging health condition and suspected Parkinson’s. This caused Andrea to worry about the impact on her future:

> You work all your life for these things, and you hear so many tales of people who retire and are stuck in the house and things like that. It was quite scary actually. (Andrea)

Whereas Diana, Sara, David and Nigel described 'brushing off' their symptoms, attributing them to common problems, such as a trapped nerve, pulled muscle, tiredness or stress. Sara described using self-help techniques such as massage, and Nigel, similarly, described trying to resolve the physical symptoms alone:

> I thought it's something, you know, I couldn't understand what was going on and I just yeah, I just thought it was for myself to resolve as opposed to reaching out to others. And I didn't want to worry anyone to be brutally honest, especially in the family. Certainly, didn't want the girls or the you know, my wife to be, you know, to be upset. (Nigel)

Nigel’s words provide an insight into what was common among four participants: a sense of symptoms signalling a problem in themselves that they felt they had to hide. Nigel described his symptoms as a ‘sign of weakness’, an appraisal which further bolstered his strategy of hiding symptoms and coping alone. This method of coping may have been self-protective, as putting off help-seeking meant that Nigel could delay the acknowledgement of a developing health problem.

Sara, who spoke of being a single parent, felt she had to try and hide her symptoms and carry on as normal, which she did until she ‘couldn't keep it together and act normal’ any longer. Whilst, David described the use of laborious strategies of holding his hand on his neck to correct the head tilt. Similarly, Jane described physically compensating for the physical disablement in attempts to appear ‘normal’.

In minimising and hiding symptoms, participants gained some control over what was happening to their bodies until such a point that they could no longer be ignored.

As the frequency and intensity of symptoms increased, they began to take on a life of their own as they began to impact participants' daily lives and those around them. Nigel and David experienced their symptoms becoming more visible, which prompted their family members to become concerned. David's family worried that his symptoms were caused by the same condition that caused his father's death; a rare stroke. Jane described a process of becoming more aware of other people noticing her symptoms which resulted in a gradual loss of confidence. Similarly, Andrea began to feel embarrassed by her symptoms: ‘I was aware when people were looking at me’, which inhibited her interactions with work colleagues.

David's symptoms intensified to the point that he could no longer safely work, which led to taking time off sick and further difficulties: ‘being at home was almost spiralling me into depression’. Sara’s symptoms had a serious impact on her livelihood; she was unable to work, and her mental health was greatly affected:

> I couldn’t function, I lost my business. And I was that far in the depths of anxiety and depression. (Sara)

In order to cope with the emotional distress related to her unexplained symptoms, Sara became dependent on alcohol to ‘escape’ her situation. Having been told by health professionals that there was nothing wrong with her (see Theme 2), she and those around her struggled to understand why she could not take control of her body. As a result, friendships and her relationship with her daughter were negatively impacted. Similarly, David noticed that drinking alcohol relieved his symptoms. In the absence of effective treatment, a vicious cycle developed where he used alcohol and higher than prescribed amounts of medication (diazepam) to experience some comfort. David explained that his coping strategies worried his partner, and they began falling out. Similarly, Nigel's symptoms did not improve, and they began to affect his mood, sleep, and overall wellbeing, and he reflected on how this impacted on his close family members:

> I was becoming more and more fatigued obviously it was probably coming more and more irritable. That's probably you know that that's not fair on the family. (Nigel)

Theme 2: Feeling powerless in response to the health system: ‘I had this change and I couldn't do anything about it, and nobody seemed to have any kind of solution’

As described in Theme 1, symptoms impacted the lives of participants. Each wished for an explanation and hoped that treatment could be provided. A common theme across some participants’ narratives were the barriers they experienced during their attempts to access healthcare. These barriers induced distress and participants felt the need to take matters into their own hands. Comparatively, those who received a prompt and appropriate referral to a specialist seemed to retain confidence and trust in health professionals’ ability to help them.

The wide-reaching impact on participants' lives led to seeking help via their GP. For some participants, the experience of accessing support from a health professional seemed to increase their distress. Having coped alone for as long as she could, Diana approached her GP for help and advice. She experienced several scans and assessments to be told that nothing was wrong, despite continuing to experience difficulties:

> I just didn't know what was happening to me. Because I knew in my head that something wasn't right. But nobody helped me. (Diana)

The isolation and powerlessness that these experiences evoked made Diana question herself: 'I thought I was going crazy.' Similarly, Sara felt that she was not taken seriously, as until her GP was able to see physical signs of symptoms, she felt that she was not believed. Sara expressed the belief that her symptoms were attributed to mental distress by healthcare professionals, and as such, she felt penalised for having previously accessed support with her mental health. Not holding an explanation for her difficulties led to further problems such as anxiety and depression:

> And then there is people staring and the questions, and you don't even know what's going on yourself. And then you get into this vicious cycle then, with the anxiety, the depression and not wanting to go out. But then being trapped in the house made all of them things worse but then not wanting to go out. (Sara)

Similarly, David felt like he was not being listened to, which resulted in frustration and a sense of powerlessness in response to the GP's decision that a trapped nerve likely caused his symptoms. With no explanation and no treatment, David remained unable to work. Quickly, his mood and wellbeing were affected. To cope, he said that he used unhelpful coping strategies, such as alcohol and medication (see Theme 1), to control symptoms and escape his situation. On approaching his GP again, David seemed to receive more explanation about the

diagnostic process than he had previously. Although remaining frustrated, this was comparatively easier to bear as he was seemingly able to make sense of the need to wait:

> The second doctor wanted to do it his way. Which I don't have any problem, I understand that the problem is it is very, very difficult condition to diagnose. I know that GPs don't have the skills to diagnose somebody with cervical dystonia. So, it was probably more frustrating for my partner than it was for me. Because I stood back and said, "that's how I would diagnose a problem on a machine". I was frustrated with the process. (David)

Nigel explained that his GP seemed to be bewildered by his symptoms, which he felt hindered further investigations. Nigel felt resigned to cope alone, which extended to a sense of helplessness: 'I couldn't see a future and I could only see things getting worse.' Nigel's words highlight his powerlessness in response to the health system.

Meanwhile, Sara and Diana continued to fight for a diagnosis; at a loss of what to do, they repeatedly visited their GP, which Diana felt strained her relationships with staff members: 'the receptionists weren't nice with me either. Because I kept going in'. Meanwhile, Nigel and Diana sought to find out what might be causing their symptoms by researching and learning about dystonia through the information available on the internet. In doing so, they became experts. Consequently, Nigel felt frustrated at the continued sense of not feeling listened to whilst feeling that he knew the help that he needed:

> I was off put because I had mentioned it to the GP and I did mention it to the physiotherapist, but none of them knew anything about it. It wasn't like it wasn't the kind of response I was expecting or needed at all. (Nigel)

Whilst four participants' help-seeking experience exacerbated their distress, two found that help-seeking led to acknowledgement and validation. Interestingly, those individuals received prompt and appropriate referrals to neurology which seemed to reduce their distress.

For example, Andrea's GPs promptly referred her to a neurological specialist, providing a much-needed sense of control and certainty which was reflected in Andrea's ongoing confidence in the role of healthcare professionals: 'Yeah, just reach out to people. You have to, don't you?'. When Jane sought support, she, too, received a referral to a movement disorder specialist. Jane's experience was not one of distress; instead, the tone of her account was one of trust and faith in health professionals. For example, when she spoke of her thoughts about the future treatment she suggested, 'I just listen to the experts'.

Theme 3: Mixed feelings upon diagnosis: 'It's kinda of like a relief, but scary at the same time'

This theme captures the mixed feelings participants experienced on receiving a diagnosis of dystonia. For some, the diagnosis represented regaining control; for others, the diagnosis of CD was unfamiliar, representing a further loss of control. Across participants, receiving a diagnosis seemed to signify the end of one journey and the beginning of the next. Some participants were left with uncertainties about the next step, which seemed to be partly influenced by the experiences they had during their journey to diagnosis.

For those who experienced a difficult journey to diagnosis, when they finally saw a neurologist, there was a sense of disbelief at the speed and ease at which specialists provided a diagnosis. Sara seemed surprised that the specialist doctor looked at her for 'five minutes' and concluded that she most likely had dystonia, exclaiming: 'He diagnosed me as soon as he saw me.' Similarly, David's words expressed his shock at being diagnosed so quickly: 'It was just unbelievably quick; it can't have been more than 30 minutes.' However, Jane, who had experienced a secure and reassuring ongoing relationship with health professionals, did not express surprise. Instead, she received the information that she had hoped for and expected, an explanation for her experiences. Diana expressed anger at not having received an explanation sooner: 'Oh, I'm mad, because I think I could have got help years ago.'

Similarly, David felt that the extent of his physical impairment and the impact on his work and the subsequent impact on his financial security could have been avoided.

Meanwhile, a sense of relief was common for some participants. Nigel described feeling reassured by the specialists' expertise: 'he put me at ease straight away, and he started talking immediately about dystonia'. Diana experienced relief related to feeling that her difficulties were finally acknowledged and validated: 'I was happy because I thought – I'm not crazy. Somebody believed me.' Sara experienced relief for similar reasons: 'I'm not a hypochondriac.' Most participants suggested that their close family members were also relieved to receive the diagnosis as it meant no longer fighting for answers (Diana, David, & Nigel) and an explanation for their loved one's difficulties (Sara & Andrea). In contrast, Jane felt that her family were not affected by her symptoms or diagnosis as she worked hard not to worry others: 'We just joke about it. I try not to be too down when I go to see him [son]'. Jane's practice of making light of her symptoms seemed to help minimise the gravity of her diagnosis.

Further difficulties expressed by participants in the time that followed diagnosis were experiences of anxiety and uncertainty. Sara and Diana had not heard of dystonia before, which provoked anxiety about what dystonia meant for their future. Diana and Nigel worried if treatment would become ineffective over time or whether dystonia may progress and cause disfigurement. Nigel and Diana also expressed isolation at not knowing anyone else with the condition. Sara's concerns were twofold; she worried about the long-term side effects of her treatment and the effect that this chronic health condition would continue to have on her life:

> If I do go back to work, what happens on the days when I can't go in? What happens on the days when it's bad? (Sara)

Sara described difficulties with sharing her concerns with the specialist doctor treating her for dystonia, suggesting that this was due to worries that her physical needs may be

dismissed as being related to her mental health, as they had been throughout her journey to diagnosis (see Theme 2). Nigel reflected on having difficulties being truthful about how dystonia was affecting him during appointments with his specialist doctor, suggesting that he often downplayed the level of pain and physical impairment due to worries about the nature of future treatment (e.g. DBS).

For some participants, diagnosis provided a sense of acceptance which seemed to help them to begin to adjust to life with dystonia. For Jane, receiving the diagnosis meant being informed and, therefore, better able to cope:

> Like anything in life, an answer is better than no answer at all. Yeah, it's got to be, and you can move on from there. (Jane)

Similarly, David expressed how the certainty of diagnosis enabled coming to terms with living with a chronic health condition as it allowed him to connect with other people with the same diagnosis via social media which helped him by providing hopefulness about living well with dystonia. Andrea, who had feared that she was developing Parkinson's, felt a sense of encouragement that the diagnosis of dystonia meant she could retain her independence, which seemed to be important to her personally: 'To me, it's not the worst. It's not recognised as a disability that you would need help for or anything.' What is more, the diagnosis came hand in hand with prompt treatment, which, although it was a process of trial and error, provided many benefits. These included an improvement in emotional wellbeing (Diana, Sara, Jane), reduced pain (Nigel, Diana), reduced physical impairment (Diana, Andrea), improved sleep (David), and feeling able to return to work (David). Sara suggested that respite from her symptoms allowed her to process and begin to recover from the experiences of the previous months and years: 'Now I feel like I've been able to take a breath. To be able to get my head in order and just to be able to think.' Which also gave some insight into the lasting emotional impact of her difficult experiences of arriving at a diagnosis.

Discussion

This study explores the impact of CD symptoms on the lives of those affected during the pre-diagnosis phase and how help-seeking experiences may exacerbate distress. This study also sheds light on how experiences during the pre-diagnosis phase may affect the experience of receiving a diagnosis. The findings will now be discussed with reference to the existing literature on the subject.

An undiagnosed symptomatic phase is common in CD due to difficulties validating symptoms in conditions reliant on clinical assessment. This phase is included within 'stage 1' of the recently developed CD Patient Journey Map (CDPJM; Benson et al., 2022). The CDPJM suggests that during this phase, symptoms are recognised, and help is sought. However, help-seeking behaviour is a complex decision-making process whereby symptom appraisal and views on help-seeking may lead to different actions (Cornally & McCarthy, 2011; Hurt et al., 2019; Yousaf et al., 2015). This study evidences a variety of complex emotional and psychological processes that impacted participants' help-seeking behaviours. For example, those who attributed their symptoms to serious illness sought advice more rapidly, and those who were bemused or embarrassed by their symptoms attempted to cope alone.

Participants experienced distress related to issues such as pain, physical impairment, impact on relationships, daily activities, anxiety and depression, seemingly exacerbated by the uncertain and unexplained nature of their symptoms. The pre-diagnosis phase of chronic illness is often associated with high levels of uncertainty leading to distress (Bailey et al., 2009). Uncertainty at any stage of a chronic health condition is difficult to avoid. However, it is essential to consider how it may be managed as uncertainty about issues such as treatment and healthcare systems, and a lack of information has been linked with several difficulties

such as poorer physical and mental wellbeing and poorer coping (Etkind et al., 2017; Mishel M. H, 1988; Mullins et al., 2001; Webster & Christman, 1988).

This study highlighted the impact of participants' experiences of reaching out for help and feeling dismissed, which seemed further to erode participants' trust in health professionals. The term ‘trust’ is used here to represent the essential relationship between patient and practitioner, whereby the patient feels they are able to depend upon and have hopeful faith in their practitioner's future actions, which is influenced by actual experiences (Birkhäuer et al., 2017; Brennan et al., 2013; Gordon et al., 2006). This is evidenced by this study by contrasting experiences, whereby participants received an explanation and felt acknowledged, and seemingly helped to contain their distress and maintain trust in health care professionals. Furthermore, a sense of being believed is a central factor in enabling trust in the patient-physician relationship, which is vital to consider as decreased trust in health professionals is associated with poorer patient outcomes (Birkhäuer et al., 2017; Murphy & Salisbury, 2020). Due to the negative impact of uncertainty (Etkind et al., 2017; Mishel M. H, 1988; Mullins et al., 2001; Webster & Christman, 1988), it was unsurprising that during the pre-diagnosis phase, participants engaged in methods to regain control. For some, this looked like hiding the physical symptoms, attempting to ignore their physical health problem or relying on other substances such as alcohol to ease their symptoms, particularly when they sought help and support and did not receive the help they needed. Lazarus and Folkman (1984) suggest that feelings of uncertainty are a stressor and increase the likelihood of developing negative perceptions about one’s ability to cope with an illness. Such beliefs are referred to as illness perceptions, which are beliefs about one's illness that influence coping strategies and physical and mental health (Leventhal et al., 1984). It was, therefore, understandable that upon receiving the diagnosis, participants experienced relief and acceptance. Receiving information via diagnosis can help patients make informed decisions

and thus feel they have greater control which can lead to more positive illness perceptions (Lazarus & Folkman, 1984; Leventhal et al., 1984; Warnock et al., 2017). However, much uncertainty was also expressed related to participants' worries about how CD might affect them in the future. This was seemingly increased for those whose journey to diagnosis had been more difficult. Therefore, the findings from this study suggest that a difficult journey to diagnosis may be associated with poorer illness perceptions. This is important to consider due to illness perceptions' role in coping and their influence over psychological adjustment and outcomes in CD (O'Connor et al., 2022).

Clinical implications

As discussed, difficulties with diagnosis are common in dystonia, yet this phase is associated with significant distress. For example, information processing biases and thinking errors which are common when faced with uncertainty, are likely to be present (Tversky & Kahneman (1978). Examples illustrated within the present study include participants who had familial experience of health conditions and feared a similar diagnosis (e.g., stroke or Parkinson's). Although it is not always possible to remove uncertainty from the diagnostic pathway, particularly with rare conditions, such as dystonia, prolonged time spent within the pre-diagnostic phase should be avoided, as the emotional difficulties this provokes may negatively impact adjustment to chronic illness (de Ridder et al, 2008). As such, it would be beneficial to see an increased awareness of this poorly understood condition within primary care settings and the general population. More knowledge about CD would better equip GPs and other health professionals who encounter individuals presenting with symptoms of CD (e.g., physiotherapists) to identify neurological symptoms and thus make appropriate onward referrals.

Regarding the management of diagnostic uncertainty, it is suggested that GPs experience uncomfortable emotions, such as frustration and inadequacy, when faced with

patients whose symptoms are unexplained (Houwen et al., 2017). In order to maintain the relationship with the patient, they should aim to avoid displaying prejudices and communicate honestly regarding their uncertainty about the cause of symptoms, as transparency regarding diagnostic uncertainty is key in developing and maintaining trust between patients and health professionals (Berger et al., 2017; Houwen et al., 2017). Such practices are complex as they require self-awareness and reflexivity and often pose a challenge for health professionals (Alam et al., 2017). However, clinical psychologists (CPs) may be well placed to support GPs in reflecting upon their interactions with patients who provoke strong emotions to prevent this from impacting their care and communication towards such patients. CPs are trained to deliver supervision to support health professionals to reflect on emotional challenges within their work to enhance practice (British Psychological Society, 2014).

As highlighted by this and previous studies (Medina Escobar et al., 2021a; Medina Escobar et al., 2021b; Worthley & Simonyan, 2021), psychological distress is common in dystonia. This study adds to this by highlighting the particular needs of those with a difficult journey to diagnosis, as this may compound their distress. Therefore, upon diagnosis and subsequent treatment, it would be helpful for healthcare professionals to seek to understand the patient's experiences prior to diagnosis. This insight may help to understand and negotiate ongoing barriers to effective treatment, such as barriers to trusting healthcare professionals. Such conversations may be enough to meet the emotional and psychological needs of patients, whilst others may require further support. For example, CPs may provide support as part of the multi-disciplinary team, providing clinical supervision or indirect systemic formulations to understand and support patients' interactions with healthcare services to overcome barriers to accessing healthcare (British Psychological Society, 2014; Murphy et al., 2013).

Others may benefit from direct psychological interventions, the current evidence for which is currently sparse for dystonia. However, recent studies have reported the benefits of cognitive behavioural therapy (CBT) and mindfulness interventions (Sandhu et al., 2016; Wadon et al., 2021). In other conditions, such as Parkinson's, CBT and mindfulness interventions effectively support people with various difficulties, including anxiety and depression (Simpson et al., 2021). Furthermore, this study highlights the impact of CD symptoms in the pre-diagnosis phase that impede participants' work and relationships and provides evidence to suggest that ongoing uncertainty about the impact of CD might continue to affect patients in these areas. Appropriate psychological approaches may include metacognitive therapy, which is effective in the treatment of unhelpful thinking styles common when faced with uncertainty and has been effectively applied to other illnesses, such as cancer (Fisher et al., 2019; McEvoy, 2019). Alternatively, acceptance and commitment therapy (ACT), is a mindfulness-based approach which promotes acceptance of difficulties and the commitment to actions which support the movement towards one's values (Hayes & Smith, 2005). ACT is used effectively with individuals with long-term health conditions, with benefits such as improved quality of life, symptom control, and reduced distress (Graham et al., 2016).

Limitations

A sample of six to 10 is recommended in IPA (Murray & Wilde, 2020), and whilst this allows for individual experiences to be retained and communicated, by its nature, the findings from this small sample are not directly generalisable to the broader population of people with a CD diagnosis. Furthermore, study recruitment was conducted via partnership with a specialist movement disorder clinic. On the one hand this strengthened the sample's homogeneity and reliability as a specialist physician confirmed all diagnoses. However, the participants were recruited from the same specialist clinic. This recruitment mode excludes

individuals living in different areas of the UK who may not have access to such a service and, therefore, may have different experiences and needs.

Future research

A similar study completed with a sample of individuals who did not have access to a specialist movement disorder clinic or were perhaps inhibited from accessing such due to mobility or financial constraints may provide further insights into the experience of diagnosis in dystonia. Understanding the impact of diagnosis experience on subsequent adjustment would be advanced by longitudinal research, with interviews conducted at the time of diagnosis and then again sometime later. In addition, studies to understand diagnosis experiences in other types of dystonia may be beneficial as separate issues may be encountered that are presently ill-understood and would deepen insights currently held about the lived experience of dystonia diagnoses collectively.

The quantitative evidence base would benefit from further research to consider the impact of diagnosis experience on issues such as illness perceptions or wellbeing. Also, further studies to consider the efficacy of psychological therapies such as CBT and ACT, particularly in randomised control trials, would strengthen the evidence for the routine use of such interventions within this population.

Conclusion

This study sheds light on the experience of the journey to diagnosis in dystonia, including experiences of noticing symptoms, help-seeking with a poorly understood condition and experiences of receiving a diagnosis. Highlighted are the potential emotional and psychological needs of individuals diagnosed with CD and an appreciation that experiences may vary. For some, the experience may be significant and associated with extreme distress leading to a range of physical and psychological difficulties that are important to consider in terms of how they impact future coping. However, for others, the

experience of diagnosis may be straightforward. The findings highlight how understanding the experiences of those who have come to be diagnosed with CD may support health professionals to effectively navigate their interactions with patients during the diagnosis and initial treatment phase and lead to improved outcomes for patients. The current study adds important insights to the evidence base considering psychological wellbeing and distress in CD.

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Tables and Figures

Table 1

Participant demographics

Participant	Gender	Age	Ethnicity	Time since diagnosis	Employment status	Living situation
Diana	Female	53	White British	2 years	Full time employed	With husband
Andrea	Female	56	White British	18 months	Retired	With husband
Sara	Female	46	Mixed ethnicity	3 years	Unemployed due to health	With daughter
Jane	Female	73	White British	3 years	Retired	Lives alone
David	Male	42	White British	1 year	Full time employed	With partner and children
Nigel	Male	57	White British	1 year	Full time employed	With partner and adult child

Appendices

Appendix 2-A: Interview Topic Guide

Topic area 1: Noticing symptoms
Example questions
- Can you tell me when you first noticed symptoms?
- Can you tell me about your thoughts and feelings related to noticing symptoms? What did you think it could be?
- What did others in your family/social group make of them?

Topic area 2: Accessing support or advice
Example questions
- Can you tell me about how you went about accessing support or advice about the symptoms you had noticed?
- How was this experience for you?
- What impact, if any, did this have on you?
- If appropriate – what did others make of this advice/support?

Topic area 3: Receiving a diagnosis
Example questions
- Can you tell me about how you came to receive a diagnosis?
- Who gave the diagnosis? What happened in that meeting? What happened next?
- What impact, if any, did this have on you/your partner/family/loved ones?

Topic area 4: Coming to terms with the diagnosis
Example questions
- Can you tell me about your experience of receiving a diagnosis of neck dystonia?
- What do you think about the diagnosis now?
- Can you tell me about any impact on how you feel/your mood?
- If applicable, did you feel able to access any support related to your mood?
- How does the diagnosis affect you now/how might it affect you in the future?

Topic area 5: Other issues
Example questions
- Is there anything else that you feel is important to talk about?

Appendix 2-B: Transcript extract with initial codes – Diana

Line No.	Verbatim transcript	Initial codes
1.	I: Can you tell me a little bit about when you first notice symptoms?	
2.	P1: Yeah, it started just over twenty years ago.	Symptoms first noticed 20 years ago
3.	They used to treat me for MS and then I went to Australia and came back.	
4.	Well, what happened sorry, do you know when I lie down, my head would turn on its own.	Noticing alarming symptoms
5.	So, they thought it was MS, but it wasn't.	They thought it was something else (MS)
6.	This went on for years.	Frustration with experience of trying to find out what was causing this
7.	I kept having x-rays, they kept saying there's nothing wrong.	Repeated/ongoing investigations and being told there was nothing wrong
8.	I spent thousands on chiropractors to try and fix it.	Spending money on funding own treatments
9.	Because it (my head) would just turn to the side, and it wouldn't move.	Symptoms that were noticeable/hard to ignore
10.	I: OK. So, you said you went to a chiropractor, did you go to anybody else for any advice?	
11.	P1: No, oh, my boss at work brought his private doctor in and he looked at my scans. He thought it was scoliosis. Because my body was right over to the side.	Accessing private medical assessments
12.	I: Right	
13.	I thought I was a hypochondriac because nobody would believe me.	Feelings that nobody believed me led to feeling like I was making it up
14.	P1: So, I used to see this doctor in Leigh, a neurologist, and he didn't know whether it was MS, you know, coming back.	
15.	But he found stuff on my brain there. But he said everybody could have it. But he sent me to Salford Royal and as soon as that doctor saw me, he told me what was wrong.	Sent to specialist neurologist by chance who identified dystonia quickly.
16.	And ever since I have been getting Botox.	
17.	My body's straighter but my neck is still a bit skew whiff. But my body's a lot straighter.	Treatment led to physical improvements.
18.	I: Ok, so once you got to the right place you got the answers you were looking for. Before that, it sounds like you spent a number of years going to different people for help.	
19.	P1: Yeah, and I had some really bad times because nobody would believe me about my neck. Because, I don't know, they just wouldn't believe me and give me help.	
20.	I: And what sort of impact did that have on you?	
21.	P1: Well, I thought I was going crazy.	Feeling like they were going crazy.
22.	I thought, am I making this up? I really did.	
23.	But luckily, my husband was really good. He believed me. He kept taking me to the chiropractors three times a week.	Believed by those closest. Supported to keep accessing treatment for symptoms.
24.	Because the pain was unbearable before the botox.	Unbearable pain before treatment.
25.	I: OK, so he supported you to keep going and getting the treatment from the chiropractor.	
26.	P1: And the neurologist in Leigh did a full blood test on me.	Ongoing investigations.
27.	And he found I had a blood disease. He said I will refer you to my friend in Manchester.	Referred to the specialist clinic by chance.
28.	And he diagnosed me as soon as he saw me.	Dr with specialist knowledge made diagnosis of dystonia quickly.
29.	I: OK	
30.	P1: I was really low; I was tired, and I couldn't be bothered doing anything.	Not feeling believed led to difficulties.
31.	I: Right.	
32.	P1: I was addicted to tramadol for the pain.	

33.	I: Really, you found yourself addicted to tramadol.	Addicted to pain relief medication.
34.	P1: Yeah, but I've been off them now for four years, I think. Three or four.	
35.	P1: Yeah so, I stopped taking them. I didn't get any help from the doctor to stop taking them. I did it on my own.	
36.	I: OK, that sounds like a whole difficulty in itself, really, the medication that you were having to take to cope with the symptoms that you were experiencing.	
37.	P1: I do feel tons better now though. Some days I'm bad but overall, I'm ok.	
38.	I: That's really good news.	
39.	I: Thinking back to the years that went by where you were having the symptoms that you mentioned. What did you think it might be? Did you have a sense of what it might have been?	
40.	P1: Yeah, well, this is another thing, while I was going to the chiropractor, I googled it and I knew what was wrong with me.	
41.	I was going to go to Australia to see a special chiropractor.	Feeling that they knew it was dystonia because of extensive research.
42.	I emailed him. Because he was like this wizard; he could fix anything.	
43.	But I don't know what happened, why I didn't go. But I did go to one over here.	Self-seeking further help and support searching for answers.
44.	I: So, did you think it was dystonia that you had?	
45.	P1: Yeah, I knew it was that because I've watched a lot of videos, and that's how my neck went.	
46.	I: Right	
47.	P1: And it was weird too, before I had Botox, when I would walk, my head would go to the side. But now I can't move my neck at all. It's rock solid.	
48.	I: OK	Mobility problems post treatment.
49.	P1: And I can't drive anymore, I can't turn my head.	
50.	I: Yeah, I was going to ask you – what sort of impact does the diagnosis have on you now? Does it stop you from doing things?	
51.	P1: Yeah, I can't drive. But I must say with this treatment, I'm a lot better. At my desk I used to have to hold my head up, but I don't anymore.	
52.	I: Right	Symptoms have not completely gone.
53.	P1: The only thing is, when I lie down, it shakes a lot now.	
54.	I: OK	A sense of relief when diagnosis was received.
55.	P1: When he told me, I was so relieved. Because I thought, I'm not a hypochondriac.	
56.	I: So, there was a sense of relief.	Relief as they were not going mad

Line No.	Verbatim transcript	Initial codes
57.	I: Can you tell me a little bit about when you first notice symptoms?	
58.	P1: Yeah, it started just over twenty years ago.	Symptoms first noticed 20 years ago
59.	They used to treat me for MS and then I went to Australia and came back.	
60.	Well, what happened sorry, do you know when I lie down, my head would turn on its own.	Noticing alarming symptoms
61.	So, they thought it was MS, but it wasn't.	They thought it was something else (MS)
62.	This went on for years.	Frustration with experience of trying to find out what was causing this
63.	I kept having x-rays, they kept saying there's nothing wrong.	Repeated/ongoing investigations and being told there was nothing wrong
64.	I spent thousands on chiropractors to try and fix it.	Spending money on funding own treatments
65.	Because it (my head) would just turn to the side, and it wouldn't move.	Symptoms that were noticeable/hard to ignore
66.	I: OK. So, you said you went to a chiropractor, did you go to anybody else for any advice?	
67.	P1: No, oh, my boss at work brought his private doctor in and he looked at my scans. He thought it was scoliosis. Because my body was right over to the side.	Accessing private medical assessments
68.	I: Right	
69.	I thought I was a hypochondriac because nobody would believe me.	Feelings that nobody believed me led to feeling like I was making it up
70.	P1: So, I used to see this doctor in Leigh, a neurologist, and he didn't know whether it was MS, you know, coming back.	
71.	But he found stuff on my brain there. But he said everybody could have it. But he sent me to Salford Royal and as soon as that doctor saw me, he told me what was wrong.	Sent to specialist neurologist by chance who identified dystonia quickly.
72.	And ever since I have been getting Botox.	
73.	My body's straighter but my neck is still a bit skew whiff. But my body's a lot straighter.	Treatment led to physical improvements.
74.	I: Ok, so once you got to the right place you got the answers you were looking for. Before that, it sounds like you spent a number of years going to different people for help.	
75.	P1: Yeah, and I had some really bad times because nobody would believe me about my neck. Because, I don't know, they just wouldn't believe me and give me help.	
76.	I: And what sort of impact did that have on you?	
77.	P1: Well, I thought I was going crazy.	Feeling like they were going crazy.
78.	I thought, am I making this up? I really did.	
79.	But luckily, my husband was really good. He believed me. He kept taking me to the chiropractors three times a week.	Believed by those closest. Supported to keep accessing treatment for symptoms.
80.	Because the pain was unbearable before the botox.	Unbearable pain before treatment.
81.	I: OK, so he supported you to keep going and getting the treatment from the chiropractor.	
82.	P1: And the neurologist in Leigh did a full blood test on me.	Ongoing investigations.
83.	And he found I had a blood disease. He said I will refer you to my friend in Manchester.	Referred to the specialist clinic by chance.
84.	And he diagnosed me as soon as he saw me.	Dr with specialist knowledge made diagnosis of dystonia quickly.
85.	I: OK	
86.	P1: I was really low; I was tired, and I couldn't be bothered doing anything.	Not feeling believed led to difficulties.
87.	I: Right.	
88.	P1: I was addicted to tramadol for the pain.	
89.	I: Really, you found yourself addicted to tramadol.	Addicted to pain relief medication.

90.	P1: Yeah, but I've been off them now for four years, I think. Three or four.	
91.	P1: Yeah so, I stopped taking them. I didn't get any help from the doctor to stop taking them. I did it on my own.	
92.	I: OK, that sounds like a whole difficulty in itself, really, the medication that you were having to take to cope with the symptoms that you were experiencing.	
93.	P1: I do feel tons better now though. Some days I'm bad but overall, I'm ok.	
94.	I: That's really good news.	
95.	I: Thinking back to the years that went by where you were having the symptoms that you mentioned. What did you think it might be? Did you have a sense of what it might have been?	
96.	P1: Yeah, well, this is another thing, while I was going to the chiropractor, I googled it and I knew what was wrong with me.	
97.	I was going to go to Australia to see a special chiropractor.	Feeling that they knew it was dystonia because of extensive research.
98.	I emailed him. Because he was like this wizard; he could fix anything.	
99.	But I don't know what happened, why I didn't go. But I did go to one over here.	Self-seeking further help and support searching for answers.
100.	I: So, did you think it was dystonia that you had?	
101.	P1: Yeah, I knew it was that because I've watched a lot of videos, and that's how my neck went.	
102.	I: Right	
103.	P1: And it was weird too, before I had Botox, when I would walk, my head would go to the side. But now I can't move my neck at all. It's rock solid.	
104.	I: OK	Mobility problems post treatment.
105.	P1: And I can't drive anymore, I can't turn my head.	
106.	I: Yeah, I was going to ask you – what sort of impact does the diagnosis have on you now? Does it stop you from doing things?	
107.	P1: Yeah, I can't drive. But I must say with this treatment, I'm a lot better. At my desk I used to have to hold my head up, but I don't anymore.	
108.	I: Right	Symptoms have not completely gone.
109.	P1: The only thing is, when I lie down, it shakes a lot now.	
110.	I: OK	A sense of relief when diagnosis was received.
111.	P1: When he told me, I was so relieved. Because I thought, I'm not a hypochondriac.	
112.	I: So, there was a sense of relief.	Relief as they were not going mad

Appendix 2-C: Audit trail of analysis for Sara

Initial codes/emergent themes	Narrative summary	Supportive Quotes
Cluster 1: Inexplicable loss of control over the body and it's catastrophic effect on life		
Attempts to get on with it and hide symptoms from others, symptoms attributed to common ailments, brushed it off until it was so bad it could no longer be ignored. Loss of control over one's body, symptoms worsened over time, difficulty getting on with life whilst experiencing painful and alarming symptoms, experiencing people staring and asking questions, vicious cycle of not wanting to go out due to social anxiety leading to feeling trapped and increased anxiety, Friends frustrated due to lack of ability to gain control over symptoms, impact on relationship with child, sense of being alone with what was happening, very distressed, thoughts of not wanting to live, difficulties with day to day functioning, anxiety/depression as a result of dystonia symptoms led to inability to function, loss of income and inability to continue payments (e.g. home payments), isolation, lost job, lost home, Using maladaptive coping strategies to cope, Coping alone, Continuing to cope alone after diagnosis, still have bad days where it is difficult to leave the house which contributes to an ongoing sense of isolation.	This theme reflects Sara's turbulent experience of noticing symptoms and help seeking which was characterised by an overall lack of control over her body and the health services on which she was reliant. Sara was scared by her symptoms, and she found they worsened over time to the point of no control. Unexplained and confusing symptoms which were causing significant pain and disrupting sleep led to several significant losses; she was unable to work and friendships ended because others were unable to understand why she couldn't take control and do something to improve her symptoms. Sara recognised that her relationship with her daughter was affected due to the unexplained changes that her daughter witnessed, suggesting that the security of the attachment between them had been affected due to effects of symptoms and related distress on parenting. The global impact of unexplained symptoms of dystonia led Sara to experience great personal distress to the level that she wished to end her life.	"I tried to hide it for it for months." (P3, line 37) "It started with a pain in my shoulders. Like a burning pain in my shoulders and in my neck. And just kept putting it down to stress." (P3, line 26) "it's just stress, it's just stress. Until I just couldn't ignore it." (P3, line 39) "I just couldn't keep it together. Couldn't keep it together and act normal." (P3, line 48) "I started to feel really isolated. Because everybody knew me as someone who kept it together." (P3, line 42) "It steadily got worse and then I couldn't stop the movements. It kept moving to the right by itself which really scared me." (P3, line 43) "I was scared. I'll be honest, I was scared. 'cause I just had the fact is that not having the control." (P3, line 29) "At first it was just slight. It would kinda like just turn and I could catch it and hold it. But as it got worse I couldn't control it. And it was scary. I just didn't know what was happening." (P3, line 34) "It would literally turn to the right by itself. Which really freaked me out." (P3, line 27) "You don't know what's going on and your whole life is kind of falling apart." (P3, line 16) "Because it's constant, there's no let up. You can't even think because the whole focus is on the pain, on the spasms, on the movements. It consumes you because it's just always there. There's no let up." (P3, line 137) "And then there is people staring and the questions, and you don't even know what's going on yourself" (P3, line 91) "And then you get into this vicious cycle then, with the anxiety, the depression and not wanting to go out. But then being trapped in the house made all of them things worse but then not wanting to go out. You feel like you are trapped, like a vicious cycle. " (P3, line 92) "And then I was just horribly depressed. There was a point where I just didn't even want to be here anymore." (P3, line 88) "And I was that far in the depths of anxiety and depression. I just couldn't function, could not function." (P3, line 159) "So I don't go out because people just find it really funny. People are horrible,

horrible. I've learnt that. That's contributed to the anxiety being just off the wall." (P3, line 186)

Cluster 2: It's not all in my head

When symptoms worsened went to GP, Felt like a long process due to relentlessness of symptoms, multiple GP visits, pre-diagnosis treatment made symptoms worse, Repeated GP appointments until GP saw symptoms for themselves, feeling a need to prove it's not in my head, impact of feeling not believed – suffering alone, frustration, long process where they felt like they weren't believed, not feeling listened to was the hardest part, regretting having sought support for MH previously, symptoms were attributed to MH feeling better mentally after receiving support/diagnosis/treatment, had to develop relationship before being able to talk about distress caused by dystonia, worry that symptoms will continue to be put down to psychological causes, needing to be seen to be believed, still have sense that most believe it is in my head.

Help seeking was a long and arduous process for Sara, there were repeated GP appointments and long waiting times. Until the GP was able to see her symptoms, she felt she was not taken seriously or believed. This process was difficult to bear because of the impact that symptoms were having on her life. The impact of these experiences stayed with Sara whereby they influenced her sense of trust in health professionals; being so distressed at the point of meeting with a movement disorder specialist she felt sure she would be dismissed once again. Having accessed support but not felt believed or provided with any treatment for effective relief, Sara resigned herself to coping alone with distress caused by ongoing symptom. She started using alcohol as a method of escaping what was going on and coping with the distress of not being believed.

"It started with a pain in my neck and my shoulders which I put down to stress. Which steadily got worse. So I went to the dr's, he said, you're just stressed, the usual." (P3, Line 3)

"Went back to the doctors. They put me on medication to try and help. Which just seemed to make things worse and make the movements worse." (P3, line 8)

"I was back and forth to the doctors but obviously there's waiting times, there's referrals. And in the meantime you're living with it day in day out." (P3, Line 16)

"And it's alright then saying you know there's waiting lists and stuff but when you wait 3-6 months you don't know what's going on and your whole life is kind of falling apart. I ended up losing my home." (P3, Line 19)

"Yeah, but even with the doctors it was such a long process. Or at least to me it felt really long.

"Because it's day in day out, 24 hours a day, seven days a week. Even when you're sleeping, you're constantly waking up with it." (P3, Line 66)

"I was just overwhelmed at that point and it got to the point where I went to doctors and he was like and he said I never seen you like this before and I think at that point he knew that he needed to do something more." (P3, Line 68)

"And the first it was like, oh we'll send you to see a psychiatrist. I was like this is not in my head. This is not in my head." (P3, Line 70)

"I felt like I was being penalized because over the years I had been for depression" (P3, Line 71)

"By that point he had seen it for himself, he'd felt my neck. And I think he knew that he had to do something more than just wait for a psychiatrist appointment." (P3, Line 75)

"It was horrible. It was horrible because, like, you're suffering. And you know that is real. You know it's not in your head because who the hell would want to make that up in their head?" (P3, Line 81)

"And then I started drinking more so than usual - because it's a relaxant. And in the moment it helped, not even necessarily so much in terms of the movement and stuff. Erm, it kind of made me relax and my brain wasn't going 100 miles an hour.

		Thinking - what am I gonna do? What am I gonna do? Nobody's believing me, what am I gonna do? And I suppose it was just like an escape from what was going on." (P3, Line 83)
Cluster 3: A mixture of feelings on finally receiving a diagnosis Scary and relief that it's not all in my head, uncertainty about the unknown, thoughts about what the future will be like with this diagnosis, not feeling like me, unsupported, others didn't understand, continuing to feel distressed, anxious, depressed about inability to control symptoms, feeling alone, nobody to talk to, wanting a solution and to go back to normal, realising that it is a case of living with symptoms, feelings of having adjusted to diagnosis, treatment has helped with adjustment, treatment has helped to improve symptoms providing relief and restored ability to cope/function, trial and error with treatment, starting to be effective, provided relief to be able to think and reflect on this experience, feel more like self now that symptoms are more under control, piecing life back together after a turbulent few years since symptoms began, rethinking work, can't do what they were doing before, hopes that relationship will repair now symptoms are more under control and feeling more like themselves, a sense of having something to pin it on – an explanation, quick examination and decision of probable dystonia once seen by a neurologist, had an explanation for what was happening	Sara held mixed feelings about receiving a diagnosis; on one hand she felt relieved and vindicated for her struggles, however on the other hand receiving the diagnosis of dystonia felt scary and unknown. The diagnosis had acknowledged that there was something seriously wrong which would impact on her life whilst not feeling believed it was possible to deny the need to adjust to a chronic illness. However, prompt treatment supported the process of adjustment, allowing some relief from symptoms to process her experiences. Life post-diagnosis for Sara entailed rebuilding of many aspects of her life including work, friendships, and familial relationships.	"it was then it was like there's a name for it. I've got something." (P3, line 122) "I just can't get my head around it, for ages. I never really had any serious illnesses" (P3, line "And it's a case of right. This is the rest of my life." (P3, line 115) "I just wanted them to fix it. Yeah, I'm just like let me get back to normal." (P3, line 114) "it's kinda like a relief, but scary at the same time because I'd never heard of it, didn't know what it was" "And then, obviously it's just waiting again. Waiting, waiting, waiting. All the time you're living with it every single day, every minute of the day." (P3, line 113) "Again, it's the unknown. The unknown is always scary. If it's something familiar that you've heard of before, would that have made it better, easier, I don't know." (P3, line 120) "And all I kept thinking is, is this for the rest of my life? Are they going to be able to do anything, or is it literally this? Is it now for the rest of my life? And I couldn't imagine that I just couldn't imagine. Because I wanted my life carry on as it was, yeah." (P3, line 123) "And it was, oh my God things might never be the same again. Is this it now for the next? How many years until I die? Will I ever work again?" (P3, line 123) "And not have anybody to speak to that understands." (P3, line 130) "Well, I guess I have come to terms with it, because it took a while for them to kind of like get treatment started." (P3, line 133) "But in the meantime, from getting that diagnosis to where you've got the treatment right enough for me to be able to function and think straight. Because it's constant, there's no let up. You can't even think because the whole focus is on the pain, on the spasms, on the movements. It consumes you because it's just always there. There's no let up." (P3, line 136) "now I feel like I've been able to take a breath. To be able to get my head in order and just to be able to think." "Now I've started to get a bit myself back. We've started to talk and stuff and hopefully we'll be able to repair the last

few years. It will take time." (P3, line 139)

"And I've started to get my thoughts back together, and even the medications that I was on, I've reduced them to the bare minimum. And especially the clonazepam, I think that kind of, pardon my French, but it f***** me up big time." (P3, line 165)

"So I'm trying to get my life back together now. Thinking about what work I could do." (P3, line 166)

"And there's still some days, some days it is still bad even though I have the Botox. I'm not willing to take a higher dose than what I'm taking already, because it's already a high dose. Which long term is going to have its own problems from having so much Botox injected." (P3, line 169)

"I'm trying not to think about that far down the road yet. But I'm thinking about if I do go back to work, what happens on the days when I can't go in." (P3, line 170)

"There are still days where I literally can't leave the house. And then again it's that isolation." (P3, line: 175)

"I'm thinking start something of my own and like when you said like in terms of support and stuff, I'm going down that route, wondering if I should do a counselling course or something along them lines. Just to be able to give that to support to people. Because there's a lot a definite lack of people understanding and getting like you're coming from." (P3, line 205)

"because it's something that's never gonna go away. Uh, is it more under control now? Yes? But then there's a long term effects of such high doses of Botox. Then there's the dread of it spreading because it can move."

"I try not to think that hard even though it is hard not to think about it, 'cause it's human nature, isn't it? But I try really hard not to think about it. Because I've always been a planner and learning to take life day by day has been hard." (P3, line 207)

"Just stepping out that door, you have to think about everything. What am I going to do if this happens. It's exhausting. So just trying to get into that right - Just think of you know living in the moment and just take one day at a time. I mean, it sounds so easy. But when you've never been that person it's really hard. And like not being able to do spur of the moment stuff. I always have to have notice now where I've got to prepare." (P3, line 209)

	"Yeah, I think losing a little bit of yourself is hard." (P3, line 211)

Appendix 2-D: Process of theme development across participants

Emergent themes/initial codes from across participants	Refined themes	Final themes	Participants contributing to theme
Symptoms were hard to ignore, symptoms impact on life, what if this is Parkinson's, lack of control over the body, not knowing what this is or what is happening to me, wondering what is happening to my body, embarrassment at the lack of control over my body, distressing first symptoms, getting on with it, worries about what other's think of me, coping alone until it could no longer be ignored, coping alone with symptoms, isolated and coping alone, impact on family and wellbeing, Coping alone until it could no longer be ignored, embarrassment at what other's may think, impact on life and other's around me, sign of weakness.	Coping alone with debilitating symptoms, getting on with it, widespread impact of symptoms, ontological concerns and fears.	Experiences of losing control over one's body and attempts to regain control	Diana, Andrea, Sara, Jane, David, Nigel
Desperate for answers, I can't go on like this, appraisals of symptoms could inhibit or disinhibit help seeking, being vulnerable and asking for help is a big step, repeated assessments, incorrect diagnoses, incorrect treatments, fighting to be listened to, becoming the expert, taking it into my/our own hands, going mad, powerless and despondent leading to depression, not believed, delayed appointments due to covid, quickly recognised symptoms and appropriate referral maintained trust in professionals, not believed and needing to prove there is a real problem, trust in health professionals having felt believed and listened to, waiting and a process of elimination led to fear, waiting, reduced trust in professionals, complying despite little hope this will work, a sense of powerlessness and isolation due to a lack of communication from health professionals, fighting to be listened to, concerns that specialists would put symptoms down to distress	Searching for answers. Frustration at not getting the help I needed, feeling ignored and not believed, taking it into my own hands, becoming the expert, impact of experiences of help seeking on mental health and wellbeing	Feeling powerless in response to the health system	Diana, Andrea, Sara, Jane, David, Nigel
Continue to cope alone after diagnosis, relief – not the feared diagnosis, hopes of being fixed, a sense of having a diagnosis and doing something about this facilitates adjustment , promptly receiving answers meant holding hope about the future, acceptance and adjustment, relief at getting the help I need, feeling believed – the relief of diagnosis, feeling acknowledged due to diagnosis , dystonia diagnosis is unknown , receiving an unknown diagnosis/ diagnosis provided relief and vindication , informed and reassured, relief at being in the right place, being informed was reassuring, reassured by expertise, worries that treatment won't work in the future, future fears about losing control again, needing to re-build life, ongoing difficulties, ongoing isolation.	Relief at finally receiving a diagnosis, Scared to receive the diagnosis, ongoing frustration and anger, uncertainty about the future, continuing to cope alone, ongoing difficulties	Mixed feelings upon diagnosis	Diana, Andrea, Sara, Jane, David, Nigel

Appendix 2-E: Psychology and Health: Instructions for Authors

Contents

About the Journal

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Psychology & Health accepts the following types of article: Original Research Articles, Reviews, Commentaries and Registered Reports.

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Original Research Articles include reports of Randomized Controlled Trials (RCTs), experimental studies, observational studies, qualitative research studies, and other investigations. All submissions must follow the appropriate reporting guidelines and instructions for reporting statistics. All RCTs and experimental studies must be pre-registered. The pre-registered details should be given in the methods section and, if possible, blinded for peer review (i.e., 'the review was preregistered at [BLINDED]') ; the details can be added at proof stage.

Reviews are systematic reviews and meta-analyses that are thorough, critical assessments of the literature and data sources pertaining to topics within the scope of Psychology and Health. Per PRISMA guidelines, systematic reviews and meta-analyses

must be identified as such in the article title and must be pre-registered. The pre-registered details should be given in the methods section and, if possible, blinded for peer review (i.e., 'the review was preregistered at [BLINDED]'); the details can be added at proof stage.

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Your paper should be compiled in the following order: title page; abstract; keywords; main text introduction, materials and methods, results, discussion; acknowledgments; declaration of interest statement; references; appendices (as appropriate); table(s) with caption(s) (on individual pages); figures; figure captions (as a list).

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Updated 11th August 2022

Section 3: Critical Appraisal

Research decisions, challenges, professional issues and personal reflections

Word Count (excluding references, tables and appendices): 3,956

Louise Glover

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

All correspondence should be sent to:
Louise Glover
Health Innovation Campus
Lancaster University
Sir John Fisher Drive
Lancaster,
LA1 4AT

Email: l.harris7@lancaster.ac.uk

Critical Appraisal

This critical appraisal aims to supplement the reports within sections one and two and provides an opportunity to discuss broader research issues. It is hoped that this section will contextualise the research for the reader and provide reflections which may be helpful for future research with similar populations. First a summary of the literature review and empirical paper are provided. Then a discussion of the relevant decisions made, challenges, professional issues and personal reflections. Throughout, will be a focus on the potential influence on the findings, strengths, limitations, and implications for future research.

Summary of findings

Literature review

The systematic literature review is a meta-ethnography informed by Noblit and Hare (1988), aimed at synthesising the relevant qualitative literature about couples' experiences of Parkinson's. Relevant findings from 19 studies are included, analysis of which produced three themes: 1) Disruption to the couple's roles and responsibilities and attempting to maintain the relationship; 2) Challenges to communication and closeness leading to increased resilience, and 3) Increased cognitive and behavioural symptoms leading to grief, burden and isolation. The findings highlight some of the issues encountered by couples when one member has Parkinson's. It highlights that further study to provide appropriate support for couples may have a positive impact on the well-being of both people with Parkinson's and their caregiving partners, thus, supporting the notion that healthcare professionals may benefit from assessing and supporting the needs of the couple, rather than regarding them as separate individuals with different needs.

The systematic literature review provides insight into the current literature, which highlighted the need for further high-quality qualitative studies about couples' experiences of Parkinson's. The review suggests that due to the heterogeneous nature of the condition, future

studies should seek to recruit a homogenous sample by focusing on specific dimensions of the Parkinson's experience, for example, a couple's experiences of diagnosis. Despite this, the review provided a broad view of the lived experience of couples living with Parkinson's more so than can be gained from the studies in isolation. Therefore, this review provides an opportunity to support healthcare professionals' understanding of the issues that couples may face based on the existing literature. This may assist healthcare professionals in having increased confidence in the identification of a couple's needs. It is suggested that tailored psychological support for couples living with a chronic health condition, such as Parkinson's, may support well-being and improve the quality of life for both members of the couple. However, further study is required to consider effective psychological interventions for individuals and couples that consider the relational and systemic issues that may present.

Empirical paper

The empirical paper is a qualitative study informed by the qualitative method, interpretative phenomenological analysis (IPA). This study aimed to explore the experience of diagnosis in cervical dystonia (CD), as whilst the quantitative literature highlights common difficulties in obtaining a diagnosis of CD, the individual lived experience is less well understood. The analysis produced three themes: 1) Losing control over one's body and attempts to regain control: 'You don't know what's going on and your whole life is kinda falling apart'; 2) Feeling powerless in response to the health system: 'I had this change and I couldn't do anything about it, and nobody seemed to have any kind of solution'; 3) Mixed feelings upon diagnosis: 'It's kinda like a relief, but scary at the same time'. The findings highlight the significance of the pre-diagnosis phase, which can be a distressing experience whereby accessing support may exacerbated distress. Conversely, the pre-diagnosis phase was straightforward for some participants, which seemed to help contain their distress and

retain trust and faith in health professionals as they went forward into a subsequent journey of receiving targeted therapy for CD.

The findings of this IPA study highlight how understanding the experiences of those who have come to be diagnosed with CD may support healthcare professionals to effectively navigate their interactions with patients during the diagnosis and treatment phases, and lead to improved outcomes for patients. For example, healthcare professionals may be more equipped to understand the personal toll of a difficult journey to diagnosis which may lead to difficulties that affect adjustment. Such understandings may further equip health professionals to explore the nature of previous experiences and consider how this may influence barriers to future care and the use of effective coping mechanisms. This study highlights a potential link between diagnosis experience and illness perceptions that warrants further study, and the need for further study regarding the impact of diagnosis experience on adjustment to illness.

Research decisions, challenges, professional issues and personal reflections

Empirical paper

Selecting a research focus and methodology

The focus for the empirical paper was selected in negotiation with the research supervisors to ensure that there was appropriate supervision available, and to ensure that the area of focus would be feasible. The health psychology focus was particularly interesting to me due to an interest in the false dichotomy that often exists within healthcare, whereby physical and mental health matters have been treated as being mutually distinct. Although this narrative is slowly shifting given more recent health initiatives which have aimed to raise the priority of mental health in line with physical health within UK healthcare (Millard & Wessely, 2014), there remains a relative dearth of research about the emotional and psychological impact of dystonia, particularly qualitative literature. This is despite the

reportedly high rates of mental health difficulties within this population (Medina Escobar et al., 2021a; Medina Escobar et al., 2021b; Worthley & Simonyan, 2021). However, one previous qualitative study (Morgan et al., 2019) identified a potential connection between diagnosis experience in dystonia, and psychological and emotional distress. Hence, it was identified that qualitative research into experiences of diagnosis in dystonia was a pertinent area for further research.

Having identified a lack of qualitative research in dystonia, IPA was selected as an appropriate qualitative methodology. IPA is concerned with the detailed psychological and emotional processes involved in individual experiences of certain phenomena, for example, meaning making (Smith et al., 2009). As such, it was consistent with the epistemological position of the research, which was interested in exploring individual experiences of diagnosis in cervical dystonia as this had been identified as a gap in the evidence base. However, several qualitative methods exist, each of which has different aims and approaches. An alternative method that may have been applied to this research due to the time-sensitive nature of the experience in question, is narrative analysis (Bingley, 2020). Narrative analysis methodology examines experiences from the point of view of a story, whereby the experience has a beginning, middle, and end, also assuming that there are patterns to be aggregated from experience (Bingley, 2020). Conversely, IPA explores how people perceive, organise and experience phenomena, taking a subjective perspective to identify individual emotional and psychological processes (Smith et al., 2009). Hence, it was determined that IPA would be the best fit for the research aims. Furthermore, being a novice researcher was a factor in deciding on an appropriate methodology. Materials are available to guide researchers in the necessary steps to take to undertake their work from an IPA perspective (Murray & Wilde, 2020; Smith et al., 2009). The materials guide the research process, including the planning, data collection, analysis and writing phases. Due to the rigorous nature of IPA methodology and

guidance, it is suggested that the approach enables the fulfilment of Yardley's criteria for quality in qualitative research, despite the level of research experience (Smith et al., 2009; Yardley, 2000).

Recruitment

A key decision when designing the empirical study was the recruitment of an appropriate sample of participants. In IPA, obtaining a small and homogenous sample is a high priority (i.e., 4-10 participants) which allows for in-depth analysis considering personal meaning-making and psychological processes of each participant (Murray & Wilde, 2020; Smith et al., 2009). A homogenous sample is often obtained by ensuring that participants are demographically similar, for example (e.g., age or gender). However, the key homogeneity must be relative to the phenomenon in question (Smith et al., 2009). For the present study, dystonia is a rare condition, hence the experience of the condition itself, promotes relative homogeneity of experience. As diagnosis experience was the key issue it was considered how to ensure that this experience was salient to the recruited sample This prompted the decision to limit the time since diagnosis. It is therefore recognised that although the participants differ demographically, they are homogenous concerning the discreet experience of receiving a diagnosis of dystonia within the three years prior to interview. Hence, the participants are homogenous with respect to the factors that are relevant to the study (Smith et al., 2009).

A common issue relevant to psychological research is the problem of self-selection. That is, the likelihood that those who put themselves forward for participation in a study, may differ from the general population which introduces bias (Rosnow & Rosenthal, 1976). Rosnow and Rosenthal (1976) suggest that to minimise volunteer bias, researchers should open recruitment as widely as possible. During this study, recruitment was attempted through two routes to minimise volunteer bias and maximise the possibility of sufficient recruitment from this small population. The routes included via a specialist NHS movement disorder

clinic, and through community means via collaboration with a charitable organisation and recruitment through their social media channels. Unfortunately, the community route was not successful, as those who approached me to participate did not meet the inclusion and exclusion criteria of the study, for example, some had other diagnoses of dystonia, such as dystonia of the eye (i.e., blepharospasm), and diagnosis received beyond the three-year window. This process of ensuring participants fit the study requirements was time intensive, and on several occasions, participants felt they did fit the criteria and were disappointed when they were unable to take part.

In comparison, the specialist clinic recruitment route was far simpler as potential participants' diagnosis and the time frame were confirmed by their healthcare team which negated such difficulties. However, it is also possible that this recruitment method may have introduced bias. For example, this cohort may have had more positive experiences and relationships with their clinical team upon diagnosis compared to others. Furthermore, there is a chance that those supporting recruitment could have hand selected those they wished to take part who they felt had had a good experience. Overall there were benefits of using the specialist recruitment method, namely, it allowed for sufficient recruitment to the study, which was a challenge to achieve due to the small cohort of people that were being accessed. Hence, further studies with similar populations may also consider establishing a connection with a specialist clinic to allow access to a small and specific cohort of people. This route may be particularly effective with time-sensitive projects such as within the research requirements of the clinical psychology doctorate programme.

Participants

Whilst collaboration with a specialist movement disorder clinic supported the effective recruitment of a homogenous group of participants, it is also possible that limiting recruitment to this site only is a weakness of this study. Recruitment from this site meant that

all six participants resided in the northwest region of the UK. Access to such a clinic is not available throughout the UK, therefore, it is quite likely that individuals without this access may have different experiences to those presented within the empirical paper. Furthermore, two participants gained access to private medical assessment during their journey to diagnosis, which then provided them with a route into NHS services. Of course, access to private healthcare is costly and not accessible to all. Therefore, interviews with individuals from a lower socio-economic status may yield different findings.

Data collection

Due to constraints that existed regarding COVID-19 and social contact during the design phase of this study, remote methods for interviews were selected. As such, the participants were provided with a choice of mode for interview either telephone or video-conferencing software (Microsoft Teams). Studies regarding the acceptability and effectiveness of remote means for qualitative data gathering have suggested that the use of telephone and video-conferencing software is an effective alternative to in-person interviews and even beneficial in certain populations (Archibald et al., 2019; Drabble et al., 2016). Video-conferencing, specifically, is suggested to be effective in widening participation, being practical and accessible to many, rather than restriction being placed upon travel or geographical area, without sacrificing researchers' ability to build rapport with participants, thus not impeding the gathering of rich data (Archibald et al., 2019; Smith et al., 2009). Indeed, in this study, I felt that remote methods were advantageous as they helped facilitate the recruitment of a homogenous group from a relatively small population, thereby strengthening the quality of the analysis (Smith et al., 2009). However, five participants opted for interview by telephone, and one opted to interview by video call. Carrying out qualitative interviews via telephone receives some criticism as on one hand, it is thought to challenge rapport building (Deakin & Wakefield, 2014; Lo Iacono et al., 2016. Yet, on the other hand, it is suggested that telephone

interviews may support the discussion of sensitive subjects, particularly in populations, such as CD, where issues such as stigma are prevalent (Drabble et al., 2016; Rinnerthaler et al., 2006). On reflection, telephone interviews did not seem to overly impact upon rapport and richness of data gathered. On the contrary, it seemed a helpful mode to enable people with CD to take part as it avoided concerns about visual differences caused by the condition. Thus, it may be beneficial to consider remote means, and certainly not to rule out interviews conducted by phone, in future studies with similar populations.

Reflexivity and personal reflections

The double hermeneutic process is a central feature of IPA (Smith et al., 2009). That is, the awareness of the simultaneous processes of sensemaking taking place by both the participant and the researcher (Smith et al., 2009). Within this, IPA asks that the researcher attempts to suspend their pre-conceptions throughout the design and data collection phases for the salient issues to be brought by participants (Smith et al., 2009). As a researcher embarking upon an IPA study, I reflected upon my pre-conceptions of the research focus and as I held no prior personal experience of dystonia, and therefore no personal pre-conceptions of the experience in question, I wondered if this would be a benefit to me whilst undertaking this study. Being an 'outsider' of the population in focus, on the one hand, allows an objective observer stance which supports the reduction of bias in the interpretation of data (Holmes et al., 2020; Smith et al., 2009). Whilst on the other hand, it is said that it is difficult to gain a representative understanding of phenomena where this is not personally relevant (Holmes, 2020). Therefore, there was a delicate balance to strike in order to produce a good quality piece of work whilst also meeting the requirements of IPA.

To ensure that the research was sensitive to context and improve the accessibility of study materials, I worked with an expert from the clinical field and an expert by experience (Duncan & Oliver, 2017; Oliver et al., 2015; Yardley, 2000). Furthermore, I needed to

engage with the pre-existing literature to identify an appropriate gap for worthwhile research, and to ensure again that that the present study was appropriately placed within the current evidence base. Therefore, despite being a relative 'outsider' to the population in focus, I noticed that through these necessary steps to produce a good quality piece of work pre-conceptions were introduced. For example, I experienced noticed that I felt growing empathy for people with difficult experiences of diagnosis and I felt motivated to improve experiences for those diagnosed with dystonia. I noticed that these preconceptions meant the experiences of participants who were more distressed stood out to me more during the analysis. Keeping a reflective log (see Figure 1) and using supervision to reflect on and maintain awareness of the influence of these preconceptions helped in my attempt to set this aside and focus on the data at hand.

I also found it difficult at times, to navigate the dual role of trainee clinical psychologist whilst also conducting IPA research. This meant, at times I was drawn to sense-making during the interviews, particularly when participants seemed distressed whilst reflecting on their experiences. Supervision and supervisor feedback from interviews enabled an awareness of this to prevent impact on gathering relevant and rich data. However, I reflected on the possible strengths of holding this dual role, for example, transferable skills such as having in-depth emotional conversations and picking up on verbal and emotional cues that require further exploration, are essential skills for rich data gathering in IPA (Smith et al., 2009). I reflected upon how these skills indeed appeared to support the gathering of rich data during interviews, despite being new to IPA, yet it was important to hold an awareness of when I was tipping too far into clinical mode as opposed to researcher mode. The use of the interview topic guide to structure participant interviews helped with this.

A further challenge I experienced during this work was the rigorous process of data analysis. A personal challenge was handling the large amounts of data during the analysis,

whilst trying to identify experiences that were relevant across participants and retain the individual stories. The thorough guidance on completing an IPA analysis provided by both Murray and Wilde (2020) and Smith et al., (2009) was a great basis on which to begin, however, there was a process of personally engaging with the analysis that I found difficult to capture and put into words. This resulted in a felt sense of connections existing between the accounts and how each built upon the experiences of the other. I was grateful for the invaluable supervision that I received to help to make sense of the data. Supervision also supported the process of continuous awareness of setting aside pre-conceptions during the analysis phase which resulted in reliance on certain participants over others. Finally, the analysis was produced and due to the small number of participants included in this study, it was possible for each participant to 'speak' in the form of quotes within each theme.

[Figure 1 about here]

Systematic Literature Review

Selecting a focus

With the focus of the empirical paper selected, it was important for the literature review to be a related subject and not too far removed. Due to the scarcity of qualitative literature in the field of dystonia, the literature was examined on related disorders, including essential tremor, multiple sclerosis and Parkinson's. It seemed that the first-hand experiences in these conditions had been thoroughly explored and synthesised. For example, experiences of Parkinson's from the point of view of both caregivers and individuals diagnosed with Parkinson's had been previously reviewed (Soundy et al., 2014; Theed et al., 2017). However, no review existed considering the experience of couples whereby one is diagnosed with Parkinson's. The couple's experience of illness is an important dimension to explore given that illness is not experienced in isolation, rather, when an individual experiences illness it affects them personally, those close to them such as partners and family members and the

relationships between them (Rosland et al., 2012). A recent review on dementia highlighted the need for greater emphasis on supporting togetherness and couple hood due to the centrality of this to both the couple and individual experience of the condition (Wadham et al., 2016). A brief initial review of the available qualitative literature regarding couples' experiences of Parkinson's revealed several available studies that focused to some extent on the couple's experience of Parkinson's, therefore, this was selected as the focus for the systematic literature review. Underpinning the link between the two papers presented in this thesis is the assumption that mental health is a central part of health, and that health is influenced by individual, social and societal factors, and their interaction with each other (Millard & Wessely, 2014; Eriksson & Hammarström, 2018). Therefore, research that considers issues at different levels has an essential connection to the other, serving to broaden and deepen understandings leading to more effective healthcare.

Quality appraisal

The mode of quality appraisal was an important decision of the literature review. The tool selected was the CASP as this is one of the most used quality appraisal tools (Hannes & Macaitis, 2012; Noyes et al., 2018). The tool comprises 10 items which aim to prompt and guide the researcher in the assessment of quality. To supplement this, scores were applied using a system introduced by Duggleby et al. (2010), whereby a score of 1-3 was given for each item on the CASP, resulting in a total score out of 24 for each paper. However, the use of quality appraisal is debated within qualitative research due to their epistemologically and ontologically separate aims (Malterud, 2019). That is, qualitative methods aim to explore and understand nuanced experiences, whilst quality appraisal is concerned with assessing the validity of research studies; a better fit with the positivist philosophy of quantitative study (Malterud, 2019). Furthermore, critics suggest that tools such as the CASP are too simplistic and prescriptive to capture the nuanced features of differing qualitative research

methodologies (Malterud, 2019). Yet, quality appraisal remains an important juncture in qualitative reviews. Tools such as the Critical Appraisal Skills Programme for qualitative studies (CASP; 2018) have several benefits, including helpfulness in guiding novice researchers in appraising studies and ability to discriminate between studies in terms of their quality to ensure their contribution to a synthesis is weighted accordingly (Hannes & Macaitis, 2012; Noyes et al., 2018).

Upon reflection, and now that my working knowledge of one qualitative approach (IPA) has increased, I believe if I was to repeat the quality appraisal phase of the literature review my responses may differ. This difference highlights the need for qualitative expertise in order to reliably appraise qualitative research (Long et al., 2020). One alternative to the CASP that may have supported the quality appraisal are four guiding principles outlined by Yardley (2000). The principles include sensitivity to context, commitment and rigour, transparency and coherence, and impact and importance. Yardley (2000) suggested that her principles for quality in qualitative research are purposefully broad to capture the characteristics of quality that may differ within separate qualitative methodologies. Applying Yardley's principles to the present review may have yielded different findings (2000). However, as the principles are broad and therefore subjective, they assume the researcher to have a good working knowledge of qualitative research. To support robust quality appraisal for novice researchers, Long et al. (2020) suggest that the CASP should be supplemented by a further item, which is influenced by Yardley's principles (2000). That is, a further item to prompt assessment of the clarity and coherence of the study's ontological and epistemological underpinnings. Therefore, future novice researchers completing a qualitive review may consider such methods to strengthen their quality appraisal methods.

Conclusions

This critical appraisal further discusses issues pertinent to the reports presented in sections one and two. In doing so, this has highlighted some of the relevant strengths and limitations of the work presented and provided useful insights for researchers completing future work of a similar nature. The strengths and limitations discussed illustrate attempts to engage critically and reflexively throughout the research process. This means that the systematic literature review and empirical paper have the potential to provide useful and important insights into the emotional and psychological experience of movement disorders. Such insights are a welcome addition to the evidence base which I hope will provide further avenues for research and have the potential to support the clinical practice of health professionals through improved understandings of issues faced by individuals and couples diagnosed and living with movement disorders such as Parkinson's and dystonia.

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Tables and Figures

Figure 1

Extract from reflective journal during interviews and analysis

Interview 1	Conversation flowed; rapport achieved. Chosen interview mode – helped achieve rapport? Noting feelings of empathy toward difficult experiences, attention drawn here. Need to ask more about early journey, symptoms. How did that impact you? How did it affect you? How did it make you feel? thinking about time since first noticing symptoms and memory for detail.
Interview 2	It was a less rich and slightly different feel than the previous interview. Potentially an example of when things go well during diagnosis journey – the experience is simplified? Noticing people are at different stages – mindful to not put my meanings on it. Wanted to focus more/ sit with the noticed symptoms waiting for appointment acute phase. What was this like?
Interview 3	I felt like I was more confident with p3 compared to p1 making annotations and groupings during coding phase. I found this an easier process than previously. Because the participant provided more depth in their interview responses, and it shows in my groupings?
Interview 4	Still some reflection, sense making coming in from my side. More able to listen and respond openly. Although it appeared less 'rich' still used full hour?

Section 4: Ethics Section

Word Count (excluding references, tables and appendices): 5,353

Louise Glover

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

All correspondence should be sent to:
Louise Glover
Health Innovation Campus
Lancaster University
Sir John Fisher Drive
Lancaster,
LA1 4AT

Email : l.harris7@lancaster.ac.uk

Ethics Cover Page and Contents

The following section contains the documents relevant to the ethics application for the empirical paper in section 2. Please note, in the following documents the name Louise Harris is used as this is the researcher's maiden name.

Contents

Ethics Proposal

NHS Health Research Authority Application

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)
The experience of being diagnosed with neck dystonia v 0.1

1. Is your project research?

(•) Yes ( ) No

2. Select one category from the list below:

( ) Clinical trial of an investigational medicinal product
( ) Combined trial of an investigational medicinal product and an investigational medical device
( ) Clinical investigation or other study of a 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)*

[x] 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 the research e.g. NHS support costs) for this study provided by a NIHR Biomedical Research Centre (BRC), NIHR Applied Research Collaboration (ARC), NIHR Patient Safety Translational Research Centre (PSTRC), or an NIHR Medtech and In Vitro Diagnostic Co-operative (MIC) 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 (CRN) provides researchers with the practical support they need to make clinical studies happen in the NHS in England 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, information from your IRAS submission will automatically be shared with the NIHR CRN. Submission of a Portfolio Application Form (PAF) is no longer required.

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 is taking part in doctoral-level study while employed by Lancaster and South Cumbria Foundation Trust.
The study is part of the thesis activity for the Doctoral programme in Clinical Psychology.

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 familar to lay reviewers of the application.

Short title and version number: (maximum 70 characters - this will be inserted as header on all forms)
The experience of being diagnosed with neck dystonia v 0.1

PART A: Core study information

1. ADMINISTRATIVE DETAILS

A1. Full title of the research:

The experience of being diagnosed with neck dystonia: An interpretative phenomenological analysis study

A2-1. Educational projects

Name and contact details of student(s):

Student 1

	Title	Forename/Initials	Surname
	Miss	Louise	Harris
Address	Health Innovation One Sir John Fisher Drive Lancaster University		
Post Code	LA1 4AT		
E-mail	l.harris7@lancaster.ac.uk		
Telephone	0000		
Fax	0000		

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	Health Innovation One		

	Sir John Fisher Drive
	Lancaster University
Post Code	LA1 4AT
E-mail	f.eccles@lancaster.ac.uk
Telephone	01524 592897
Fax	0000

Academic supervisor 2

	Title	Forename/Initials	Surname
	Dr	Clare	Dixon
Address	Health Innovation One		
	Sir John Fisher Drive		
	Lancaster University		
Post Code	LA1 4AT		
E-mail	c.dixon3@lancaster.ac.uk		
Telephone	01524 593492		
Fax	0000		

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 Louise Harris	☑ Dr Fiona Eccles ☑ Dr Clare Dixon

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
	Dr	Fiona	Eccles
Post	Lecturer		
Qualifications	2012 Postgraduate Certificate in Academic Practice, Lancaster University		
	2011 Foundation Course in Systemic Practice, University of Leeds		
	2009 Doctorate in Clinical Psychology, Lancaster University		
	2005 Graduate Diploma in Psychology (Distinction), Oxford Brookes University		
	2004 Qualifying Certificate in Psychology, Oxford Brookes University		

	2004 DPhil Atmospheric, Oceanic and Planetary Physics, University of Oxford
	1999 Master of Physics, 1st class, University of Oxford
ORCID ID	0000 0003 1484 2703
Employer	Lancaster University
Work Address	Health Innovation One
	Sir John Fisher Drive
	Lancaster University
Post Code	LA1 4AT
Work E-mail	f.eccles@lancaster.ac.uk
* Personal E-mail	N/A
Work Telephone	01524592807
* Personal Telephone/Mobile	01524592807
Fax	0000

** 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.*

	Title	Forename/Initials	Surname
	Mrs	Becky	Gordon
Address	Head of Research Quality and Policy		
	Lancaster University		
	Lancaster		
Post Code	LA1 4YT		
E-mail	sponsorship@lancaster.ac.uk		
Telephone	0000		
Fax	0000		

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:	09/11/2020
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
N/A	N/A

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.
N/A

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.*

Dystonia is the name for a group of rare movement disorders which lead to tightening of muscles in certain areas of the body that restrict movement and activity. The most common type, usually experienced in adulthood, is neck dystonia, also known as cervical dystonia or spasmodic torticollis. The cause of neck dystonia is unclear, and no cure exists. It involves tightening of the muscles in the neck and head and can cause significant pain and disablement. There is often a difficult journey to diagnosis involving assessment by a number of health professionals, a long wait (five to seven years) and misdiagnosis. Research in other conditions suggests that a difficult experience during the pre-diagnosis phase can make adjusting to living well with the condition more of a challenge and in dystonia it is suggested that a difficult journey to diagnosis may discourage individuals from accessing support, particularly in relation to their mood. This study aims to carry out in-depth interviews with people who have been recently diagnosed with neck dystonia to understand the meaning that people make from this experience. This will help in considering how services may better support people.

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.

The study will require participants to reflect and talk about in-depth, what may have been a difficult experience. To limit the distress this may cause, the study will exclude anyone who has received a diagnosis of cervical dystonia within the previous six months as it is felt that this would in most cases be sufficient time to have had some emotional adjustment to the diagnosis yet still be able to recall their experience with accuracy. I have discussed this with an expert by experience who is advising the project about any issues that may be relevant to the participant group. The expert by experience felt that 6 months should be sufficient time for individuals to have somewhat come to terms with their diagnosis and not be in the midst of distress this may have caused.

While it is not expected that participation in this study should cause any significant distress, there is the potential for sensitive material to arise during the interview. The interviewer will also be mindful of the wellbeing of the participant throughout the interview. Participants will be given time to think about their answers and will be encouraged to take a break if the need arises. Participants will also be able to stop the interview at any time and offered the opportunity to reschedule if they so wish.

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
- [x] Cross-sectional study
- [ ] Database analysis
- [ ] Epidemiology
- [ ] Feasibility/ pilot study
- [ ] Laboratory study
- [ ] Metanalysis
- [x] Qualitative research
- [ ] Questionnaire, interview or observation study
- [ ] Randomised controlled trial
- [ ] Other (please specify)

N/A

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

How do people experience the journey to diagnosis in neck dystonia?

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

N/A

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

Dystonia is the name given to a group of neurological movement disorders that cause muscular contractions in one or more parts of the body which can lead to abnormal posture, repetitive or jerking movements and it is the third most common movement disorder diagnosis. Many people experience significant pain, disablement, depression, anxiety, fatigue and sleep disturbance. The impact on quality of life can be significant in relation to the physical disability including effects on daily activities, work and social activity. Effective treatments are available that can significantly improve quality of life. But often patients experience difficulties and delays arriving at an accurate diagnosis. Patients are assessed by an average of three health professionals and the journey to diagnosis can go on for between one and seven years, and even longer in some cases.

The majority of research in the area of patient experience of diagnosis in dystonia has been completed using quantitative methods. Studies have shown that the process of diagnosis is often delayed, patients are repeatedly assessed and may receive incorrect diagnoses initially before themselves prompting further investigations. Often individuals' symptoms are explained as being related to a mood disorders such as anxiety or depression. The only available qualitative study focused on experiences of living with dystonia. An important theme emerged which was the difficulties participants experienced in their journey to diagnosis and the impact this had had for them. The researcher found that participants felt dismissed and stigmatised as a result of symptoms being explained as related to issues such as anxiety and depression. Having finally received a diagnosis of dystonia, participants suggested they were wary of accessing support related to their mood due to a perceived risk that they may be further stigmatised. In other long term conditions such as Parkinson's disease, motor neuron disease and cancer it is suggested that difficulties in the journey to diagnosis can lead to distress and difficulties with adjusting to the condition. It is therefore important to address the gap in understanding that exists in dystonia; a lack of understanding of the patient's perspective of the journey to diagnosis of dystonia using qualitative methodology. The existing qualitative study which aimed to understand the experience of living with dystonia included participants with differing diagnoses of dystonia who had therefore had different experiences. This study will include participants with a diagnosis of neck dystonia only as this is the most common form.

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.

This study will use the qualitative methodology, interpretative phenomenological analysis (IPA) which aims to produce an in-depth understanding of the meaning making and sense making that individuals experience in relation to a particular experience. This is a suitable methodology for the current study as it fits with the aim of understanding the experience of the journey to diagnosis in neck dystonia. This style of study intends to be guided by the participants, as to what about their experience they found to be important. Therefore, the research question is broad and open; "how do people experience the journey the diagnosis in neck dystonia?".

This study requires a small number of participants (8-12 people), who will be asked to provide some demographic information and take part in a semi-structured interview which are expected to take between 60 and 90 minutes per interview. Interviews will be conducted remotely, either by telephone or video call and to be arranged at a convenient time for the participant. A topic guide will provide some structure for the interview. Topic areas will focus on the experience of initially noticing symptoms, the experience of accessing support or advice related to the symptoms, the experience of being diagnosed, the experience of coming to terms with the diagnosis and the impact of the diagnosis journey on the person. As this is a guide only, the interviewer will also be guided by the issues that are important to the participant. An expert by experience with a diagnosis of neck dystonia has been consulted regarding the appropriateness of the interview topics and structure.

There will be two modes of recruitment, the second is to be activated if the first mode does not gather a sufficient number of participants. Initially, potential participants will be identified via the Salford Royal Foundation Trust (SRFT) Manchester Centre for Clinical Neuroscience. Potential participants will either be sent a research pack through the post or be handed this during routine clinic appointments. This will include a letter introducing the study from their clinician, a Participant information sheet (PIS) and copy of the consent form which will be obtained verbally before commencing the interview. All materials have been produced in consultation with an expert by experience to ensure that they are appropriate and accessible. Potential participants will consent to being contacted by the researcher or they will be able to contact the student directly by email. Potential participants will be able to discuss the study further and ask questions with the student or with the academic supervisor if they wish to. ollowing this, if potential participants are interested in taking part, the student will arrange a convenient time for the interview to take place and negotiate as to whether this will take place by phone or video call.

If the previously stated mode of recruitment does not gather the participant numbers required for the study, the student will advertise the study via Dystonia UK's website and social media channels using pre-prepared wording. A link will be provided which will direct the potential participants to an electronic version of the PIS and a copy of the consent form. Potential participants will be asked to contact the student directly by email to register their interest in taking part.

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?

- [x] 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.
An expert by experience who is a volunteer for Dystonia UK, has agreed to consult on the design and materials for this study. This input has led to alterations around consent procedures and a change of wording in the title and throughout the study, from cervical dystonia to the more commonly used among patients - 'neck dystonia'.

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
- [x] Neurological
- [ ] Oral and Gastrointestinal
- [ ] Paediatrics
- [ ] Renal and Urogenital
- [ ] Reproductive Health and Childbirth
- [ ] Respiratory
- [ ] Skin
- [ ] Stroke

Gender:		Male and female participants
Lower age limit:	35	Years
Upper age limit:	75	Years

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

To be eligible to take part in this study participants must be between age 35 and 75. They must have a diagnosis of idiopathic adult onset isolated cervical dystonia (neck dystonia) which they have received within the previous three years and no sooner that the previous six months. This is in order for participants to have had sufficient time to have moved past the initial impact of the diagnosis and be able to reflect as accurately as possible on their experience. This time frame was agreed in consultation with an expert by experience based on their own experience of diagnosis. Participants must be able to speak fluently in English, be willing to participate, to be interviewed by telephone or video call and to be able to provide informed consent.

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

Participants will not be able to take part if they have any additional physical or mental health difficulties which for any reason may have significantly altered their experience of diagnosis. For example, another movement disorder diagnosis, cognitive impairment or a significant level of psychological distress, for example severe depression or psychosis.

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
Introduce study and provide the PIS and consent to contact form.	1	0	5 minutes	SRFT Manchester Centre for Clinical Neuroscience. The person's clinician will either hand them the research pack during a clinic appointment or write to inform them of the study.
Further discussion about the study with participant and answering questions.	1	0	15 minutes	Either student contacts to answer questions and discuss the study further or individual contacts student directly with questions.
Seeking consent	1	0	10 minutes	Student to read out consent form at the beginning of remote interview arranged at a convenient time outside of any clinic time either by phone or video call.
Interview	1	0	60-90 minutes	Student to conduct interview. To take place remotely by phone or video call.

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

Between 60 and 90 minutes

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.

While it is not expected that participation in this study should cause any significant distress, there is the potential for sensitive material to arise during the interview. The interviewer will also be mindful of the well being of the participant throughout the interview. Participants will be given time to think about their answers and will be encouraged to take a break if the need arises. Participants will also be able to stop the interview at any time and offered the opportunity to reschedule if they so wish.

Participants will be advised that they are able to withdraw their consent up to two weeks after the interview if they wish to. After this time the information will have been anonymised and coded.

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:

The interviewer will also be mindful of the wellbeing of the participant throughout the interview. Participants will be given time to think about their answers and will be encouraged to take a break if the need arises. Participants will also be able to stop the interview at any time and offered the opportunity to reschedule if they so wish. If participants experience significant distress, they will be encouraged to raise appropriate issues with their health care professional, their GP or to access relevant local support services, the information of which will be provided as part of their participant information sheet.

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

There will be no direct benefit to participants. However, it is hoped that the opportunity to contribute to the development of health services for people with dystonia and to register their views and experience will justify any cost or risk of harm to participants.

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

Interviews will be conducted remotely, therefore there should be no risks involved in the undertaking of interviews. If content of interviews is of emotional relevance to the student, they are encouraged to discuss this during supervision with the academic supervisor.

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).*

Potential participants will be identified by accessing patient information held by the Manchester Centre for Clinical Neuroscience at Salford Royal Foundation Trust. The patient information will be accessed by a member of the clinical team. Once identified, potential participants will either be given information about the study during clinic appointments or sent in the post - this will include a letter from the clinician introducing the study, the participant information sheet and a copy of the consent form. They will be asked to provide consent to be contacted by the research team or potential participants may contact the student directly.

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:
Yes, personal information will be required to identify potential participants to approach who fit the study criteria. Details such as diagnosis, age, and name will be needed in order to identify suitable participants.

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.*

A member of the patients care team only will have access to patient records in order to identify potential participants, check whether they meet the inclusion criteria and make the initial approach to patients (by mail or via clinic appointment). Explicit consent will be provided before the research team are able to access personal contact details or the patient will contact the research team directly.

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

○ Yes ◉ No

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).

A second mode of recruitment will go ahead if the initial route through SRFT movement disorder service does not gather the number of participants required for this study. The Dystonia Society UK have agreed, pending ethical approval, to advertise the study on their website and through their social media channels including facebook and twitter.

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

Participants recruited by the SRFT route will be approached by their clinician initially or they will be posted a research pack from their clinician. If participants are recruited by Dystonia UK they will see the advert and contact the student 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 be able to view either a hard copy or electronic version of the participant information sheet. They will be offered an opportunity to discuss the participant information sheet with the student either verbally over the phone or by email where any questions will be answered. At the beginning of the remote interview, the student will read out the consent form and participants will be able to respond yes or no to the statements. The consent conversation will be recorded by use of a digital recording device and stored separately to the interview recording.

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

N/A

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

If No, how will it be recorded?

As interviews will take place remotely it will not be possible to obtain written informed consent. It was considered as to whether electronic consent could be obtained by using a Qualtrics survey that could be sent to participants. This was discussed with an expert by experience who expressed concern this may require too many interactions and thus participants may be discouraged from taking part. It was therefore decided that consent would be given verbally and the student would record and store this separately to the interview recording, in order to streamline the process.

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

Potential participants will have as much time as they like to consider whether they would like to take part during the recruitment phase of the study.

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)*

Unfortunately there is no funding available for this study to enable the use of interpreters. Therefore, participants will be required to speak fluently in English.

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:
N/A

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
- [x] 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
- [x] Use of personal addresses, postcodes, faxes, emails or telephone numbers
- [x] Publication of direct quotations from respondents
- [ ] Publication of data that might allow identification of individuals
- [x] Use of audio/visual recording devices
- [x] 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
  - [x] University computers
  - [ ] Private company computers
  - [ ] Laptop computers

Further details:
N/A

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

All personal contact information will be stored separately to other data in a file space on the Lancaster University server or on a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). This information will be destroyed once the project has been completed and once summary reports have been disseminated if requested by participants.

Audio recording will be encrypted (that is no-one other than the researchers will be able to access them) and stored separately in a secure file space on the Lancaster University server or on a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). After this they will be deleted from the recording device. Audio recordings will be permanently deleted once the project has been examined. Audio recording of consent, however, will be similarly stored in a secure file space on the Lancaster University server or on a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). They will be stored for ten years by the Clinical Psychology Programme, following assessment or publication, whichever is longer.

The audio recording of the interview will be typed up and stored in a file space on the Lancaster University server or on a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). They will be encrypted (that is no-one other than the researcher will be able to access them) and the computer itself password protected. The typed version of the interview will be stored securely by the Doctorate in Clinical Psychology programme for 10 years after the project has been examined, or 10 years following publication, whichever is longer.

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.*

The typed version of the interviews will be made anonymous by removing any identifying information including names which will be relaced with pseudonyms. Anonymised direct quotations from the interviews may be used in the reports or publications from the study and names will not be attached to them. All reasonable steps will be taken to protect the anonymity of the participants involved in this project.

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.*

Participants who are recruited through Manchester Centre for Clinical Neuroscience will be required to provide consent for their personal contact information to be shared with the research team.

All personal contact information will be available to the student, the chief investigator and academic supervisor only.

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?

The data will be analysed by the student with support from the academic supervisor. The analysis will take place on a secure file space on the Lancaster University server or on a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One rive).

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	2012 Postgraduate Certificate in Academic Practice, Lancaster University 2011 Foundation Course in Systemic Practice, University of Leeds 2009 Doctorate in Clinical Psychology, Lancaster University 2005 Graduate Diploma in Psychology (Distinction), Oxford Brookes University		

	2004 Qualifying Certificate in Psychology, Oxford Brookes University
	2004 DPhil Atmospheric, Oceanic and Planetary Physics, University of Oxford
	1999 Master of Physics, 1st class, University of Oxford
Work Address	Health Innovation One Sir John Fisher Drive Lancaster University
Post Code	LA1 4AT
Work Email	ecclesf@lancaster.ac.uk
Work Telephone	01524592807
Fax	0000

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

- ○ Less than 3 months
- ○ 3 – 6 months
- ◉ 6 – 12 months
- ○ 12 months – 3 years
- ○ Over 3 years

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.*

Transcripts will be stored electronically, they will be stored until12 months encrypted and stored separately to the recordings in a file space on the Lancaster University server or to a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). Access will be to the student, research supervisors, and the research coordinator. The anonymised transcripts will be stored for ten years, or ten years following publication (whichever is longer), by the research coordinator of the Doctorate in Clinical Psychology at Lancaster University, overseen by the chief investigator. After this time, they will be destroyed.

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.
No appropriate database has been identified.

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 5-1.

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

N/A

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

When audio recordings are transcribed participant names will be replaced with pseudonyms. If other individuals are named during the interview such as health professionals or significant others, these details will also be replaced. During the publication process, care will be taken to try to avoid any combination of incidental details which could lead to individuals being identifiable.

A53. How and when will you inform participants of the study results?

If there will be no arrangements in place to inform participants please justify this.
Participants will be offered a copy of summary results from this study. The study findings will be available via Dystonia UK.

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:
The project has been reviewed by the research staff at the host institution and by the educational supervisors. The sponsor has also reviewed the project before providing sponsorship.

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:	0

Further details:
N/A

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.*

This is a typical sample size for a qualitative IPA study, which aims to produce an in-depth analysis of a small number of participants' experiences.

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 an inductive double hermeneutic process, whereby, the student will use IPA to make sense of and understand the meaning behind rich accounts of meaning and sense making provided by the participants. This study will follow the steps for IPA analysis set out in the 2009 book 'Interpretative Phenomenological Analysis: Theory, Method and Research' by Smith, Flowers & Larkin (2009). Each interview recording will be transcribed verbatim by the student and analysed individually. Individual transcripts will be broken down and coded in appropriate 'chunks' with initial observations recorded. Next, emergent themes will be identified followed by the identification of super-ordinate themes and themes across transcripts. The researcher will return to the original data regularly to retain the participant voice. The use of supervision will allow the student to hold an awareness of their own biases and influences on the meaning they make of the participant's accounts.

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	Christopher	Kobylecki
Post	Consultant Neurologist		
Qualifications	MB ChB BSc (Hons) PhD FRCP		
Employer	Salford Royal Foundation Trust		
Work Address	Stott Lane Salford Greater Manchester		
Post Code	M6 8HD		
Telephone	01612062574		
Fax	0000		
Mobile	0000		
Work Email	christopher.kobylecki@srft.nhs.uk		

A64. Details of research sponsor(s)

A64-1. Sponsor

Lead Sponsor

Status: ○ NHS or HSC care organisation
◉ Academic
○ Pharmaceutical industry
○ Medical device industry
○ Local Authority
○ Other social care provider (including voluntary sector or private organisation)
○ Other

Commercial status: Non-Commercial

If Other, please specify: N/A

Contact person

Name of organisation	Lancaster University
Given name	Becky
Family name	Gordon
Address	Lancaster University

Town/city	Lancaster
Post code	LA1 4YT
Country	United Kingdom
Telephone	+44 (0)1524 592981
Fax	0000
E-mail	sponsorship@lancaster.ac.uk

Legal representative for clinical investigation of medical device (studies involving Northern Ireland only)
Clinical investigations of Medical Devices that take place in Northern Ireland must have a legal representative of the sponsor that is based in Northern Ireland or the EU

Contact person

Name of organisation	
Given name	
Family name	
Address	
Town/city	
Post code	
Country	
Telephone	
Fax	
E-mail	

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
- [x] No application for external funding will be made

What type of research project is this?

- (•) Standalone project
- ( ) 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:
N/A

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
	Mrs	Katie	Doyle
Organisation	Salford Royal NHS Foundation Trust		
Address	Summerfield House, 1st Floor		
	544 Eccles New Road		
	Salford		
Post Code	M5 5AP		
Work Email	katie.doyle@srft.nhs.uk		
Telephone	0161 206 4734		
Fax	0000		
Mobile	0000		

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: 01/04/2021
Planned end date: 31/12/2021
Total duration:
Years: 0 Months: 8 Days: 31

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 1

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:*

- [x] NHS organisations in England 1
- [ ] NHS organisations in Wales
- [ ] NHS organisations in Scotland
- [ ] HSC organisations in Northern Ireland
- [ ] GP practices in England
- [ ] GP practices in Wales
- [ ] GP practices in Scotland
- [ ] GP practices in Northern Ireland
- [ ] Joint health and social care agencies (eg community mental health teams)
- [ ] Local authorities
- [ ] Phase 1 trial units
- [ ] Prison establishments
- [ ] Probation areas
- [ ] Independent (private or voluntary sector) organisations
- [ ] Educational establishments
- [ ] Independent research units
- [ ] Other (give details)

N/A

Total UK sites in study: 1

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 chief investigator and academic supervisors will monitor the conduct of this research study by working closely with the student who will carry out the research activities. The CI and academic supervisors will meet with the student for regular supervision. The student's academic supervisors will listen to an early interview recording to provide feedback about the interview style and questioning. Academic supervisors will oversee the data analysis and write up of research findings.

A76. Insurance/ indemnity to meet potential legal liabilities

Note: In this question '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)

N/A

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?

○ Yes ◉ No ○ Not sure

9. Has the study been the subject of a scientific review/opinion (Expert Panel)?

○ Yes ○ No

If yes, please provide a copy of the review as part of your application.

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			
	◉ NHS/HSC Site	Forename	Christopher
	○ Non-NHS/HSC Site	Middle name	unknown
		Family name	Kobylecki
Organisation name	SALFORD ROYAL NHS FOUNDATION TRUST	Email	christopher.kobylecki@srft.nhs.uk
Address	SALFORD ROYAL STOTT LANE SALFORD GREATER MANCHESTER	Qualification (MD...)	MB ChB BSc (Hons) PhD FRCP
Post Code	M6 8HD		
Country	ENGLAND	Country	United Kingdom
IN2			
	○ NHS/HSC Site	Forename	Victoria
	◉ Non-NHS/HSC Site	Middle name	unknown
		Family name	Wareham
		Email	victoria@dystonia.org.uk
Institution name	Dystonia UK	Qualification (MD...)	unknown
Department name	Head Office		
Street address	3rd Floor, 89 Albert Embankment	Country	United Kingdom
Town/city	London		
Post Code	SE1 7TP		
Country	United Kingdom		

Appendices

Appendix 4-A: Research Protocol

Introduction

Dystonia is the name given to a group of neurological movement disorders that cause muscular contractions in one or more parts of the body which can lead to abnormal posture, repetitive or jerking movements (Albanese et al., 2013; Jinnah & Factor, 2015). It is the third most common movement disorder diagnosis affecting approximately 16.43 per 100,000 people worldwide (Steeves et al., 2012).

Classification of dystonia has changed over time, highlighting the evolution of understanding of this heterogenous condition (Bressman, 2004; Fahn, 1988; Fahn & Eldridge, 1976; Phukan et al., 2011). Currently classification is based upon two axes (Albanese et al., 2013). The first relates to clinical characteristics, including age of onset, bodily location and distribution, temporal pattern, coexistence of other movement disorders and other neurological symptoms. Axis II relates to aetiology; which remains a developing area as the ability to determine this is limited. The most prevalent form is isolated focal dystonia, which may develop as a result of genetic aetiology, but in many cases of those that develop in adulthood, the cause is idiopathic. Different diagnoses exist within the classification of idiopathic focal dystonia, depending on the area affected and the most common is dystonia involving the neck muscles, which is also referred to as spasmodic torticollis and cervical dystonia (Defazio et al., 2004, 2013). Individual differences in presentation occur but symptoms include tightening of the muscles in the neck and head, causing twisting of the head and jerking movements (Albanese, 2013; Defazio 2004; Tiderington, 2013). There are no significant differences in presentation between genders, however, it is more prevalent in females (LeDoux et al., 2016; LaHue et al., 2020). The most common age of onset is mid-

forties, but it can occur anywhere between ... (LeDoux et al., 2016; LaHue et al., 2020; Ndukwe et al, 2020).

The symptoms of neck dystonia can have a significant impact on the lives of those affected. Many people experience significant pain, disablement, depression, anxiety, fatigue and sleep disturbance (Charles et al, 2014; Lewis et al., 2008; Novaretti et al., 2019). The impact on quality of life can be significant in relation to the physical disability including effects on daily activities, work and social activity (De Pauw et al., 2017; Page et al., 2007). There is developing understanding of symptoms including anxiety, depression and sleep disorders which have been found be prevalent and potentially primary or preceding motor symptoms of neck dystonia (Eichenseer et al., 2014; Fabbrini et al., 2010; Ndukwe et al, 2020; Stamelou et al., 2011), although psychosocial factors may also play a role. These symptoms are among the highest indicators for impact on quality of life along with a perceived stigma of this ill-understood condition (Ben-Shlomo et al., 2002; De Pauw et al., 2017; Girach et al., 2019; Krzak-Kubica et al., 2018; Mordin et al., 2014; Pekmezovic et al., 2009; Tomic et al., 2016). This has led several researchers and practitioners in the field to stress that assessment should be holistic and capture impact on physical disability as well as patient's mental health and wellbeing in order to consider appropriate treatment and support (Ben-Shlomo et al., 2002; Girach et al., 2019; Jinnah & Factor, 2015; Tomic et al., 2016; Page et al., 2007;).

Various symptom relieving treatments are available which include oral medications, botulinum toxin injections and surgical treatments such as deep brain stimulation (DBS) and selective peripheral denervation surgery (Adam & Jankovic, 2007; Cacciola et al., 2010; Jankovic, 2013; Krauss, 2010; Termsarasab et al., 2016; Zoons et al., 2012). These treatments can have a significant impact on improving the quality of life for those affected by dystonia (Adam & Jankovic, 2007; Chou et al., 2012; Girach et al., 2019; Skogseid et al.,

2007; Zoons et al., 2012). Good effect is also reported from physiotherapeutic intervention particularly when applied in addition to other treatments, however, further large-scale studies are required (Girach et al., 2019; Isabel Useros-Olmo et al., 2020; Pauw et al 2014). Increasing attention has been paid to the benefits of psychosocial interventions for dystonia such as education, counselling and more formal psychological therapies (Ben-Shlomo, 2002; Bernstein et al., 2016; Comella & Bhatia, 2015; Jinnah & Factor, 2015; Lewis et al., 2008). Potential benefits include, increased treatment acceptance, decreased severity of physical symptoms, reduced anxiety, reduction in perceived stigma and improved quality of life (Ben-Shlomo, 2002; Bernstein et al., 2016 Jinnah & Factor, 2015).

It is crucial that individuals are accurately diagnosed in order to receive the available and effective treatments. However, despite neck dystonia being the most common form, patients often experience difficulties arriving at an accurate diagnosis and many cases are undiagnosed (LaHue et al., 2020; Macerollo et al., 2015; Steeves et al., 2012; Tiderington et al., 2013). On average, patients with neck dystonia are assessed by three health professionals before receiving a correct diagnosis (Betram & Williams, 2016). Some researchers estimate at least a one-year delay before receiving an accurate diagnosis while others suggest an average wait time of between five and seven years, or even longer (Betram & Williams, 2016; Jog et al., 2011; Tiderington et al., 2013). Many initially receive incorrect diagnoses, including other movement disorders such as Parkinson's disease, musculoskeletal conditions, psychogenic explanations, depression and anxiety (Bertram & Williams, 2016; Jinnah & Factor, 2015).

There are many potential reasons for the delays in diagnosis and incorrect diagnoses that patients commonly experience. There is a lack of reliable diagnostic tests and biomarkers, therefore, diagnosis is made clinically by observing symptoms, clinical examination and history taking (Betram & Williams, 2012; Camargos, & Cardoso, 2016).

Difficulties with identification can occur when there is an overlap of symptoms. For example, tremor can be present in dystonia and is often misdiagnosed as essential tremor or Parkinson's disease (Camargos & Cardoso, 2016; Gazewood et al., 2013; Schrag at al., 2000). It is recommended that suspected cases are referred to a specialist for assessment where even so, accuracy of diagnosis is below the desired level (Jinnah & Factor, 2015). Referral to a specialist neurologist is dependent on the general practitioner (GP) having some knowledge of the condition in order to consider it as a possibility rather than attribute complaints to more common ailments such as musculoskeletal complaints, anxiety or depression. Neck dystonia is under-recognised publicly which contributes further to delays because individuals may seek alternative avenues of support rather than visiting their GP (LaHue et al., 2020; Macerollo et al., 2015; McNeill et al., 2004; Tiderington et al., 2013). Protracted diagnosis pathways have been a focus of researchers in other long-term conditions, such as cancer. Miles et al. (2017) found that one third of patients undergoing investigations for suspected cancer have clinically significant levels of distress which was comparable to those with a confirmed diagnosis. Other studies have shown that people with rare cancers, where diagnosis is often delayed and involves multiple referrals and visits to a GP, are likely to be dissatisfied with their experience and have a lack of trust and confidence in health professionals (Larsen et al., 2011; Mendonca et al., 2016). Having trust in your healthcare professional is associated with fewer symptoms and more constructive health behaviours (Birkhäuer et al., 2017). Research in motor neuron disease (MND) suggests that a difficult pre-diagnostic phase increases uncertainty which may lead to experiences of anxiety and depression (Remm et al., 2019). Anxiety and depression can impact negatively on adjustment to illness (de Ridder et al., 2008).

Adjustment is the necessary process required to live well with long-term illness (Taylor & Aspinwall, 1996). Several models have attempted to conceptualise this process.

Cognitive models highlight the need to make sense of and understand the illness to discover new ways of coping and gain a sense of control (Lazarus & Folkman, 1984; Taylor, 1983). The health belief model (HBM, Irwin et al., 1988) suggests that behaviours that support adjustment are more likely, if the perceived benefit is high and barriers are perceived as low. The self-regulation model (SRM, Leventhal & Nerenz, 1984) considers maintaining or gaining a new sense of self-identity as a central agent to successful adjustment. To integrate the many contributors to adjustment it is suggested that individuals experience a complex interplay of factors such as personality, stressors, appraisals, psychosocial functioning, coping strategies, affect, disease activity and physical adjustment; hence it is suggested that successful adjustment depends on achieving an equilibrium of the physical, social and psychological impact of the condition (Moss-Morris, 2013; Walker 2004).

The physical and prognostic implications of diagnostic delay in dystonia are well documented whereas there is limited evidence of the psychological implications. Delay to accessing effective treatments and unnecessary healthcare costs for inappropriate and costly tests and treatment are well understood (Bertram & Williams, 2016; Camargos & Cardoso, 2016; Jinnah & Factor, 2015; LaHue et al., 2020; Mcneill et al., 2004). One hypothesis related to the scarcity of research as to the psychological implications is due to a poor uptake of psychological support in this condition, which is thought to be connected to the common experience of patients receiving incorrect diagnoses and feeling that symptoms are ‘all in their head’ (Bernstein et al., 2016). One qualitative study by Morgan et al. (2019) discovered that an important theme for those living with dystonia was the difficulties they experienced during their journey to diagnosis. This led to patients feeling dismissed and stigmatized and in turn, leads to wariness to access psychological support at the risk of being further stigmatized.

Research in the field of diagnostic delay in dystonia has been largely made up of quantitative studies with comparatively little qualitative evidence available. Through quantitative means, researchers have suggested that diagnostic delay leads to months or years of frustration and uncertainty that can lead to mistrust of health providers and reduced uptake of suggested treatments (Abdo et al., 2010; Jinnah & Factor, 2015; Jog et al., 2011; LaHue et al., 2020). Such studies contained limitations in time since diagnosis and resultant participant recall bias (Jinnah & Factor 2015; Macerollo et al., 2015). Qualitative literature has aided clinical understandings and appropriate support for the psychological impact of experiences of diagnosis in other conditions, such as multiple sclerosis and Parkinson's disease (Borkoles et al., 2016; Strickland et al., 2017; Warren et al., 2016). The only available qualitative study in dystonia used interpretative phenomenological analysis (IPA) to gain an in-depth understanding of individual experiences of living with dystonia (Morgan et al., 2019). From this emerged a dominant theme across accounts related to the difficult journey to diagnosis. There were some limitations to this study such as reduced homogeneity of the sample which included individuals with different subtypes of dystonia and therefore, differing experiences. Furthermore, the study did not intend to understand the experience of diagnosis, rather it was a theme that emerged from an attempt to understand what it is like to live with dystonia.

IPA was the methodology for the related qualitative research and remains the most suitable approach for this study. IPA is a methodological framework which seeks to gain an in depth understanding of the meaning made of life experiences (Smith & Osborn, 2008). To increase homogeneity of the sample this study will focus solely on patients who have a diagnosis of neck dystonia, as this is the most common subtype of dystonia. The diagnosis will aim to be confirmed by a clinical specialist in the field by recruiting patients from a specialist movement disorder. To increase validity further, the participants included are to be

within the most common age range for diagnosis to further reduce variance of experience. Furthermore, a restriction on the time since diagnosis will be set to increase recall accuracy.

Research Aim

The aim of this study is to gain an in depth understanding of what it is like for patients to experience the journey to diagnosis in neck dystonia and therefore answer the following research question: how do people experience the journey to diagnosis in neck dystonia? This will provide direction for future research and recommendations for future care delivery.

Method

Design

IPA was chosen as it is the most suitable methodological framework for this study. In consultation with an expert by experience it was suggested that interviews would be a suitable format to ask a person about their diagnosis experience because it would likely be an important experience that individuals would value talking in detail about. As a phenomenological approach, IPA is a framework that allows for deep reflection on a particular experience (Smith et al., 2009). From previous studies, there is some understanding about the potential impact of the journey to diagnosis and the physical, psychological and social challenges. However, we do not yet understand this from a qualitative perspective and the meanings individuals make of this experience is unknown. IPA analyses rich and emotional data, verbal accounts, descriptions and stories in an attempt to make sense of and understand the meanings made of personal experiences (Smith & Osborne, 2008).

IPA studies commonly involve a purposive sample, as is the case with this study. The sample will be selected in line with the eligibility criteria outlined. This is to achieve, as much as is possible, a homogenous sample, which is particularly important in IPA as it allows for a sample for whom the research question is of significance and personal concern (Smith et al., 2009). As such, participants will be in a position to reflect and talk in detail on their experiences, in doing so providing rich data for analysis.

Participants

Since this is a qualitative IPA study, which aims to produce an in-depth analysis of participants' experiences, a sample of 8-12 participants are required (Smith et al., 2009). Participants will be recruited with adherence to the following criteria.

Inclusion criteria

To be eligible for inclusion, participants must:

- Be of any gender
- Be between age 35 and 75 inclusive.
- Have a diagnosis of idiopathic adult onset isolated cervical dystonia (neck dystonia).
- Have received the diagnosis within the previous three years and no sooner than the previous six months. This time frame is to allow for initial adjustment to have taken place while allowing recall to be as accurate as possible and has been decided on in consultation with an expert by experience and with reference to appropriate research evidence.
- Be able to speak fluently in English to enable participation in the interview.
- Be willing to participate, be interviewed by telephone or video call and able to provide informed consent.

Exclusion criteria

Participants will be excluded if they:

- Have any additional physical or mental health difficulties which may have significantly altered their experience of diagnosis, for example, another movement disorder diagnosis, cognitive impairment or a significant level of psychological distress, for example severe depression or psychosis.

Recruitment

There will be two possible routes for recruitment. The second will be instigated if insufficient patients are recruited via the first route or if this route cannot be followed due to COVID-19 restrictions.

i. Recruiting in collaboration with the Manchester Centre for Clinical Neurosciences, Salford Royal NHS Foundation Trust (SRFT): the patient database will be accessed by a member of the clinical team to identify individuals who meet the criteria for this study. Potential participants will be sent a research pack through the post which will include: a covering letter from the local collaborator, Chris Kobylecki, the participant information sheet (PIS), and a copy of the consent form. Alternatively, potential participants will be advised of the study and provided with the research pack during clinic appointments. Potential participants will either provide consent to be contacted to the clinical team or they may contact the student directly to register their interest in taking part in the study.

ii. A second mode of recruitment will be facilitated by Dystonia UK, who have given permission to advertise the study via their social media channels and via their website, pending ethical approval. A link will be included that will direct potential participants to the Qualtrics PIS and consent form. The study will be advertised using specific wording (Appendix 2).

Consent

Once consent to contact is obtained the student will contact potential participants, or the student will be contacted directly by potential participants. There will be opportunity to discuss the nature and objectives of the study by telephone or email. They will have already read the PIS, but the student will ensure this has been read and understood and provide opportunity for further explanation and questions. At this point the student will ensure that the potential participants meet the required inclusion and exclusion criteria for the study. A

convenient time for interview will be arranged with those who would like to take part. Consent will be obtained verbally prior to beginning the interview. The student will read out the consent statements for the participants to answer either yes or no to signify their consent. This will be recorded separately to the interview.

During the recruitment phase, potential participants will have as much time as they like to consider whether they would like to take part. All interviews will take place remotely by telephone or video call. At the end of the interview, participants will be directed to the PIS which will include details of relevant support agencies. Participants will be able to withdraw their consent for up to two weeks following the interview.

Data collection

Demographic data collection

Before the interview commences, participants will be asked for some demographic information by the student. This will include age, gender, time since diagnosis, ethnicity, living situation (e.g. with partner/children/alone) and employment status.

Interviews

Individual semi-structured interviews will be conducted by the student, Louise Harris. It is expected that interviews will take between 60-90 minutes. Semi- structured interviews are the most appropriate choice for this study as they allow for participants' experiences to guide the investigation and the interview to be flexible to issues raised. The interviewer's role is to ask questions that prompt reflection at a deeper level rather than a surface level summary of what happened (Smith et al., 2009). The interviewer will also need to gently re-orientate the participant if they stray away from the topic area. The topic guide has been structured around existing theory and issues raised through quantitative and qualitative research into dystonia, similar related conditions and broader health psychology theoretical understandings (Appendix 1). An expert by experience with a diagnosis of neck dystonia has been consulted

regarding the appropriateness of the interview topics and structure. Topic areas will focus on the experience of initially noticing symptoms, the experience of accessing support or advice related to the symptoms, the experience of being diagnosed, the experience of coming to terms with the diagnosis and the impact on the person of their diagnosis journey. The student's academic supervisors will listen to an early interview recording to provide feedback about the interview style and questioning. All interviews will take place remotely by telephone or Microsoft Teams as this is the approved online video conferencing software for Lancaster University. The audio from each interview will be recorded on a digital recording device. Audio recordings will be transcribed manually by the student.

Data Analysis

Analysis will be an inductive double hermeneutic process, whereby, the student will use IPA to make sense of and understand the meaning behind rich accounts of meaning and sensemaking provided by participants (Murray & Wild, 2020). This study will follow the steps for IPA analysis identified by Smith, Flowers & Larkin (2009). Each interview recording will be transcribed verbatim by the student and analysed individually. Individual transcripts will be broken down and coded in appropriate 'chunks' with initial observations recorded. Next, emergent themes will be identified followed by the identification of super-ordinate themes and themes across transcripts. The researcher will return to the original data regularly to retain the participants' voice. The use of supervision will allow the student to hold an awareness of their own biases and influences on the meaning they make of the participants accounts.

Data storage

All data generated from this study will be accessible to the student, chief investigator and academic supervisors only. The below specifies the recording and storage processes for each form of data or secure information which will be derived from this study:

i. *Personal contact details*: participants personal contact information will be stored securely in a file space on the Lancaster University server or on a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). This information will be stored separately and destroyed once the project has been completed and once summary reports have been disseminated.

ii. *Consent forms and demographics*: Consent and demographic data audio recordings will be stored separately from the interview transcripts. They will be stored in a file space on the Lancaster University server or on a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). They will be saved in this location as encrypted files and stored for ten years or ten years following publication (whichever is longer), by the research coordinator of the Doctorate in Clinical Psychology at Lancaster University, overseen by the chief investigator. After this time, they will be destroyed.

iii. *Audio recordings:* Each participant interview will be recorded onto a digital recording device and transferred as soon as possible to a file space on the Lancaster University server or to a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). They will be saved in this location as encrypted files and then deleted from the audio recorder. The audio recordings will be deleted following the student's viva voce examination.

iv. *Transcribed interviews:* The audio recordings will be transcribed by the student at which point participant names will be replaced with pseudonyms and any identifiable details removed. Transcripts will be stored electronically, they will be encrypted and stored separately to the recordings in a file space on the

Lancaster University server or to a cloud-based service approved by Lancaster University with similar secure credentials (e.g. One Drive). The anonymised transcripts will be stored for ten years, or ten years following publication (whichever is longer), by the research coordinator of the Doctorate in Clinical Psychology at Lancaster University, overseen by the chief investigator. After this time, they will be destroyed.

Participants will be advised that they can withdraw their consent for up to two weeks after the interview. This information will be contained in the consent form.

Dissemination

It is anticipated that findings will be submitted for publication in a relevant peer reviewed academic journal. If the opportunity arises, this information would also be presented at academic conferences and special interest groups. The research findings will be presented at Lancaster University's Doctorate for Clinical Psychology thesis presentation day. Participants will be given a copy of results and they will be shared with the dystonia community through the research page of the Dystonia UK website and/or newsletters and/or social media.

Practical issues

Costs

It is not expected that any financial cost to the participant will be incurred by participating in this study. There will be no monetary reward for involvement. For participants there will be a time cost of approximately 60 to 90 minutes of their time, plus time required to complete consent forms and answer a demographic questionnaire. The student plans to conduct interviews at a convenient time for participants.

Potential Limitations

As funding is not available for a translator, only participants who speak English will be considered eligible to participate. Further to this, the process of interpretation could prove problematic for in depth IPA analysis.

Patient & Public Involvement

Public involvement has been present in this study from the outset. The study focus, design and materials were developed in consultation with an expert by experience who is a volunteer for Dystonia UK.

Ethical considerations

While it is not expected that participation in this study should cause any significant distress, there is the potential for sensitive material to arise during the interview. The interviewer will also be mindful of the wellbeing of the participant throughout the interview. Participants will be given time to think about their answers and will be encouraged to take a break if the need arises. Participants will also be able to stop the interview at any time and offered the opportunity to reschedule if they so wish. It is hoped that the opportunity to contribute to the development of health services for people with dystonia and to register their views and experience will justify any cost or risk of harm to participants. A summary report of overall findings of the study will be made available to participants, if they wish.

Anonymity

When audio recordings are transcribed participant names will be replaced with pseudonyms. If other individuals are named during the interview such as health professionals or significant others, these details will also be replaced. During the publication process, care will be taken to try to avoid any combination of incidental details which could lead to individuals being identifiable.

Confidentiality

All information will be treated confidentially, however, participants will be made aware of the duty of the investigator to report information that has implications for the safety

of the participant or others to health services (e.g. health care professional or GP, if recruited via the specialist movement disorder clinic or) or the police as appropriate. See below for further details regarding risk management.

Risk Management

i. *Participants recruited from the Manchester Centre for Clinical Neurosciences:* If participants experience significant distress, they will be encouraged to raise appropriate issues with their health care professional where appropriate, their GP, or to access relevant local support services, the information of which will be provided as part of their participant information sheet and the participant debrief sheet.

ii. *Participants recruited from the community (through Dystonia UK):* If participants experience significant distress, it will be suggested that they access support via their GP or access relevant local support services, the information of which will be provided as part of their participant information sheet and the participant debrief sheet.

In all cases, If the student identifies any immediate risk of harm or where this is reported by the participant, the participant will be advised to attend A&E. If any risk to others is evident then the student will contact the appropriate authorities (e.g. police). Efforts will be made to communicate with the participant in such cases prior to taking any action.

Estimated Timescale

April 2021 – July 2021

Initial mode of recruitment begins in collaboration with the SRFT movement disorder clinic and student begins data collection

June – July 2021

If initial mode of recruitment yields insufficient participant numbers, the second mode of recruitment will be initiated (community recruitment via Dystonia UK)

July – August 2021

Data collection ends

August-October 2021

Analyse and write-up data

November – December 2021

Complete final version of research paper

March 2022

Submit thesis

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Appendix 4-B: HRA Approval Letter

Ymchwil Iechyd a Gofal Cymru
Health and Care Research Wales

NHS
Health Research Authority

Dr Fiona Eccles
Health Innovation One
Sir John Fisher Drive
Lancaster University
LA1 4AT

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

11 March 2021

Dear Dr Eccles

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

Study title:	**The experience of being diagnosed with neck dystonia: An interpretative phenomenological analysis study**
IRAS project ID:	**287998**
REC reference:	**21/SC/0065**
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 "Inform tion to support study set up" section tow rds 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 **287998**. Please quote this on all correspondence.

Yours sincerely,

Maeve Groot Bluemink
Approvals Manager

Email: approvals@hra.nhs.uk

Copy to: *Mrs Becky Gordon*

List of Documents

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

Document	*Version*	*Date*
Copies of materials calling attention of potential participants to the research [Study Poster]	0.1	09 November 2020
Copies of materials calling attention of potential participants to the research [Social Media Advert Wording]	0.1	09 November 2020
Cover Letter [Response email incl completed table]		08 March 2021
Evidence of Sponsor insurance or indemnity (non NHS Sponsors only) [Sponsor's Indemnity Insurance]	0.1	01 March 2021
Interview schedules or topic guides for participants [Interview Topic Guide]	0.1	09 November 2020
IRAS Application Form [IRAS_Form_17122020]		17 December 2020
Letters of invitation to participant [Participant Invite Letter]	0.1	09 November 2020
Other [Proof of Peer Review]	0.1	23 October 2020
Participant consent form [NHS Consent Form]	0.1	09 November 2020
Participant information sheet (PIS) [NHS PIS 0.2]	0.2	01 March 2021
Research protocol or project proposal [Research Protocol]	0.1	09 November 2020
Summary CV for Chief Investigator (CI) [Chief Investigator CV]		07 October 2020
Summary CV for student [Student CV]	0.1	17 December 2020
Summary CV for supervisor (student research) [CDixon CV]	0.1	17 December 2020
Summary CV for supervisor (student research) [Chief Investigator CV]	0.1	17 December 2020
Summary of any applicable exclusions to sponsor insurance (non-NHS sponsors only) [EL PL 2020-21]	0.1	01 March 2021
Summary, synopsis or diagram (flowchart) of protocol in non technical language [Flow chart]	0.1	09 November 2020

IRAS project ID	287998

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
The single NHS organisation will act as a Participant Identification Centre.	PIC activities should not commence until a PIC Agreement is in place. HRA and HCRW recommend use of the standard Participating NHS Organisation to PIC agreement available here.	HRA and HCRW recommend use of the standard Participating NHS Organisation to PIC agreement, available here.	No external funding has been sought.	A Principal Investigator (PI)/Local Collaborator (LC) is not expected for PIC activity.	Individuals will be identified and approached by a member of the care team, therefore, it is not expected that any additional HR arrangements will be necessary.

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.
- Some participants may also be recruited outside the NHS and some activity may take place outside the NHS. HRA & HCRW Approval does not cover activity outside the NHS. Before recruiting or undertaking activity outside the NHS the research team must follow the procedures and governance arrangements of responsible organisations.

Appendix 4-C: Amendment Application

Amendment Tool
v1.6 08 December 2021

For office use
QC: No

Section 1: Project information

Short project title*:	The experience of being diagnosed with neck dystonia v 0.1
IRAS project ID* (or REC reference if no IRAS project ID is available):	287998
Sponsor amendment reference number*:	Amendment 1
Sponsor amendment date* (enter as DD/MM/YY):	25 April 2022
Briefly summarise in lay language the main changes proposed in this amendment. Explain the purpose of the changes and their significance for the study. If the amendment significantly alters the research design or methodology, or could otherwise affect the scientific value of the study, supporting scientific information should be given (or enclosed separately). Indicate whether or not additional scientific critique has been obtained (note: this field will adapt to the amount of text entered)*:	1) To extend the end date of recruitment to 21st October 2022. This is allow for recruitment to continue in order to gather a full sample of clients. Currently 5 interviews have been completed. 2) To change the number of participants from 8-12 to 6-10 based on most recently available IPA literature which suggests a smaller sample of about 6-10 people Murray & Wilde (2020). 3) Change to student researcher name due to marriage - Miss Louise Harris is now Mrs Louise Glover

Project type (select):	**Specific study**			
	Research tissue bank			
	Research database			
Has the study been reviewed by a UKECA-recognised Research Ethics Committee (REC) prior to this amendment?:	**Yes**		No	
What type of UKECA-recognised Research Ethics Committee (REC) review is applicable? (select):	**NHS/HSC REC**			
	Ministry of Defence (MoDREC)			
Is all or part of this amendment being resubmitted to the Research Ethics Committee (REC) as a **modified amendment** (i.e. a substantial amendment previously given an unfavourable opinion)?	Yes		**No**	
Where is the NHS/HSC Research Ethics Committee (REC) that reviewed the study based?:	England	Wales	Scotland	Northern Ireland
	No	**Yes**	No	No
Was the study a clinical trial of an investigational medicinal product (CTIMP) OR does the amendment make it one?:	Yes		**No**	
Was the study a clinical investigation or other study of a medical device OR does the amendment make it one?:	Yes		**No**	
Did the study involve the administration of radioactive substances, therefore requiring ARSAC review, OR does the amendment introduce this?:	Yes		**No**	
Did the study involve the use of research exposures to ionising radiation (not involving the administration of radioactive substances) OR does the amendment introduce this?:	Yes		**No**	
Did the study involve adults lacking capacity OR does the amendment introduce this?:	Yes		**No**	
Did the study involve access to confidential patient information outside the direct care team without consent OR does the amendment introduce this?:	Yes		**No**	
Did the study involve prisoners or young offenders who are in custody or supervised by the probation service OR does the amendment introduce this?:	Yes		**No**	
Did the study involve children OR does the amendment introduce this?:	Yes		**No**	
Did the study involve NHS/HSC organisations prior to this amendment?:	**Yes**		No	
Did the study involve non-NHS/HSC organisations OR does the amendment introduce them?:	**Yes**		No	
	England	Wales	Scotland	Northern Ireland
Lead nation for the study:	**Yes**	No	No	No
Which nations had participating NHS/HSC organisations prior to this amendment?	**Yes**	No	No	No
Which nations will have participating NHS/HSC organisations after this amendment?	**Yes**	No	No	No
Was this a "single site, self sponsored" study in England or Wales prior to this amendment?	Yes		**No**	
Which nations had participating non-NHS/HSC organisations prior to this amendment?	**Yes**	No	No	No
Which nations will have participating non-NHS/HSC organisations after this amendment?	**Yes**	No	No	No

Section 2: Summary of change(s)

Please note: Each change being made as part of the amendment must be entered separately. For example, if an amendment to a clinical trial of an investigational medicinal product (CTIMP) involves an update to the Investigator's Brochure (IB), affecting the Reference Safety Information (RSI) and so the information documents to be given to participants, these should be entered into the Amendment Tool as three separate changes. A list of all possible changes is available on the "Glossary of Amendment Options" tab. To add another change, click the "Add another change" box.

Change 1				
Area of change (select)*:	Study Design			
Specific change (select - only available when area of change is selected first)*:	Extension to study duration that will not have any additional resource implications for participating organisations - Please specify in the free text below			
Further information In particular, please describe why this change can be implemented within the existing resource in place at the participating organisations (free text - note that this field will adapt to the amount of text entered)*	Recruitment period to be extended to 21st October 2022 to ensure recruitment aims are met. This is supported by the research site who are happy to continue recruitment.			
Applicability:	England	Wales	Scotland	Northern Ireland
Where are the participating NHS/HSC organisations located that will be affected by this change?*:	**Yes**	No	No	No
Will all participating NHS/HSC organisations be affected by this change, or only some? (**please note** that this answer may affect the categorisation for the change):	**All**		Some	
Where are the participating non-NHS/HSC organisations located that will be affected by this change?*:	**Yes**	No	No	No
			Remove all changes below	

Change 2				
Area of change (select)*:	Study Design			
Specific change (select - only available when area of change is selected first)*:	Participant numbers - Minor change to sample size			
Further information (free text - note that this field will adapt to the amount of text entered):	Change number of participant aim from 8-12 to 6-10 in line with most recent literature about IPA sample size (Murray & Wilde, 2020). This change does not impact the scientific value of the project in any way.			
Applicability:	England	Wales	Scotland	Northern Ireland
Where are the participating NHS/HSC organisations located that will be affected by this change?*:	**Yes**	No	No	No
Will all participating NHS/HSC organisations be affected by this change, or only some? (**please note** that this answer may affect the categorisation for the change):	**All**		Some	
Where are the participating non-NHS/HSC organisations located that will be affected by this change?*:	**Yes**	No	No	No
			Remove all changes below	

Change 3				
Area of change (select)*:	Researchers			
Specific change (select - only available when area of change is selected first)*:	Changes to the research team (other than CIs or PIs)			
Further information (free text - note that this field will adapt to the amount of text entered):	The student researcher was previously named Miss Louise Harris, but has now changed her name to Mrs Louise Glover. This amendment does not change the researcher, instead just notifies of the change of contact name.			
Applicability:	England	Wales	Scotland	Northern Ireland
Where are the participating NHS/HSC organisations located that will be affected by this change?*:	**Yes**	No	No	No
Will all participating NHS/HSC organisations be affected by this change, or only some? (**please note** that this answer may affect the categorisation for the change):	**All**		Some	
Where are the participating non-NHS/HSC organisations located that will be affected by this change?*:	**Yes**	No	No	No
			Add another change	

Declaration by the Sponsor or authorised delegate

- I confirm that the Sponsor takes responsibility for the completed amendment tool
- I confirm that I have been formally authorised by the Sponsor to complete the amendment tool on their behalf

*Name (first name and surname)**:	Becky Gordon
*Email address**:	sponsorship@lancaster.ac.uk

Look for submission

Please note: This button will only become available when all mandatory (*) fields have been completed. When the button is available, clicking it will generate a locked PDF copy of the completed amendment tool which must be included in the amendment submission. Please ensure that the amendment tool is completed correctly before looking it for submission.

Look for submission

After looking the tool, proceed to submit the amendment online. The "Submission Guidance" tab provides further information about the next steps for the amendment.

Section 4: Review bodies for the amendment

Please note: This section is for **information only**. Details in this section will complete automatically based on the options selected in Sections 1 and 2.

	Review bodies																	
	UK wide:						England and Wales:				Scotland:				Northern Ireland:			
	REC	Competent Authority MHRA - Medicines	Competent Authority MHRA - Devices	ARSAC	Radiation Assurance	UKSW Governance	REC (IRCA)	CAG	HMPPS	HRA and HCRW Approval	REC (AWIA)	PBPP	SPS (RAEC)	National coordinating function	HSC REC	HSC Data Guardians	Prisons	National coordinating function	Category:
Change 1:						(Y)				(Y)									C
Change 2:						(Y)				(Y)									A
Change 3:						N				N									N/A
Overall reviews for the amendment:																			
Full review:						N				N									
Notification only:						Y				Y									
Overall amendment type:	Non-substantial, no study-wide review required																		
Overall Category:	A																		

Appendix 4-D: Amendment Approval Email

Dear Dr Eccles,

IRAS Project ID:	287998
Short Study Title:	The experience of being diagnosed with neck dystonia v 0.1
Amendment No./Sponsor Ref:	Amendment 1
Amendment Date:	25 April 2022
Amendment Type:	Non Substantial Non-CTIMP

Thank you for submitting the amendment. Please note that as a non-substantial no study wide review required amendment (per the amendment tool) this change can be implemented without any further correspondence from the HRA.

Please note that in future, this type of amendment can be filed as 'non-substantial no study wide review required ' instead of 'non-substantial'. No correction is required for this amendment- this will provide automatic confirmation that the change can be implemented.

Kind Regards

Simon Fisher (he/him)
Approvals Administrator
Health Research Authority
Bristol HRA Centre, Ground Floor, Temple Quay House, 2 The Square, Bristol, BS1 6PN
T. 020 7104 8256
E. simon.fisher@hra.nhs.uk
W. www.hra.nhs.uk

Appendix 4-E: Interview Topic Guide

Interview Topic Guide

This topic guide indicates the areas to be covered and some example questions. However, the interview will also be guided by the areas salient to the participant.

Topic area 1: Noticing symptoms

Example questions

- Can you tell me when you first noticed symptoms?
- Can you tell me about your thoughts and feelings related to noticing symptoms? What did you think it could be?
- What did others in your family/social group make of them?

Topic area 2: Accessing support or advice

Example questions

- Can you tell me about how you went about accessing support or advice about the symptoms you had noticed?
- How was this experience for you?
- What impact, if any, did this have on you?
- If appropriate – what did others make of this advice/support?

Topic area 3: Receiving a diagnosis

Example questions

- Can you tell me about how you came to receive a diagnosis?
- Who gave the diagnosis? What happened in that meeting? What happened next?
- What impact, if any, did this have on you/your partner/family/loved ones?

Topic area 4: Coming to terms with the diagnosis

Example questions

- Can you tell me about your experience of receiving a diagnosis of neck dystonia?
- What do you think about the diagnosis now?
- Can you tell me about any impact on how you feel/your mood?

- If applicable, did you feel able to access any support related to your mood?
- How does the diagnosis affect you now/how might it affect you in the future?

Topic area 5: Other issues

Example questions

- Is there anything else that you feel is important to talk about?

Appendix 4-F: Invitation to Participate

Doctorate in Clinical Psychology | Lancaster University

Salford Royal NHS Foundation Trust
University Teaching Trust

Invite for research study: 'The experience of being diagnosed with neck dystonia'

Dear patient,

We have identified that you may be eligible to take part in a study. The purpose of the study is to understand how people experience the journey to being diagnosed with neck dystonia. For full details, please read the enclosed Participant Information Leaflet.

If you have any questions, or might be interested in taking part, you may let your nurse or doctor know that you are happy for your details to be passed to the research team, or you may contact the main researcher directly:

Louise Harris
Email: l.harris7@lancaster.ac.uk
Tel: 07508375663

Best Wishes,

Dr Christopher Kobylecki

On behalf of 'The experience of being diagnosed with neck dystonia' study team

Appendix 4-G: Study Information Sheet

Doctorate in Clinical Psychology | Lancaster University

Salford Royal NHS Foundation Trust
University Teaching Trust

Participant Information Sheet
Study Title: The experience of being diagnosed with neck dystonia

My name is Louise Harris, and I am conducting this research as a student of the Doctoral in Clinical Psychology programme at Lancaster University. I'd like to invite you to take part in this research study.

What is the study about?
We know that the journey to receiving a diagnosis of dystonia can take a long time and many people are given different explanations for their symptoms at first. But we know very little about the impact of this experience on people's mood and wellbeing. This study aims to speak to people who have experienced the journey to receiving a diagnosis of neck dystonia first-hand. This will enhance our understanding of what this experience is like for people and to begin to consider what support people need.

Why have you been approached?
You have been approached because a member of your care team at the Manchester Centre for Clinical Neurosciences, has identified that you may be able to take part. This study requires information from people who:

- Have received the diagnosis within the previous three years and no sooner than the previous six months (this is so that there has been some chance to come to terms with the diagnosis whilst being able to remember what happened as accurately as possible).
- Are aged between 35 and 75 years old
- Can speak fluently in English.
- Have no additional physical or mental health difficulties which may have significantly impacted your experience of diagnosis. For example, if you have another movement disorder diagnosis, cognitive impairment or a significant level of psychological distress, for example severe depression or psychosis, you would not be able to take part.

Do you have to take part?
No. It's completely up to you to decide whether you would like to take part.

What will you be asked to do if you take part?
If you decide you would like to take part, you would be asked to take part in an interview. The interview will ask about early symptoms, where you went for help, the journey to diagnosis and the impact of this on your mood.

Doctorate in Clinical Psychology | Lancaster University

Salford Royal NHS Foundation Trust
University Teaching Trust

Where will the study take place?
The interview will take place remotely by phone or video call.

How long will the study last?
The interview will take 60-90 minutes.

When will it be?
The interview will take place at a convenient time for you.

How will we use information about you?
We will need to use information from you for this research project.

- Your name and contact details will be held to allow the main researcher to contact you to arrange a convenient time for the interview. After the research is complete this information will be used to send you a summary of the results. After this, your contact details will be permanently deleted.
- Consent to take part will be taken at the beginning of the interview. This will be recorded and stored separately to the interview recording. The recording will be encrypted and stored securely on a computer (that is no-one other than the researcher will be able to access them). After the project has been examined, the consent recording will be stored by Lancaster University where it will be stored securely for 10 years.
- Your interview will be recorded. The recording will be encrypted and stored securely on a computer (that is no-one other than the researcher will be able to access them). After it is stored, the recording will be deleted from the recording device. Audio recordings will be permanently deleted once the project has been examined.
- The audio recording of the interview will be typed up by the main researcher and stored on a computer. This will be encrypted, that is no-one other than the researcher 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 reasonable steps will be taken to protect the anonymity of the participants' involved in this project.
- After the project has been examined, the typed versions will be stored by Lancaster University where they will be stored securely for 10 years

Are there any circumstances where your information may not remain confidential?

There are some limits to confidentiality: if what is said in the interview makes me think that you, or someone else, is at significant risk of harm, I will have to break confidentiality and speak to a member of staff about this. If possible, I will tell you if I must do this.

What are your choices about how your information is used?
Your participation is voluntary, and you are free to withdraw at any time during or up to two weeks after the interview, without giving any reason and without your medical care or legal rights being affected.

What are your rights about your personal information?
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 anonymised direct quotations from your interview may be used. The results will be reported in a thesis, will be presented at conferences and may be submitted for publication in an academic or professional journal. The findings will be available online at Dystonia Society UK's research page. You will be sent a copy of the results if you wish.

Are there any risks?
Some distress may arise from reflecting in detail on your experiences. If you experience any distress following participation you are encouraged to inform the researcher and contact the resources provided at the end of this sheet.

Are there any benefits to taking part?
Often people find that having time and space to reflect on your experiences is beneficial. Also, the findings of this project may mean in the future, that others who share your experience are better supported.

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Who has reviewed the project?

This study has been reviewed and approved by an ethics committee of the NHS Health Research Authority.

Where can you obtain further information about the study and how can you take part?

If you have any questions about the study, or might be interested in taking part please let your nurse or doctor know, or contact the main researcher directly:

Louise Harris
Email: l.harris7@lancaster.ac.uk
Tel: 07508375663
Health Innovation One, Sir John Fisher Drive, Lancaster University, Lancaster, LA1 4AT

If you wish to speak to the chief investigator/research supervisor, you can contact:

Dr Fiona Eccles
Tel: 01524 592897
Email: f.eccles@lancaster.ac.uk
Address: Health Innovation One, Sir John Fisher Drive, Lancaster University, Lancaster, LA1 4AT

Or, if you wish to speak to the research supervisor, you can contact:
Dr Clare Dixon
Tel: 01524 593492
Email: c.dixon3@lancaster.ac.uk

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:

Dr Ian Smith
Tel: 07507857069
Email: i.smith@lancaster.ac.uk
Doctorate in Clinical Psychology
Health Innovation One, Sir John Fisher Drive, Lancaster University, Lancaster, LA1 4AT

If you wish to speak to someone outside of the Doctorate in Clinical Psychology programme, you may also contact:

Professor Roger Pickup
Chair of FHM REC
Email: r.pickup@lancaster.ac.uk
Faculty of Health and Medicine
(Lancaster Medical School), Lancaster University, Lancaster, LA1 4YG

Resources in the event of distress

Should you feel distressed either as a result of taking part, or in the future, please discuss this with your nurse or doctor, or contact your general practitioner (GP).

The following resources may also be of assistance:

The Samaritans (Available 24/7)
Tel: 116 123
Email: jo@samaritans.org
Address: Chris
Freepost RSRB-KKBY-CYJK
PO Box 9090
STIRLING FK8 2SA

The Dystonia Society
Open Mon-Thurs 10:00-16:00 (not for emergencies)
Tel: 020 7793 3650
Email: support@dystonia.org.uk

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

Appendix 4-H: Copy of Consent Form

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Consent Form

Study Title: The experience of being diagnosed with neck dystonia

We are asking if you would like to take part in a research project aimed at understanding the experience of being diagnosed with neck dystonia.

Before you consent to participating in the study, we ask that you read the participant information sheet carefully. **This is a copy of the consent form**, the researcher will ask you to confirm your name and answer yes or no to the following questions, before starting the interview. Your consent will be recorded and stored in a separate location to your interview recording. If you have any questions or queries before answering the consent questions, please speak to the main researcher: Louise Harris (l.harris7@lancaster.ac.uk Tel: 07508375663).

Name of Participant _____

		Y/N
1.	Do you confirm that you have read the information sheet and fully understand what is expected of you within this study?	
2.	Do you confirm that you have had the opportunity to ask any questions and to have them answered?	
3.	Do you understand that your interview will be audio recorded and then made into an anonymised written transcript?	
4.	Do you understand that audio recordings will be kept until the research project has been examined?	
5.	Do you understand that your participation is voluntary and that you are free to withdraw at any time during or up to two weeks after the interview, without giving any reason and without your medical care or legal rights being affected?	
7.	Do you understand that the information from your interview will be pooled with other participants' responses, anonymised and may be published; all reasonable steps will be taken to protect the anonymity of the participants involved in this project.	
8.	Do you consent to information and quotations from your interview being used in reports, conferences and training events?	
9.	Do you understand that the researcher will discuss data with their supervisors as needed?	
10.	Do you understand that any information you give will remain confidential and anonymous unless it is thought that there is a risk of harm to you or	

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	others, in which case the researcher will need to share this information with their research supervisors?	
11.	Do you consent to Lancaster University keeping written transcriptions of the interview for 10 years after the study has finished or 10 years from publication, whichever is longer?	
12.	Do you consent to take part in the above study?	