**Doctoral Thesis**

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

# Psychological Experiences in Neurological Conditions: Multiple Sclerosis and Cervical Dystonia

Doctorate in Clinical Psychology

Lancaster University

Derval McCormack

2021 Intake

# Statement of Total Word Count

| Section | Main Text | Appendices including References, Figures and Tables | Total |
|---|---|---|---|
| Thesis Abstract | 286 | -- | 286 |
| Systematic Literature Review | 7473 | 13,860 | 21,333 |
| Research Paper | 7985 | 11,065 | 19,050 |
| Critical Appraisal | 3744 | 1062 | 4806 |
| Ethics | 4850 | 3266 | 8116 |
| **Total** | **24,338** | **29,253** | **53,591** |

# Thesis Abstract

This thesis focuses on psychological experiences in two distinct neurological conditions: multiple sclerosis (MS), and cervical or neck dystonia (ND).

Section one outlines a systematic literature review, carried out to examine whether a relationship exists between body image and psychological outcome in MS. Four academic databases were searched systematically using key words relating to body image and MS. Manual searches of returned studies were conducted to find papers that examined psychological outcome. Although limited research has been carried out previously in this area, the findings offer preliminary evidence for a relationship between body image and psychological outcome in MS. The review suggests that positive body image is associated with better mood, lower anxiety, increased self-esteem, and greater quality of life. This has important clinical implications, suggesting that body image could be a target for future intervention. Future research should explore the association between body image and psychological outcome in more detail.

Section two details an empirical study examining lived experiences of psychological distress in ND. Eleven individuals with ND were interviewed using a semi-structured schedule, and data was analysed using interpretative phenomenological analysis. The findings suggested that uncertainty, abandonment, loss, stigma, and isolation were central to people's experiences of distress. This research contributes to wider literature regarding beliefs about psychological distress in dystonia. The results suggest that distress arises from living with ND, with some individuals also experiencing distress prior to onset of symptoms. The study provides valuable insight into gaps in clinical psychology provision, at direct and indirect levels. Future research including individuals with complex presentations of ND is recommended.

Section three presents the critical appraisal, which evaluates the process of conducting this thesis. It considers important decisions, challenges encountered, and personal reflections.

# 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. This work has not been submitted for any other academic award.

Name: Derval McCormack

Signature:

Date: 11/03/2024

# Acknowledgements

Firstly, I would like to thank the eleven individuals who kindly gave up their time to participate in this project. It was a privilege to speak with you all, and to listen to your experiences first-hand. I hope that the research presented in section two treats your experiences respectfully and truthfully, and that it will be beneficial in improving experiences of people with dystonia.

Secondly, I would like to take this opportunity to thank Dystonia UK for their support with recruitment. I would also like to thank David Ward for participating as an expert by experience, and supporting with the design of the research study.

I would like to thank my research supervisor Dr Fiona Eccles, and my field supervisor Dr Fiadhnait O'Keeffe for their continued support and guidance throughout this project. Thank you for sharing your knowledge and expertise with me, and for your kindness and understanding throughout the process.

Finally, I would like to extend my gratitude to Christina Seery for her assistance as an independent reviewer for the systematic review. Many thanks for your dedication and commitment to this task.

## Contents

**Section One: Literature Review**

**Section Two: Research Paper**

**Section Three: Critical Appraisal**

**Section Four: Ethics**

# Section One: Systematic Literature Review

# The Association between Body Image and Psychological Outcome in Multiple Sclerosis. A Systematic Literature Review

Word count (excluding references, tables and appendices): 7473

Derval McCormack

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

Prepared for submission to *Multiple Sclerosis and Related Disorders*

**All correspondence should be sent to:**

Derval McCormack
Doctorate in Clinical Psychology
Lancaster University
Furness College
United Kingdom
LA1 4GY

Email: d.mccormack@lancaster.ac.uk

Tel: 01524 59297

## Abstract

**Background:**

Multiple sclerosis (MS) is a demyelinating autoimmune condition, in which body image may be altered due to reduced mobility and neurobiological factors. The aim of this review was to examine whether there is a relationship between body image and psychological outcome in MS, in order to target clinical interventions.

**Methods:**

PsycInfo, Medline, CINAHL and Scopus databases were searched systematically for eligible studies, using terms relating to MS and body image. Returned studies were checked manually for psychological outcome. Quantitative studies that examined the relationship between body image and psychological outcome in MS were included.

**Results:**

Thirteen studies met all inclusion criteria and were included in the final review. Quality appraisal was conducted for each of these studies. A narrative synthesis suggested that positive body image was associated with improved mood, lower anxiety, increased self-esteem, and better quality of life, however the causal nature of these relationships was unclear.

**Conclusion:**

The findings offer preliminary evidence to suggest that a more positive body image is associated with lower levels of psychological distress in MS, indicating that body image may be a target for intervention. Further research is necessary to provide a greater understanding of this association, and to inform future clinical practice.

*Keywords:* Multiple Sclerosis; MS; body image; mood

## Introduction

Multiple sclerosis (MS) is a chronic demyelinating autoimmune condition affecting the central nervous system (Bobholz & Gremley, 2011; Gunnersen et al., 2022). An estimated 2.8 million people worldwide live with MS, and prevalence rates are increasing steadily (Campbell et al., 2020; Walton et al., 2020). Proposed hypotheses for such trends include early diagnosis, greater ascertainment, and increased life expectancy (Walton et al., 2020). MS usually develops in early adulthood, with a mean age of onset of approximately 30 years, and it is two to four times more common in females than males (Hosseini et al., 2022). Although the cause of MS remains unknown, the development of the condition has been shown to be associated with a range of biological, environmental, and genetic factors (Kamińska et al., 2017). The condition is unevenly distributed throughout the world, with higher prevalence rates in temperate regions of northern latitude (Amezcua & McCauley, 2020). There have been previous reports suggesting a prevalence of 140 per 100,000 in North America, and 2.2 per 100,000 in parts of Africa (Leray et al., 2016). Some researchers have suggested that exposure to vitamin D may be a factor in explaining these geographical differences (Ramagopalan et al., 2010; Simpson et al., 2011), while others have proposed that genetic components may be at play (Ebers, 1992; Milo & Kahana, 2010). Climate, diet, and infection exposure have been proposed as other plausible factors to explain regional differences in disease distribution (Bobholz & Gremley, 2011; Dobson & Giovannoni, 2018).

The most common presenting symptoms of MS include sensory disturbances, walking difficulties, visual problems, urinary or bowel dysfunction, dizziness, vertigo, sexual dysfunction, and cognitive difficulties (Ghasemi et al., 2017). In terms of cognitive difficulties, the domains most commonly affected in MS are working memory and processing speed (Bobholz & Gremley, 2011), with such difficulties experienced by approximately 50 to 60% of individuals (Amato et al., 2008). The frequency of symptoms and the severity of disease progression varies greatly across individuals with MS. Four main disease courses have been identified: clinically isolated syndrome (CIS), relapsing-remitting MS (RRMS), secondary progressive MS (SPMS), and primary progressive MS (PPMS) (Lublin et al., 2020). CIS refers to the first time a person experiences the symptoms of MS, and this episode must last at least 24 hours to meet criteria. RRMS is the most common form of MS, and is characterised by clear periods of ‘attacks’, where people experience new or increasing symptoms, followed by periods of remission. Some people with RRMS will progress to SPMS, whereby disease severity worsens progressively over time. With PPMS, disability accumulates more rapidly from the onset of symptoms, and does not involve periods of remission (Bobholz & Gremley, 2011).

MS is associated with high levels of psychological distress, with a recent meta-analysis estimating prevalence rates of 30.5% and 22.1% for depression and anxiety, respectively (Boeschoten et al., 2017). Reports suggest that the psychological effects of MS occur as a result of both psychosocial and biological factors underlying the condition (Sá, 2008). In terms of psychosocial factors, increased mood difficulties have been reported to be associated with younger age, shorter disease duration, symptom severity, and sexual dysfunction (Hanna & Strober, 2020; Zavoreo et al., 2016). A number of studies have reported reduced quality of life (QoL) in people with MS, compared with the general population (Karatepe et al., 2011; McCabe & McKern, 2002). A large-scale systematic review by Gil-González et al. (2020) identified level of disability, unemployment, fatigue, psychological wellbeing, and cognitive decline as factors associated with lower QoL in MS. A number of qualitative studies have examined people’s experiences of living with MS, and its impact upon activities of daily living. Hosseini et al. (2022) reported that MS was associated with reduced social interactions, inability to continue working, and financial difficulty. A further study found that people with MS were less likely to exercise due to increased fatigue, reduced control over movements, and feelings of shame when exercising in front of other people (Borkoles et al., 2008). The English National Institute for Health and Care Excellence (NICE) guidelines (2009) currently recommend a cognitive behavioural therapy (CBT) approach to managing psychological distress in people with MS, with previous evidence suggesting that such interventions can be effective (Hind et al., 2014). There is preliminary support for the effectiveness of third-wave interventions such as mindfulness-based cognitive therapy (MBCT) in addressing psychological difficulties in MS (Zarotti et al., 2023), however further research is needed to consider other models and long-term effectiveness.

Body image may be an important factor to consider when understanding the psychological experiences of people with MS. While there is no commonly accepted definition for the term ‘body image’ (Badoud & Tsakiris, 2017), it has frequently been described as a multifaceted construct, encompassing how individuals subjectively perceive and appraise their own bodies (Cash, 2004). It consists of perceptive, affective and cognitive components, which together take into account how individuals view their own bodies, the positive or negative feelings they have about their appearance, and the beliefs they hold about certain aspects of their body, including features or movements (Badoud & Tsakiris, 2017; Stokes & Frederick-Recascino, 2003). The general schematic model (Slade, 1994) proposes that body image is influenced by seven factors, including history of sensory inputs to the body, history of weight change, cultural and social norms, individual attitudes to shape, cognitive and affective variables, individual psychological difficulties, and biological variables. The internal representation of a person’s external appearance is important in developing a sense of self-awareness and personal identity (Przezdziecki et al., 2012; Tsakiris, 2010). Dissatisfaction in one’s own body image can lead to the development of psychological difficulties such as anxiety and depression, as well as sexual dysfunction (Di Cara et al., 2019).

The term ‘body image’ is often used interchangeably within literature to describe another concept called interoception, which focuses on an individual’s awareness and appraisal of internal bodily sensations or experiences (Mehling et al., 2012; Todd et al., 2019). From a psychological perspective, an individual’s sensitivity to bodily signals closely links with their experiences and regulation of emotions (Badoud & Tsakiris, 2017). The extent to which interoceptive awareness contributes to body image is not fully understood, however there are indications in the literature that body image and interoception are related. Literature on eating disorders, for example, suggests that poorer interoceptive sensitivity may be associated with bodily self-consciousness (Costantini, 2014). There is evidence to suggest a neural connection between body image and interoception, with the insula being reported as a specific region involved in the cortical representation of both interoceptive and body image signals (Badoud & Tsakiris, 2017). Furthermore, it has been suggested that lower levels of interoceptive awareness may predispose individuals to greater body image dissatisfaction (Badoud & Tsakiris, 2017).

An individual’s perception of their body can fluctuate over time due to a variety of factors including societal expectations, comparison with others, relationships and disability (Samonds & Cammermeyer, 1989). When a person acquires an illness that alters the functioning or appearance of their body, their body image can also be altered, which in turn can create challenges for psychological wellbeing (Cash, 2004). Previous systematic reviews have reported increased dissatisfaction in body image due to physical changes arising from other health conditions, including cancer (Lehmann et al., 2015) and burn injuries (Connell et al., 2013). The representation of body image in MS may be further altered due to specific neurobiological processes arising from the condition (Di Cara et al., 2019). The development of body image relies on the integration of multisensory signals that are compromised in autoimmune conditions such as MS (Costantini, 2014). Furthermore, people with MS display differences in the brain regions responsible for interoception, the process by which people perceive physiological conditions in the body (Ware et al., 2023). To the researcher's best knowledge, only one review has examined body image in MS previously (Di Cara et al., 2019). This review took a descriptive approach rather than synthesising results, and the association between body image and psychological outcome was not examined. Their findings suggested that body image is an important construct in MS, which may have a significant impact on a person's self-esteem or psychological wellbeing.

The aim of this systematic review was to examine whether there is a relationship between body image and psychological outcome in MS. For the purpose of this review, psychological outcome will be treated broadly, including but not limited to concepts such as anxiety, depression, and QoL. A greater understanding of the relationship between body image and psychological outcome in MS will allow clinicians to target psychological interventions appropriately, in order to provide person-centred support for people living with this condition.

## Materials and Methods

The author referred to the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) checklist (Moher, 2019) and published Cochrane guidance for conducting systematic reviews (Higgins & Thomas, 2023) throughout the review process.

### Defining the Research Question

An initial scoping search was conducted using an academic library database, OneSearch to support with refinement of the research question. The research question provides the critical foundations of a systematic review, for which the development of a search strategy can follow (Eriksen & Frandsen, 2018). The development of the question was guided by the FINER criteria, which suggest that research questions should be feasible, interesting, novel, ethical, and relevant (Cummings et al., 2007). The initial scoping search suggested that not much is known about the psychological impact of body image in MS, therefore a broad research question was deemed most suitable.

The author registered the current review on the PROSPERO website, which can be accessed via crd.york.ac.uk/prospero/display_record.php?RecordID=477640, registration number CRD42023477640.

### Inclusion and Exclusion Criteria

Papers were considered suitable if they met the following inclusion criteria:

- Participants were adults aged eighteen or over, with a diagnosis of MS. Studies with younger participants were excluded because adolescents tend to experience a number of changes during puberty that can affect their body image in a unique way to that experienced by adults (Borzekowski & Bayer, 2005).
- Were peer-reviewed, quantitative empirical papers, written in English. While limiting included studies to one specific language can introduce bias, the alternative relies on resources that were outside the scope of this review (Welch et al., 2023).
- Included a measure of both body image and psychological outcome e.g. depression, anxiety, QoL etc. The measures used in the studies to examine body image and psychological outcomes must have been established within research to be reliable and valid. For the purpose of this review, the term ‘body image’ includes studies assessing individuals’ perception or appraisal of external or internal aspects of their bodies. Studies using an external objective measure of body functioning were not included within the scope of this review, for example body mass index.
- The relationship between body image and psychological outcome was examined in some way, for example using correlation or regression studies. Intervention studies

that measured both body image and psychological outcome pre and post intervention were included.

## Search Strategy

Studies were identified by searching across four databases: PsycInfo, Medline, CINAHL, and Scopus. These databases were selected following guidance from an academic librarian to capture journals within the fields of psychology, neurology, medicine, and related health-care disciplines. These databases have been used in previous systematic reviews examining the concepts of MS (Allen & Walter, 2016) and body image (Luca et al., 2022). Subject headings were identified by searching key words related to both MS and body image using Medical Search History (MeSH) terms and database thesauruses. The search strategy incorporated a free text option to capture other relevant studies. The search focused on two main concepts, MS and body image, which were then combined. Full details of the final search string and related key terms can be found in Table 1-1. Returned papers were then hand searched for any relevant psychological outcome. The final search was conducted on 2nd November 2023.

[Insert Table 1-1 here]

## Screening Process

The search returned an initial 1024 studies, which were exported into a data management software package. Duplicates were removed, and the remaining 555 papers were then screened for suitability. The author and a second reviewer independently screened the title and abstracts of each of these papers, using the inclusion and exclusion criteria. Inconsistencies were discussed together and resolved without the need for a third reviewer. Following screening of title and abstracts, 21 papers remained for full screening. The author searched the reference lists of each of these papers for further suitable studies. They identified one further study to screen in this process. Forward referencing identified a further two studies. This left 24 studies for full text screening. The author and second reviewer independently screened each of these papers for eligibility. No inconsistencies were found, leaving 13 studies for inclusion in the final review. A flow diagram of the selection process can be seen in Figure 1-1.

[Insert Figure 1-1 here]

## Data Extraction and Quality Assessment

The author extracted the data from the studies. Information extracted from the studies included the sample size, gender, mean age, type of MS, disease duration, study design and analysis, measures of body image and psychological variable, outcomes of body image and psychological variable, relationship between body image and psychological outcome, and correlation coefficient as a measure of effect size.

The quality of the studies was assessed using the QualSyst tool (Appendix 1-B), a 14-item survey for assessing and evaluating quantitative research papers for inclusion in systematic reviews (Kmet et al. 2004). The tool is suitable for assessing a broad range of study design types and was therefore considered most suitable for this review, which includes both cross-sectional and intervention studies. Each item in the survey is scored based on one of three responses: yes, partial or no, and is worth two, one or zero points, respectively. Three of the 14 items in this scale were questions relating to intervention studies only, and were therefore not applicable to all studies included in this review. Items that were not applicable to a particular study were excluded from the calculation of the summary score, in line with published guidelines (Kmet et al., 2004). The summary score was calculated by dividing the total score from all applicable items by the total possible score. The author assessed the quality of each study at the stage of data extraction. A second reviewer performed this independently to minimise the risk of bias. Discrepancies were discussed together. The final summary scores were agreed by both parties.

### Data Synthesis and Presentation

The extracted data was presented in tables, and a narrative synthesis of studies was used to report patterns in the findings (Popay et al.,2006). To answer the research question, the studies were grouped based on psychological outcome reported. Given the differences in study design, results from cross-sectional studies were presented separately to those of intervention studies.

## Results

### General Study Characteristics

Ten of the studies included in this review were cross-sectional in design, and three were intervention studies. Two of the studies were carried out in the USA (Hernandez-Reif et al., 1998; Stevens et al., 2018), two in Iran (Farnam et al., 2017; Ghodusi & Heidari, 2014), two in Austria (Pfaffenberger et al., 2011; Tesar et al., 2003 ), two in Italy (Lo Buono et al., 2023; Paolucci et al., 2022), one in Canada (Kindrat, 2007), one in Portugal (Barbosa et al. 2023), one in Turkey (Sengul et al., 2019), one in Israel (Barak et al. 1998), and one in Poland (Wilski et al., 2016). The sample size for studies ranged from 24 (Hernandez-Reif et al., 1998) to 395 (Ghodusi & Heidari, 2014), totalling 1533 participants across all studies. Eight of the studies specified MS type. In five studies, 100% of the participants had RRMS (Barak et al. 1998; Kindrat, 2007; Lo Buono et al., 2023; Sengul et al., 2019; Paolucci et al., 2022). In the other three studies, the percentage of participants with RRMS was 21.5%, 68.9%, and 34%, the percentage of participants with SPMS was 6.2%, 17.2% and 22%, and the percentage of participants with PPMS was 5.6%, 11.3% and 20%, respectively (Barbosa et al. 2023; Stevens et al., 2018; Wilski et al., 2016). All studies reported on age, with a pooled mean age of 41.8 across each of these studies. Seven studies used the Expanded Disability Severity Scale (EDSS) to report on disease severity, with results ranging from 2.63 to 5.37, with higher scores indicating greater levels of disability. One of these studies did not report a mean score, however the majority of participants (73%) scored between 3.5 and 6.5, indicating moderate disability (Wilski et al., 2016). Nine studies reported disease duration, with a pooled mean of 8.2 years across these studies. A summary of studies included in the review are shown in Table 1-2.

[Insert Table 1-2 here]

### Quality Appraisal

The scores from the quality appraisal ranged from 0.77 to 1, out of a possible 1. The main reasons for reduced quality scores were low sample sizes, or the characteristics of the sample or the research question not being sufficiently described. Some intervention studies lost points where, if feasible, randomisation was not done, or investigators were not blinded, as per tool guidelines. Table 1-3 provides further information on the quality appraisal of included studies.

[Insert Table 1-3 here]

### Body Image Outcome Measures

A total of 14 self-report questionnaires were used to measure body image across the studies. There was much heterogeneity in these measures, summarised in Table 1-4. The number of items in each measure ranged from seven (Barbosa et al., 2023) to 52 (Pfaffenberger et al., 2011). The majority of measures used a Likert scale, ranging from three (Lo Buono et al., 2023; Paolucci et al., 2022) to seven (Barbosa et al., 2023). One measure asked for a true or false response (Pfaffenberger et al., 2011). Most measures examined satisfaction in external body appearance, and two focused on interoceptive awareness (Barbosa et al., 2023; Paolucci et al., 2022). One of the measures examined how individuals think other people perceive their appearance (Barbosa et al., 2023). This was considered relevant because positive perceptions of body acceptance by others may foster positive body image in self (Wang et al., 2022). All studies were reported to be psychometrically robust.

[Insert Table 1-4 here]

## Psychological Outcomes

A total of nine measures were used to examine psychological outcome across the studies, summarised in Table 1-5. Five self-report questionnaires were used to measure depression in seven studies. These measures ranged from nine (Stevens et al., 2018) to 21 items (Sengul et al., 2019; Pfaffenberger et al., 2011). The Symptom Checklist-90-Revised (SCL-90-R) scale was used by one study (Lo Buono et al., 2023) to measure depression, with one nine-item dimension designed to assess this variable. All other measures were primarily focused on depression. Two self-report questionnaires were used to measure self-esteem in five studies. These measures ranged from 10 (Barak et al. 1998) to 30 items (Ghodusi & Heidari, 2014; Hernandez-Reif et al., 1998; Lo Buono et al., 2023; Wilski et al., 2016). Three self-report measures were used to examine anxiety in four studies. These measures ranged from nine (Lo Buono et al., 2023) to 21 items (Sengul et al., 2019). Four studies looked at QoL, each using a different measure. These measures ranged from 12 (Paolucci et al., 2022) to 29 items (Wilski et al., 2016). Three of these measures focused on neurological or health-related aspects of QoL (Paolucci et al., 2022; Stevens et al., 2018; Wilski et al., 2016). Emotion regulation, aggression, and stress were measured in one study (Farnam et al., 2017), and coping was measured by another study (Tesar et al., 2003). The majority of measures to assess psychological outcomes were evidenced to be psychometrically reliable and robust.

[Insert Table 1-5 here]

## Relationship between Body Image and Psychological Outcome

### *Depression*

Four studies looked at the relationship between body image and depression in MS, using either Pearson’s or Spearman’s correlation analysis (Kindrat, 2007; Lo Buono et al., 2023; Pfaffenberger et al., 2011; Sengul et al., 2019). All of these studies reported a significant positive relationship between the two concepts, indicating that a positive body image is associated with lower levels of depression. Strong effect sizes were reported across these studies, ranging from 0.53 (Pfaffenberger et al., 2011; Sengul et al., 2019) to 0.81 (Kindrat, 2007).

Two studies examined this relationship further using regression analysis. Lo Buono et al. (2023) used Poisson regression analysis to examine which factors predicted body image in MS. They reported that the depression dimension of the SCL-90-R was a significant predictor of body image ($p < .01$). Stevens et al. (2018) used a hierarchical linear regression to examine predictors of body image. In the first step of this analysis, researchers reported that sex, BMI, and depression were all significant predictors of body image ($p < .01$).

### ***Anxiety***

Two studies examined the relationship between body image and anxiety in MS using correlation analysis (Lo Buono et al., 2023; Sengul et al., 2019). Both studies found a positive significant relationship between the two variables, with higher levels of body dissatisfaction associated with higher levels of anxiety ($p<.001$). Similar to previous findings regarding depression, both of these studies reported strong effect sizes: 0.5 (Lo Buono et al., 2023) and 0.57 (Sengul et al., 2019).

### ***Self-esteem***

Three studies used Spearman’s correlation to examine the relationship between body image and self-esteem in MS. Two studies (Ghodusi & Heidari, 2014; Wilski et al., 2016) reported significant positive correlations, indicating that high levels of body satisfaction were associated with increased self-esteem. The effect sizes were 0.48 and 0.63. While this relationship did not reach significance in the study by Barak et al. (1998), the effect size was similar, at 0.43. The study may not have reached significance due to the sample size being lower than that of the other two.

### *Quality of Life*

Two studies used correlation analysis to examine the relationship between body image and QoL (Barbosa et al., 2023; Wilski et al., 2016). Both studies reported significant correlations between these variables, with effect sizes of 0.31 and 0.51. Results indicated that positive body image was associated with better QoL. Both studies used regression analysis to explore this relationship further.

Hierarchical multiple regression was used by Wilski et al. (2016) to examine predictors of body-esteem. All aspects of QoL, other than the physical dimension, were found to be significant predictors of body-esteem ($p \leq .001$). Stevens et al. (2018) used hierarchical linear regression to analyse predictors of body image. Findings demonstrated that QoL was not a significant predictor of body image in this particular study. Significant predictors of body image were found to be sex, body mass index, and low mood. Barbosa et al. (2023) used multiple linear regression to identify variables that predicted QoL. They found that 40.6% of the variance in general QoL was explained by all independent variables in the predictor model, which consisted of body functionality, meaning of life, body appreciation by others, body appreciation by self, and emotional regulation. The model reported that body appreciation accounted for 13% of variance in psychological QoL. Body acceptance by others accounted for 3.7% and 1.3% of variance in social QoL and environmental QoL, respectively. All of these studies used different measures to assess QoL (see Table 1-5).

### *Other Psychological Outcomes*

Farnam et al. (2017) used Pearson's correlation to examine the relationship between body image and emotion regulation, aggression, and stress. The researchers reported a significant negative correlation between body image and aggression ($p<.05$) and stress ($p<.05$), indicating that higher levels of body satisfaction are associated with lower levels of both aggression and stress. Similar effect sizes (0.26) were reported. No significant correlation was reported between body image and emotion regulation ($p > 0.05$).

The researchers conducted a stepwise regression analysis, which showed that body image was a predictor of stress. Across the six subscales of body image, appearance evaluation, body area satisfaction (an individual's satisfaction in specific aspects of their body), and fitness orientation (the extent to which an individual values being physically fit) accounted for 27% of variance in stress. The researchers conducted another regression analysis to identify variables predicting aggression, with body image found to be a significant predictor. Fitness evaluation was entered in the first step of the analysis, and accounted for 17% of variance in aggression; fitness evaluation and body area satisfaction were entered in the second step of the analysis, and they accounted for 23% of variance in aggression.

## Intervention Studies

### *Aim of Interventions*

Three studies administered body image and psychological outcome measures before and after a given intervention. While these studies did not directly examine the relationship between body image and psychological outcome, variables that move together may be indicative of possible correlation (Bewick et al., 2003). The first of these studies aimed to assess whether a five-week massage intervention would improve body image and self-esteem in individuals with MS (Hernandez-Reif et al., 1998). While the study did not provide theoretical evidence for a link between body image and self-esteem, it suggested that the intervention would show improvements in both, given the positive effects that massage has had on both mood and range of motion in other long-term health conditions (Field et al., 1997). A second intervention study by Tesar et al. (2003) examined the effects of a seven-week psychological therapy on anxiety and body image. The researchers integrated elements of CBT with specialised techniques such as relaxation and visualisation to increase participants’ body awareness. Similar to the previous study, the researchers did not provide evidence for a link between body image and psychological outcome, and assessed these post-therapy improvements independently of one another. Paolucci et al. (2022) assessed whether an eight-week programme consisting of physiotherapy and neuro-cognitive exercises would improve body image and QoL. The intervention was based on posture, proprioception, body image, breathing, body relaxation, motor skills, coordination, visuospatial coordination, and balance. The researchers proposed that encouraging individuals with MS to focus on these areas would have a positive impact on interoceptive awareness. They hypothesised that this intervention would improve QoL due to previous research demonstrating the effectiveness of physiotherapy and exercise in people with MS (Coulter at al., 2020).

### ***Results of Intervention Studies***

Two of the studies reported that there was an improvement in both body image and psychological outcome following intervention. Following massage therapy, participants reported improved self-esteem ($p=.02$) and body satisfaction ($p=.011$) (Hernandez-Reif et al., 1998). The physiotherapy and neurocognitive intervention (Paolucci et al., 2022) saw improvements in body image, with regards to body satisfaction ($p < .001$) and interoceptive awareness ($p= .0038$), as well as QoL ($p=.0179$). While Tesar et al. (2003) reported a significant reduction in depression and depressive coping for participants following the psychological therapy intervention, there were no significant changes found in body image scores. While the researchers concluded that the effects of the therapy were weaker for body image compared with psychological outcome, the reasons for this were not proposed. It might be that the exercises employed by the intervention to focus on body awareness and muscle relaxation were more associated with interoception, and less helpful for external aspects of body image.

## Discussion

The aim of this systematic review was to examine the association between body image and psychological wellbeing in MS. The limited number of studies included in this review (n=13) suggests that there has been little focus on this relationship to date. However, findings provide preliminary evidence to suggest that body image has important psychological implications for this population. Overall, the findings from this review suggest that positive body image is associated with lower psychological distress, although the causal direction of this relationship remains unclear.

The relationship most frequently examined was between body image and depression. A relationship between body image and depression in other long-term health conditions such as diabetes (Lee & Song, 2002) and head and neck cancer (Rhoten et al., 2014) has been demonstrated. The effect sizes yielded by these existing studies were 0.32 (Rhoten et al., 2014) and 0.68 (Lee & Song, 2002). The present review supports these earlier findings, demonstrating that a more positive image is associated with lower levels of depression, with similar moderate to large effect sizes reported across the studies. The direction of this relationship, in terms of cause and effect, however, remains unclear. There have been suggestions that depression can exacerbate body image dissatisfaction (Marsella et al., 1981), therefore it might be that people experiencing lower mood are more likely to appraise themselves negatively. Conversely, it might be that greater concern about one's body image may lead to lower mood (Di Cara et al., 2019). While the specific body image measures differed across the studies assessing the relationship between body image and depression, they were all focused on the appraisal or satisfaction of external appearance rather than interoceptive awareness. All of these studies reported a strong effect size, however Kindrat (2007) found a larger effect. This may be due to the sample in this study being the only one to consist solely of women. It has been suggested that women are more likely to have concerns about their bodies (Demarest & Allen, 2000), with media and societal pressures proposed as factors associated with these concerns (Bedford & Johnson, 2006; Choate, 2005). This increased pressure experienced by women may offer an explanation as to why body image was found to have a greater impact on depression within the Kindrat (2007) study. Given that MS is more prevalent in women, this finding provides further information regarding the relevance of body image for individuals with MS.

The findings from this review suggest that a more positive body image is associated with reduced levels of anxiety in MS, however the direction was again unclear. A similar finding between body image and anxiety in women post-menopause has been reported (Simbar et al., 2020). The measures used to assess anxiety in the present review viewed anxiety broadly, rather than specify the type of anxiety that people with MS were experiencing. There has been previous research evidencing an association between body concern and social anxiety in student populations (Bakhtiarpoor et al., 2011). It might be that perceived stigma associated with body image changes in MS may increase a person's sense of social anxiety, however further research is needed to better understand the nature of this relationship.

The findings from this review suggest that a more positive body image is associated with higher levels of self-esteem. This supports earlier findings reporting an association between body image and self-esteem in both people with MS (Di Cara et al., 2019) and the general population (Kostanski & Gullone, 1998). The link between body image and self-esteem is perhaps not surprising given the role that body image plays in a person's identity and sense of self (O'Dea, 2012). While this review provides further evidence for an association between body image and self-esteem in MS, as before, the direction of this relationship remains unclear. Existing literature reports that the progression of MS has a negative effect on an individual's self-esteem (Robati & Shareh, 2018). It is possible that lower levels of self-esteem arising from MS may lead to individuals experiencing their bodies in a more negative way. On the other hand, it is possible that physical changes in the body arising from MS may lead to lower levels of self-esteem.

The findings from this review suggest that positive body image is associated with higher levels of QoL. These findings support earlier research demonstrating an association between body image and QoL, both within the general population (Nayir et al., 2016) and in other long-term health conditions (Akkaya et al., 2012). While the factors associated with this relationship are not made fully clear within the context of this review, a number of plausible explanations exist. It is possible, for instance, that disease severity may affect this relationship in some way. Individuals who experience lower levels of disease severity may be able to engage more in activities of daily living, such as exercise, and therefore may not experience physical changes to their bodies in the same way that those living with higher levels of disease severity do. Increased disease severity may be associated with increased gait difficulties or sensory problems, which may negatively affect QoL, and may cause individuals to view their bodies in a less positive way. Stigma may also play a role in this relationship; individuals with higher levels of body dissatisfaction may be more likely to withdraw from social activities due to perceived stigma or social anxiety (Lillis et al., 2011). It is likely that the psychological processes underlying the relationship between body image and QoL will differ across varying aspects of QoL, such as environmental, physical or social. Future qualitative research may be helpful in disentangling these subtle differences.

There was a high level of heterogeneity across the studies in terms of measures used to assess body image. The term ‘body image’ in this review mirrored its use in literature, encompassing two distinct concepts: appraisal of external appearance, and interoceptive awareness. The variability in findings may reflect the different dimensions of body image under investigation. Barbosa et al. (2023) found significant relationships between QoL and body image, using three distinct body image measures, however the effect size was lower when the Body Responsiveness Scale (BRS) was used. The BRS focuses on interoceptive awareness, and therefore differences in effect may indicate differences in the relationship between psychological outcome and distinct dimensions of body image. Interestingly, the studies that used similar body image measures were most consistent in their findings. Two of the studies examining the relationship between body image and self-esteem used the Body Esteem Scale (BES) to assess body image (Barak et al. 1998; Wilski et al., 2016). The third study examining this relationship used the Physical Disability Body Esteem Questionnaire (PDBEQ), an alternative measure to assess the same construct (Ghodusi & Heidari, 2014). The results from these studies were consistent in terms of effect size reported. Similar consistencies were found in the studies that examined the relationship between body image and anxiety. Although the measures used were different, the key concepts being measured in both scales were similar.

There was diversity across the studies in terms of culture. Seven studies were published in Europe, three in North America, and three in Asia. It is likely that participants from different cultural backgrounds related to their bodies in different ways, with previous research indicating differences in body image across diverse ethnic groups (Ricciardelli et al., 2007). This echoes the aforementioned schematic model of body image (Slade, 1994), which describes how cultural factors can affect body image. People from collectivist cultures have been reported to have higher levels of body dissatisfaction than those from individualist cultures (Çakıcı et al., 2021). This may be due to a person's body being representative of more than just the individual (Gillen & Lefkowitz, 2012), or the result of differences in body ideals across cultures (Grabe et al., 2008; Shoraka et al., 2019). These cultural differences in body image may have accounted for some of the variation in effect sizes reported within this review, however strong conclusions cannot be made due to the small number of studies. It must also be said that despite the heterogeneity in countries that the studies were published in, this review excluded non-English studies, thus introducing a risk of bias (Lefebvre et al., 2023), and a potential lack of cultural representation.

## Clinical Implications

This present review suggests an association between body image and psychological outcome in MS. Importantly, the findings indicated that a more positive body image was associated with better mood, lower anxiety, increased self-esteem, greater QoL, and lower levels of stress. While the causal nature of these associations was not clear, it is likely that they may be bidirectional. Therefore, psychological interventions that target body image may be helpful in improving psychological outcome for people with MS.

There has been little focus in the literature on interventions aimed at improving body image in MS. While evidence is limited, the majority of research regarding the efficacy of psychological interventions for body image comes from oncology literature. A review by Sebri et al. (2021) reported that interventions including cognitive, social, mindfulness, and art therapy, were effective in reducing body image concerns among individuals with breast cancer. It is possible that such interventions may be also be helpful in targeting body image in MS. As mentioned previously, the English NICE (2009) guidelines currently recommend individual or group CBT for the management of psychological distress across all chronic physical conditions, however these do not take into account the specific challenges experienced by individuals with MS. Previous reviews have indicated that CBT may be helpful in managing depression for people struggling with adjustment in MS (Thomas et al., 2006).

There has been some evidence suggesting that CBT may be helpful in improving body image among the general population (Rosen et al., 1989). Approaches that use CBT for body image have found that techniques such as cognitive restructuring, psychoeducation, guided imagery, and exposure are helpful in improving body image and reducing unhelpful behaviours such as body checking (Alleva et al., 2015; Jarry & Ip, 2005). CBT has also been shown to have a positive effect on both image and sexual functioning in people with breast cancer (Hummel et al.,2018). While research regarding the efficacy of other psychological models for body image remain largely understudied, it is possible that such interventions may be helpful in MS populations. For example, Compassion Focused Therapy (CFT) supports individuals with high levels of shame to develop a more compassionate relationship with themselves (Gilbert, 2014), and has been proposed as a suitable model to use when working with individuals with neurological conditions (Ashworth & Murray, 2019). Shame may be particularly relevant when working with individuals with lower body image, as they can often experience negative feelings for not reaching socially desirable body standards (Duarte et al., 2015). Another psychological therapy, Acceptance and Commitment Therapy (ACT), may also be helpful as it supports acceptance of difficult life events, by encouraging people to actively attend to negative experiences (Hayes et al., 2006). The model supports individuals to move away from a place of cognitive fusion, where they find it difficult to separate themselves from negative thoughts or feelings, toward a more values-orientated way of life (Hayes et al., 2006). This model may be particularly helpful in supporting people to come to terms with changes in their bodies as a result of MS, with evidence suggesting an association between acceptance and better adjustment in this population (Pakenham & Fleming, 2011). Further evidence for use of this model comes from reports that cognitive fusion mediates the relationship between psychological distress and impact of illness in MS (Valvano et al., 2016). While further research is necessary to provide a greater understanding of the relationship between body image and psychological outcome in people with MS, the findings from this review highlight the need for a person-centred, holistic approach to working with people with this condition.

## Limitations

### *Limitations of the Literature*

There were limitations to the present review. Firstly, there has been little research conducted looking specifically at the relationship between body image and psychological outcome in MS, and the included studies varied greatly in terms of both methodology and analysis. The differences across these studies, such as differences in outcome measures or psychological concepts under examination, posed challenging to creating a meaningful narrative synthesis. Secondly, there were limitations in terms of study design and statistical analysis employed by the individual studies, to examine the relationship between body image and psychological outcome. The majority of included studies were cross-sectional in design, which can make it difficult to infer causation and directionality of associations (Chirico, 2023). Furthermore, a number of the studies in this review examined the relationship between body image and a range of different factors, rather than focusing on one specific psychological outcome. As a result, the associations reported were not explored in great depth. The results are indicative of underlying psychological processes mediating the relationship between body image and psychological outcome, however these processes are not yet fully understood. Possible mediating factors that require further investigation might be adjustment to illness, stigma, sexual functioning, and disease severity.

### *Limitations of the Review Process*

Due to the limited research in this area, three intervention studies were included, which did not directly examine the relationship between body image and psychological outcome. These studies varied, both in terms of intervention used and psychological concepts studied. While the findings offered preliminary evidence for a relationship between body image and psychological outcome, the results were inconsistent. It should be noted that these studies provide weaker evidence for associations than the studies that employed correlation or regression analysis. Furthermore, these studies all lacked any strong explanation of how body image theory informed the interventions.

The term ‘body image’ is used interchangeably to describe two rather distinct concepts: appraisal of external appearance and interoceptive awareness. The author made the decision early in the review process to include studies that examined both of these concepts. This decision was made to allow the researcher to understand how body image has been studied in MS previously. While this was helpful due to limited research in this area, the broad definition of body image created challenges in terms of heterogeneity across studies, and data synthesis.

## Future Research

Future research would benefit from longitudinal study designs, which have been reported to be more valid for examining cause and effect relationships (Caruana et al., 2015). This type of design would be helpful in providing greater understanding about the impact that disease progression has upon body image and psychological wellbeing in MS, as well as enabling understanding of how each concept predicts the other. This review identified studies that examined the relationship between body image and a wide range of psychological variables. Further investigation would benefit from more in-depth analysis of these relationships, taking into account any mediating factors underlying these associations. For instance, the studies that examined the relationship between body image and anxiety did not explore different facets of anxiety. Future research may consider the different types of anxiety that people with MS experience, for example social anxiety, panic, health anxiety or generalised anxiety. It would be helpful to then explore the relationships between each of these variables with different components of body image. A more complex statistical model may then be used to examine other factors that may be associated with this relationship, such as adjustment, support, stigma, or cognitive fusion. More detailed analysis would also be helpful to explore the direction of associations between body image and psychological outcome. This present review did not include qualitative studies. A future review may examine the lived experiences of body image among individuals with MS. This may be helpful in providing greater understanding about how people with MS experience body image, and how this occurs across different aspects of their lives. Finally, this review highlighted heterogeneity of body image measures used across studies. Further research to compare the psychometric properties of body image measures would be helpful to ensure consistency going forward.

## Conclusion

This review highlights the limited research into body image and MS. It offers preliminary evidence to suggest that a more positive body image is associated with better mood, lower anxiety, increased self-esteem, and greater QoL. Further research is recommended to explore these associations further, and to understand psychological processes underlying the relationship between body image and psychological outcome in MS. Increased understanding will allow clinicians to tailor interventions appropriately in support of a holistic and person-centred approach to care.

## Declaration of Interest

'Declarations of interest: none'

## Funding

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

## References

Abell, N. (1991). The Index of Clinical Stress: A brief measure of subjective stress for practice and research. *Social Work Research and Abstracts*, *27*(2), 12–15. https://doi.org/10.1093/swra/27.2.12

Akkaya, N., Akkaya, S., Atalay, N. S., Balci, C. S., & Sahin, F. (2012). Relationship between the body image and level of pain, functional status, severity of depression, and quality of life in patients with fibromyalgia syndrome. *Clinical Rheumatology*, *31*, 983-988. https://doi.org/10.1007/s10067-012-1965-9

Allen, M. S., & Walter, E. E. (2016). Personality and body image: A systematic review. *Body Image*, *19*, 79–88. https://doi.org/10.1016/j.bodyim.2016.08.012

Alleva, J. M., Sheeran, P., Webb, T. L., Martijn, C., & Miles, E. (2015). A meta-analytic review of stand-alone interventions to improve body image. *PLoS One*, *10*(9), e0139177. https://doi.org/10.1371/journal.pone.0139177

Amato, M. P., Zipoli, V., & Portaccio, E. (2008). Cognitive changes in multiple sclerosis. *Expert Review of Neurotherapeutics*, *8*(10), 1585–1596. https://doi.org/10.1586/14737175.8.10.1585

Amezcua, L., & McCauley, J. L. (2020). Race and ethnicity on MS presentation and disease course. *Multiple Sclerosis Journal*, *26*(5), 561–567. https://doi.org/10.1177/1352458519887328

Ashworth, F., & Murray, C. (2019). Compassion focused therapy for neurological conditions. In *Psychological therapies in acquired brain injury* (pp. 75-94). Routledge. https://doi.org/10.4324/9780429506796-5

Badoud, D., & Tsakiris, M. (2017). From the body's viscera to the body's image: Is there a link between interoception and body image concerns?. *Neuroscience & Biobehavioral Reviews*, *77*, 237-246. https://doi.org/10.1016/j.neubiorev.2017.03.017

Bakhtiarpoor, S., Heidarie, A., & Khodadadi, S. A. (2011). The relationship of the self-focused attention, body image concern and generalized self-efficacy with social anxiety in students. *Life Science Journal*, *8*(4), 704-713.

Balogun, J. A. (1986). Reliability and construct validity of the Body Cathexis Scale. *Perceptual and Motor Skills*, *62*(3), 927-935. https://doi.org/10.2466/pms.1986.62.3.927

Barak, Y., Lampl, Y., Sarova-Pinchas, I., & Achiron, A. (1998). Self and body esteem perception in multiple sclerosis. *Behavioural neurology*, *11*(3), 159-161. https://doi.org/10.1155/1999/976453

Barbosa, M. C., Torres, S., Barbosa, R., Vieira, F., Lencastre, L., & Guerra, M. P. (2023). Quality of Life in Multiple Sclerosis: The Predictive Roles of Positive Body Image and Meaning in Life. *Sclerosis*, *1*(2), 76-90. https://doi.org/10.3390/sclerosis1020010

Bedford, J. L., & Johnson, C. S. (2006). Societal influences on body image dissatisfaction in younger and older women. *Journal of Women & Aging*, *18*(1), 41-55. https://doi.org/10.1300/J074v18n01_04

Bewick, V., Cheek, L., & Ball, J. (2003). Statistics review 7: Correlation and regression. *Critical care*, *7*, 1-9. https://doi.org/10.1186/cc2401.

Bobholz, J. A., & Gremley, S. (2011). Multiple Sclerosis and Other Demyelinating Disorders. In *The Little Black Book of Neuropsychology. A Syndrome Based Approach* (pp. 647–662). Springer. https://doi.org/10.1007/978-0-387-76978-3_20

Boeschoten, R. E., Braamse, A. M. J., Beekman, A. T. F., Cuijpers, P., van Oppen, P., Dekker, J., & Uitdehaag, B. M. J. (2017). Prevalence of depression and anxiety in Multiple Sclerosis: A systematic review and meta-analysis. *Journal of the Neurological Sciences*, *372*, 331–341. https://doi.org/10.1016/j.jns.2016.11.067

Borkoles, E., Nicholls, A. R., Bell, K., Butterly, R., & Polman, R. C. J. (2008). The lived experiences of people diagnosed with multiple sclerosis in relation to exercise. *Psychology & Health*, *23*(4), 427–441. https://doi.org/10.1080/14768320701205309

Borzekowski, D. L., & Bayer, A. M. (2005). Body image and media use among adolescents. *Adolesc Med Clin*, *16*(2), 289-313. https://doi.org/10.1016/j.admecli.2005.02.010

Brown, T. A., Cash, T. F., & Mikulka, P. J. (1990). Attitudinal body-image assessment: Factor analysis of the Body-Self Relations Questionnaire. *Journal of personality assessment*, *55*(1-2), 135-144. https://doi.org/10.1080/00223891.1990.9674053

Buss, A. H., & Perry, M. (1992). The aggression questionnaire. *Journal of personality and social psychology*, *63*(3), 452. https://doi.org/ 10.1037//0022-3514.63.3.452

Çakıcı, K., Mercan, Z., & Nazlıgül, M. D. (2021). A Systematic Review of Body Image and Related Psychological Concepts: Does Ethnicity Matter? *Psikiyatride Guncel Yaklasimlar*, *13*(4), 707-725. https://doi.org/10.18863/pgy.869244

Campbell, J. A., Simpson Jr, S., Ahmad, H., Taylor, B. V., van der Mei, I., & Palmer, A. J. (2020). Change in multiple sclerosis prevalence over time in Australia 2010–2017 utilising disease-modifying therapy prescription data. *Multiple Sclerosis Journal, 26*(11), 1315-1328. https://doi.org/10.1177/1352458519861270

Caruana, E. J., Roman, M., Hernández-Sánchez, J., & Solli, P. (2015). Longitudinal studies. *Journal of thoracic disease*, *7*(11), E537. https://doi.org/ 10.3978/j.issn.2072-1439.2015.10.63

Cash, T. F. (2004). Body image: past, present, and future. *Body Image*, *1*(1), 1–5. https://doi.org/10.1016/s1740-1445(03)00011-1

Cash, T. F., & Szymanski, M. L. (1995). The Development and Validation of the Body-Image Ideals Questionnaire. *Journal of Personality Assessment*, *64*(3), 466–477. https://doi.org/10.1207/s15327752jpa6403_6

Cheli, S., Agostini, A., Herd-Smith, A., Caligiani, L., Martella, F., & Fioretto, L. (2016). The Italian version of Body Image Scale Reliability and Sensitivity in a sample of breast cancer patients. *Psicoterapia Cognitiva e Comportamentale*, *22*(1). https://doi.org/10.1037/t56642-000

Chirico  F. (2023). Cross-sectional studies in occupational health research: An overview of strengths and limitations. *G Ital Psicol Med Lav*, *3*(3), 86-93.

Choate, L. H. (2005). Toward a theoretical model of women's body image resilience. *Journal of Counseling & Development*, *83*(3), 320-330. https://doi.org/10.1002/j.1556-6678.2005.tb00350.x

Connell, K. M., Coates, R., Doherty-Poirier, M., & Wood, F. M. (2013). A Literature Review to Determine the Impact of Sexuality and Body Image Changes Following Burn Injuries. *Sexuality and Disability*, *31*(4), 403–412. https://doi.org/10.1007/s11195-013-9321-9

Costantini, L., Pasquarella, C., Odone, A., Colucci, M. E., Costanza, A., Serafini, G., ... & Amerio, A. (2021). Screening for depression in primary care with Patient Health Questionnaire-9 (PHQ-9): A systematic review. *Journal of affective disorders*, *279*, 473-483. https://doi.org/10.1016/j.jad.2020.09.131

Costantini, M. (2014). Body perception, awareness, and illusions. *Wiley Interdisciplinary Reviews: Cognitive Science*, *5*(5), 551–560. https://doi.org/10.1002/wcs.1309

Coulter, E. H., Bond, S., Dalgas, U., & Paul, L. (2020). The effectiveness of interventions targeting physical activity and/or sedentary behaviour in people with Multiple

Sclerosis: a systematic review. *Disability and rehabilitation*, *42*(5), 594-612. https://doi.org/10.1080/09638288.2018.1503737

Cummings, S., Browner, W., & Hulley, S. (2007). Conceiving the research question and developing the study plan. In S. Hulley, S. Cummings, & W. Browner (Eds.), *Designing Clinical Research: An Epidemiological Approach* (pp. 14–22). Lippincott Williams & Wilkins.

Demarest, J., & Allen, R. (2000). Body image: Gender, ethnic, and age differences. *The Journal of social psychology*, *140*(4), 465-472. https://doi.org/10.1080/00224540009600485

Derogatis, L. R. (1992). SCL-90-R: Administration, scoring & procedures manual-II for the (revised) version and other instruments of the psychopathology rating scale series. *Clinical Psychometric Research.*, 1-16.

Di Cara, M., Lo Buono, V., Corallo, F., Cannistraci, C., Rifici, C., Sessa, E., D'Aleo, G., Bramanti, P., & Marino, S. (2019). Body image in multiple sclerosis patients: a descriptive review. *Neurological Sciences*, *40*(5), 923–928. https://doi.org/10.1007/s10072-019-3722-1

Dobson, R., & Giovannoni, G. (2018). Multiple sclerosis - a review. *European Journal of Neurology*, *26*(1), 27–40. https://doi.org/10.1111/ene.13819

Dozois, D. J., Dobson, K. S., & Ahnberg, J. L. (1998). A psychometric evaluation of the Beck Depression Inventory–II. *Psychological assessment*, *10*(2), 83. https://doi.org/10.1037/1040-3590.10.2.83

Duarte, C., Pinto-Gouveia, J., Ferreira, C., & Batista, D. (2015). Body image as a source of shame: A new measure for the assessment of the multifaceted nature of body image shame. *Clinical psychology & psychotherapy*, *22*(6), 656-666. https://doi.org/10.1002/cpp.1925

Ebers, G. C. (1992). The geography of MS reflects genetic susceptibility. *Journal of Tropical and Geographical Neurology*, *2*, 66-72.

Eriksen, M. B., & Frandsen, T. F. (2018). The Impact of patient, intervention, comparison, Outcome (PICO) as a Search Strategy Tool on Literature Search quality: a Systematic Review. *Journal of the Medical Library Association*, *106*(4), 420–431. NCBI. https://doi.org/10.5195/jmla.2018.345

Evans, C., & Dolan, B. (1993). Body Shape Questionnaire: derivation of shortened "alternate forms". *International Journal of Eating Disorders*, *13*(3), 315-321. https://doi.org/10.1002/1098-108X(199304)13:3<315::AID-EAT2260130310>3.0.CO;2-3

Farnam, A., Marashi, F., & Sana'tnama, M. (2017). The relationship of body image with emotion regulation, stress, and aggression and their comparison between males and females with multiple sclerosis. *Jundishapur Journal of Chronic Disease Care*, *6*(3). https://doi.org/10.5812/jjcdc.13818

Field, T. M., Sunshine, W., Hernandezreif, M., Quintino, O., Schanberg, S., Kuhn, C., & Burman, I. (1997). Massage therapy effects on depression and somatic symptoms in chronic fatigue syndrome. *Journal of chronic fatigue syndrome*, *3*(3), 43-51. https://doi.org/10.1300/J092v03n03_03

Franzoi, S. L., & Shields, S. A. (1984). The Body Esteem Scale: Multidimensional structure and sex differences in a college population. *Journal of personality assessment*, *48*(2), 173-178. https://doi.org/10.1207/s15327752jpa4802_12

Ghasemi, N., Razavi, S., & Nikzad, E. (2017). Multiple sclerosis: pathogenesis, symptoms, diagnoses and cell-based therapy. *Cell Journal (Yakhteh)*, *19*(1), 1. https://doi.org/ 10.22074/cellj.2016.4867

Ghodusi, M., & Heidari, M. (2014). The relationship between body image and selfesteem among multiple sclerosis patients.

Gilbert, P. (2014). The origins and nature of compassion focused therapy. *British Journal of Clinical Psychology*, *53*(1), 6–41. https://doi.org/10.1111/bjc.12043

Gil-González, I., Martín-Rodríguez, A., Conrad, R., & Pérez-San-Gregorio, M. Á. (2020). Quality of life in adults with multiple sclerosis: a systematic review. *BMJ Open*, *10*(11), e041249. https://doi.org/10.1136/bmjopen-2020-041249

Gillen, M. M., & Lefkowitz, E. S. (2012). Gender and racial/ethnic differences in body image development among college students. *Body image*, *9*(1), 126-130. https://doi.org/10.1016/j.bodyim.2011.09.004

Grabe, S., Ward, L. M., & Hyde, J. S. (2008). The role of the media in body image concerns among women: a meta-analysis of experimental and correlational studies. *Psychological bulletin*, *134*(3), 460. https://doi.org/10.1037/0033-2909.134.3.460

Gunnersen, S. R., Hanehøj, K., Gro, S., Petersen, C. M., & Skovgaard, L. (2022). The effects of a 12-week yoga intervention on body awareness in people with multiple sclerosis: A non-controlled pilot study. *Advances in Integrative Medicine*, *9*(3), 167-172. https://doi.org/10.1016/j.aimed.2022.06.001

Hanna, M., & Strober, L. B. (2020). Anxiety and Depression in Multiple Sclerosis (MS): Antecedents, Consequences, and Differential Impact on Well-being and Quality of Life. *Multiple Sclerosis and Related Disorders*, *44*, 102261. https://doi.org/10.1016/j.msard.2020.102261

Hayes, S. C., Luoma, J. B., Bond, F. W., Masuda, A., & Lillis, J. (2006). Acceptance and Commitment Therapy: Model, processes and outcomes. *Behaviour Research and Therapy*, *44*(1), 1–25. https://doi.org/10.1016/j.brat.2005.06.006

Hernandez-Reif, M., Field, T., Field, T., & Theakston, H. (1998). Multiple sclerosis patients benefit from massage therapy. *Journal of Bodywork and Movement therapies*, *2*(3), 168-174. https://doi.org/10.1016/S1360-8592(98)80009-0

Higgins, J., & Thomas, J. (2023). *Cochrane Handbook For Systematic Reviews Of Interventions.* (2nd ed.). Wiley-Blackwell.

Hind, D., Cotter, J., Thake, A., Bradburn, M., Cooper, C., Isaac, C., & House, A. (2014). Cognitive behavioural therapy for the treatment of depression in people with multiple sclerosis: a systematic review and meta-analysis. *BMC psychiatry*, *14*(1), 1-13. https://doi.org/10.1186/1471-244X-14-5

Hosseini, Z., Homayuni, A., & Etemadifar, M. (2022). Barriers to quality of life in patients with multiple sclerosis: a qualitative study. *BMC Neurology*, *22*(1). https://doi.org/10.1186/s12883-022-02700-7

Hummel, S. B., van Lankveld, J. J., Oldenburg, H. S., Hahn, D. E., Kieffer, J. M., Gerritsma, M. A., ... & Aaronson, N. K. (2018). Internet-based cognitive behavioral therapy realizes long-term improvement in the sexual functioning and body image of breast cancer survivors. *Journal of sex & marital therapy*, *44*(5), 485-496. https://doi.org/10.1080/0092623X.2017.1408047

Ibánez, M. I., Ruipérez, M. A., Moya, J., Marqués, M. J., & Ortet, G. (2005). A short version of the self-regulation inventory (SRI-S). *Personality and individual differences*, *39*(6), 1055-1059. https://doi.org/10.1016/j.paid.2005.02.029

Jarry, J. L., & Ip, K. (2005). The effectiveness of stand-alone cognitive-behavioural therapy for body image: A meta-analysis. *Body image*, *2*(4), 317-331. https://doi.org/10.1016/j.bodyim.2005.10.001

Kamińska, J., Koper, O. M., Piechal, K., & Kemona, H. (2017). Multiple sclerosis - etiology and diagnostic potential. *Postepy Higieny I Medycyny Doswiadczalnej (Online)*, *71*(0), 551–563. https://doi.org/10.5604/01.3001.0010.3836

Karatepe, A. G., Kaya, T., Günaydn, R., Demirhan, A., Ce, P., & Gedizlioglu, M. (2011). Quality of life in patients with multiple sclerosis: the impact of depression, fatigue, and disability. *International Journal of Rehabilitation Research*, *34*(4), 290-298. https://doi.org/10.1016/S0022-510X(02)00312-X

Kindrat, S. (2007). The relationship between body image and depression in women diagnosed with relapsing remitting multiple sclerosis. *Can J Neurosci Nurs*, *29*(1), 8-13. https://doi.org/10.3390/jcm12103606

Kmet, L. M., Cook, L. S., & Lee, R. C. (2004). Standard quality assessment criteria for evaluating primary research papers from a variety of fields. https://doi.org/10.7939/R37M04F16

Kostanski, M., & Gullone, E. (1998). Adolescent body image dissatisfaction: Relationships with self-esteem, anxiety, and depression controlling for body mass. *The Journal of Child Psychology and Psychiatry and Allied Disciplines*, *39*(2), 255-262. https://doi.org/10.1017/S0021963097001807

Lee, M. H., & Song, M. S. (2002). Relationship between body image and depression of diabetes mellitus patients. *The Korean Journal of Rehabilitation Nursing*, *5*(1), 49-59.

Lefebvre, C., Glanville, J., Briscoe, S., Featherstone, R., Littlewood, A., Metzendorf, M.-I., Noel-Storr, A., Paynter, R., Rader, T., Thomas, J., & Wieland, L. S. (2023). Chapter 4: Searching for and selecting studies. In J. Higgins & J. Thomas (Eds.), *Cochrane Handbook for Systematic Reviews of Interventions Version 6.4*.

Lehmann, V., Hagedoorn, M., & Tuinman, M. A. (2015). Body image in cancer survivors: a systematic review of case-control studies. *Journal of Cancer Survivorship*, *9*, 339-348. https://doi.org/10.1007/s11764-014-0414-y

Lemoine, J. E., Konradsen, H., Jensen, A. L., Roland-Levy, C., Ny, P., Khalaf, A., & Torres, S. (2018). Factor structure and psychometric properties of the Body Appreciation Scale-2 among adolescents and young adults in Danish, Portuguese, and Swedish. *Body image*, *26*, 1-9. https://doi.org/10.1016/j.bodyim.2018.04.004

Leray, E., Moreau, T., Fromont, A., & Edan, G. (2016). Epidemiology of multiple sclerosis. *Revue neurologique*, *172*(1), 3-13. https://doi.org/10.1016/j.neurol.2015.10.006

Lillis, J., Levin, M. E., & Hayes, S. C. (2011). Exploring the relationship between body mass index and health-related quality of life: A pilot study of the impact of weight self-stigma and experiential avoidance. *Journal of health psychology*, *16*(5), 722-727. https://doi.org/10.1177/13591053103883

Lo Buono, V., Bonanno, L., Corallo, F., Cardile, D., D'Aleo, G., Rifici, C., ... & De Cola, M. C. (2023). The Relationship between Body Image, Disability and Mental Health in Patients with Multiple Sclerosis. *Journal of Clinical Medicine*, *12*(10), 3606.https://doi.org/10.3390/jcm12103606

Lublin, F. D., Coetzee, T., Cohen, J. A., Marrie, R. A., & Thompson, A. J. (2020). The 2013 clinical course descriptors for multiple sclerosis. *Neurology*, *94*(24), 1088–1092. https://doi.org/10.1212/wnl.0000000000009636

Luca, M., Eccles, F., Perez Algorta, G., & Patti, F. (2022). Illness perceptions and outcome in multiple sclerosis: A systematic review of the literature. *Multiple Sclerosis and Related Disorders*, *67*, 104180. https://doi.org/10.1016/j.msard.2022.104180

Marsella, A. J., Shizuru, L., Brennan, J., & Kameoka, V. (1981). Depression and body image satisfaction. *Journal of cross-cultural psychology*, *12*(3), 360-371. https://doi.org/10.1177/00220221811230

McCabe, M. P., & McKern, S. (2002). Quality of life and multiple sclerosis: Comparison between people with multiple sclerosis and people from the general population. *Journal of Clinical Psychology in Medical Settings*, *9*, 287-295. https://doi.org/10.1023/A:1020734901150

Mehling, W. E., Price, C., Daubenmier, J. J., Acree, M., Bartmess, E., & Stewart, A. (2012). The multidimensional assessment of interoceptive awareness (MAIA). *PloS one*, *7*(11), e48230. https://doi.org/10.1371/journal.pone.0208034

Milo, R., & Kahana, E. (2010). Multiple sclerosis: geoepidemiology, genetics and the environment. *Autoimmunity reviews*, *9*(5), A387-A394. https://doi.org/10.1016/j.autrev.2009.11.010

Moher, D. (2019). Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *Annals of Internal Medicine*, *151*(4), 264. https://doi.org/10.7326/0003-4819-151-4-200908180-00135

National Institute for Health and Care Excellence. (2009). Depression in adults with a chronic physical health problem: recognition and management. https://www.nice.org.uk/guidance/cg91/chapter/Recommendations#step-4-complex-and-severe-depression

Nayir, T., Uskun, E., Yürekli, M. V., Devran, H., Çelik, A., & Okyay, R. A. (2016). Does body image affect quality of life?: A population based study. *PloS one*, *11*(9), e0163290. https://doi.org/10.1371/journal.pone.0163290

O'Dea, J. A. (2012). Body image and self-esteem. In T. F. Cash (Ed.), *Encyclopedia of body image and human appearance* (pp. 141–147). Elsevier Academic Press.

Pakenham, K. I., & Fleming, M. (2011). Relations between acceptance of multiple sclerosis and positive and negative adjustments. *Psychology & health*, *26*(10), 1292-1309. https://doi.org/10.1080/08870446.2010.517838

Paolucci, T., de Sire, A., Agostini, F., Bernetti, A., Salomè, A., Altieri, M., ... & Paoloni, M. (2022). Efficacy of interoceptive and embodied rehabilitative training protocol in patients with mild multiple sclerosis: A randomized controlled trial. *Frontiers in Neurology*, *13*, 1095180. https://doi.org/10.3389/fneur.2022.1095180

Pfaffenberger, N., Gutweniger, S., Kopp, M., Seeber, B., Stürz, K., Berger, T., & Günther, V. (2011). Impaired body image in patients with multiple sclerosis. *Acta Neurologica Scandinavica*, *124*(3), 165-170. https://doi.org/10.1111/j.1600-0404.2010.01460.x

Popay, J., Roberts, H. M., Sowden, A., Petticrew, M., Arai, L., Rodgers, M., & Britten, N. (2006). Guidance on the conduct of narrative synthesis in systematic reviews.

Przezdziecki, A., Sherman, K. A., Baillie, A., Taylor, A., Foley, E., & Stalgis-Bilinski, K. (2012). My changed body: breast cancer, body image, distress and self-compassion. *Psycho-Oncology*, *22*(8), 1872–1879. https://doi.org/10.1002/pon.3230

Ramagopalan, S. V., Dobson, R., Meier, U. C., & Giovannoni, G. (2010). Multiple sclerosis: risk factors, prodromes, and potential causal pathways. *The Lancet Neurology*, *9*(7), 727–739. https://doi.org/10.1016/S1474-4422(10)70094-6

Reynolds, W. M., & Gould, J. W. (1981). A psychometric investigation of the standard and short form Beck Depression Inventory. *Journal of consulting and clinical psychology*, *49*(2), 306. https://doi.org/10.1037/0022-006X.49.2.306

Rhoten, B. A., Deng, J., Dietrich, M. S., Murphy, B., & Ridner, S. H. (2014). Body image and depressive symptoms in patients with head and neck cancer: an important relationship. *Supportive Care in Cancer*, *22*, 3053-3060. https://doi.org/ 10.1007/s00520-014-2312-2

Ricciardelli, L. A., McCabe, M. P., Williams, R. J., & Thompson, J. K. (2007). The role of ethnicity and culture in body image and disordered eating among males. *Clinical psychology review*, *27*(5), 582-606. https://doi.org/10.1016/j.cpr.2007.01.016

Robati, Z., & Shareh, H. (2018). The effectiveness of cognitive-behavioral group therapy in anxiety and self-esteem in patients with multiple sclerosis. *Journal of Fundamentals of Mental Health*, *20*(6).

Rosen, J. C., Saltzberg, E., & Srebnik, D. (1989). Cognitive behavior therapy for negative body image. *Behavior therapy*, *20*(3), 393-404. https://doi.org/10.1016/S0005-7894(89)80058-9

Sá, M. J. (2008). Psychological aspects of multiple sclerosis. *Clinical Neurology and Neurosurgery*, *110*(9), 868–877. https://doi.org/10.1016/j.clineuro.2007.10.001

Samonds, R. J., & Cammermeyer, M. (1989). Perceptions of Body Image in Subjects with Multiple Sclerosis: A Pilot Study. *Journal of Neuroscience Nursing*, *21*(3), 190–194. https://doi.org/10.1097/01376517-198906000-00010

Schmalbach, I., Schmalbach, B., Zenger, M., Berth, H., Albani, C., Petrowski, K., & Brähler, E. (2020). A brief assessment of body image perception: Norm Values and Factorial Structure of the Short Version of the FKB-20. *Frontiers in Psychology*, *11*, 579783. https://doi.org/10.3389/fpsyg.2020.579783

Sebri, V., Durosini, I., Triberti, S., & Pravettoni, G. (2021). The efficacy of psychological intervention on body image in breast cancer patients and survivors: a systematic-review and meta-analysis. *Frontiers in Psychology*, *12*, 611954. https://doi.org/10.3389/fpsyg.2021.611954

Sengul, H. S., Sengul, Y., Tak, A. Z. A., Kocak, M., & Tunc, A. (2019). Evaluation of body image perception in multiple sclerosis patients without neurological deficit. *Ideggyogy Sz*, *72*(1-2), 49-54. http://dx.doi.org/10.18071/isz.71.0049

Shoraka, H., Amirkafi, A., & Garrusi, B. (2019). Review of body image and some of contributing factors in Iranian population. *International journal of preventive medicine*, *10*. https:// 10.4103/ijpvm.IJPVM_293_18

Simbar, M., Nazarpour, S., Alavi Majd, H., Dodel Andarvar, K., Jafari Torkamani, Z., & Alsadat Rahnemaei, F. (2020). Is body image a predictor of women's depression and anxiety in postmenopausal women? *BMC psychiatry*, *20*(1), 1-8. https://doi.org/ 10.1186/s12888-020-02617-w

Simpson, S., Blizzard, L., Otahal, P., Van der Mei, I., & Taylor, B. (2011). Latitude is significantly associated with the prevalence of multiple sclerosis: a meta-analysis. *Journal of Neurology, Neurosurgery & Psychiatry*, *82*(10), 1132–1141. https://doi.org/10.1136/jnnp.2011.240432

Slade, P. D. (1994). What is body image? *Behaviour research and therapy*. https://doi.org/10.1016/0005-7967(94)90136-8

Steer, R. A., & Beck, A. T. (1997). Beck Anxiety Inventory. In C. P. Zalaquett & R. J. Wood (Eds.), *Evaluating stress: A book of resources* (pp. 23–40). Scarecrow Education.

Stevens, S. D., Thompson, N. R., & Sullivan, A. B. (2018). Prevalence and Correlates of Body Image Dissatisfaction in Patients with Multiple Sclerosis. *International Journal of MS Care*. https://doi.org/10.7224/1537-2073.2018-066

Stokes, R., & Frederick-Recascino, C. (2003). Women's Perceived Body Image: Relations with Personal Happiness. *Journal of Women & Aging*, *15*(1), 17–29. https://doi.org/10.1300/j074v15n01_03

Swami, V., Todd, J., Stieger, S., & Tylka, T. L. (2020). The Body Acceptance by Others Scale: An assessment of its factorial validity in adults from the United Kingdom. *Body image*, *35*, 71-74. https://doi.org/10.1016/j.bodyim.2020.08.006

Tesar, N., Baumhackl, U., Kopp, M., & Günther, V. (2003). Effects of psychological group therapy in patients with multiple sclerosis. *Acta Neurologica Scandinavica*, *107*(6), 394-399. https://doi.org/10.1034/j.1600-0404.2003.00077.x

Thomas, P. W., Thomas, S., Hillier, C., Galvin, K., & Baker, R. (2006). Psychological interventions for multiple sclerosis. *Cochrane database of systematic reviews*, (1). https://doi.org/10.1002/14651858.CD004431.pub2

Tinakon, W., & Nahathai, W. (2012). A comparison of reliability and construct validity between the original and revised versions of the Rosenberg Self-Esteem Scale. *Psychiatry investigation*, *9*(1), 54. https://doi.org/10.4306/pi.2012.9.1.54

Todd, J., Aspell, J. E., Barron, D., & Swami, V. (2019). Multiple dimensions of interoceptive awareness are associated with facets of body image in British adults. *Body Image*, *29*, 6-16. https://doi.org/10.1016/j.bodyim.2019.02.003

Torres, S., Vieira, A. I., Vieira, F. M., Lencastre, L., Guerra, M. P., Miller, K., & Barbosa, M. R. (2023). Boosting multidimensional assessment of body image: Psychometric analysis of the Body Responsiveness Questionnaire in the Portuguese population. https://doi.org/10.21203/rs.3.rs-3097874/v1

Tsakiris, M. (2010). My body in the brain: A neurocognitive model of body-ownership. *Neuropsychologia*, *48*(3), 703–712. https://doi.org/10.1016/j.neuropsychologia.2009.09.034

Valvano, A., Floyd, R. M., Penwell-Waines, L., Stepleman, L., Lewis, K., & House, A. (2016). The relationship between cognitive fusion, stigma, and well-being in people with multiple sclerosis. *Journal of Contextual Behavioral Science*, *5*(4), 266-270. https://doi.org/10.1016/j.jcbs.2016.07.003

Victorson, D., Cavazos, J. E., Holmes, G. L., Reder, A. T., Wojna, V., Nowinski, C., ... & Cella, D. (2014). Validity of the Neurology Quality-of-Life (Neuro-QoL) measurement system in adult epilepsy. *Epilepsy & Behavior*, *31*, 77-84. https://doi.org/10.1016/j.yebeh.2013.11.008

Walton, C., King, R., Rechtman, L., Kaye, W., Leray, E., Marrie, R. A., Robertson, N., La Rocca, N., Uitdehaag, B., van der Mei, I., Wallin, M., Helme, A., Angood Napier, C., Rijke, N., & Baneke, P. (2020). Rising prevalence of multiple sclerosis worldwide: Insights from the Atlas of MS, third edition. *Multiple Sclerosis Journal*, *26*(14), 1816–1821. https://doi.org/10.1177/1352458520970841

Wang, J., Shaw, A., & Jackson, T. (2022). Validation of the Body Acceptance by Others Scale-2 (BAOS-2) and assessment of its associations with prospective changes in body image among young adults in China. *Body Image*, *43*, 112-124. https://doi.org/10.1016/j.bodyim.2022.08.017

Ware, J. E., Kosinski, M., & Keller, S. D. (1996). A 12-Item Short-Form Health Survey: construction of scales and preliminary tests of reliability and validity. *Medical care*, *34*(3), 220-233. http://doi.org/ 10.1097/00005650-199603000-00003

Ware, M., O'Connor, P., Bub, K., Backus, D., & McCully, K. (2023). Investigating Relationships Among Interoceptive Awareness, Emotional Susceptibility, and Fatigue in Persons With Multiple Sclerosis. *International Journal of MS Care*, *25*(2), 75-81. https://doi.org/10.7224/1537-2073.2022-007

Welch, V., Petkovic, J., Jull, J., Hartling, L., Klassen, T., Kristjansson, E., Pardo Pardo, J., Petticrew, M., J Stott, D., Thomson, D., Ueffing, E., Williams, K., Young, C., & Tugwell, P. (2023). Equity and specific populations. In J. Higgins & J. Thomas (Eds.), *Cochrane Handbook for Systematic Reviews of Interventions version 6.4*. Cochrane.

Wilski, M., T. Tasiemski, and A. Dąbrowski. Body esteem among women with multiple sclerosis and its relationship with demographic, clinical and socio-psychological factors. *International journal of behavioral medicine* 23 (2016): 340-347. https://doi.org/10.1007/s12529-015-9527-6

Zarotti, N., Eccles, F., Broyd, A., Longinotti, C., Mobley, A., & Simpson, J. (2023). Third wave cognitive behavioural therapies for people with multiple sclerosis: a scoping review. *Disability and Rehabilitation*, *45*(10), 1720-1735. https://doi.org/10.1080/09638288.2022.2069292

Zavoreo, I., Gržinčić, T., Preksavec, M., Madžar, T., & Bašić Kes, V. (2016). Sexual Dysfunction and Incidence of Depression in Multiple Sclerosis Patients. *Acta Clinica Croatica*, *55*(3), 402–406. https://doi.org/10.20471/acc.2016.55.03.08

## Tables and Figures

**Table 1-1**

**Database Search Strategy**

CINAHL [128 hits]

| Concept 1 | Concept 2 |
|---|---|
| Multiple Sclerosis<br>• MH "Multiple Sclerosis+"<br>• MH "Demyelinating Diseases+"<br>• Free text: “multiple sclerosis” OR MS OR “demyelinating disease” OR “relapsing remitting” OR “sclerosis” OR (“autoimmune disease” N3 “nervous system”) | Body Image<br>• MH "Body Image+"<br>• MH "Body Dissatisfaction"<br>• MH "Intraoperative Awareness"<br>• Free text: "body image" OR "body percep*" OR "body dissatisfac*" OR "body satisfac* OR "body esteem" OR "body aware*" OR “body interocept*” OR “body-image” OR “body-percep*” OR “body-dissatisfac*” OR “body-satisfac*” OR “body-esteem” OR “body-aware” OR “body-interocept*” OR “body concept” OR (body N5 (image OR percep* OR satisfac* OR disatisfact* OR esteem OR aware* OR self-percep* OR self-percep* OR concept) |

MEDLINE [339 hits]

| Concept 1 | Concept 2 |
|---|---|
| Multiple Sclerosis<br>• MH "Multiple Sclerosis<br>• MH "Multiple Sclerosis, Chronic Progressive"<br>• MH "Multiple Sclerosis, Relapsing-Remitting"<br>• Free text: “multiple sclerosis” OR “MS” OR “demyelinating disease” OR “relapsing remitting” OR “sclerosis” OR (“autoimmune disease” N3 “nervous system”) | Body Image<br>• MH "Body Image+"<br>• MH "Body Dissatisfaction"<br>• MH "Intraoperative Awareness"<br>• Free text: "body image" OR "body percep*" OR "body dissatisfac*" OR "body satisfac* OR "body esteem" OR "body aware*" OR “body interocept*” OR “body-image” OR “body-percep*” OR “body-dissatisfac*” OR “body-satisfac*” OR “body-esteem” OR “body-aware” OR “body-interocept*” OR “body concept” OR (body N5 (image |

| | |
|---|---|
| | OR percep* OR satisfac* OR disatisfact* OR esteem OR aware* OR self-percep* OR self-percep* OR concept) |

PSYCINFO [169 hits]

| Concept 1 | Concept 2 |
|---|---|
| Multiple Sclerosis<br>• DE "Multiple Sclerosis"<br>• DE "Sclerosis (Nervous System)”<br>• DE "Demyelination"<br>• Free text: “multiple sclerosis” OR MS OR “demyelinating disease” OR “relapsing remitting” OR “sclerosis” OR (“autoimmune disease” N3 “nervous system”) | Body Image<br>• DE "Body Image"<br>• DE "Body Awareness"<br>• DE "Body Dissatisfaction"<br>• DE “Body Esteem”<br>• Free text: "body image" OR "body percep*" OR "body dissatisfac*" OR "body satisfac* OR "body esteem" OR "body aware*" OR “body interocept*” OR “body-image” OR “body-percep*” OR “body-dissatisfac*” OR “body-satisfac*” OR “body-esteem” OR “body-aware” OR “body-interocept*” OR “body concept” OR (body N5 (image OR percep* OR satisfac* OR disatisfact* OR esteem OR aware* OR self-percep* OR self-percep* OR concept) |

SCOPUS [388 hits]

| Concept 1 | Concept 2 |
|---|---|
| Multiple Sclerosis<br>(TITLE-ABS ({multiple sclerosis} OR {MS} OR {demyelinating disease} OR {relapsing remitting} OR {sclerosis} OR ({autoimmune disease} W/3 {nervous system}))) | Body Image<br>(TITLE-ABS ({body image} OR {body percep*} OR {body dissatisfac*} OR {body satisfac*} OR {body esteem} OR {body aware*} OR {body interocept*} OR {body-image} OR {body-percep*} OR {body-dissatisfac*} OR {body-satisfac*} OR {body-esteem} OR {body-aware} OR {body-interocept*} OR (body W/5 (image OR percep* OR satisfac* OR disatisfact* OR esteem OR aware* OR self-percep* OR self-percep*)))) |

**Table 1-2**

**Study Characteristics**

| Study | Country | Sample Size | % Female | Mean Age (SD) | MS Type % RRMS (% SPMS; % PPMS) | Disease Severity (EDSS) | Disease Duration | Study Type | Body Image Measures | Psychological Measures | Effect size | Quality Appraisal |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Barak et al. 1998 | Israel | 76 | 66% | 38.9 | 100% | 4.1 (1.4) | NR | Cross-sectional | Body Esteem Scale (BES) | Eysenck Self Esteem Scale | 0.43 | 0.77 |
| Barbosa et al. 2023 | Portugal | 305 | 70% | 46.13 (11.81) | 21.5% (6.2%; 5.6%) | NR | 10.31 (9.59) | Cross-sectional | Body Appreciation Scale-2 (BAS-2); Body Acceptance by Others | The World Health Organization Quality of Life–BREF; Meaning of Life Scale; Difficulties of Emotional Regulation Scale | 0.31*-0.46* | 0.91 |
| Farnam et al., 2017 | Iran | 60 | 50% | 50%<br>*18-28*;<br>28.3%<br>*29-39;*<br>21.7%<br>*40-50* | NR | NR | NR | Cross-sectional | Body Image Questionnaire | Index of Clinical Stress; The Self-Regulation Inventory; The Aggression Questionnaire | 0.26 (Stress, Aggression)**<br>0.16 (Emotional Regulation) | 0.85 |
| Ghodusi & Heidari, 2014 | Iran | 395 | 70% | 36.68 (8.84) | NR | NR | 3.51 (1.40) | Cross-sectional | Physical Disability Body Esteem Questionnaire | Rosenberg self-esteem questionnaire | 0.63* | 0.85 |
| Kindrat, 2007 | Canada | 30 | 100% | 39.5 (8.7) | 100% | NR | 7 (5.8) | Cross-sectional | Body-Image Ideals Questionnaire | The Beck Depression Inventory-Short Form | 0.81* | 0.9 |
| Lo Buono et al., 2023 | Italy | 100 | 74% | 40.3 (11.6) | 100% | 2.9 (1.6) | 10 (7.4) | Cross-sectional | Body Image Scale | Rosenberg Self-Esteem Scale; Symptom Checklist-90-Revised | 0.52 (Self-esteem)*<br>0.57 (Depression)*<br>0.50 (Anxiety)* | 0.95 |
| Pfaffenberger et al., 2011 | Austria | 68 | 73% | 37.35 (9.41) | NR | 2.63 (1.66) | 7.3 (5.16) | Cross-sectional | Questionnaire on body image | Beck Depression Inventory | 0.14-0.59* | 0.85 |
| Sengul et al., 2019 | Turkey | 50 | 62% | 31.66 (7.88) | 100% | NR | NR | Cross-sectional | Body Cathexis Scale | Beck Anxiety Inventory; Beck Depression Inventory | 0.57 (Anxiety)*<br>0.53 (Depression)* | 0.9 |

| | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|
| Stevens et al., 2018 | USA | 151 | 66% | 46.1 (11.8) | 68.9% (17.2%; 11.3%) | NR | 10 | Cross-sectional | Body Shape Questionnaire | The Quality of Life in Neurological Disorders scale; Patient Health Questionnaire-9 | NR | 1 |
| Wilski et al., 2016 | Poland | 185 | 100% | 49 | 34% (22%; 20%) | Majority moderate (73%) | 14 (9.7) | Cross-sectional | Body Esteem Scale | Rosenberg Self-Esteem Scale; Actually Received Support Scale; Multiple Sclerosis Impact Scale | 0.48 (Self-esteem)* | 1 |
| Hernandez-Reif et al., 1998 | USA | 24 | 75% | 47.5 (10.9) | NR | 5.25 (1.45) | NR | Intervention | Multidimensional Body-Self Relations Questionnaire (modified for MS) | State Anxiety Inventory; Profile of Mood States Depression Scale; Rosenberg Self-Esteem Scale | NR | 0.85 |
| Paolucci et al., 2022 | Italy | 60 | 72% | 41.84 (10.28) | 100% | 5.37 (3.22) | 5.37 (3.22) | Intervention | Multidimensional Assessment of Interoceptive Awareness; Tinetti Mobility Test; Trunk Appearance Perception Scale; Body Image Scale. | 12-Item Short From Health Surey | NR | 0.96 |
| Tesar et al., 2003 | Austria | 29 | 86% | 36.9 (6.7) | NR | 2.7 (1) | 4.6 (3.2) | Intervention | Body image questionnaire | Beck Depression Inventory; Freiburg disease coping questionnaire; State anxiety inventory | NR | 0.77 |

*$p \le 0.01$
**$p < 0.05$

**Table 1-3**

**Quality Appraisal Summary**

| **Criteria** | | Barak et al. (1998) | Barbosa et al. (2023) | Farnam et al., (2017) | Ghodusi & Heidari (2014) | Hernandez-Reif et al. (1998) | Kindrat (2007) | Lo Buono et al. (2023) | Paolucci et al. (2022) | Pfaffenberger et al. (2011) | Sengul et al. (2019) | Stevens et al. (2018) | Tesar et al. (2003) | Wilski et al. (2016) |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| **1** | Question/objective sufficiently described. | 1 | 2 | 2 | 1 | 2 | 2 | 2 | 1 | 1 | 2 | 2 | 2 | 2 |
| **2** | Study design evident and appropriate? | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 |
| **3** | Method of subject/comparison group selection *or* source of information/input variables described and appropriate? | 2 | 1 | 1 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 |
| **4** | Subject (and comparison group, if applicable) characteristics sufficiently described? | 2 | 1 | 0 | 1 | 1 | 1 | 2 | 2 | 2 | 1 | 2 | 1 | 2 |
| **5** | If intervention and random allocation was possible, was it described? | N/A | N/A | N/A | N/A | 2 | N/A | N/A | 2 | N/A | N/A | N/A | 0 | N/A |
| **6** | If interventional and blinding of investigators was possible, was it reported? | N/A | N/A | N/A | N/A | 0 | N/A | N/A | N/A | N/A | N/A | N/A | 0 | N/A |

| | | | | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|---|---|---|
| **7** | If interventional and blinding of investigators was possible, was it reported? | N/A | N/A | N/A | N/A | N/A | N/A | N/A | 2 | N/A | N/A | N/A | N/A | N/A |
| **8** | Outcome and (if applicable) exposure measure(s) well defined and robust to measurement/misclassification bias? Means of assessment reported? | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 1 | 2 | 2 | 2 | 2 |
| **9** | Sample size appropriate? | 1 | 2 | 2 | 2 | 1 | 1 | 1 | 2 | 1 | 1 | 2 | 1 | 2 |
| **10** | Analytic methods described/justified and appropriate? | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 |
| **11** | Some estimate of variance is reported for the main results? | 2 | 2 | 2 | 1 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 |
| **12** | Controlled for confounding? | 1 | 2 | N/A | N/A | 2 | N/A | N/A | 2 | N/A | N/A | N/A | 2 | N/A |
| **13** | Results reported in sufficient detail? | 1 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 |
| **14** | Conclusions supported by results? | 1 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 | 2 |
| **Total Scores** | | **17** | **20** | **17** | **17** | **22** | **18** | **19** | **25** | **17** | **18** | **20** | **20** | **20** |
| **Total Possible Score** | | **22** | **22** | **20** | **20** | **26** | **20** | **20** | **26** | **20** | **20** | **20** | **26** | **20** |
| **Summary Score** | | **0.77** | **0.91** | **0.85** | **0.85** | **0.85** | **0.90** | **0.95** | **0.96** | **0.85** | **0.9** | **1** | **0.77** | **1** |

**Table 1-4:**

**Body Outcome Measures**

| Questionnaire | Study used in | Items | Key Areas Assessed | Language | Reliability and Validity |
|---|---|---|---|---|---|
| Body-Image Ideals Questionnaire (BIQ) | Kindrat, 2007 | 11 (4 point Likert scale) | Importance and ideal of different physical attributes | English | 0.77<br>Further reliability and validity information reported (Cash & Szymanski, 1995) |
| Body Image Scale (BIS) | Lo Buono et al., 2023; Paolucci et al., (2022) | 10 (3 point Likert scale) | Feelings and satisfaction about body image | Translated into Italian | 0.92<br>Further reliability and validity information reported (Cheli et al., 2016) |
| Body Shape Questionnaire (BSQ-SF) | Stevens et al. (2018) | 16 (6 point Likert scale) | Feelings about body over recent weeks | English | 0.95<br>Further reliability and validity information reported (Evans & Dolan, 1993) |
| Questionnaire for assessing one's own body (FBeK) | Pfaffenberger et al. (2011) | 52 (true v untrue) | Attractiveness; external appearance; worry about physical difficulties; sexual problems | German | Not reported |
| Questionnaire on body image (FKB) | Pfaffenberger et al. (2011); Tesar et al., 2003 | 20 (5 point Likert scale) | Negative body judgement and vitality | German | Original articles examining the psychometric properties of these scales were not available in English, there are reports that the scales have been demonstrated to be psychometrically satisfactory (Schmalbach et al., 2020). |
| Body Cathexis Scale (BCS) | Sengul et al., 2019 | 40 (5 point Likert scale) | Satisfaction in body appearance | English | 0.87<br>Further reliability and validity information reported |
| Multidimensional Body-Self Relations Questionnaire (MBSRQ) | Hernandez-Reif et al. (1998) | Not reported | Satisfaction about body; effect of condition on body | English | 0.75-0.91 (Balogun, 1986)<br>Further reliability and validity information reported (Brown et al., 1990) |
| Body Esteem Scale (BES) | Barak et al. (1998); Wilski et al. (2016) | 35 (5 point Likert scale) | Different aspects of body-esteem | English | 0.78-0.87<br>Further reliability and validity information reported (Franzoi & Shields, 1984) |
| Physical Disability Body Esteem Questionnaire (PDBEQ) | Ghodusi & Heidari (2014) | 13 (5 point Likert scale) | Body attraction; sense of comfort with body; comparison with a healthy body; sexual attraction | Translated into Persian | 0.91<br>Further reliability and validity information reported (Ghodusi & Heidari, 2014) |
| Body Appreciation Scale-2 (BAS-2) | Barbosa et al., 2023 | 10 (5 point Likert scale) | Appreciation of own body | Translated to Portuguese | 0.95<br>Further reliability and validity information reported (Lemoine et al., 2018) |

| | | | | | |
|---|---|---|---|---|---|
| Body Acceptance by Others Scale (BAOS) | Barbosa et al., 2023 | 10 (5 point Likert scale) | How individuals think others perceive them | Translated to Portuguese | 0.94<br>Further reliability and validity information reported (Barbosa et al., 2023); Swami et al., 2020) |
| Body Responsiveness Scale (BRS) | Barbosa et al., 2023 | 7 (7 point Likert scale) | Interoceptive awareness | Translated to Portuguese | 0.63<br>Further reliability and validity information reported (Torres et al., 2023) |
| Body Image Questionnaire (BIQ) | Farnam et al. (2017) | 46 (5 point Likert scale) | Evaluation and orientation of appearance; fitness; concern with weight and body satisfaction | English | 0.72<br>Further reliability and validity information reported |
| Multidimensional Assessment of Interoceptive Awareness (MAIA) | Paolucci et al., (2022) | 32 (6 point Likert scale) | Interoceptive awareness | English | 0.66-0.82<br>Further reliability and validity information reported (Mehling et al., 2012) |

**Table 1-5**

**Psychological Outcome Measures**

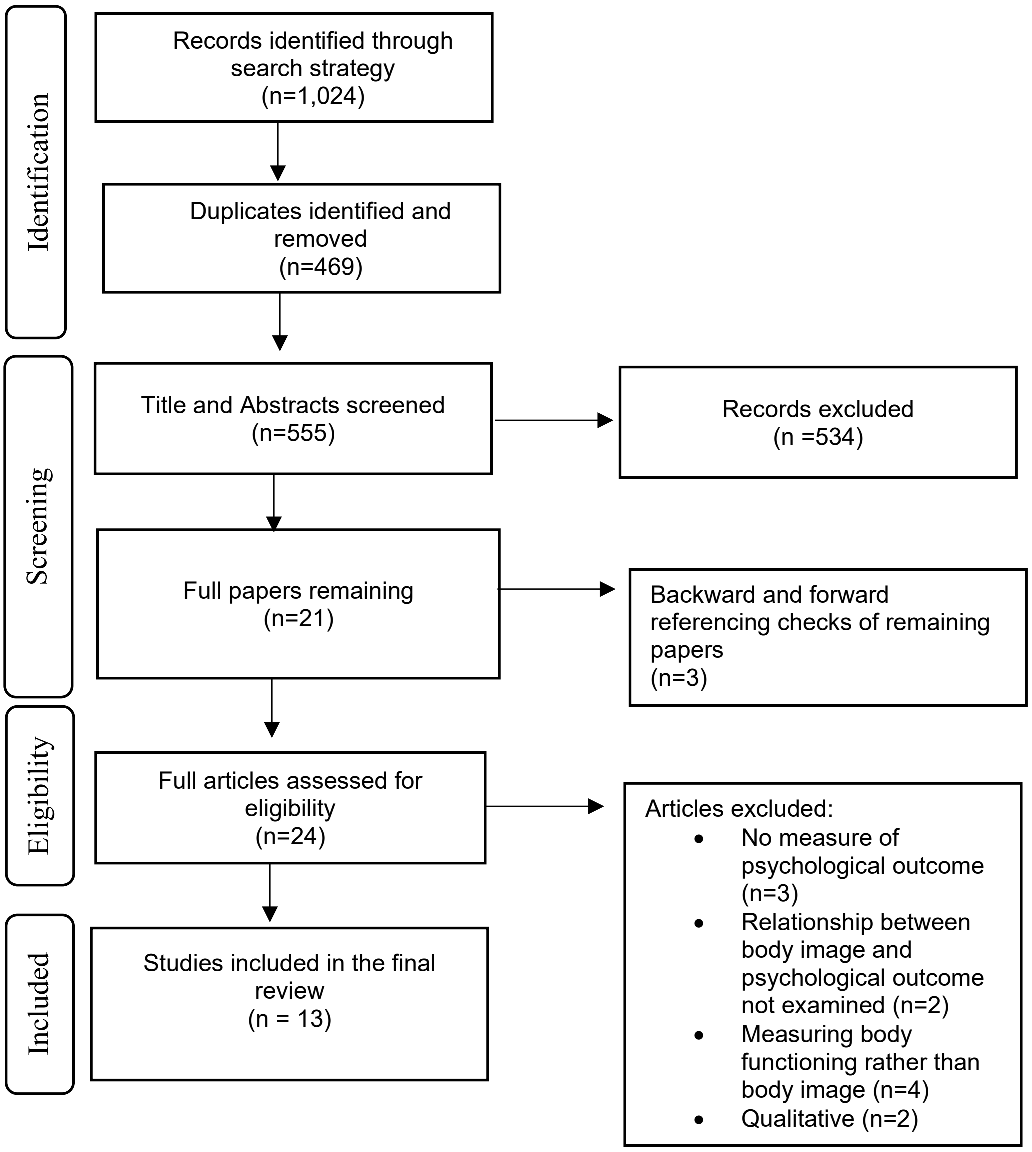
| Psychological Outcome | Questionnaires | Brief Explanation of studies |
|---|---|---|
| Depression | Beck Depression Inventory-II (Sengul et al., 2019; Pfaffenberger et al., 2011) | Consists of 21 statements, with higher scores indicative of higher levels of depression. Psychometrically robust (Dozois et al., 1998). |
| | Beck Depression Inventory Short-Form (Kindrat, 2007; Tesar et al., 2003) | A 13-item measure, evidenced to be a reliable and valid alternative to the original form (Reynolds & Gould, 1981). |
| | Patient Health Questionnaire-9 (Stevens et al., 2018) | A nine item questionnaire, psychometrically robust questionnaire, to measure depression (Costantini et al., 2021) |
| | Profile of Mood States Depression Scale (Hernandez-Reif et al., 1998) | A 19-item questionnaire with adjectives to reflect present mood. Original validation paper unavailable, however reports of strong internal consistency (r=0.05) and good validity (Hernandez-Reif et al., 1998). |
| | Symptom Checklist-90-Revised (Lo Buono et al., 2023) | A validated 90-item measure consisting of nine dimensions including depression, measured by 13 items (Derogatis, 1992). |
| Self-Esteem | Eysenck Self Esteem Scale (Barak et al. 1998) | A 30-item measure. Psychometric properties of the scale could not be found for purpose of this review. |

| | | |
|---|---|---|
| | Rosenberg self-esteem questionnaire (Ghodusi & Heidari, 2014; Hernandez-Reif et al., 1998; Lo Buono et al., 2023; Wilski et al., 2016) | A ten-item measure, interested in two main dimensions: self-competence and self-liking, which has been demonstrated to be psychometrically robust (Tinakon & Nahathai, 2012). |
| Anxiety | Beck Anxiety Inventory (Sengul et al., 2019) | A 21-item psychometrically sound questionnaire developed to measure anxiety (Steer & Beck, 1997) |
| | State Anxiety Inventory (Hernandez-Reif et al., 1998; Tesar et al., 2003) | A 20-item measure interested in individuals current feelings of anxiety. Original validation paper unavailable, however reports of strong internal consistency (r=0.83) and good validity (Hernandez-Reif et al., 1998). |
| | Symptom Checklist-90-Revised (Lo Buono et al., 2023) | See above for more details. The anxiety dimension encompasses 9 items. |
| Quality of Life | The World Health Organization Quality of Life–BREF (Barbosa et al. 2023) | A 26-item scale examining physical, psychological, social and environmental aspects of QoL, as well as overall perception. The Portuguese version was used, with internal consistency reported to be 0.94 in the included study (Barbosa et al., 2023). |
| | The Quality of Life in Neurological Disorders scale (Stevens et al., 2018) | A 13-item measure of physical, mental and social aspects of QoL., evidenced to be reliable and valid (Victorson et al., 2014) |
| | The SF-12 (Paolucci et al., 2022) | A 12-item measure examining the effects of someone's health on their QoL (Ware et al., 1996). |
| | Multiple Sclerosis Impact Scale (Wilski et al., 2016) | This 29-item survey consists of two subscales: a physical scale with 20 items and a psychological scale |

| | | |
|---|---|---|
| | | with 9 items. High internal consistency rate (0.97) in present study. |
| Emotion regulation | The Self-Regulation Inventory (Farnam et al.,, 2017) | A 25-item survey examining positive actions, controllability, expression of feelings, assertiveness and wellbeing, with evidence of reliability and validity (Ibánez et al., 2005). |
| Aggression | The Aggression Questionnaire (Farnam et al., 2017) | This item consists of 29 items, examining physical and verbal aggression, anger and hostility (Buss & Perry, 1992) |
| Stress | Index of Clinical Stress (Farnam et al., 2017) | A 25-item scale measuring stress-related issues, demonstrated to be reliable and valid (Abell, 1991) |
| Coping | Freiburg disease coping questionnaire (Tesar et al., 2003) | This item consists of 35-item German measure, psychometric properties unavailable in English. |

**Figure 1-1**

**Study Flow Chart**



# Appendices

## Appendix 1-A

### Highlights

- Body image is an important psychological construct within MS
- Positive body image is associated with increased psychological wellbeing
- Body image may be a target for psychological intervention in MS
- Further research is needed to explore reported associations in more detail

## Appendix 1-B

## QualSyst Critical Appraisal Tool

| **Criteria** | | **Yes (2)** | **Partial (1)** | **No (0)** |
|---|---|---|---|---|
| **1** | Question/objective sufficiently described. | | | |
| **2** | Study design evident and appropriate? | | | |
| **3** | Method of subject/comparison group selection *or* source of information/input variables described and appropriate? | | | |
| **4** | Subject (and comparison group, if applicable) characteristics sufficiently described? | | | |
| **5** | If intervention and random allocation was possible, was it described? | | | |
| **6** | If interventional and blinding of investigators was possible, was it reported? | | | |
| **7** | If interventional and blinding of investigators was possible, was it reported? | | | |
| **8** | Outcome and (if applicable) exposure measure(s) well defined and robust to measurement/misclassification bias? Means of assessment reported? | | | |
| **9** | Sample size appropriate? | | | |
| **10** | Analytic methods described/justified and appropriate? | | | |
| **11** | Some estimate of variance is reported for the main results? | | | |
| **12** | Controlled for confounding? | | | |
| **13** | Results reported in sufficient detail? | | | |
| **14** | Conclusions supported by results? | | | |
| **Total Scores** | | | | |

## Appendix 1-C

## Author Guidelines for Multiple Sclerosis and Related Disorders

### Types of article

*Original Research Articles*

Full length research papers will not normally be more than 3500 words in length from the Introduction through the Discussion section and will preferably be shorter. Submission of a paper to Multiple Sclerosis and Related Disorders will be held to imply that it represents original research not previously published (except in the form of an abstract or preliminary report), that it is not being considered for publication elsewhere, and that if accepted by Multiple Sclerosis and Related Disorders it will not be published elsewhere in the same form in any language without the consent of the Publisher. Major papers of topical content will be given priority in publication.

*Book Reviews*

These are normally submitted by the Book Review Editors, but they welcome suggestions of books for review.

*Case Reports*

Please note we are no longer accepting case reports as such. These are now incorporated under `Correspondence?. Please refer to the section below

*Correspondence*

The Editors will consider for publication Correspondence, Editorials, Letters or Short Reports (including Case Reports) that illustrate important points. These should not exceed 1000 words in length, have a title page, a summary of about 100 words and up to 10 references, one figure and one table. The word length restriction is not rigid and, in the case of Case Reports only, if the authors wish to write a detailed discussion there will be no limit on length unless advised otherwise by the reviewers.

*Clinical Trial papers*

Manuscripts detailing the results of clinical trials in MS and related disorders are encouraged. The trial methodology should account for all screened participants, and analyses should observe an intention-to-treat model where appropriate. All sources of funding for the study must be disclosed, and the involvement of the study sponsor must be detailed. Clinical trial manuscripts should be a maximum of 3500 words.

*Review Articles*

Review papers are normally 4000-5000 words in total. Authors are advised to consult one of the Editors with an outline before submitting a review.

### Contact details for submission

Authors may send queries concerning the submission process, manuscript status, or journal procedures to our **Support Centre**

### Submission checklist

You can use this list to carry out a final check of your submission before you send it to the journal for review. Please check the relevant section in this Guide for Authors for more details.

**Ensure that the following items are present:**

One author has been designated as the corresponding author with contact details:

- E-mail address
- Full postal address

All necessary files have been uploaded:

*Manuscript*:

- Include keywords
- All figures (include relevant captions)
- All tables (including titles, description, footnotes)
- Ensure all figure and table citations in the text match the files provided
- Indicate clearly if color should be used for any figures in print

*Graphical Abstracts / Highlights files* (where applicable)
*Supplemental files* (where applicable)
Further considerations

- Manuscript has been 'spell checked' and 'grammar checked'
- All references mentioned in the Reference List are cited in the text, and vice versa
- Permission has been obtained for use of copyrighted material from other sources (including the Internet)
- A competing interests statement is provided, even if the authors have no competing interests to declare
- Journal policies detailed in this guide have been reviewed
- Referee suggestions and contact details provided, based on journal requirements

**Ethics in publishing**

Please see our information on Ethics in publishing.

**Studies in humans and animals**

If the work involves the use of human subjects, the author should ensure that the work described has been carried out in accordance with The Code of Ethics of the World Medical Association (Declaration of Helsinki) for experiments involving humans. The manuscript should be in line with the Recommendations for the Conduct, Reporting, Editing and Publication of Scholarly Work in Medical Journals and aim for the inclusion of representative human populations (sex, age and ethnicity) as per those recommendations. The terms sex and gender should be used correctly.

The author should ensure that the manuscript contains a statement that all procedures were performed in compliance with relevant laws and institutional guidelines and have been approved by the appropriate institutional committee(s). This statement should contain the date and reference number of the ethical approval(s) obtained. Authors should also include a statement in the manuscript that informed consent was obtained for experimentation with human subjects. The privacy rights of human subjects must always be observed.

The journal will not accept manuscripts that contain data derived from unethically sourced organs or tissue, including from executed prisoners or prisoners of conscience, consistent with recommendations by Global Rights Compliance on Mitigating Human Rights Risks in Transplantation Medicine. For all studies that use human organs or tissues authors must provide sufficient evidence that they were procured in line with WHO Guiding Principles on Human Cell, Tissue and Organ Transplantation. The source of the organs or tissues used in clinical research must be transparent and traceable. Authors of manuscripts describing organ transplantation must additionally declare within the manuscript:

1. that autonomous consent free from coercion was obtained from the donor(s) or their next of kin; and
2. that organs/tissues were not sourced from executed prisoners or prisoners of conscience.

All animal experiments should comply with the ARRIVE guidelines and should be carried out in accordance with the U.K. Animals (Scientific Procedures) Act, 1986 and associated guidelines, EU Directive 2010/63/EU for animal experiments, or the National Research Council's Guide for the Care and Use of Laboratory Animals and the authors should clearly indicate in the manuscript that such guidelines have been followed. The sex of animals must be indicated, and where appropriate, the influence (or association) of sex on the results of the study.

*Checklist for reporting and reviewing studies of experimental animal models of multiple sclerosis and related disorders*

The guide, reported here, is intended to act as a checklist to aid both authors and referees of manuscripts, just as the Consolidated Standards of Reporting Trials (CONSORT) guidelines are a compulsory part of reporting clinical trials.

Please click here for the checklist and the complete article by Sandra Amor and David Baker.

**Informed consent and patient details**

Studies on patients or volunteers (including organ/tissue donors) require informed consent, which should be documented in the paper. Appropriate consents, permissions and releases must be obtained where an author wishes to include case details or other personal information or images of patients and any other individuals in an Elsevier publication. Written consents must be retained by the author, but copies should not be provided to the journal.

Only if specifically requested by the journal in exceptional circumstances (for example if a legal issue arises) the author must provide copies of the consents or evidence that such consents have been obtained. For more information, please review the Elsevier Policy on the Use of Images or Personal Information of Patients or other Individuals.

Unless the author has written permission from the patient (or, where applicable, the next of kin), the personal details of any patient included in any part of the article and in any supplementary materials (including all illustrations and videos) must be removed before submission.

**Declaration of interest**

All authors must disclose any financial and personal relationships with other people or organizations that could inappropriately influence (bias) their work. Examples of potential competing interests include employment, consultancies, stock ownership, honoraria, paid expert testimony, patent applications/registrations, and grants or other funding. Authors must disclose any interests in two places: 1. A summary declaration of interest statement in the title page file (if double anonymized) or the manuscript file (if single anonymized). If there are no interests to declare then please state this: 'Declarations of interest: none'. 2. Detailed disclosures as part of a separate Declaration of Interest form, which forms part of the journal's official records. It is important for potential interests to be declared in both places and that the information matches. More information.

**Declaration of generative AI in scientific writing**

The below guidance only refers to the writing process, and not to the use of AI tools to analyse and draw insights from data as part of the research process.

Where authors use generative artificial intelligence (AI) and AI-assisted technologies in the writing process, authors should only use these technologies to improve readability and language. Applying the technology should be done with human oversight and control, and authors should carefully review and edit the result, as AI can generate authoritative-sounding output that can be incorrect, incomplete or biased. AI and AI-assisted technologies should not be listed as an author or co-author, or be cited as an author. Authorship implies responsibilities and tasks that can only be attributed to and performed by humans, as outlined in Elsevier’s AI policy for authors.

Authors should disclose in their manuscript the use of AI and AI-assisted technologies in the writing process by following the instructions below. A statement will appear in the published work. Please note that authors are ultimately responsible and accountable for the contents of the work.

***Disclosure instructions***

Authors must disclose the use of generative AI and AI-assisted technologies in the writing process by adding a statement at the end of their manuscript in the core manuscript file, before the References list. The statement should be placed in a new section entitled 'Declaration of Generative AI and AI-assisted technologies in the writing process'. *Statement: During the preparation of this work the author(s) used [NAME TOOL / SERVICE] in order to [REASON]. After using this tool/service, the author(s) reviewed and edited the content as needed and take(s) full responsibility for the content of the publication.* This declaration does not apply to the use of basic tools for checking grammar, spelling, references etc. If there is nothing to disclose, there is no need to add a statement.

**Submission declaration and verification**

Submission of an article implies that the work described has not been published previously (except in the form of an abstract, a published lecture or academic thesis, see 'Multiple, redundant or concurrent publication' for more information), that it is not under consideration for publication elsewhere, that its publication is approved by all authors and tacitly or explicitly by the responsible authorities where the work was carried out, and that, if accepted, it will not be published elsewhere in the same form, in English or in any other language, including electronically without the written consent of the copyright-holder. To verify compliance, your article may be checked by Crossref Similarity Check and other originality or duplicate checking software.

*Preprints*

Please note that preprints can be shared anywhere at any time, in line with Elsevier's sharing policy. Sharing your preprints e.g. on a preprint server will not count as prior publication (see 'Multiple, redundant or concurrent publication' for more information).

**Use of inclusive language**

Inclusive language acknowledges diversity, conveys respect to all people, is sensitive to differences, and promotes equal opportunities. Content should make no assumptions about the beliefs or commitments of any reader; contain nothing which might imply that one individual is superior to another on the grounds of age, gender, race, ethnicity, culture, sexual orientation, disability or health condition; and use inclusive language throughout. Authors should ensure that writing is free from bias, stereotypes, slang, reference to dominant culture and/or cultural assumptions. We advise to seek gender neutrality by using plural nouns ("clinicians, patients/clients") as default/wherever possible to avoid using "he, she," or "he/she." We recommend avoiding the use of descriptors that refer to personal attributes such as age, gender, race, ethnicity, culture, sexual orientation, disability or health condition unless they are relevant and valid. When coding terminology is used, we recommend to avoid offensive or exclusionary terms such as "master", "slave", "blacklist" and "whitelist". We suggest using alternatives that are more appropriate and (self-) explanatory such as "primary", "secondary", "blocklist" and "allowlist". These guidelines are meant as a point of reference to help identify appropriate language but are by no means exhaustive or definitive.

**Reporting sex- and gender-based analyses**

***Reporting guidance***

For research involving or pertaining to humans, animals or eukaryotic cells, investigators should integrate sex and gender-based analyses (SGBA) into their research design according to funder/sponsor requirements and best practices within a field. Authors should address the sex and/or gender dimensions of their research in their article. In cases where they cannot, they should discuss this as a limitation to their research's generalizability. Importantly, authors should explicitly state what definitions of sex and/or gender they are applying to enhance the precision, rigor and reproducibility of their research and to avoid ambiguity or conflation of terms and the constructs to which they refer (see Definitions section below).

Authors can refer to the Sex and Gender Equity in Research (SAGER) guidelines and the SAGER guidelines checklist. These offer systematic approaches to the use and editorial review of sex and gender information in study design, data analysis, outcome reporting and research interpretation - however, please note there is no single, universally agreed-upon set of guidelines for defining sex and gender.

***Definitions***

Sex generally refers to a set of biological attributes that are associated with physical and physiological features (e.g., chromosomal genotype, hormonal levels, internal and external anatomy). A binary sex categorization (male/female) is usually designated at birth (""sex assigned at birth""), most often based solely on the visible external anatomy of a newborn. Gender generally refers to socially constructed roles, behaviors, and identities of women, men and gender-diverse people that occur in a historical and cultural context and may vary across societies and over time. Gender influences how people view themselves and each other, how they behave and interact and how power is distributed in society. Sex and gender are often incorrectly portrayed as binary (female/male or woman/man) and unchanging whereas these constructs actually exist along a spectrum and include additional sex categorizations and gender identities such as people who are intersex/have differences of sex development (DSD) or identify as non-binary. Moreover, the terms ""sex"" and ""gender"" can be ambiguous—thus it is important for authors to define the manner in which they are used. In addition to this definition guidance and the SAGER guidelines, the resources on this page offer further insight around sex and gender in research studies.

**Author contributions**

For transparency, we require corresponding authors to provide co-author contributions to the manuscript using the relevant CRediT roles. The CRediT taxonomy includes 14 different roles describing each contributor’s specific contribution to the scholarly output. The roles are: Conceptualization; Data curation; Formal analysis; Funding acquisition; Investigation; Methodology; Project administration; Resources; Software; Supervision; Validation; Visualization; Roles/Writing - original draft; and Writing - review & editing. Note that not all roles may apply to every manuscript, and authors may have contributed through multiple roles. More details and an example.

**Changes to authorship**

Authors are expected to consider carefully the list and order of authors **before** submitting their manuscript and provide the definitive list of authors at the time of the original submission. Any addition, deletion or rearrangement of author names in the authorship list should be made only **before** the manuscript has been accepted and only if approved by the journal Editor. To request such a change, the Editor must receive the following from the **corresponding author**: (a) the reason for the change in author list and (b) written confirmation (e-mail, letter) from all authors that they agree with the addition, removal or rearrangement. In the case of addition or removal of authors, this includes confirmation from the author being added or removed.

Only in exceptional circumstances will the Editor consider the addition, deletion or rearrangement of authors **after** the manuscript has been accepted. While the Editor considers the request, publication of the manuscript will be suspended. If the manuscript has already been published in an online issue, any requests approved by the Editor will result in a corrigendum.

**Clinical trial results**

In line with the position of the International Committee of Medical Journal Editors, the journal will not consider results posted in the same clinical trials registry in which primary registration resides to be prior publication if the results posted are presented in the form of a brief structured (less than 500 words) abstract or table. However, divulging results in other circumstances (e.g., investors' meetings) is discouraged and may jeopardise consideration of the manuscript. Authors should fully disclose all posting in registries of results of the same or closely related work.

*Reporting clinical trials*

Randomized controlled trials should be presented according to the CONSORT guidelines. At manuscript submission, authors must provide the CONSORT checklist accompanied by a flow diagram that illustrates the progress of patients through the trial, including recruitment, enrollment, randomization, withdrawal and completion, and a detailed description of the randomization procedure. The CONSORT checklist and template flow diagram are available online.

*Registration of clinical trials*

Registration in a public trials registry is a condition for publication of clinical trials in this journal in accordance with International Committee of Medical Journal Editors recommendations. Trials must register at or before the onset of patient enrolment. The clinical trial registration number should be included at the end of the abstract of the article. A clinical trial is defined as any research study that prospectively assigns human participants or groups of humans to one or more health-related interventions to evaluate the effects of health outcomes. Health-related interventions include any intervention used to modify a biomedical or health-related outcome (for example drugs, surgical procedures, devices, behavioural treatments, dietary interventions, and process-of-care changes). Health outcomes include any biomedical or health-related measures obtained in patients or participants, including pharmacokinetic measures and adverse events. Purely observational studies (those in which the assignment of the medical intervention is not at the discretion of the investigator) will not require registration.

*Article transfer service*

This journal uses the Elsevier Article Transfer Service to find the best home for your manuscript. This means that if an editor feels your manuscript is more suitable for an alternative journal, you might be asked to consider transferring the manuscript to such a journal. The recommendation might be provided by a Journal Editor, a dedicated Scientific Managing Editor, a tool assisted recommendation, or a combination. If you agree, your manuscript will be transferred, though you will have the opportunity to make changes to the manuscript before the submission is complete. Please note that your manuscript will be independently reviewed by the new journal. More information.

**Copyright**

Upon acceptance of an article, authors will be asked to complete a 'Journal Publishing Agreement' (see more information on this). An e-mail will be sent to the corresponding author confirming receipt of the manuscript together with a 'Journal Publishing Agreement' form or a link to the online version of this agreement.

Subscribers may reproduce tables of contents or prepare lists of articles including abstracts for internal circulation within their institutions. Permission of the Publisher is required for resale or distribution outside the institution and for all other derivative works, including compilations and translations. If excerpts from other copyrighted works are included, the author(s) must obtain written permission from the copyright owners and credit the source(s) in the article. Elsevier has preprinted forms for use by authors in these cases.

For gold open access articles: Upon acceptance of an article, authors will be asked to complete a 'License Agreement' (more information). Permitted third party reuse of gold open access articles is determined by the author's choice of user license.

***Author rights***

As an author you (or your employer or institution) have certain rights to reuse your work. More information.

*Elsevier supports responsible sharing*

Find out how you can share your research published in Elsevier journals.

**Role of the funding source**

You are requested to identify who provided financial support for the conduct of the research and/or preparation of the article and to briefly describe the role of the sponsor(s), if any, in study design; in the collection, analysis and interpretation of data; in the writing of the report; and in the decision to submit the article for publication. If the funding source(s) had no such involvement, it is recommended to state this.

**Open access**

Please visit our Open Access page for more information about open access publishing in this journal.

*Elsevier Researcher Academy*

Researcher Academy is a free e-learning platform designed to support early and mid-career researchers throughout their research journey. The "Learn" environment at Researcher Academy offers several interactive modules, webinars, downloadable guides and resources to guide you through the process of writing for research and going through peer review. Feel free to use these free resources to improve your submission and navigate the publication process with ease.

*Language (usage and editing services)*

Please write your text in good English (American or British usage is accepted, but not a mixture of these). Authors who feel their English language manuscript may require editing to eliminate possible grammatical or spelling errors and to conform to correct scientific English may wish to use the Language Editing service available from Elsevier's Language Services.

**Submission**

Our online submission system guides you stepwise through the process of entering your article details and uploading your files. The system converts your article files to a single PDF file used in the peer-review process. Editable files (e.g., Word, LaTeX) are required to typeset your article for final publication. All correspondence, including notification of the Editor's decision and requests for revision, is sent by e-mail.

*Submit your article*

Please submit your article via https://www.editorialmanager.com/msard/default.aspx

*Suggesting reviewers*

Please submit the names and institutional e-mail addresses of several potential reviewers. You should not suggest reviewers who are colleagues, or who have co-authored or collaborated with you during the last three years. Editors do not invite reviewers who have potential competing interests with the authors. Further, in order to provide a broad and balanced assessment of the work, and ensure scientific rigor, please suggest diverse candidate reviewers who are located in different countries/regions from the author group. Also consider other diversity attributes e.g. gender, race and ethnicity, career stage, etc. Finally, you should not include existing members of the journal's editorial team, of whom the journal are already aware.

Note: the editor decides whether or not to invite your suggested reviewers.

**Queries**

For questions about the editorial process (including the status of manuscripts under review) or for technical support on submissions, please visit our Support Center.

**NEW SUBMISSIONS**

Submission to this journal proceeds totally online and you will be guided stepwise through the creation and uploading of your files. The system automatically converts your files to a single PDF file, which is used in the peer-review process.

As part of the Your Paper Your Way service, you may choose to submit your manuscript as a single file to be used in the refereeing process. This can be a PDF file or a Word document, in any format or lay-out that can be used by referees to evaluate your manuscript. It should contain high enough quality figures for refereeing. If you prefer to do so, you may still provide all or some of the source files at the initial submission. Please note that individual figure files larger than 10 MB must be uploaded separately.

*References*

There are no strict requirements on reference formatting at submission. References can be in any style or format as long as the style is consistent. Where applicable, author(s) name(s), journal title/book title, chapter title/article title, year of publication, volume number/book chapter and the article number or pagination must be present. Use of DOI is highly encouraged. The reference style used by the journal will be applied to the accepted article by Elsevier at the proof stage. Note that missing data will be highlighted at proof stage for the author to correct.

*Formatting requirements*

There are no strict formatting requirements but all manuscripts must contain the essential elements needed to convey your manuscript, for example Abstract, Keywords, Introduction, Materials and Methods, Results, Conclusions, Artwork and Tables with Captions.

If your article includes any Videos and/or other Supplementary material, this should be included in your initial submission for peer review purposes.

Divide the article into clearly defined sections.

*Figures and tables embedded in text*

Please ensure the figures and the tables included in the single file are placed next to the relevant text in the manuscript, rather than at the bottom or the top of the file. The corresponding caption should be placed directly below the figure or table.

**Peer review**

This journal operates a single anonymized review process. All contributions will be initially assessed by the editor for suitability for the journal. Papers deemed suitable are then typically sent to a minimum of two independent expert reviewers to assess the scientific quality of the paper. The Editor is responsible for the final decision regarding acceptance or rejection of articles. The Editor's decision is final. Editors are not involved in decisions about papers which they have written themselves or have been written by family members or colleagues or which relate to products or services in which the editor has an interest. Any such submission is subject to all of the journal's usual procedures, with peer review handled independently of the relevant editor and their research groups. More information on types of peer review.

**Article structure**

*Subdivision - numbered sections*

Divide your article into clearly defined and numbered sections. Subsections should be numbered 1.1 (then 1.1.1, 1.1.2, ...), 1.2, etc. (the abstract is not included in section numbering). Use this numbering also for internal cross-referencing: do not just refer to 'the text'. Any subsection may be given a brief heading. Each heading should appear on its own separate line.

*Introduction*

State the objectives of the work and provide an adequate background, avoiding a detailed literature survey or a summary of the results.

*Material and methods*

Provide sufficient details to allow the work to be reproduced by an independent researcher. Methods that are already published should be summarized, and indicated by a reference. If quoting directly from a previously published method, use quotation marks and also cite the source. Any modifications to existing methods should also be described.

*Results*

Results should be clear and concise.

*Discussion*

This should explore the significance of the results of the work, not repeat them. A combined Results and Discussion section is often appropriate. Avoid extensive citations and discussion of published literature.

*Conclusions*

The main conclusions of the study may be presented in a short Conclusions section, which may stand alone or form a subsection of a Discussion or Results and Discussion section.

*Appendices*

If there is more than one appendix, they should be identified as A, B, etc. Formulae and equations in appendices should be given separate numbering: Eq. (A.1), Eq. (A.2), etc.; in a subsequent appendix, Eq. (B.1) and so on. Similarly for tables and figures: Table A.1; Fig. A.1, etc.

**Essential title page information**

- ***Title.*** Concise and informative. Titles are often used in information-retrieval systems. Avoid abbreviations and formulae where possible.
- ***Author names and affiliations.*** Please clearly indicate the given name(s) and family name(s) of each author and check that all names are accurately spelled. You can add your name between parentheses in your own script behind the English transliteration. Present the authors' affiliation addresses (where the actual work was done) below the names. Indicate all affiliations with a lower-case superscript letter immediately after the author's name and in front of the appropriate address. Provide the full postal address of each affiliation, including the country name and, if available, the e-mail address of each author.
- ***Corresponding author.*** Clearly indicate who will handle correspondence at all stages of refereeing and publication, also post-publication. This responsibility includes answering any future queries about Methodology and Materials. **Ensure that the e-mail address is given and that contact details are kept up to date by the corresponding author.**
- ***Present/permanent address.*** If an author has moved since the work described in the article was done, or was visiting at the time, a 'Present address' (or 'Permanent address') may be indicated as a footnote to that author's name. The address at which the author actually did the work must be retained as the main, affiliation address. Superscript Arabic numerals are used for such footnotes.

**Highlights**

Highlights are mandatory for this journal as they help increase the discoverability of your article via search engines. They consist of a short collection of bullet points that capture the novel results of your research as well as new methods that were used during the study (if any). Please have a look at the example Highlights.

Highlights should be submitted in a separate editable file in the online submission system. Please use 'Highlights' in the file name and include 3 to 5 bullet points (maximum 85 characters, including spaces, per bullet point).

**Abstract**

A structured abstract that includes the headings: Background, Methods, Results, and Conclusion is required. The abstract must not exceed 500 words. **Background.** The background should provide a brief and concise description of the background and reason for the study citing relevant literature overview, and a clear statement of hypothesis, and study objectives. The introduction should not be an exhaustive review of the literature. **Methods.** This section provides a concise description of the procedural, experimental, and statistical methods in sufficient detail to allow other investigators to reproduce the study. **Results.** State concisely the results of the study. **Conclusion.** Briefly summarize the major conclusions of the investigation.

*Graphical abstract*

Although a graphical abstract is optional, its use is encouraged as it draws more attention to the online article. The graphical abstract should summarize the contents of the article in a concise, pictorial form designed to capture the attention of a wide readership. Graphical abstracts should be submitted as a separate file in the online submission system. Image size: Please provide an image with a minimum of 531 × 1328 pixels (h × w) or proportionally more. The image should be readable at a size of 5 × 13 cm using a regular screen resolution of 96 dpi. Preferred file types: TIFF, EPS, PDF or MS Office files. You can view Example Graphical Abstracts on our information site.

**Keywords**

Immediately after the abstract, provide a maximum of 6 keywords, using American spelling and avoiding general and plural terms and multiple concepts (avoid, for example, 'and', 'of'). Be sparing with abbreviations: only abbreviations firmly established in the field may be eligible. These keywords will be used for indexing purposes.

*Abbreviations*

Define abbreviations that are not standard in this field in a footnote to be placed on the first page of the article. Such abbreviations that are unavoidable in the abstract must be defined at their first mention there, as well as in the footnote. Ensure consistency of abbreviations throughout the article.

*Acknowledgements*

Collate acknowledgements in a separate section at the end of the article before the references and do not, therefore, include them on the title page, as a footnote to the title or otherwise. List here those individuals who provided help during the research (e.g., providing language help, writing assistance or proof reading the article, etc.).

*Formatting of funding sources*

List funding sources in this standard way to facilitate compliance to funder's requirements:
Funding: This work was supported by the National Institutes of Health [grant numbers xxxx, yyyy]; the Bill & Melinda Gates Foundation, Seattle, WA [grant number zzzz]; and the United States Institutes of Peace [grant number aaaa].

It is not necessary to include detailed descriptions on the program or type of grants and awards. When funding is from a block grant or other resources available to a university, college, or other research institution, submit the name of the institute or organization that provided the funding.

If no funding has been provided for the research, it is recommended to include the following sentence:

This research did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

*Math formulae*

Please submit math equations as editable text and not as images. Present simple formulae in line with normal text where possible and use the solidus (/) instead of a horizontal line for small fractional terms, e.g., X/Y. In principle, variables are to be presented in italics. Powers of e are often more conveniently denoted by exp. Number consecutively any equations that have to be displayed separately from the text (if referred to explicitly in the text).

*Footnotes*

Footnotes should be used sparingly. Number them consecutively throughout the article. Many word processors build footnotes into the text, and this feature may be used. Should this not be the case, indicate the position of footnotes in the text and present the footnotes themselves separately at the end of the article.

*Figure captions*

Ensure that each illustration has a caption. A caption should comprise a brief title (**not** on the figure itself) and a description of the illustration. Keep text in the illustrations themselves to a minimum but explain all symbols and abbreviations used.

**Tables**

Please submit tables as editable text and not as images. Tables can be placed either next to the relevant text in the article, or on separate page(s) at the end. Number tables consecutively in accordance with their appearance in the text and place any table notes below the table body. Be sparing in the use of tables and ensure that the data presented in them do not duplicate results described elsewhere in the article. Please avoid using vertical rules and shading in table cells.

**References**

*Citation in text*

Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Any references cited in the abstract must be given in full. Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.

*Reference management software*

This journal has standard templates available in key reference management packages EndNote http://www.endnote.com/support/enstyles.asp and Reference Manager http://refman.com/support/rmstyles.asp. Using plug-ins to wordprocessing packages, authors only need to select the appropriate journal template when preparing their article and the list of references and citations to these will be formatted according to the journal style which is described below.

*Reference links*

Increased discoverability of research and high quality peer review are ensured by online links to the sources cited. In order to allow us to create links to abstracting and indexing services, such as Scopus, Crossref and PubMed, please ensure that data provided in the references are correct. Please note that incorrect surnames, journal/book titles, publication year and pagination may prevent link creation. When copying references, please be careful as they may already contain errors. Use of the DOI is highly encouraged.

A DOI is guaranteed never to change, so you can use it as a permanent link to any electronic article. An example of a citation using DOI for an article not yet in an issue is: VanDecar J.C., Russo R.M., James D.E., Ambeh W.B., Franke M. (2003). Aseismic continuation of the Lesser Antilles slab beneath northeastern Venezuela. Journal of Geophysical Research, https://doi.org/10.1029/2001JB000884. Please note the format of such citations should be in the same style as all other references in the paper.

*Web references*

As a minimum, the full URL should be given and the date when the reference was last accessed. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired, or can be included in the reference list.

*Data references*

This journal encourages you to cite underlying or relevant datasets in your manuscript by citing them in your text and including a data reference in your Reference List. Data references should include the following elements: author name(s), dataset title, data repository, version (where available), year, and global persistent identifier. Add [dataset] immediately before the reference so we can properly identify it as a data reference. The [dataset] identifier will not appear in your published article.

*Preprint references*

Where a preprint has subsequently become available as a peer-reviewed publication, the formal publication should be used as the reference. If there are preprints that are central to your work or that cover crucial developments in the topic, but are not yet formally published, these may be referenced. Preprints should be clearly marked as such, for example by including the word preprint, or the name of the preprint server, as part of the reference. The preprint DOI should also be provided.

*References in a special issue*

Please ensure that the words 'this issue' are added to any references in the list (and any citations in the text) to other articles in the same Special Issue.

*Reference management software*

Most Elsevier journals have their reference template available in many of the most popular reference management software products. These include all products that support Citation Style Language styles, such as Mendeley. Using citation plug-ins from these products, authors only need to select the appropriate journal template when preparing their article, after which citations and bibliographies will be automatically formatted in the journal's style. If no template is yet available for this journal, please follow the format of the sample references and citations as shown in this Guide. If you use reference management software, please ensure that you remove all field codes before submitting the electronic manuscript. More information on how to remove field codes from different reference management software.

*Reference formatting*

There are no strict requirements on reference formatting at submission. References can be in any style or format as long as the style is consistent. Where applicable, author(s) name(s), journal title/book title, chapter title/article title, year of publication, volume number/book chapter and the article number or pagination must be present. Use of DOI is highly encouraged. The reference style used by the journal will be applied to the accepted article by Elsevier at the proof stage. Note that missing data will be highlighted at proof stage for the author to correct. If you do wish to format the references yourself they should be arranged according to the following examples:

*Reference style*

*Text:* All citations in the text should refer to:

1. *Single author:* the author's name (without initials, unless there is ambiguity) and the year of publication;
2. *Two authors:* both authors' names and the year of publication;
3. *Three or more authors:* first author's name followed by 'et al.' and the year of publication.

Citations may be made directly (or parenthetically). Groups of references can be listed either first alphabetically, then chronologically, or vice versa.

Examples: 'as demonstrated (Allan, 2000a, 2000b, 1999; Allan and Jones, 1999)…. Or, as demonstrated (Jones, 1999; Allan, 2000)… Kramer et al. (2010) have recently shown …'

*List:* References should be arranged first alphabetically and then further sorted chronologically if necessary. More than one reference from the same author(s) in the same year must be identified by the letters 'a', 'b', 'c', etc., placed after the year of publication..

*Journal abbreviations source*

Journal names should be abbreviated according to the List of Title Word Abbreviations.

**Video**

Elsevier accepts video material and animation sequences to support and enhance your scientific research. Authors who have video or animation files that they wish to submit with their article are strongly encouraged to include links to these within the body of the article. This can be done in the same way as a figure or table by referring to the video or animation content and noting in the body text where it should be placed. All submitted files should be properly labeled so that they directly relate to the video file's content. In order to ensure that your video or animation material is directly usable, please provide the file in one of our recommended file formats with a preferred maximum size of 150 MB per file, 1 GB in total. Video and animation files supplied will be published online in the electronic version of your article in Elsevier Web products, including ScienceDirect. Please supply 'stills' with your files: you can choose any frame from the video or animation or make a separate image. These will be used instead of standard icons and will personalize the link to your video data. For more detailed instructions please visit our video instruction pages. Note: since video and animation cannot be embedded in the print version of the journal, please provide text for both the electronic and the print version for the portions of the article that refer to this content.

*Patient Details*

Every video submission must consist of high-resolution images and a consent form for publication for educational purposes signed by the patient see form, please see the **Patient Details** section below. The Editors reserve the right to ask for additional video/s or video modifications.

**Patient Details** Studies on patients or volunteers require ethics committee approval and informed consent, which should be documented in your paper.

If you wish to include images of patients or case details in an Elsevier publication, you will need to adhere to the following requirements:

In order to comply with data protection and privacy rules, each individual who appears in any video, recording, photograph or case report must be made aware in advance of the fact that such photographs are being taken or such video, recording or report is being made and of all of the purposes for which you wish to use them and that individual (or next of kin in the case of children) must give his/her explicit written consent. If such consent is made subject to any conditions (for example, adopting measures to prevent personal identification of the person concerned), Elsevier must be made aware of all such conditions. Written consents must be provided to Elsevier on request.

The author is responsible for obtaining all necessary consents from patients for (i) the performance of any medical procedure involved, as well as (ii) a release permitting our use of the relevant material. It is our insurers' preference that we do not have any direct contractual relationship with the patients themselves. Please download the Patient consent form here

**Data visualization**

Include interactive data visualizations in your publication and let your readers interact and engage more closely with your research. Follow the instructions here to find out about available data visualization options and how to include them with your article.

**Supplementary material**

Supplementary material such as applications, images and sound clips, can be published with your article to enhance it. Submitted supplementary items are published exactly as they are received (Excel or PowerPoint files will appear as such online). Please submit your material together with the article and supply a concise, descriptive caption for each supplementary file. If you wish to make changes to supplementary material during any stage of the process, please make sure to provide an updated file. Do not annotate any corrections on a previous version. Please switch off the 'Track Changes' option in Microsoft Office files as these will appear in the published version.

**Research data**

This journal encourages and enables you to share data that supports your research publication where appropriate, and enables you to interlink the data with your published articles. Research data refers to the results of observations or experimentation that validate research findings, which may also include software, code, models, algorithms, protocols, methods and other useful materials related to the project.

Below are a number of ways in which you can associate data with your article or make a statement about the availability of your data when submitting your manuscript. If you are sharing data in one of these ways, you are encouraged to cite the data in your manuscript and reference list. Please refer to the "References" section for more information about data citation. For more information on depositing, sharing and using research data and other relevant research materials, visit the research data page.

*Data linking*

If you have made your research data available in a data repository, you can link your article directly to the dataset. Elsevier collaborates with a number of repositories to link articles on ScienceDirect with relevant repositories, giving readers access to underlying data that gives them a better understanding of the research described.

There are different ways to link your datasets to your article. When available, you can directly link your dataset to your article by providing the relevant information in the submission system. For more information, visit the database linking page.

For supported data repositories a repository banner will automatically appear next to your published article on ScienceDirect.

In addition, you can link to relevant data or entities through identifiers within the text of your manuscript, using the following format: Database: xxxx (e.g., TAIR: AT1G01020; CCDC: 734053; PDB: 1XFN).

*Mendeley Data*

This journal supports Mendeley Data, enabling you to deposit any research data (including raw and processed data, video, code, software, algorithms, protocols, and methods) associated with your manuscript in a free-to-use, open access repository. Before submitting your article, you can deposit the relevant datasets to *Mendeley Data*. Please include the DOI of the deposited dataset(s) in your main manuscript file. The datasets will be listed and directly accessible to readers next to your published article online.

For more information, visit the Mendeley Data for journals page.

*Data statement*

To foster transparency, we encourage you to state the availability of your data in your submission. This may be a requirement of your funding body or institution. If your data is unavailable to access or unsuitable to post, you will have the opportunity to indicate why during the submission process, for example by stating that the research data is confidential. The statement will appear with your published article on ScienceDirect. For more information, visit the Data Statement page.

# Section Two: Research Paper

# Lived Experience of Psychological Distress in Neck Dystonia

Word count (excluding references, tables and appendices): 7985

Derval McCormack

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

Prepared for submission to *Disability and Rehabilitation*

**All correspondence should be sent to:**

Derval McCormack
Doctorate in Clinical Psychology
Lancaster University
Furness College
United Kingdom
LA1 4GY

Email: d.mccormack@lancaster.ac.uk
Tel: 01524 59297

## Abstract

**Purpose:**

Neck dystonia (ND) is a movement disorder affecting the head and neck regions. A high prevalence of psychological distress has been reported in ND, however little is known about the experiences of those living with this condition. The aim of this study was to explore lived experiences and understandings of distress for people with ND.

**Materials and Methods:**

Interpretative phenomenological analysis (IPA) was used to analyse data obtained through semi-structured interviews with eleven individuals with ND, recruited through a UK based charity.

**Results:**

Four group experiential themes were constructed from the data: 1) Uncertainty in the lead up to diagnosis; “I was so full of fear” 2) The isolating nature of the condition; “you don't know who to turn to” 3) Mind and body relationship; “is my head turning because I’m thinking about it?” and 4) A challenging new way of life; “living life sideways”.

**Conclusion:**

Uncertainty, abandonment, loss, stigma and isolation were central to experiences of distress for people with ND. Most participants believed that psychological difficulties were the result of living with ND, while some wondered if their distress also preceded the onset of their condition. The study highlighted the need for greater psychological input and support for people with ND.

*Keywords:* Cervical dystonia, spasmodic torticollis, wellbeing, mood, distress

# Introduction

Dystonia is a hyperkinetic movement disorder characterised by sustained or intermittent muscle contractions, resulting in abnormal, and often painful movements and postures (Di Biase et al., 2022). It is the third most common movement disorder after Parkinson's disease and essential tremor (Grütz & Klein, 2021). Most types of dystonia present as idiopathic (cause unknown), however the condition can occur secondary to acquired brain injury or neurological conditions (Richter & Löscher, 1998). Adult-onset idiopathic focal dystonia (affecting a specific body region) is the most common form, of which neck dystonia (ND), often referred to as cervical dystonia or spasmodic torticollis, is the most common phenotype (Ben-Shlomo et al., 2002). ND typically affects the head and neck regions, however it is possible for symptoms to spread to adjacent body areas (Costanzo et al., 2021). The involuntary movements that characterise this condition are thought to be associated with loss of inhibition within neural circuits in the brainstem, basal ganglia, cerebellum, and cortex (Albanese et al., 2023). Despite being the most common type of focal dystonia, diagnosis of ND is based upon clinical examination rather than standard criteria, and therefore often results in misdiagnosis or delays to diagnosis (Albanese et al., 2023; Defazio et al., 2023). Previous reports on the prevalence of ND have provided inconsistent results, with recent reports across a multi-ethnic population suggesting 1.8/100,000 (LaHue et al., 2020). The mean age of onset has been reported as 42, with a 2:1 female to male ratio (Defazio et al., 2013).

The main symptoms of ND including head twisting, tremor, and pulling of the neck, tend to develop slowly (Ben-Shlomo et al., 2002), with progression mostly happening within the first five to ten years following initial onset (Albanese et al., 2023). While there is no cure for the condition, symptoms may be alleviated using botulinum toxin intramuscular injections, typically delivered every three months (Moll et al., 2018). Deep brain stimulation

(DBS) may be considered as an alternative treatment in severe cases, and involves the surgical insertion of electrical stimulators to control brain signals (Rodrigues et al., 2019). The physical symptoms that characterise ND have been shown to interfere with a person's ability to continue activities of daily living. The prevalence of pain is higher in ND than all other types of dystonia (Williams et al., 2017), with reports that it affects approximately two-thirds of individuals (Marciniec et al., 2020). The condition is associated with uncontrollable spasmodic movements, which can cause the neck to rotate, flex, extend, tilt, or shift laterally (Velickovic et al., 2001). It has been reported that approximately one-third of individuals report a sensation of pulling in the neck (Rosales et al., 2021). Some people with ND may experience other physical symptoms including headache and tremor (Rosales et al., 2021). Such symptoms are a source of disability for individuals with ND (Velickovic et al., 2001), interfering with daily tasks including the ability to stay in employment (Ortiz et al., 2019).

While ND was considered predominantly a motor disorder for many years, there has been growing interest in recent decades in the non-motor aspects of the condition, in particular the psychological component (Costanzo et al., 2021). A number of studies have examined the prevalence of psychological distress in ND, with results suggesting that anxiety and depression are present in approximately 25 to 60 % of people (Ceylan et al. 2019; Comella & Bhatia, 2015; Müller et al., 2002; Ndukwe et al., 2020; Tomic et al., 2016). Emerging literature suggests a cognitive component to ND, with a recent systematic review reporting subtle difficulties in the domains of processing speed, verbal and visual memory, visuospatial function, executive function, and social cognition (O'Connor et al., 2023a). While ND does not reduce life expectancy, the impact that the condition has upon quality of life (QoL) has been reported to be comparable to that of Parkinson's disease, multiple sclerosis (MS), and stroke (Camfield et al., 2002). In one study examining 201 individuals with ND, pain, disability, and mood were all found to be significant factors in reducing a person's QoL, with mood being reported as the most predominant predictor (Ndukwe et al., 2020). Furthermore, stigma arising from postural abnormalities and changes in body image (Papathanasiou et al., 2001; Paracka et al., 2020) has been found to have a negative impact on personal, social, and professional domains of life (Tomic et al., 2016).

There is an ongoing debate within the literature regarding why people with ND experience psychological distress. Some researchers believe that mood difficulties are a primary symptom of the condition, intrinsic to the neurobiology driving the motor difficulties, while others believe that mood difficulties are secondary to living with the debilitating symptoms. In support of the former argument, studies have found that approximately 43% of people with ND experience mood difficulties prior to onset of motor symptoms (Moraru et al., 2002; Wenzel et al., 1998), suggesting that distress is a primary symptom of the condition. A study examining 86 individuals with focal dystonia, the majority of whom had ND, reported a mean age of 24.3 for onset of mood difficulties, and 42.5 for onset of motor symptoms (Lencer et al., 2009). This finding supports the argument that psychological difficulties precede motor difficulties in ND. Furthermore, recent longitudinal research has demonstrated stability of mood difficulties despite changing severity of motor symptoms, indicating that distress is an independent component of ND (Berardelli et al., 2015). On the other side of this debate, Mueller et al. (2008) found that mood improved with successful treatment of the physical symptoms, indicating that psychological distress is likely to be secondary to living with ND. Similarly, Skogseid et al. (2007) reported an association between severity of symptoms and low mood. In addition, psychosocial variables such as body image, stigma, illness perception, self-esteem, and cognitive ability have all been reported to be associated with psychological distress (Gowling et al., 2024; Jahanshahi & Marsden, 1990; Monaghan et al., 2022; O'Connor et al., 2023b) and QoL (Ben-Shlomo et al., 2002; Tomic et al., 2016). Importantly, little is known about the perspectives of individuals with ND regarding how they view and understand their distress.

Developing a greater understanding of the psychological aspect of living with ND has important clinical implications. Currently, interventions tend to prioritise the management of physical symptoms of ND, with other aspects typically being neglected (Ndukwe et al., 2020). This prioritisation of motor symptoms in ND is mirrored in the literature, with little research examining psychological interventions for people with ND. A systematic review into the effectiveness of behavioural therapies in the management of dystonia was unable to draw clear conclusions due to limited research (Bernstein et al., 2016). The English National Institute for Health and Care Excellence (NICE) guidelines (2009) currently recommend a cognitive behavioural therapy (CBT) approach to managing distress in long-term health conditions. However, this pathway is based on recommendations developed for non-dystonic populations, and does not take into account the specific needs of this particular group (Zurowski et al., 2013). The majority of research investigating psychological distress in ND has been quantitative in design, with few studies exploring lived experiences. A recent study by Morgan et al. (2021) used phenomenological methods to explore the experiences of people with different types of dystonia. Their results indicated that the psychological aspect of ND was particularly salient for participants, with recommendations for future phenomenological studies to focus on one particular type of dystonia.

Consequently, this study will examine the lived experiences of psychological distress in people with ND through qualitative methods. The purpose of this research is to develop greater insight into how individuals experience and understand mood difficulties in ND. Greater understanding of people’s perceptions of their distress will guide the development of interventions, which can be tailored appropriately by clinicians to support person-centred care. The study aimed to answer the following research questions:

1. What are the experiences of psychological distress in ND?
2. How do people with ND understand their psychological difficulties?

## Materials and Methods

### Design

Interpretative phenomenological analysis (IPA) was the methodological approach chosen for this study because it focuses on people's sense-making of their experiences (Smith et al., 2022). There are three main theoretical underpinnings of IPA: phenomenology, hermeneutics, and idiography (Smith et al., 2022). Unlike more descriptive qualitative approaches, the phenomenological element of IPA focuses not just on a person's experience of what happened, but on the meaning they place on that experience. This, along with the double hermeneutic process of interpretation, and the focus on the particular, allows IPA research to understand the perspective of the person, as much as possible (Smith et al., 2022). These theoretical underpinnings place IPA as a good fit for clinical psychology research, where it is important to develop an understanding of how people perceive and understand significant events in their life (Smith & Eatough, 2015). A review of 293 empirical IPA studies demonstrated that this method is mostly used for research exploring illness experiences (Smith, 2011). IPA has previously been employed to examine the psychological aspects of living with movement disorders including dystonia (Morgan et al., 2021; Simpson et al., 2015; Todd et al., 2010). This present study followed recommendations by Smith et al. (2022) for carrying out IPA research.

In line with published guidelines regarding service user involvement in clinical psychology research, an individual with lived experience of ND was consulted throughout the design process (Sheldon & Harding, 2010). They provided input regarding terminology, interview schedule and supporting documentation.

Ethical approval for this research was granted by the Faculty of Health and Medicine Research Ethics Committee at Lancaster University (Ethics Appendix 4-B).

**Participants**

In line with Smith et al. (2022) guidelines for sampling, participants were selected purposively through Dystonia UK, a national charity to support people with dystonia. A research invitation (Ethics Appendix 4-C) was published on both the charity website and in its newsletter, providing information about the aims of the research and the inclusion criteria for participation. Individuals aged 18 to 69, with adult-onset ND, were included. The sample targeted those of working age to reduce likelihood of additional age-related health problems. Individuals with significant concurrent health difficulties affecting daily functioning were excluded, to ensure homogeneity of the sample (Smith et al., 2022). Individuals must have been living with ND for at least six months to allow for a period of adjustment. Participants were required to have experience of psychological distress, either currently, since their diagnosis, or up to six months prior to diagnosis.

There are no specific requirements for sample size within IPA, however some recommendations have been made. Smith et al. (2022) report that sample sizes within doctoral IPA studies typically range from six to ten participants, while other reports suggest that up to 15 participants may be required for publication (Pietkiewicz & Smith, 2014). Twenty-four individuals initially responded to the research invitation. Participant information sheets (Ethics Appendix 4-D) and available interview times were sent to each of these individuals. Six people did not respond to the invite; three were excluded prior to interview due to older age, concurrent health conditions, or having multiple types of dystonia; one individual was excluded following telephone interview due to fault with the recording; three more individuals were excluded following interview due to significant concurrent health conditions that came to light during the interview. This left a final sample of 11. The researcher consulted with supervisors regarding eligibility criteria, and excluded participants were contacted regarding this decision process.

Demographic data was collected prior to interview. The final sample consisted of six females and five males, ranging in age from 52 to 69. Ten participants were white British and one was white Irish. Eight participants were still working at the time of interview, two were retired, and one was no longer working. Of the eight still working, two were in part-time employment. The duration of ND ranged from six months to 40 years. Nine participants were managing the symptoms of ND with botulinum toxin injections or DBS, and two participants were currently not on any treatment. One participant was taking anti-depressants for psychological distress, and had been prior to diagnosis of ND. Eight participants were married, two were single, and one was in a relationship. Table 2-1 outlines the characteristics of the sample. Pseudonyms have been used to ensure anonymity, and were chosen by the researcher to reflect the age, gender and culture of participants.

[Insert Table 2-1 here]

## Procedure

### *Data Collection*

Interviews were carried out using a semi-structured approach. This is the most common form of data analysis in IPA research as it allows the researcher to follow up on salient topics that the participant brings up in the interview (Smith & Eatough, 2015). It allows for a rapport to develop between the researcher and the participant, supporting the hermeneutic roots of IPA (Smith et al., 2022). A semi-structured schedule (Ethics Appendix 4-E) was developed to guide the research. In line with recommendations (Smith et al., 2022), open questions were used to ensure that participants had the opportunity to provide their own accounts. Following recommendations by an expert by experience, simple language was used to maximise accessibility and inclusivity. The schedule opened with a question that asked participants to recall the lead up to their diagnosis. The purpose of this question was to encourage a descriptive response that would allow the participants to settle into the interview setting. As the interview progressed, the interviewer guided participants to discuss the impact of ND on daily living, their experiences and understandings of psychological distress, and support for distress. Topics appearing salient to participants were explored more fully.

Seven interviews took place via video, and four by telephone, with participants choosing their preferred method to ensure comfort during the process. The interviews ranged in duration from 40 to 68 minutes. The researcher read the consent form (Ethics Appendix 4-F) aloud, and obtained verbal consent before beginning the interview. The form was emailed to participants beforehand, and following interview they were sent a debrief form (Ethics Appendix 4-G). All interviews were recorded and transcribed verbatim. For video interviews, the inbuilt transcribe function on MS Teams was checked and edited, with phone interviews being fully transcribed by the researcher.

### ***Data Analysis***

The first stage of the analysis involved immersive reading and re-reading of the first transcript to focus the analysis on the particular, in line with an idiographic approach. This was followed by exploratory noting, whereby the researcher made initial comments of anything that was felt to be particularly noteworthy within the transcript. This process is demonstrated using an extract from Michael's transcript (Appendix 2-B). The third stage involved the construction of descriptive and interpretative experiential statements, arising from these exploratory notes (Appendix 2-B). The statements were then refined and clustered together based on connections or similarities (Appendix 2-C). In the final stage, each cluster of statements was given a name, and these personal experiential themes (PETS) were consolidated into a table representing that participant's unique experiences (Appendix 2-D). The first five stages outlined above were repeated, until all 11 participants had a PET table, each with relevant statements and supporting quotes. The next stage of the analysis involved the development of group experiential themes (GETS), with the aim of identifying shared and unique experiences of psychological distress in ND. The researcher used a colour coding system to find initial connections between the PETS (Appendix 2-E). These final GETS (Appendix 2-F) were then written up into a narrative report, which involved an iterative process of writing and re-writing. Given the relatively large sample size in this study, the researcher chose supporting quotes that were particularly rich or emotive, whilst also ensuring that each participant's voice was heard (Smith et al., 2022). The write-up explicitly states the number of contributors for each theme.

The process of analysis was inherently iterative throughout, with the researcher moving backwards and forwards, thinking about the data in different ways. This process allowed for a deep and interpretative analysis that remained grounded within the participants' own experiences (Murray & Wilde, 2020). To ensure that this research was of high quality and validity, the researcher referred to the core principles of qualitative research, outlined by Yardley (2000). The researcher continually evaluated their own assumptions or biases throughout the interpretative process through use of a reflective journal, detailing each stage of the analytical process (Vicary et al., 2017). To ensure transparency of the process, the researcher included a personal statement, identifying their standpoint in approaching the study (Levitt et al., 2018).

## Personal Statement

I had previous experience working as part of a multidisciplinary research team on a study involving people with ND (Monaghan et al., 2022). The medical professionals that I worked with previously were very open to research into the psychological aspect of ND. I understood that this could not be assumed of all medical teams, and recognised the importance of remaining open to hearing participants' experiences of this.

I carried out this research as part of a clinical psychology doctorate. As a trainee psychologist, I had previous clinical experience of working with distress. This stance helped me to develop rapport with participants quickly, and to offer a comfortable space to discuss emotive topics.

I had no personal or clinical experience of ND prior to this study. Considering the principle of sensitivity of context (Yardley, 2000), I consulted with an expert by experience during the study design stage to ensure that this project was person-centred. I also worked closely with an experienced clinical neuropsychologist to ensure that this research was clinically relevant.

## Results

The analysis produced four GETS: 1) Uncertainty in the lead up to diagnosis; "I was so full of fear" 2) The isolating nature of the condition; "you don't know who to turn to" 3) Mind and body relationship; "is my head turning because I'm thinking about it?", and 4) A challenging new way of life; "living life sideways". These themes will be explored in more detail below.

### Uncertainty in the lead up to Diagnosis; "I was so full of fear"

Nine participants contributed to this first theme, which encompasses people's experiences of distress prior to diagnosis. There was a sense that the journey to diagnosis was long, and shrouded in uncertainty. People worried about why they were experiencing their new symptoms, with anxiety perpetuated by dismissive healthcare services. The theme also captures the experiences of ambiguity upon receiving a diagnosis.

#### *Worrying about symptoms*

The majority of participants recalled the period prior to diagnosis as being particularly "awful" (Barbara) and "unnerving" (Sharon). Christopher believed his worry came from not knowing why he had his symptoms: "There was so many things going through my head, you know. Is this a tumour in my head causing the problems?" It appeared that uncertainty was leading to Christopher thinking of worst-case scenarios, a thought process mirrored by Barbara: "If it wasn't motor, it had to have been neurological…and that's the first thing that you think about". There was a sense of uncontrollability in the distress felt by some participants during this period. Steve described not being able to recognise himself due to the levels of anxiety he was experiencing:

> I'm not normally a very anxious person or whatever, but I'd got myself into, into such a state that I, I mean, I didn't recognise, you know. And it was so intense. And so…er I mean, I was just so full of fear and anxiety, it was nuts.

### ***Feeling dismissed by healthcare providers***

There was a sense among participants that the distressing period prior to diagnosis was perpetuated by an unresponsive or dismissive healthcare system. Participants used language such as "frustrating" (Michael) and "angry" (Barbara) to recall how they felt toward services at that time. Donna described the impact that dismissal had in terms of how she saw herself: "My doctor had said to me it's probably a habit, like a nervous habit I'd got into...and I felt a bit stupid because I was like why can't I control this, do you know?" Sharon also recalled her physical symptoms being dismissed:

> So I think that was, you know, obviously that was probably initially something I mentioned as well, just feeling quite anxious about it. And er, as I say that's what was treated at that point. So that was antidepressants, I think at that time.

Sharon believed that her anxiety came from not knowing why she was experiencing the physical symptoms. It appeared that her understanding of distress was misaligned with that of her doctor's. Brian had a similar experience of physical symptoms being dismissed as anxiety, which led to feelings of hopelessness, increased psychological difficulty, and the development of unhelpful coping strategies: “I didn't know what was wrong with me. They weren't gonna do anything about it and em, not much I could do about it, so I just resorted to drink and drugs.”

### *Unrealistic expectations and false hope*

Participants spoke about the relief they felt on receiving their diagnosis, which Michael described as a “light at the end of the tunnel”, emphasising the difficult period prior to this. There was an additional sense of validation in Audrey’s relief: “I just was so relieved and just so elated and just felt finally, you know, I've actually got a condition that I, I'm not making it up.” There was a feeling of hope among many participants that life would return to normal. Sandra described how she assumed the botulinum treatment would allow her to return to work. Her language, “my own little like world”, suggested a false hope, perhaps hinting at the later disappointment she felt when life did not return to normal: “I…in my, my own little like world was thinking ‘oh, I can have the injections...I'll be coming straight back to work’”. Sandra believed that unrealistic expectations upon diagnosis worsened the experiences of distress for her later: “if someone had explained it to me like that at the beginning…I might not be quite in my mental state as I am now.”

## The Isolating Nature of the Condition; “you don’t know who to turn to”

The second theme focuses on the sense of isolation arising from living with an uncommon condition, with ten participants contributing. The subthemes capture how isolation arises across different levels, considering relationships with healthcare professionals, friends and family, and wider society. Feelings of abandonment, hopelessness, shame, and stigma were central to this theme.

### *Being left to manage the condition alone*

There was a shared experience among participants of receiving a diagnosis, and a perception of then being left to deal with it themselves. Paul believed that his neurologist was “unwilling” to answer his questions, which led to him feeling like nobody could support him. He used emotive language that suggested a sense of abandonment and hopelessness: “I felt kind of absolute despair when I came out, out of that meeting and it was like, well, what am I gonna do now, right you know. If he can't help me, who's gonna help me?” There was a feeling of disappointment among participants, with some spending a lot of time and effort seeking out further support or alternative treatments, perhaps in an attempt to regain some control: “It took me frustratingly nine months to access the right person to get the…head up brace” (Barbara). Sandra spoke about her feelings of disappointment when questions she raised with her neurologist were left unanswered. There was a similar sense of abandonment here, with Sandra feeling let down by healthcare professionals:

> …’cause you sort of think the guy who is supposed to be trying to give me some help, a little bit of hope maybe, just doesn’t even want to discuss anything, it’s almost like well I can’t cure it, it’s this or nothing, off you go so, and… he sort of flippantly does the injections, goes see you in three months, and I sort of walk out of the room, and I, I sometimes wonder, I just feel like crying, so I just think yeah...he said that, but I actually have to live the next three months… with my head on sideways.

Sandra believed this dismissal or abandonment triggered negative beliefs about herself, such as “I’m not important.”

Participants described feeling alone in their distress, with many commenting on the medical focus of healthcare professionals: “it’s just a case of, well look you’ve got cervical dystonia, go and have the botox injections” (Paul). The lack of focus on the emotional side led to participants questioning the validity of their distress. The language used by Donna was suggestive of shame or self-criticism as she described how the research interview itself was the first time she felt her feelings were validated: “you sort of validated that for me in this conversation here ‘cause I, when I get like that, I think ‘oh why don’t I, I need to just snap out of it, and get out of this mood.'" Michael expressed a similar sense of shame in his distress:

> I kind of chastise myself em, em, there are people out there with far worse…things wrong with it…them than I've got wrong with me physically or, or whatever, em and that I shouldn't you know, I shouldn't kind of feel they way that I do about it.

It appeared that the shame felt by Donna and Michael was in relation to the distress they felt internally. For others, shame came from showing distress, with Brian reporting how he tries to mask such feelings around other people: "I try and breakthrough that or this feeling of anxiety by being happy, smiley and 'meety greety.'"

### ***Nobody else understands this***

Participants spoke about the difficulties of living with a condition that few people around them understand. Sandra described family members questioning her management of her symptoms. Her repetition of the phrase "you're not listening" was indicative of her frustration, and her emphasis on "close" family further highlighted the isolating nature of ND: "I've got family that em, close family that actually then sort of go em oh haven't they done anything about your neck yet? And you think, you're not listening, you're not listening." Similarly, Audrey described the loneliness that arose from this lack of understanding:

> …other conditions that people suffer from…you, you get a lot of….people feel sympathy or they offer support, or…it just felt in this particular condition, you just feel very, you do feel very isolated and that you don't know who to turn to.

### ***The embarrassment of navigating the social world***

There was a sense of self-consciousness among several participants in terms of their appearance. Sandra discussed physical changes in her body, which caused her to view herself more negatively: "I don't feel nice at all about myself. I can feel, I can physically see that my er shoulders have become more rounded.” Donna experienced similar physical changes: “my body’s all misshapen”, however unlike Sandra, found that humour reduced the impact this had on her sense of self: “I do, like, joke about it.”

Several participants spoke about feeling self-conscious and embarrassed in public due to the physical features of ND, such as neck pulling or twisting. Participants used words such as “odd” (Michael) “abnormal” (Audrey) and “weird” (Sandra), to describe perceptions of how others see them. These negative feelings appeared to have a significant impact on participants’ sense of self, with Michael reporting his self-esteem to be “through the floor”. Such stigma also appeared to have a significant impact on the social lives of many participants. Christopher emotively described how ND had “totally trashed” his social life. This was echoed by Kathryn, who avoided social situations due to discomfort around other people: “I don't want to bump into people who I knew before who haven't seen me like this because I feel embarrassed and self-conscious.”

### Mind and Body Relationship; “Is my head turning because I'm thinking about it?”

This theme focuses on how individuals made sense of the relationship between mind and body, with six participants contributing to this theme. This theme explores participants’ beliefs about the role of trauma in the onset of ND, and the role of the mind in controlling the physical symptoms.

#### *Curiosity about the role of earlier trauma*

Several participants wondered if the onset of their dystonia was related to an earlier trauma (Paul, Brian, Steve, Audrey). There was a sense of certainty in Paul’s language about the role of trauma in his ND:

> This is what I believe are the root causes. And for me it, it's been to do with trauma right...over the years and literally this, this trauma, whether it was physical injury, head trauma, or emotional trauma right. Em I can take this right back to when I was a child.

For others, like Brian, this was a more tentative wondering: “Is it something that happens to us psychologically that brings it on? Or is it something, or do we get it, and then we get psychologically sick. You know what I mean?” There was a sense of curiosity here for Brian, as he pondered with the possibility that psychological difficulties preceded his physical difficulties. Brian recalled a difficult time in his life prior to the onset of symptoms, when he had moved from a job he loved to live overseas. He described how he regretted the decision and felt he had made a mistake:

> I was very, em I suppose you could say anxious about er living there, working there, and having left a great job and a great way of life, and em ending up in this strange place working for a very tiny outfit that didn't, er that I hadn't been....not necessarily trained, but I’d been over-trained, should I say so. It was a big step back.

A similar sense of wondering was experienced by Steve, who recalled a stressful period in his life shortly before his symptoms began. He described making the decision to return to the UK after building a life abroad, a move that was associated with significant change: “…basically taken 25 years of, of our life out there and turned it upside down and ended up back in the UK”. He described how his stress at that time was “through the roof”, and wondered if there was a connection between this difficult period in his life and the onset of his ND: “I'm not sure in myself whether I actually have a medical problem and whether I just, you know, it's just another manifestation, physical manifestation of, of the, you know, the anxiety and the fears that I had.”

### ***Regaining control of body***

Some participants noticed that re-focusing their attention away from their physical symptoms helped to alleviate the intensity of those symptoms. There was a sense of curiosity among participants as to whether this was indicative of psychological processes playing a role in ND. Audrey tried to make sense of why her physical symptoms seem less prominent when her mind is distracted:

> Now whether it, it's just because I know that dystonia can sometimes improve a bit or, or whether it was because so many other things were going on in my life that I just didn't have time to think about er….for this condition to rear up, I don't know.

Brian echoed the helpfulness of distraction in reducing the impact of physical symptoms. It appeared that refocusing his attention was providing Brian with a sense of control over his symptoms: "It's also a case of when you're directing your attention at something outside of yourself… wholly and completely, you, you forget you've got the problem." Steve also wondered about the relationship between his mind and body, considering the possibility that problems in his neck were the result of thought processes in his mind: "It makes me really wonder whether, you know…is my head turning because I'm thinking about it."

## A Challenging New Way of Life; "Living life Sideways"

The final theme explores participants' understanding that psychological difficulties arise from the long-term nature of ND. The subthemes all consider distress arising from adjusting to a chronic illness. They capture the difficulties people have in accepting a condition that is associated with constant discomfort, pain and loss. All participants contributed to this theme.

### *The incessant discomfort brings you down*

Many participants talked about the constant discomfort and pain associated with living with ND. Donna described her belief that pain was the "key" to her distress. It appeared that the unpleasant nature of pain causes distress for Donna, and it also prevents her from being able to work, resulting in her staying at home alone, which causes further isolation: "The pain makes me feel low, just like the actual pain does…and then I can't work which makes me feel low, 'cause I'm on my own." The belief that distress comes from living with the physical symptoms was echoed by Sandra: "I think it’s living with it…and the stress is caused by then the pain creeping in.” It seemed that pain appears later for Sandra, perhaps exacerbating the distress she already feels from living with ND, whereas for Donna pain is more central to her distress.

Participants talked about the lack of “escape” (Kathryn) from ND, and the constant need to make adaptations to manage symptoms. Sharon talked about planning ahead when going to social events: “I probably would go, but I would be worried about it, I’d be worried about…I’d probably be wanting to get there early so I could position myself.” Kathryn emphasised this by describing how she repositions her body in response to her neck turning: “That's how I have to live my life: sideways.” The effort and time that is required to plan ahead and make adaptations was felt among most participants. Michael summarised this: “It's just that it occupies my mind every day, all of the time…whatever situation I'm going into.”

### ***Struggling to accept that this is not going away***

Several participants found it difficult to accept that their dystonia was “not going away” (Kathryn). There was a sense of annoyance and perhaps resentment toward dystonia for some individuals, with Michael expressing his reluctance to accept the condition “Just kind of go away and leave me alone and, and let me do the things I want to do.” Sandra echoed this. There was a sense of disbelief in Sandra’s language here, and injustice given her previous healthy lifestyle: “There's a little bit of you know, god, why me? I, I’ve been making the effort, I’ve been exercising...” There were some participants (Steve, Paul, Audrey) who found that accepting ND as part of their life helped them to cope better with the condition. Audrey described that changing her mind-set helped her to adjust to the condition more:

> Do you know what, this is me, this is, this is what I am. I'm not gonna try and make excuses anymore, I’m not gonna be embarrassed, I'm not gonna sort of hold my head in shame. If I have em flare-ups then people will just have to get over it...And that in

> itself…taking that mind-set…just makes the whole, the condition just much more bearable and much more easy to cope with.

### *Grieving for loss of self*

For many participants, there was a sense that dystonia had taken away a part of their identity. Kathryn described a loss of self, and a yearning for the person she was prior to dystonia: “I’ve, I’ve lost so much of who I was…and although I am adapting now, I'm, I'm still angry and I, I'm, I'm ashamed to admit I'm jealous of like people who still have the free movement.” Michael echoed this loss of identity, and his desire to be defined as more than this condition: "I’m not just a guy who, who holds his head in a strange way. I’m, you know, I’m kind of me, that’s it". Several participants spoke about the loss of being able to do things that they once loved, and the sadness they have felt with that loss: “It actually makes me want to cry, to think that I can’t do it anymore.” (Sandra). There was a sense among participants that ND had been “imposed” upon them, and taken away so much. Paul understood this process as a type of grief:

> I've been robbed of some of the things that I used to take for granted. And I, I suppose you go through something of a grieving process because you've lost a part of something, you’ve lost a part of you."

## Discussion

This study explored the lived experiences of psychological distress in people with ND. The findings indicated that uncertainty, abandonment, loss, stigma, and isolation were central factors in participants’ experiences of distress. The majority of participants understood their distress as being the result of living with the debilitating nature of ND. Some participants tentatively questioned whether distress occurred more independently of their motor symptoms, with recollections of distressing periods also preceding the onset of physical difficulties.

Findings are supported by a large body of literature aimed at understanding adjustment difficulties in long-term conditions. A working model of adjusting to chronic illness by Moss-Morris (2013) suggests that illness-specific factors such as disability, disfigurement, treatment, and uncertainty, along with contextual factors such as early life, social support, and relationships with healthcare services, can all disrupt emotional equilibrium and QoL. The model suggests that adjustment difficulties may arise from a number of cognitive and behavioural factors including ‘wishful thinking’, suppression of negative emotions, and withdrawal from activities. Such factors were prevalent in the themes of the present study, with feelings of shame and experiences of stigma creating challenges for participants to interact socially, and for many this resulted in masking feelings and social withdrawal.

Illness uncertainty describes ambiguity that a person may hold toward their condition (Mishel, 1988), and has been reported to predict worse adjustment and increased psychological difficulties in MS (Dennison et al., 2009). The common sense model provides a theoretical framework for understanding the relationship between illness representation and psychological outcome (Leventhal et al., 2016). This sociocognitive model suggests that healthcare professionals play an important role in reducing ambiguity, with information-giving supporting the development of a person’s beliefs and understandings about their condition (Leventhal et al., 2016). The findings from this present study suggested a sense of abandonment by healthcare professionals, with many participants expressing disappointment in the lack of information and support received. Previous literature has identified a relationship between illness beliefs and psychological wellbeing (Hagger & Orbell, 2022). A recent study demonstrated that illness beliefs play a significant role in adjusting to ND, with threat-based beliefs being associated with increased distress and reduced QoL (O’Connor et al., 2023b).

The present study indicated the importance of stigma in the psychological experiences of individuals, echoing previous movement disorder research in people with MS (Cadden et al., 2018) and Parkinson's disease (Ma et al., 2016). Existing literature suggests that stigma arises from the co-occurrence of factors such as labelling, negative stereotyping, and discrimination, and is a key determinant in predicting psychological outcome (Link & Phelan, 2001). Participants in the present study described experiences of staring and name-calling, in addition to fears regarding how they may be perceived, suggesting the presence of both enacted and felt stigma (Scambler & Hopkins, 1986). Participants reported feelings of embarrassment and self-consciousness regarding their appearance, indicating that body image may play an important role in experiences of stigma. These findings echo previous research identifying a relationship between body image and psychological distress in ND (Jahanshahi & Marsden, 1990; Lewis et al., 2008), with negative views of body being associated with lower mood. The present study also supports earlier IPA research which identified coping strategies to manage stigma in dystonia, such as masking symptoms (Maxwell-Scott, 2023), and withdrawal from social interactions (Morgan et al., 2021).

There was a sense of grief among participants in this present study, with individuals describing the profound and wide-ranging loss they experienced. Echoing existing chronic health literature, participants described how the long-term nature of ND was as a constant reminder of the loss of function, relationships, and independence (Richman, 2022). Previous qualitative studies have found similar themes of loss in other health conditions such as chronic fatigue syndrome (Dickson et al., 2008) and stroke (Pallesen, 2014). The concept of disenfranchised grief (Doka, 2008) may be useful in understanding the experiences of grief in people with ND. This concept considers the complexities of grief when people are unable to openly acknowledge their loss, and therefore may be particularly relevant to chronic illness (Gunning & Taladay-Carter, 2023). Neimeyer's (2006) constructivist model of grief has been deemed appropriate for addressing disenfranchised grief given the focus it places on meaning reconstructing in response to loss (Pillai-Friedman & Ashline, 2014). Thus, this model may be helpful in supporting individuals to rebuild their lives around living with ND, and the losses that they have incurred.

The participants' beliefs about their distress mirrored the aforementioned debate in the literature as to whether mood difficulties are an intrinsic feature of ND, or whether they are secondary to living with the debilitating symptoms. While the majority of participants believed that distress came from living with the challenging condition, there were some tentative beliefs that psychological difficulties arising from physical and emotional trauma may have preceded motor symptoms. A wide body of previous research suggests a link between peripheral physical trauma and ND, with reports from nine studies proposing that approximately five to 21% of cases are trauma-induced (O'Riordan & Hutchinson, 2004). It appeared that this information was not provided to participants in the present study, perhaps perpetuating their illness uncertainty. Some participants considered whether their physical symptoms had a psychological foundation, leading to curiosity about the connection between the mind and body in ND. While a dualism often exists within the medical model (Pagnini et al., 2014), this present finding reflects the biopsychosocial model of health, which encompasses biological, psychological, interpersonal, and contextual factors influencing a person's health (Lehman et al., 2017). This model may be helpful in understanding the reciprocal relationship between physical symptoms and psychological distress described by participants in the present study.

### *Clinical Implications*

The study identified that psychological distress remains a neglected yet salient part of living with ND. All participants reported experiences of distress, however none of these individuals had been offered support for such difficulties. Clinical psychologists could support people with ND through a combination of indirect and direct input.

Findings suggest the importance of integrating clinical psychologists into multidisciplinary teams providing care for people with ND. Through formulation, psychologists can develop a shared psychological understanding of a person's difficulties with other professionals, with the aim of providing holistic and person-centred care (Geach et al., 2018). There are reports that increased coordination between different care levels can improve the provision of care for people with complex health conditions (Tomaschek et al., 2022). Thus, increased information sharing and collaboration between health specialists, GPs and third-sector organisations may further support person-centred care in ND, and seek to decrease feelings of uncertainty and abandonment, identified in this present study.

There are a number of psychological interventions that may be effective in working with people with ND, however further research is necessary to provide evidence for specific models. The aforementioned review examining the effectiveness of behavioural interventions in dystonia was inconclusive in its findings (Bernstein et al., 2016). The adjustment literature discussed above identified cognitive and behavioural factors associated with poorer adjustment to chronic illnesses, thus suggesting the potential benefits of a CBT approach. Social withdrawal arising from feelings of embarrassment may be perpetuating feelings of low mood or loneliness for individuals, therefore supporting individuals to develop alternative behaviours may help to improve psychological outcome. However, this model focuses on challenging negative thoughts, which may be difficult in a chronic condition like ND, where thoughts are often valid and not irrational per se (Curvis & Methley, 2021). The present study indicated the salience of shame and self-criticism in ND, identifying such feelings in relation to emotions, body image, and appearance. Compassion focused therapy (CFT) is a therapeutic model that was specifically developed to address feelings of shame and self-criticism, and as such may be well placed to support people to develop a more compassionate relationship with themselves (Gilbert, 2014). The present findings highlighted the difficulty in coming to terms with the chronic nature of ND, and the continual loss that is associated with the condition. Acceptance and commitment therapy (ACT) encourages individuals to separate thoughts and feelings from difficult life events, in order to move forward, and live a meaningful and values-oriented life (Hayes et al., 2006). This model has been evidenced as effective in other chronic conditions (Jin et al., 2021). Furthermore, the present findings suggest that physical symptoms of ND may be reduced when the mind is distracted, supporting the mindfulness processes emphasised within the ACT model (Fletcher and Hayes, 2005).

### *Limitations*

ND is a heterogeneous condition, which posed challenging for an IPA study requiring homogeneity of the sample. Differences in severity of symptoms, support networks, and attitudes of healthcare teams may have impacted people's experiences of distress differently. Disease duration in the current sample ranged from six months to 40 years, thus individuals living with the condition longer may have adjusted to the condition differently to those with a more recent diagnosis. To ensure homogeneity, strict eligibility criteria were employed, meaning that certain voices were not heard in this particular study, including those above working age, and those with concurrent health conditions.

### *Further Research*

This study highlighted a number of factors that played a central role in the experiences of distress including uncertainty, abandonment, loss, stigma, and isolation. Future qualitative research may explore these experiences in more detail, for example, examining the lived experiences of stigma or body image in ND. All participants in the present study were white adults in their fifties or sixties, with the majority in employment.

Further research may wish to examine distress in other groups such as older adults. The present study offered preliminary evidence that acceptance may help individuals in managing distress. Further longitudinal studies would be helpful to follow individuals' psychological journey over time, and examine changes to acceptance, grief, and adjustment. Finally, future research would benefit from examining the effectiveness of psychological intervention in managing distress in ND populations.

## Conclusion

The study highlighted that uncertainty, abandonment, loss, stigma, and isolation were salient factors in the psychological distress experienced by individuals with ND. The majority of participants believed that distress came from living with the difficulties of ND, however there were some tentative beliefs that distress also preceded onset of physical symptoms. Research regarding interventions to reduce psychological distress would be helpful to inform further practice and support a person-centred approach to care.

## Declaration of Interest

The authors report there are no competing interests to declare.

# References

Albanese, A., Bhatia, K. P., Cardoso, F., Comella, C., Defazio, G., Fung, V. S., ... & Vidailhet, M. (2023). Isolated cervical dystonia: Diagnosis and classification. *Movement Disorders*. https://doi.org/10.1002/mds.29387

Ben-Shlomo, Y., Camfield, L., & Warner, T. (2002). What are the determinants of quality of life in people with cervical dystonia?. *Journal of neurology, neurosurgery, and psychiatry*, *72*(5), 608. https://doi.org/10.1136/jnnp.72.5.608

Berardelli, I., Ferrazzano, G., Pasquini, M., Biondi, M., Berardelli, A., & Fabbrini, G. (2015). Clinical course of psychiatric disorders in patients with cervical dystonia. *Psychiatry research*, *229*(1-2), 583-585. https://doi.org/10.1016/j.psychres.2015.07.076

Bernstein, C. J., Ellard, D. R., Davies, G., Hertenstein, E., Tang, N. K., Underwood, M., & Sandhu, H. (2016). Behavioural interventions for people living with adult-onset primary dystonia: a systematic review. *BMC neurology*, *16*, 1-14. https://doi.org/10.1186/s12883-016-0562-y

Cadden, M. H., Arnett, P. A., Tyry, T. M., & Cook, J. E. (2018). Judgment hurts: the psychological consequences of experiencing stigma in multiple sclerosis. *Social science & medicine*, *208*, 158-164. https://doi.org/10.1016/j.socscimed.2018.01.015

Camfield, L., Ben-Shlomo, Y., Warner, T. T., & Epidemiological Study of Dystonia in Europe (ESDE) Collaborative Group. (2002). Impact of cervical dystonia on quality of life. *Movement disorders*, *17*(4), 838-841.https://doi.org/10.1002/mds.10127

Ceylan, D., Erer, S., Zarifoğlu, M., Türkeş, N., & Özkaya, G. (2019). Evaluation of anxiety and depression scales and quality of LIFE in cervical dystonia patients on botulinum toxin therapy and their relatives. *Neurological Sciences*, *40*, 725-731. https://doi.org/10.1007/s10072-019-3719-9

Comella, C., & Bhatia, K. (2015). An international survey of patients with cervical dystonia. *Journal of neurology*, *262*, 837-848. https://doi.org/ 10.1007/s00415-014-7586-2

Costanzo, M., Belvisi, D., Berardelli, I., Maraone, A., D'Antonio, F., Baione, V., ... & Berardelli, A. (2021). Motor and non-motor subtypes of cervical dystonia. *Parkinsonism & Related Disorders*, *88*, 108-113. https://doi.org/10.1016/j.parkreldis.2021.06.008

Curvis, W., & Methley, A. (Eds.). (2021). *Acceptance and Commitment Therapy and Brain Injury: A Practical Guide for Clinicians*. Routledge. https://doi.org/10.4324/9781003024408

Defazio, G., Belvisi, D., Comella, C., Hallett, M., Jinnah, H. A., Cimino, P., ... & Berardelli, A. (2023). Validation of a guideline to reduce variability in diagnosing cervical dystonia. *Journal of Neurology*, *270*(5), 2606-2612. https://doi.org/10.1007/s00415-023-11585-6

Defazio, G., Jankovic, J., Giel, J. L., & Papapetropoulos, S. (2013). Descriptive epidemiology of cervical dystonia. *Tremor and Other Hyperkinetic Movements*, *3*. https://doi.org/ 10.7916/D80C4TGJ

Dennison, L., Moss-Morris, R., & Chalder, T. (2009). A review of psychological correlates of adjustment in patients with multiple sclerosis. *Clinical psychology review*, *29*(2), 141-153. https://doi.org/10.1016/j.cpr.2008.12.001

Di Biase, L., Di Santo, A., Caminiti, M. L., Pecoraro, P. M., & Di Lazzaro, V. (2022). Classification of dystonia. *Life*, *12*(2), 206. https://doi.org/10.3390/life12020206

Dickson, A., Knussen, C., & Flowers, P. (2008). 'That was my old life; it's almost like a past-life now': Identity crisis, loss and adjustment amongst people living with Chronic

Fatigue Syndrome. *Psychology and Health*, *23*(4), 459-476. https://doi.org/10.1080/08870440701757393

Doka, K. J. (2008). Disenfranchised grief in historical and cultural perspective. In M. S. Stroebe, R. O. Hansson, H. Schut, & W. Stroebe (Eds.), *Handbook of bereavement research and practice: Advances in theory and intervention* (pp. 223–240). https://doi.org/10.1037/14498-011

Fletcher, L., & Hayes, S. C. (2005). Relational frame theory, acceptance and commitment therapy, and a functional analytic definition of mindfulness. *Journal of rational-emotive and cognitive-behavior therapy*, *23*, 315-336. https://doi.org/10.1007/s10942-005-0017-7

Geach, N., Moghaddam, N. G., & De Boos, D. (2018). A systematic review of team formulation in clinical psychology practice: definition, implementation, and outcomes. *Psychology and Psychotherapy: Theory, Research and Practice*, *91*(2), 186-215. https://doi.org/10.1111/papt.12155

Gilbert, P. (2014). The origins and nature of compassion focused therapy. *British Journal of Clinical Psychology*, *53*(1), 6–41. https://doi.org/10.1111/bjc.12043

Gowling, H., O'Keeffe, F., & Eccles, F. J. (2024). Stigma, coping strategies, distress and wellbeing in individuals with cervical dystonia: a cross-sectional study. *Psychology, Health & Medicine*, 1-18. https://doi.org/10.1080/13548506.2024.2305172

Grütz, K., & Klein, C. (2021). Dystonia updates: definition, nomenclature, clinical classification, and etiology. *Journal of Neural Transmission*, *128*, 395-404. https://doi.org/10.1007/s00702-021-02314-2

Gunning, J. N., & Taladay-Carter, C. (2023). Grieving "The death of possibility": Memorable messages of (dis) enfranchised loss in invisible, physical illness. *Health Communication*, 1-11. https://doi.org/10.1080/10410236.2023.2257942

Hagger, M. S., & Orbell, S. (2022). The common sense model of illness self-regulation: A conceptual review and proposed extended model. *Health psychology review*, *16*(3), 347-377. https://doi.org/10.1080/17437199.2021.1878050

Hayes, S. C., Luoma, J. B., Bond, F. W., Masuda, A., & Lillis, J. (2006). Acceptance and Commitment Therapy: Model, processes and outcomes. *Behaviour Research and Therapy*, *44*(1), 1–25. https://doi.org/10.1016/j.brat.2005.06.006

Jahanshahi, M., & Marsden, C. D. (1990). Body concept, disability, and depression in patients with spasmodic torticollis. *Behavioural neurology*, *3*(2), 117-131. https://doi.org/ 10.3233/BEN-1990-3206

Jin, X., Wong, C. L., Li, H., Chen, J., Chong, Y. Y., & Bai, Y. (2021). Acceptance and Commitment Therapy for psychological and behavioural changes among parents of children with chronic health conditions: A systematic review. *Journal of advanced nursing*, *77*(7), 3020-3033. https://doi.org/10.1111/jan.14798

LaHue, S. C., Albers, K., Goldman, S., Lo, R. Y., Gu, Z., Leimpeter, A., ... & Tanner, C. M. (2020). Cervical dystonia incidence and diagnostic delay in a multiethnic population. *Movement Disorders*, *35*(3), 450-456.https://doi.org/10.1002/mds.27927

Lehman, B. J., David, D. M., & Gruber, J. A. (2017). Rethinking the biopsychosocial model of health: Understanding health as a dynamic system. *Social and personality psychology compass*, *11*(8), e12328. https://doi.org/10.1111/spc3.12328

Lencer, R., Steinlechner, S., Stahlberg, J., Rehling, H., Orth, M., Baeumer, T., ... & Hagenah, J. (2009). Primary focal dystonia: evidence for distinct neuropsychiatric and personality profiles. *Journal of Neurology, Neurosurgery & Psychiatry*. https://doi.org./10.1136/jnnp.2008.170191

Leventhal, H., Phillips, L. A., & Burns, E. (2016). The Common-Sense Model of Self-Regulation (CSM): a dynamic framework for understanding illness self-management. *Journal of behavioral medicine*, *39*, 935-946. https://doi.org/10.1007/s10865-016-9782-2

Levitt, H. M., Bamberg, M., Creswell, J. W., Frost, D. M., Josselson, R., & Suarez-Orozco, C. (2018). Journal article reporting standards for qualitative research in psychology: The APA publications and communications board task force report. *American Psychologist*, *73*(1), 26-46. https://doi.org/10.1037/amp0000151

Lewis, L., Butler, A., & Jahanshahi, M. (2008). Depression in focal, segmental and generalized dystonia. *Journal of neurology*, *255*, 1750-1755. https://doi.org/10.1007/s00415-008-0020-x

Link, B. G., & Phelan, J. C. (2001). Conceptualizing stigma. *Annual review of Sociology*, *27*(1), 363-385. https://doi.org/10.1146/annurev.soc.27.1.363

Ma, H. I., Saint-Hilaire, M., Thomas, C. A., & Tickle-Degnen, L. (2016). Stigma as a key determinant of health-related quality of life in Parkinson's disease. *Quality of Life Research*, *25*, 3037-3045. https://doi.org/ 10.1007/s11136-016-1329-z

Marciniec, M., Szczepańska-Szerej, A., Popek-Marciniec, S., & Rejdak, K. (2020). Pain incidence in cervical dystonia is determined by the disease phenotype. *Journal of Clinical Neuroscience*, *79*, 133-136. https://doi.org/10.1016/j.jocn.2020.07.069

Maxwell-Scott, M. (2023). *Psychosocial aspects of living with a visible neurological condition*. [Doctoral Thesis, Lancaster University]. Lancaster University. https://doi.org/10.17635/lancaster/thesis/2079

Mishel, M. H. (1988). Uncertainty in illness. *Image: The Journal of Nursing Scholarship*, *20*(4), 225-232. https://doi.org/10.1111/j.1547-5069.1988.tb00082.x

Moll, M., Rosenthal, D., & Hefter, H. (2018). Quality of life in long-term botulinum toxin treatment of cervical dystonia: Results of a cross sectional study. *Parkinsonism & Related Disorders*, *57*, 63-67. https://doi.org/10.1016/j.parkreldis.2018.07.019

Monaghan, R., McCormack, D., Ndukwe, I., O'Riordan, S., Burke, T., Pender, N., ... & O'Keeffe, F. (2022). 238 Cogniton and mood disorder in cervical dystonia. *Journal of Neurology, Neurosurgery and Psychiatry*, *93*(6), A82-A82. https://doi.org/10.1016/j.prdoa.2020.100084

Moraru, E., Schnider, P., Wimmer, A., Wenzel, T., Birner, P., Griengl, H., & Auff, E. (2002). Relation between depression and anxiety in dystonic patients: implications for clinical management. *Depression and anxiety*, *16*(3), 100-103. https://doi.org/10.1002/da.10039

Morgan, A., Eccles, F. J., & Greasley, P. (2021). Experiences of living with dystonia. *Disability and rehabilitation*, *43*(7), 944-952. https://doi.org/10.1080/09638288.2019.1645217

Moss-Morris, R. (2013). Adjusting to chronic illness: time for a unified theory. https://doi.org/10.1111/bjhp.12072

Mueller, J., Skogseid, I. M., Benecke, R., Kupsch, A., Trottenberg, T., Poewe, W., ... & Deep-Brain Stimulation for Dystonia Study Group. (2008). Pallidal deep brain stimulation improves quality of life in segmental and generalized dystonia: results from a prospective, randomized sham-controlled trial. *Movement disorders*, *23*(1), 131-134. https://doi.org/10.1002/mds.21783

Müller, J., Kemmler, G., Wissel, J., Schneider, A., Voller, B., Grossmann, J., ... & Austrian Botulinum Toxin and Dystonia Study Group. (2002). The impact of blepharospasm and cervical dystonia on health-related quality of life and depression. *Journal of neurology*, *249*, 842-846. https://doi.org/ 10.1007/s00415-002-0733-1

Murray, C. D., & Wilde, D. J. (2020). Thinking about, doing and writing up research using interpretative phenomenological analysis. In *Handbook of theory and methods in* *applied health research* (pp. 140-166). Edward Elgar Publishing. https://doi.org/10.4337/9781785363214.00015

National Institute for Health and Care Excellence. (2009). Depression in adults with a chronic physical health problem: recognition and management. https://www.nice.org.uk/guidance/cg91/chapter/Recommendations#step-4-complex-and-severe-depression

Ndukwe, I., O'Riordan, S., Walsh, C. B., & Hutchinson, M. (2020). Mood disorder affects age at onset of adult-onset cervical dystonia. *Clinical Parkinsonism & Related Disorders*, *3*, 100049. https://doi.org/10.1016/j.prdoa.2020.100049

Neimeyer, R. A. (2006). Complicated grief and the reconstruction of meaning: Conceptual and empirical contributions to a cognitive-constructivist model. *Clinical Psychology: Science and Practice*, *13*(2), 141-145. https://doi.org/10.1111/j.1468-2850.2006.00016.x

O'Connor, S., Hevey, D., & O'Keeffe, F. (2023b). Illness perceptions, coping, health-related quality of life and psychological outcomes in cervical dystonia. *Journal of Clinical Psychology in Medical Settings*, *30*(1), 129-142. https://doi.org/ 10.1007/s10880-022-09851-2

O'Connor, S., Hevey, D., Burke, T., Rafee, S., Pender, N., & O'Keeffe, F. (2023a). A Systematic Review of Cognition in Cervical Dystonia. *Neuropsychology Review*, 1-21. https://doi.org/10.1007/s11065-022-09558-z

O'Riordan, S., & Hutchinson, M. (2004). Cervical dystonia following peripheral trauma: a case-control study. *Journal of neurology*, *251*, 150-155. https://doi.org/10.1007/s00415-004-0291-9

Ortiz, R. M., Scheperjans, F., Mertsalmi, T., & Pekkonen, E. (2019). Comorbidity and retirement in cervical dystonia. *Journal of neurology*, *266*, 2216-2223. https://doi.org/ 10.1007/s00415-019-09402-0

Pagnini, F., Bosma, C. M., Phillips, D., & Langer, E. (2014). Symptom changes in multiple sclerosis following psychological interventions: a systematic review. *BMC neurology*, *14*(1), 1-9. https://doi.org/10.1186/s12883-014-0222-z

Pallesen, H. (2014). Body, coping and self-identity. A qualitative 5-year follow-up study of stroke. *Disability and rehabilitation*, *36*(3), 232-241. https://doi.org/10.3109/09638288.2013.788217

Papathanasiou, I., MacDonald, L., Whurr, R., & Jahanshahi, M. (2001). Perceived stigma in Spasmoic Torticollis. *Movement Disorders: Official Journal of the Movement Disorder Society*, *16*(2), 280-285. https://doi.org/10.1002/mds.1055

Paracka, L., Wegner, F., Escher, C., Klietz, M., de Zwaan, M., Abdallat, M., ... & Krauss, J. K. (2020). Body concept and quality of life in patients with idiopathic dystonia. *Brain Sciences*, *10*(8), 488. https://doi.org/10.3390/brainsci10080488

Pietkiewicz, I., & Smith, J. A. (2014). A practical guide to using interpretative phenomenological analysis in qualitative research psychology. *Psychological journal*, *20*(1), 7-14. https://doi.org/10.14691/CPPJ.20.1.7

Pillai-Friedman, S., & Ashline, J. L. (2014). Women, breast cancer survivorship, sexual losses, and disenfranchised grief–a treatment model for clinicians. *Sexual and Relationship Therapy*, *29*(4), 436-453. https://doi.org/10.1080/14681994.2014.934340

Richman, S. (2022). Grief and Chronic Disease. *Cinahl Information Systems: Glendale, CA, USA*.

Richter, A., & Löscher, W. (1998). Pathophysiology of idiopathic dystonia: findings from genetic animal models. *Progress in neurobiology*, *54*(6), 633-677. https://doi.org/10.1016/s0301-0082(97)00089-0

Rodrigues, F. B., Duarte, G. S., Prescott, D., Ferreira, J., & Costa, J. (2019). Deep brain stimulation for dystonia. *The Cochrane database of systematic reviews*, *2019*(1). https://doi.org/10.1002/14651858.CD012405.pub2

Rosales, R. L., Cuffe, L., Regnault, B., & Trosch, R. M. (2021). Pain in cervical dystonia: mechanisms, assessment and treatment. *Expert Review of Neurotherapeutics*, *21*(10), 1125-1134. https://doi.org/10.1080/14737175.2021.1984230

Scambler, G., & Hopkins, A. (1986). Being epileptic: coming to terms with stigma. *Sociology of health & illness*, *8*(1), 26-43. https://doi.org/10.1111/1467-9566.ep11346455

Sheldon, K., & Harding, E. (2010). Good practice guidelines to support the involvement of service users and carers in clinical psychology services. British Psychological Society.

Simpson, J., McMillan, H., Leroi, I., & Murray, C. D. (2015). Experiences of apathy in people with Parkinson's disease: a qualitative exploration. *Disability and rehabilitation*, *37*(7), 611-619. https://doi.org/10.3109/09638288.2014.939771

Skogseid, I. M., Malt, U. F., Røislien, J., & Kerty, E. (2007). Determinants and status of quality of life after long-term botulinum toxin therapy for cervical dystonia. *European journal of neurology*, *14*(10), 1129-1137. https://doi.org/10.1111/j.1468-1331.2007.01922.x

Smith, J. A. (2011). Evaluating the contribution of interpretative phenomenological analysis. *Health psychology review*, *5*(1), 9-27. https://doi.org/10.1080/17437199.2010.510659

Smith, J. A., & Eatough, V. (2015). Interpretative Phenomenological Analysis. In E.Lyons & A. Coyle (Eds.), Analysing *Qualitative Data in Psychology 2nd Edition* (pp. 50–57). SAGE. https://doi.org/10.53841/bpsqmip.2022.1.33.51

Smith, J. A., Flowers, P., & Larkin, M. (2022). *Interpretative phenomenological analysis: Theory, method and research* (2nd ed.). Sage Publications.

Todd, D., Simpson, J., & Murray, C. (2010). An interpretative phenomenological analysis of delusions in people with Parkinson's disease. *Disability and Rehabilitation*, *32*(15), 1291-1299. https://doi.org/10.3109/09638280903514705

Tomaschek, R., Lampart, P., Scheel-Sailer, A., Gemperli, A., Merlo, C., & Essig, S. (2022). Improvement strategies for the challenging collaboration of general practitioners and specialists for patients with complex chronic conditions: a scoping review. *International journal of integrated care*, *22*(3). https://doi.org/10.5334/ijic.5970

Tomic, S., Petkovic, I., Pucic, T., Resan, B., Juric, S., & Rotim, T. (2016). Cervical dystonia and quality of life. *Acta Neurologica Belgica*, *116*, 589-592. htpps://doi.org/ 10.1007/s13760-016-0634-1

Velickovic, M., Benabou, R., & Brin, M. F. (2001). Cervical dystonia: pathophysiology and treatment options. *Drugs*, *61*, 1921-1943. https://doi.org/10.2165/00003495-200161130-00004

Vicary, S., Young, A., & Hicks, S. (2017). A reflective journal as learning process and contribution to quality and validity in interpretative phenomenological analysis. *Qualitative social work*, *16*(4), 550-565. https://doi.org/10.1177/147332501663524

Wenzel, T., Schnider, P., Wimmer, A., Steinhoff, N., Moraru, E., & Auff, E. (1998). Psychiatric comorbidity in patients with spasmodic torticollis. *Journal of* *psychosomatic research*, *44*(6), 687-690. https://doi.org/10.1016/s0022-3999(97)00229-8

Williams, L., McGovern, E., Kimmich, O., Molloy, A., Beiser, I., Butler, J. S., ... & Hutchinson, M. (2017). Epidemiological, clinical and genetic aspects of adult onset isolated focal dystonia in Ireland. *European journal of neurology*, *24*(1), 73-81. https://doi.org/10.1111/ene.13133

Yardley, L. (2000). Dilemmas in qualitative health research. *Psychology and health*, *15*(2), 215-228. https://doi.org/10.1080/08870440008400302

Zurowski, M., McDonald, W. M., Fox, S., & Marsh, L. (2013). Psychiatric comorbidities in dystonia: emerging concepts. *Movement Disorders*, *28*(7), 914-920. https://doi.org/10.1002/mds.25501

# Tables and Figures

## Table 2-1

**Participant Demographics**

| Name | Age | Gender (same as assigned at birth) | Ethnicity | Employment Status | Disease Duration (years) | Currently on treatment for ND-Yes/No | Relationship Status | Living alone Yes/No | Interview Mode |
|---|---|---|---|---|---|---|---|---|---|
| Kathryn | 63 | Female (Yes) | White British | Employed | 2 | No | Married | No | Video |
| Paul | 62 | Male (Yes) | White British | Employed | 4 | No | Married | No | Video |
| Barbara | 69 | Female (Yes) | White British | Retired | 1 | Yes | Single | Yes | Telephone |
| Brian | 67 | Male (Yes) | White British | Employed | 40 | Yes | Married | No | Video |
| Sharon | 63 | Female (Yes) | White Irish | Employed | 10 | Yes | Married | No | Video |
| Michael | 69 | Male (Yes) | White British | Retired | 17 | Yes | Married | No | Video |
| Donna | 52 | Female (Yes) | White British | Employed | 3 | Yes | Relationship | No | Telephone |
| Steve | 61 | Male (Yes) | White British | Employed | 0.5 | Yes | Married | No | Video |
| Sandra | 58 | Female (Yes) | White British | Employed | 1.5 | Yes | Married | No | Telephone |
| Christopher | 60 | Male (Yes) | White British | Unemployed | 12 | Yes | Single | Yes | Telephone |
| Audrey | 64 | Female (Yes) | White British | Employed | 27 | Yes | Married | No | Video |

# Appendices

## Appendix 2-A

### Implications for Rehabilitation

- Neck dystonia is associated with debilitating and chronic symptoms that impact a person’s ability to engage in activities of everyday living
- Psychological distress in neck dystonia is common in individuals with neck dystonia, with psychosocial variables including mood, body image, and stigma associated with reduced quality of life
- Multidisciplinary interventions, with clinical psychological input, should support individuals to manage these difficulties, with the aim of improving their quality of life

## Appendix 2-B

## Process of Exploratory Noting and Constructing Personal Experiential Statements in an Extract from Michael

| Personal Experiential Statements | Interview Transcript | Exploratory Notes |
|---|---|---|
| *I find ways to disguise what I've got to avoid embarrassment* | Michael: …and now I can talk to you as if, you know, it's absolutely fine, and, and you probably wouldn't notice anything was wrong…<br>I: No, no<br>Michael: Em so, it, it was about…I mean if I sit like this for a moment (*moves hands from* chin) and, and, and don't kind of put my hands there, then em I can't actually hold my head still.<br>I: Mmm<br>Michael: And that, that's the difficulty that I have is that I had to devise ways to em, to avoid, to avoid, kind of to avoid that embarrassment.<br>I: Mmm mmm | *"now"-was this different in the past? Sense that Michael is thinking about how he is perceived by others*<br>*Michael using his hands to hold his head still*<br>*"I have had to" sense here that Michael felt he had no choice but to mask his symptoms*<br>*Devising ways to disguise symptoms to avoid embarrassment* |

| | | |
|---|---|---|
| *It occupies a lot of space in my mind*<br>*Feeling embarrassed around other people* | Michael: So, so, I suppose to answer your question, that it's a large, a large part of my thinking each day<br>I: Mmm mmm<br>Michael: If I'm meeting someone that's not met me before or I'm gonna meet someone for whatever reason is, I will always position myself so that I'm kinda turned like that if I'm stood talking to you now, I would stand kinda this way [*moves body towards left]*<br>I: Yes…okay<br>Michael:…so that I could talk to you and feel more comfortable, em…<br>I: Yes okay okay, yeah. So something about kind of that em…and thank you for sharing that because I can see that it's emotional talking about the impact that it's…has on your life, em, understandably, and I think what's coming across there is wanting to, wanting to almost disguise or…<br>Michael: Yeah, yeah<br>I: …hide it in some sense… | *Thinking about it a lot of the time*<br>*Something about meeting new people- is there a greater sense of embarrassment around someone new?*<br>*Changing body position around others –adapting to feel more comfortable* |

| | | |
|---|---|---|
| | Michael: Yeah<br>I: …em even though it's really uncomfortable. So you're going in…you're, it sounds like…from a work perspective anyway, going into these situations but kind of thinking how, how am I going to manage this, how am I going to compensate, and moving your, yeah changing your position to…<br>Michael: Yeah yeah…yeah, yeah | |
| *Developing and fine-tuning strategies to disguise symptoms* | I: …accommodate that<br>Michael: And, and…I've become very good at that, that other people that I meet will, will never notice it because I've always got into position as it were…<br>I: Yes | *There's a sense here that Michael has spent time learning best ways to disguise symptoms overtime*<br>*It appears that other people not noticing is important for Michael* |
| *There's an all-consuming nature to dystonia, it takes up a lot of space in my mind* | Michael: And em, but it, it, it's just that it occupies my mind every day, all of the time…<br>I: Yeah | *All-consuming nature, thinking about it all the time* |
| | Michael: …whatever situation I'm going into<br>I: Mmm mmm | *It's there all the time, sense of no let up* |

| | | |
|---|---|---|
| *Shame in relation to the thoughts and feelings I have about dystonia*<br>*The psychological side of dystonia is debilitating for me* | Michael: That…and, and that, it kind of feels as though it's stopped me thinking of, of things that I should be thinking about. I shouldn't have this…these, these kind of thoughts going through my head all the time about well you know, I'm gonna watch how I'm gonna stand here, you know, I better kind of sit like this, I shouldn't even be thinking things like that, em I should be thinking of you know, what am I going to do with my life, what's next, what am I going to do today, that kind of thing…<br>I: Mmm mmm<br>Michael: …and I just find it a bit em…yeah a bit debilitating really in that sense, psychologically<br>I: Yeah of course, so almost like it takes up all the thinking space and…<br>MIch ael: Yeah…well that's it, that's exactly it, that's, that's exactly it…it takes up, it does take up thinking space that it shouldn't do…<br>I: Mmm<br>Michael: Em, you know maybe if I was a stronger person then I wouldn't feel like that, you know, I, I | *No space to think about other things, again suggesting the all-consuming nature of dystonia*<br>*Language-"I shouldn't" and "I should", hints at self-criticism or shame about his own thought processes*<br>*"debilitating" strong language indicating the salience of the psychological side of dystonia for Michael*<br>*Language here again giving a sense of shame: "if I was stronger";* |

| | | |
|---|---|---|
| *Shame in relation to the thoughts and feelings I have about dystonia* | kind of chastise myself em, em, there are people out there with far worse sit…things wrong with it them than I’ve got wrong with me physically or, or whatever, em and that I shouldn’t you know, I shouldn’t kind of feel they way that I do about it… | *“chastise”; “I shouldn’t”*<br>*Emotive for Michael*<br>*Doesn’t think he should feel the way he’s feeling* |

## Appendix 2-C

## Audit Trail-Searching for Connections and Clustering of Michael’s PETs

| **Searching for Connections Across Experiential Statements** |
|---|
| **Neck dystonia does not make me feel good about myself** |
| There was a feeling of infuriation when other people commented on how I was appearing |
| The longer the condition went on, the worse it became, and the more self-conscious I was |
| I find ways to disguise what I’ve got to avoid embarrassment |
| I disguise what’s going on because I don’t have the confidence to explain to people |
| I worry that I will stand out and just want to live a “normal” life |
| I feel ashamed at how appearing odd to other people |
| I don’t want to appear unattractive to other people |
| The distress has only been since living with the dystonia |
| Neck dystonia doesn’t make me feel good about myself and has lowered my self-esteem |
| Belief that not being a confident person makes this more difficult |
| Belief that the psychological difficulties have grown more with the condition, and the constancy or permanency of finding ways to disguise |
| **Resentment for the space that dystonia takes up in my life** |
| It’s all-consuming and takes up a lot of space in my mind |
| The psychological side of dystonia is debilitating for me |
| It’s taken away a part of me |
| Finding it really difficult that this is how people define me now |
| I don’t want this to define who I am as a person because I’m more than this condition |
| The feelings of self-consciousness stop me from doing things I want to do |
| Difficulty accepting this condition, just wanting it to go away |
| I don’t do all the things I’d like to do because I’m afraid of standing out |
| The psychological side of dystonia impacts my life in a way that the physical doesn’t |
| Resentment toward dystonia |

When first diagnosed not believing that nothing could be done to treat it, a feeling of light at the end of the tunnel

**Shame in how I feel**

I shouldn't be feeling like this, I should be stronger and able to deal with this

Internal struggle where I experience shame in my feelings

Shame in relation to the thoughts and feelings I have about dystonia

Feeling guilty for experiencing distress because other people are in worse situations

I should be able to deal with this without these feelings of distress

Reluctance to open up about distress to others

I feel I should just deal with it myself and get on with it

There's a real struggle accepting the emotional side of this condition

**Frustrations in healthcare system**

The doctors suggested it was nothing untoward and so I spent two years trying to self-correct it

I felt annoyed in the lead up to the diagnosis in myself and the healthcare system

## Appendix 2-D

## Audit Trail-Michael's PET Table with Supportive Quotes

### A.NECK DYSTONIA DOESN'T MAKE ME FEEL GOOD ABOUT MYSELF

**The longer the condition goes on the more distressed I feel**

Belief that the psychological difficulties have grown more with the condition, and the constancy or permanency of finding ways to disguise

*"the longer it's gone on, I think the more I've, I've realised actually that there are probably, well there are mental health implications"*

When first diagnosed not believing that nothing could be done to treat it, a feeling of light at the end of the tunnel

*"I wasn't sure I believed that at the time, but I think at the time I thought, no, I…I can, I can sort this out, you know"*

**I disguise this condition all the time became I'm embarrassed at how I appear to others**

I worry that I will stand out and just want to live a "normal" life

*"I just want, just want to to be me, and, and, and live my life in a normal way and, and em not stand out and look different in any way"*

Belief that not being a confident person makes this more difficult

*"I think that's why I try and hide it. It's like because I, I haven't, haven't got the confidence to deal with it I guess. And, and just to say, well that's the way I am, take it or leave it, you know, and I don't care what you think. That's not how I feel"*

### B. I FEEL RESENTMENT FOR THE SPACE THAT DYSTONIA TAKES UP IN MY LIFE

**It's taken away a part of who I am**

Finding it really difficult that this is how people define me now

*"I don't want to be the guy that people look at and say, em well that's the guy who holds his head in a funny way"*

I don't want this to define who I am as a person because I'm more than this condition

*"I'm not just a guy who, who holds his head in a strange way. I'm, you know, I'm kind of me, that's it"*

**It stops me from doing things I want to do**

The feelings of self-consciousness stop me from doing things I want to do

*"...joining a gym for one thing, that's, that's definitely em something that I would have liked to have done…em…but it's the same thing…it's just…I don't, I kind of…it's like raising my head above the parapet"*

The psychological side of dystonia impacts my life in a way that the physical doesn't

*"physically it doesn't stop me doing anything at all really…em it's this kind of self-inflicted, em, you know, reluctance I guess…"*

**I just want it go away**

Difficulty accepting this condition, just wanting it to go away

*"Just kind of go away and leave me alone and, and let me do the things I want to do…"*

It's all-consuming and takes up a lot of space in my mind

*"...it's just that it occupies my mind every day, all of the time…whatever situation I'm going into…it kind of feels as though its' stopped me thinking of, of things that I should be thinking about"*

### C. THE INTERNAL STRUGGLE BETWEEN THE FEELINGS I HAVE AND THE SHAME ASSOCIATED WITH THOSE FEELINGS

**I should be strong enough to deal with this**

I shouldn't be feeling like this, I should be stronger and able to deal with this

*"...that's down to me being not a strong enough person, and being, being you know, being weaker than I should be I suppose. Em I should be strong enough to kind of deal with that, and like I say, just, just say to people, well just, you know take it or leave it. But that's, that's not what I am"*

Feeling guilty for experiencing distress because other people are in worse situations

*"I kind of chastise myself em, em, there are people out there with far worse sit…things wrong with it them than I've got wrong with me physically or, or whatever, em and that I shouldn't you know, I shouldn't kind of feel they way that I do about it…"*

**Reluctance to open up to others about the emotional distress**

I feel I should just deal with it myself and get on with it

*"...feeling as though I should be able to deal with things myself, em and yeah, I'll kind of let my wife in, em but I wouldn't talk in depth I don't think to anyone else about it."*

## D. THE LEAD UP TO DIAGNOSIS LED TO NEGATIVE FEELINGS

**Frustrations in the healthcare providers**

The doctors suggested it was nothing untoward and so I spent two years trying to self-correct it

*"It was frustrating, not, not…to think that it had been two years since I had first been to the doctor…before I actually got a diagnosis"*

Believing there was more to this than what doctors said and doing own research

*"I was diagnosing it myself by then…I, I'd realised by then there was something not quite right"*

**Annoyed at myself that I couldn't correct this**

I felt annoyed in the lead up to the diagnosis in myself

*"…a bit of annoyance with myself that I couldn't correct it myself…"*

## Appendix 2-E

## Audit Trail-Developing Group Experiential Themes using an Initial Colour Coding system to Search for Connections between PETs

| **Particular-Whole: Initial Clustering of PETs** |
|---|
| • Frustration of not knowing what was wrong (Barbara)<br>• The lead up to the diagnosis was associated with a lot of worry (Barbara)<br>• The condition wasn’t treated appropriately because nobody knew what was wrong (Brian)<br>• Resorting to and drugs was a way of coping with this for Brian<br>• Sharon was prescribed antidepressants but her anxiety was about the symptoms which weren’t being investigated<br>• Feeling uneasy and unnerved by symptoms before diagnosis (Sharon)<br>• The lead up to diagnosis was long and worrying (Sharon)<br>• When first diagnosed not believing that nothing could be done to treat it, a feeling of light at the end of the tunnel (Michael)<br>• The doctors suggested it was nothing untoward and so Michael spent two years trying to self-correct it<br>• Michael felt annoyed in himself in the lead up to the diagnosis<br>• Donna felt stupid that she couldn't control her symptoms<br>• The unpredictability of ND made it difficult to show the doctor what was happening |

- Being dismissed by her doctor made her feel stupid
- There was no doubt in Donna's mind that the injections would work
- Relief upon receiving diagnosis that it wasn’t a more serious neurological condition
- The worry came from Steve thinking there was something seriously wrong with him
- An inaccessible healthcare system with lengthy waiting times exacerbated the distress
- Lack of understanding from the healthcare system made distress worse
- The level of anxiety Steve felt at this time shocked him because he'd never felt fear like it
- Difficulty functioning due to debilitating anxiety
- Hope that injections would work and life would get back to normal
- Worrying about possible reasons for why this was happening (Christopher)
- Relief upon initial diagnosis, unsure what was to come
- Audrey found it devastating that doctors didn’t take her symptoms seriously
- Feeling abnormal for symptoms that had no explanation
- Diagnosis was a vindication for what Audrey had experienced
- There was a feeling that life could now begin
- Awareness of appearing differently around other people
- Avoidance of social situations now because it's too difficult

- Other people don't recognise the condition or understand what Kathryn is living with
- Not feeling listened to by his neurologist led to Paul experiencing feelings of despair and hopelessness
- Paul's neurologist didn’t answer his questions about his condition
- Seeking out alternatives offered Paul a sense of hope and positivity
- The feelings of anxiety were a shock to a Paul
- There was a lack of appreciation for the psychological side of condition
- The value of meeting others living with neck dystonia (Paul)
- Appreciation for other people understanding my need for support without me having to ask for it (Paul)
- Less bothered about appearance and more about social behaviour and interactions
- Loss for social connections  and interactions that working brought to Barbara
- There was no warning for what she was going to experience (Barbara)
- Barbara spent a lot of time and effort finding the answers she was searching for
- I wasn’t listened to when forthright and direct with my neurologist about how I felt
- Masking the psychological distress around other people
- There's a sense of shame around the psychological distress that Brian feels
- Feeling self-conscious about how he is appearing to others (Brian)
- Focus of healthcare on injections rather than psychological side (Brian)

- Feeling disheartened by dismissive GP led to Sharon doing her own research to find out what was wrong
- Feeling unable to raise this questions or fears with my neurologist
- Feeling more aware of how she is appearing to other people now (Sharon)
- Michael worries that he will stand out and just wants to live a “normal” life
- Belief that not being a confident person makes this more difficult
- The feelings of self-consciousness stops Michael from doing things he wants to do
- Michael thinks he shouldn’t be feeling like this, and he should be stronger and able to deal with this
- Feeling guilty for experiencing distress because other people are in worse situations
- Thinking he should be able to deal with this by himself
- Believing there was more to this than what doctors said and doing own research
- Lack of acknowledgement for distress causes Donna to question her feelings
- Donna feels embarrassed that she will look odd to other people
- Recognition that certain social situations are no longer comfortable
- Sandra had the feeling that she was left to deal with this herself
- If others don't care about her, why should she bother
- Sandra feels like nobody is listening to her
- Difficulty understanding why her brain would force her body into a position that is so unnatural

- Belief that greater explanation about what to expect in dystonia would reduce distress
- Not many doctors understand the condition
- People commenting on Christopher's symptoms is really difficult
- Not wanting to go out because of how other people see him
- Feeling isolated in that nobody else understands what this condition is
- Uncertainty whether people will empathise with a condition that’s not life-threatening
- The symptoms cause Audrey to feel embarrassed around others

- The belief that earlier traumas are connected to his condition (Paul)
- Patterns in mood associated with patterns in physical symptoms (Paul)
- Strong feelings that Brian was in the wrong place at that point in his life (Brian)
- Brian was drinking more to try to fit in with this new life he was in
- Identifying the possibility that there's a psychological role in the onset of dystonia (Brian)
- Recognising that distraction helps to re-focus attention away from difficult symptoms
- Distraction helps Brian to refocus other people’s attention away from this condition
- Brian is constantly battling between the philosophical part of me and the older version of me
- The struggle to shut out the old self can lead to feelings of depression for Brian
- Distraction can help me to get on with things

- Identifying times when his mind is distracted and the physical symptoms reduce
- Significant life changes and stressors prior to onset of symptoms
- The more Steve worried about the physical symptoms he was experiencing, the worse the symptoms were becoming
- Desire to learn more about the mind-body relationship in this condition to manage it as best as possible
- Refocusing attention away from self could reduce the prominence of symptoms
- Recognising the possibility that physical symptoms are less prominent when mind distracted with other things
- Wondering if onset of symptoms was associated to previous trauma
- Difficulty understanding whether anxiety is part of self or the dystonia, or a combination of both
- Recognition of the loss of identity that comes with this condition
- Kathryn tries to continue daily activities, but she doesn't always enjoy them
- Everything Kathryn does now is done at an angle
- Life now involves making adaptations to account for neck position
- Comparison with surviving cancer, finding it challenging that she can't fight this as much
- Losing his independence was like losing a part of himself (Paul)
- Chronic fatigue makes Barbara feel so frustrated
- The nature of everyday tasks is distressing
- It’s difficult resigning to the fact that she can’t do things she enjoyed anymore

- Belief that her distress came from coping with the pain and discomfort, and people noticing my sudden movements (Sharon)
- Spending a lot of time thinking about neck dystonia and adaptions she might need to make (Sharon)
- Sharon feels more relaxed now if someone walks with her
- Sharon worries about how this condition will progress and be impacted in later life
- Identification that it's not going anywhere so Sharon feels she needs to get on with it
- Belief that the psychological difficulties have grown more with the condition, and the constancy or permanency of finding ways to disguise
- Michael finds it difficult thinking that this is how other people define him now
- Not wanting this to define who Michael is as a person because he's more than this condition
- The psychological side of dystonia impacts Michael's life in a way that the physical side doesn’t
- Difficulty accepting this condition, just wanting it to go away
- It’s all-consuming and takes up a lot of space in his mind
- Determination not to let dystonia stop her from doing anything
- Rather than focus on things she can't do, Donna tries to stay grateful for things she can
- Worrying about the future and the progression of dystonia
- Acceptance that Steve might not be able to do things he loves as well as he used to, but he can still do them

- Steve can still appreciate life and enjoy being around other people
- The constant struggle to find comfort is exhausting
- Sandra feels her body has changed in a way that doesn't feel nice to her
- Sandra feels like it's not fair that this has happened to her
- Low mood comes from living with the pain
- Sadness in giving up things she once loved
- The physical effort required takes the pleasure out of activities that should be enjoyable
- The depression comes from living with the symptoms (Christopher)
- The permanency of the symptoms is difficult
- Not being able to work and use skills he had spent years learning
- Steve feels that dystonia makes his anxiety worse
- When other difficult things happen in life, they are exacerbated by dystonia
- DBS surgery leaving Christopher with additional physical difficulties
- Feeling that there's not many options left at the moment
- Successful treatment helped the distress
- Waiting longer than 12 weeks between injections increases stress
- Losing a sense of self because of the effects of living with dystonia

| |
|---|
| • Accepting dystonia as a part of self removes the shame and makes the condition easier to live with |

## Appendix 2-F

## Audit Trail-GET Table with Supportive Quotes

| A. UNCERTAINTY IN THE LEAD UP TO DIAGNOSIS “I WAS SO FULL OF FEAR” |
|---|
| **Worrying about symptoms** |
| The lead up to the diagnosis was associated with high levels of anxiety |
| “*I’m not normally a very anxious person or whatever, but I’d got myself into, into such a stage that I, I mean, I didn’t recognise, you know. And it was so intense” (*Steve) |
| Worrying about possible reasons for why this was happening |
| *“There were so many things going through my head, you know. Is this a tumour in my head causing the problems?”* (Christopher) |
| **Feeling dismissed by healthcare providers** |
| Symptoms treated inappropriately due to lack of understanding about what was causing them |
| *“…that was probably something I mentioned as well, just feeling quite anxious about it. And er as I say, that’s what was treated at that point…not really addressing why, what was causing this at all”* (Sharon) |
| Feeling stupid because doctors were dismissing symptoms |
| “*My doctor had said to me it’s probably a habit, like a nervous habit I’d got into…and I felt a bit stupid because I was like why can’t I control this, do you know?”* (Donna) |
| **Unrealistic expectations and false hope** |
| Vindication for experiencing the symptoms |
| *"I just was so relieved and just so elated and just felt finally, you know, I've actually got a condition that I, I'm not making it up"* (Audrey) |
| Hope that things would get back to normal |
| *“I…in my, my own little like world was thinking oh, I can have the injections...I'll be coming straight back to work”* (Sandra) |
| Belief that greater explanation about what to expect in dystonia would reduce distress |

*"If someone had explained it to me like that at the beginning…I might not be quite in my mental state as I am now"* (Sandra)

**B. THE ISOLATING NATURE OF THE CONDITION "YOU DON'T KNOW WHO TO TURN TO"**

**Being left to manage the condition alone**

Not feeling listened to or having questions answered

*"I felt kind of absolute despair when I came I out, out of that meeting and it was like, well, what am I gonna do now, right you know. If he can't help me, who's gonna help me?"* (Paul)

Having to take matters into own hands

*"It took me frustratingly nine months to access the right person to get the…head up brace"* (Barbara)

Shame around the psychological distress

*"I kind of chastise myself em, em, there are people out there with far worse sit…things wrong with it them than I've got wrong with me physically or, or whatever, em and that I shouldn't you know, I shouldn't kind of feel they way that I do about it…"* (Michael)

**Nobody else understands this**

Other people don't recognise the condition or understand

*"I've got family that em, close family that actually then sort of go em oh haven't they done anything about your neck yet? And you think, you're not listening, you're not listening"* (Sandra)

Uncertainty whether people will empathise with a condition that's not life-threatening

*"…other conditions that people suffer from…you, you get a lot of….people feel sympathy or they offer support, or…it just felt in this particular condition, you just feel very, you do feel very isolated and that you don't know who to turn to"* (Audrey)

**The embarrassment of navigating the social world**

Changes to body image affect self-esteem

*"I don't feel nice at all about myself. I can feel, I can physically see that my er shoulders have become more rounded"* (Sandra)

Feeling embarrassed and self-conscious around others

*"I don't want to bump into people who I knew before who haven't seen me like this because I feel embarrassed and self-conscious"* (Kathryn)

---

**C. MIND AND BODY RELATIONSHIP "IS MY HEAD TURNING BECAUSE I'M THINKING ABOUT IT?"**

**Curiosity about the role of early trauma**

Identifying the possibility that there's a psychological role in the onset of dystonia

*"Is it something that happens to us psychologically that brings it on? Or is it something, or do we get it, and then we get psychologically sick. You know what I mean?"* (Brian)

Belief that the psychological component may be greater than the physical

*"I'm also I'm not sure in myself whether I actually have a medical problem and whether I just, you know, it's just another manifestation, physical manifestation of, of the, you know, the anxiety and the fears that I had"* (Steve)

**Regaining control of body**

Questioning a relationship between the symptoms and the mind

*"Now whether it, it's just because I know that dystonia can sometimes improve a bit or, or whether it was because so many other things were going on in my life that I just didn't have time to think about er….for this condition to rear up, I don't know"* (Audrey)

Recognising that distraction helps to re-focus attention away from difficult symptoms

**"***it makes me really wonder whether, you know…is my head turning because I'm thinking about it"* (Steve)

**D. A CHALLENGING NEW WAY OF LIFE "LIVING LIFE SIDEWAYS"**

**The incessant discomfort brings you down**

It impacts upon everything in life

*"That's how I have to live my life: sideways"* (Kathryn)

Thinking about it all the time

*"It's just that it occupies my mind every day, all of the time…whatever situation I'm going into"* (Michael)

Pain triggers the psychological aspect

*"The pain makes me feel low, just like the actual pain does…and then I can't work which makes me feel low, 'cause I'm on my own…"* (Donna)

**Struggling to accept that this is not going away**

---

Not wanting to accept this thing that is interfering with my life

*“Just kind of go away and leave me alone and, and let me do the things I want to do"* (Michael)

Feeling like it's not fair

*“There's a little bit of you know, god, why me. I, I’ve been making the effort, I’ve been exercising, I’m looking after my weight”* (Sandra)

Learning to accept dystonia as part of identity

*“Do you know what, this is me, this is, this is what I am. I'm not gonna try and make excuses anymore, I’m not gonna be embarrassed, I'm not gonna sort of hold my head in shame. If I have em flare-ups then people will just have to get over it...And that in itself…taking that mind-set…just makes the whole, the condition just much more bearable and much more easy to cope with”* (Audrey)

**Grieving for loss of self**

Dystonia takes away a part of your identity

*“I’ve, I’ve lost so much of who I was…and although I am adapting now, I'm, I'm still angry and I I'm, I'm ashamed to admit I'm jealous of like people who still have the free movement”* (Kathryn)

There's a grief for what is lost

*"I've been robbed of some of the things that I used to take for granted. And I, I suppose you go through something of a grieving process because you've lost a part of something, you’ve lost a part of you."* (Paul)

## Appendix 2-G

## Author Guidelines for Disability and Rehabilitation

### About the journal

*Disability and Rehabilitation* is an international, peer reviewed journal, publishing high-quality, original research. Please see the journal's Aims & Scope for information about its focus and peer-review policy.

From 2018, this journal will be online only, and will no longer provide print copies.

Please note that this journal only publishes manuscripts in English.

*Disability and Rehabilitation* accepts the following types of article: Research Articles, Reviews, Letters to the Editor, Case Reports, and Editorials. Systematic Reviews including meta-syntheses of qualitative research should be submitted as Reviews. All other types of Reviews will normally be considered as Perspectives in Rehabilitation.

Special Issues and specific sections on contemporary themes of interest to the Journal's readership are published. Please contact the Editor for more information.

### Open Access

You have the option to publish open access in this journal via our Open Select publishing program. Publishing open access means that your article will be free to access online immediately on publication, increasing the visibility, readership and impact of your research. Articles published Open Select with Taylor & Francis typically receive 45% more citations* and over 6 times as many downloads** compared to those that are not published Open Select.

Your research funder or your institution may require you to publish your article open access. Visit our Author Services website to find out more about open access policies and how you can comply with these.

You will be asked to pay an article publishing charge (APC) to make your article open access and this cost can often be covered by your institution or funder. Use our APC finder to view the APC for this journal.

Please visit our Author Services website if you would like more information about our Open Select Program.

*Citations for articles published online 2018-2022. Data obtained on 23rd August 2023, from Digital Science's Dimensions platform, available at https://app.dimensions.ai **Usage in 2020-2022 for articles published online 2018-2022

### Peer review

Taylor & Francis is committed to peer-review integrity and upholding the highest standards of review. For submissions to *Disability and Rehabilitation* authors are given the option to remain anonymous during the peer-review process. Authors will be able to indicate whether their paper is 'Anonymous' or 'Not Anonymous' during submission, and should pay particular attention to the below:

- Authors who wish to remain anonymous should prepare a complete text with information identifying the author(s) removed. Authors should upload their files using the 'double anonymous peer review' article types during submission. A separate title page should be included providing the full affiliations of all authors. Any acknowledgements and the Declaration of Interest statement must be included but should be worded mindful that these sections will be made available to referees.

- Authors who wish to be identified should include the name(s) and affiliation(s) of author(s) on the first page of the manuscript. Authors should upload their files using the ‘authors made known to the reviewers’ article types during submission.

Once your paper has been assessed for suitability by the editor, it will be peer-reviewed by independent, double anonymous expert referees. If you have shared an earlier version of your Author’s Original Manuscript on a preprint server, please be aware that anonymity cannot be guaranteed. Further information on our preprints policy and citation requirements can be found on our Preprints Author Services page. Find out more about what to expect during peer review and read our guidance on publishing ethics.

**Preparing your paper**

All authors submitting to medicine, biomedicine, health sciences, allied and public health journals should conform to the Uniform Requirements for Manuscripts Submitted to Biomedical Journals, prepared by the International Committee of Medical Journal Editors (ICMJE).

We also refer authors to the community standards explicit in the American Psychological Association's (APA) Ethical Principles of Psychologists and Code of Conduct.

We encourage authors to be aware of standardised reporting guidelines below when preparing their manuscripts:

- Case reports - CARE
- Diagnostic accuracy - STARD
- Observational studies - STROBE
- Randomized controlled trial - CONSORT
- Systematic reviews, meta-analyses - PRISMA

Whilst the use of such guidelines is supported, due to the multi-disciplinary nature of the Journal, it is not compulsory.

**Structure**

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); figures; figure captions (as a list).

In the main text, an introductory section should state the purpose of the paper and give a brief account of previous work. New techniques and modifications should be described concisely but in sufficient detail to permit their evaluation. Standard methods should simply be referenced. Experimental results should be presented in the most appropriate form, with sufficient explanation to assist their interpretation; their discussion should form a distinct section.

Tables and figures should be referred to in text as follows: figure 1, table 1, i.e. lower case. The place at which a table or figure is to be inserted in the printed text should be indicated clearly on a manuscript. Each table and/or figure must have a title that explains its purpose without reference to the text.

The title page should include the full names and affiliations of all authors involved in the preparation of the manuscript. The corresponding author should be clearly designated, with full contact information provided for this person.

**Word count**

Please include a word count for your paper. There is no word limit for papers submitted to this journal, but succinct and well-constructed papers are preferred.

**Format-Free Submission**

Authors may submit their paper in any scholarly format or layout. Manuscripts may be supplied as single or multiple files. These can be Word, rich text format (rtf), open document format (odt), PDF, or LaTeX files. Figures and tables can be placed within the text or submitted as separate documents. Figures should be of sufficient resolution to enable refereeing.

- There are no strict formatting requirements, but all manuscripts must contain the essential elements needed to evaluate a manuscript: abstract, author affiliation, figures, tables, funder information, references. Further details may be requested upon acceptance.
- References can be in any style or format, so long as a consistent scholarly citation format is applied. For manuscripts submitted in LaTeX format a .bib reference file must be included. Author name(s), journal or book title, article or chapter title, year of publication, volume and issue (where appropriate) and page numbers are essential. All bibliographic entries must contain a corresponding in-text citation. The addition of DOI (Digital Object Identifier) numbers is recommended but not essential.
- The journal reference style will be applied to the paper post-acceptance by Taylor & Francis.
- Spelling can be US or UK English so long as usage is consistent.

Note that, regardless of the file format of the original submission, an editable version of the article must be supplied at the revision stage.

**Alt Text**

This journal is now including Alt Text (alternative text), a short piece of text that can be attached to your figure to convey to readers the nature or contents of the image. It is typically used by systems such as pronouncing screen readers to make the object accessible to people that cannot read or see the object, due to a visual impairment or print disability. Alt text will also be displayed in place of an image, if said image file cannot be loaded. Alt Text can also provide better image context/descriptions to search engine crawlers, helping them to index an image properly. To include Alt Text in your article, please follow our Guidelines.

**Taylor & Francis Editing Services**

To help you improve your manuscript and prepare it for submission, Taylor & Francis provides a range of editing services. Choose from options such as English Language Editing, which will ensure that your article is free of spelling and grammar errors, Translation, and Artwork Preparation. For more information, including pricing, visit this website.

**Checklist: what to include**

1. **Author details**. Please ensure everyone meeting the International Committee of Medical Journal Editors (ICJME) requirements for authorship is included as an author of your paper. Please ensure all listed authors meet the Taylor & Francis authorship criteria. All authors of a manuscript should include their full name and affiliation on the cover page of the manuscript. Where available, please also include ORCiDs and social media handles (Facebook, Twitter or LinkedIn). One author will need to be identified as the corresponding author, with their email address normally displayed in the article PDF (depending on the journal) and the online article. Authors' affiliations are the affiliations where the research was conducted. If any of the named co-authors moves affiliation during the peer-review process, the new affiliation can be given as a footnote. Please note that no changes to affiliation can be made after your paper is accepted. Read more on authorship.

2. A structured **abstract** of no more than 200 words. A structured abstract should cover (in the following order): the *purpose* of the article, its *materials and methods* (the design and methodological procedures used), the *results* and conclusions (including their relevance to the study of disability and rehabilitation). Read tips on writing your abstract.
3. You can opt to include a **video abstract** with your article. Find out how these can help your work reach a wider audience, and what to think about when filming.
4. 5-8 **keywords**. Read making your article more discoverable, including information on choosing a title and search engine optimization.
5. A feature of this journal is a boxed insert on **Implications for Rehabilitation**. This should include between two to four main bullet points drawing out the implications for rehabilitation for your paper. This should be uploaded as a separate document. Below are examples:
   *Example 1: Leprosy*
   - Leprosy is a disabling disease which not only impacts physically but restricts quality of life often through stigmatisation.
   - Reconstructive surgery is a technique available to this group.
   - In a relatively small sample this study shows participation and social functioning improved after surgery.

*Example 2: Multiple Sclerosis*

   - Exercise is an effective means of improving health and well-being experienced by people with multiple sclerosis (MS).
   - People with MS have complex reasons for choosing to exercise or not.
   - Individual structured programmes are most likely to be successful in encouraging exercise in this cohort.

6. **Acknowledgement**. Please supply all details required by your funding and grant-awarding bodies as follows: *For single agency grants*: This work was supported by the under Grant . *For multiple agency grants*: This work was supported by the under Grant ; under Grant ; and under Grant .
7. **Declaration of Interest**. This is to acknowledge any financial or non-financial interest that has arisen from the direct applications of your research. If there are no relevant competing interests to declare please state this within the article, for example: *The authors report there are no competing interests to declare.* Further guidance on what is a conflict of interest and how to disclose it.
8. **Data availability statement.** If there is a data set associated with the paper, please provide information about where the data supporting the results or analyses presented in the paper can be found. Where applicable, this should include the hyperlink, DOI or other persistent identifier associated with the data set(s). Templates are also available to support authors.
9. **Data deposition.** If you choose to share or make the data underlying the study open, please deposit your data in a recognized data repository prior to or at the time of submission. You will be asked to provide the DOI, pre-reserved DOI, or other persistent identifier for the data set.
10. **Supplemental online material.** Supplemental material can be a video, dataset, fileset, sound file or anything which supports (and is pertinent to) your paper. We publish supplemental material online via Figshare. Find out more about supplemental material and how to submit it with your article.
11. **Figures.** Figures should be high quality (1200 dpi for line art, 600 dpi for grayscale and 300 dpi for colour). Figures should be saved as TIFF, PostScript or EPS files.

12. **Tables.** Tables should present new information rather than duplicating what is in the text. Readers should be able to interpret the table without reference to the text. Please supply editable files.
13. **Equations**. If you are submitting your manuscript as a Word document, please ensure that equations are editable. More information about mathematical symbols and equations.
14. **Units.** Please use SI units (non-italicized).

**Using third-party material in your paper**
You must obtain the necessary permission to reuse third-party material in your article. The use of short extracts of text and some other types of material is usually permitted, on a limited basis, for the purposes of criticism and review without securing formal permission. If you wish to include any material in your paper for which you do not hold copyright, and which is not covered by this informal agreement, you will need to obtain written permission from the copyright owner prior to submission. More information on requesting permission to reproduce work(s) under copyright.

**Declaration of Interest Statement**
Please include a declaration of interest statement, using the subheading "Declaration of interest." If you have no interests to declare, please state this (suggested wording: *The authors report no conflicts of interest*). For all NIH/Wellcome-funded papers, the grant number(s) must be included in the disclosure of interest statement. Read more on declaring conflicts of interest.

**Clinical Trials Registry**
In order to be published in Disability and Rehabilitation , all clinical trials must have been registered in a public repository, ideally at the beginning of the research process (prior to participant recruitment). Trial registration numbers should be included in the abstract, with full details in the methods section. Clinical trials should be registered prospectively – i.e. before participant recruitment. The clinical trial registry should be publicly accessible (at no charge), open to all prospective registrants, and managed by a not-for-profit organization. For a list of registries that meet these requirements, please visit the WHO International Clinical Trials Registry Platform (ICTRP). The registration of all clinical trials facilitates the sharing of information among clinicians, researchers, and patients, enhances public confidence in research, and is in accordance with the ICMJE guidelines.

**Complying with ethics of experimentation**
Please ensure that all research reported in submitted papers has been conducted in an ethical and responsible manner, and is in full compliance with all relevant codes of experimentation and legislation. All papers which report *in vivo* experiments or clinical trials on humans or animals must include a written statement in the Methods section. This should explain that all work was conducted with the formal approval of the local human subject or animal care committees (institutional and national), and that clinical trials have been registered as legislation requires. Authors who do not have formal ethics review committees should include a statement that their study follows the principles of the Declaration of Helsinki. Please ensure that all research reported in submitted papers has been conducted in an ethical and responsible manner, and is in full compliance with all relevant codes of experimentation and legislation. All original research papers involving humans, animals, plants, biological material, protected or non-public datasets, collections or sites, must include a written statement in the Methods section, confirming ethical approval has been obtained from the

appropriate local ethics committee or Institutional Review Board and that where relevant, informed consent has been obtained. For animal studies, approval must have been obtained from the local or institutional animal use and care committee. All research studies on humans (individuals, samples, or data) must have been performed in accordance with the principles stated in the Declaration of Helsinki. In settings where ethics approval for non-interventional studies (e.g. surveys) is not required, authors must include a statement to explain this. In settings where there are no ethics committees in place to provide ethical approval, authors are advised to contact the Editor to discuss further. Detailed guidance on ethics considerations and mandatory declarations can be found in our Editorial Policies section on Research Ethics.

**Consent**

All authors are required to follow the ICMJE requirements and Taylor & Francis Editorial Policies on privacy and informed consent from patients and study participants. Authors must include a statement to confirm that any patient, service user, or participant (or that person's parent or legal guardian) in any type of qualitative or quantitative research, has given informed consent to participate in the research. For submissions where patients or participants can be potentially identified (e.g. a clinical case report detailing their medical history, identifiable images or media content, etc), authors must include a statement to confirm that they have obtained written informed consent to publish the details from the affected individual (or their parents/guardians if the participant in not an adult or unable to give informed consent; or next of kin if the participant is deceased). The process of obtaining consent to publish should include sharing the article with the individual (or whoever is consenting on their behalf), so that they are fully aware of the content of the article before it is published. Authors should familiarise themselves with our policy on participant/patient privacy and informed consent. They may also use the Consent to Publish Form, which can be downloaded from the same Author Services page.

**Health and safety**

Please confirm that all mandatory laboratory health and safety procedures have been complied with in the course of conducting any experimental work reported in your paper. Please ensure your paper contains all appropriate warnings on any hazards that may be involved in carrying out the experiments or procedures you have described, or that may be involved in instructions, materials, or formulae.

Please include all relevant safety precautions; and cite any accepted standard or code of practice. Authors working in animal science may find it useful to consult the International Association of Veterinary Editors' Consensus Author Guidelines on Animal Ethics and Welfare and Guidelines for the Treatment of Animals in Behavioural Research and Teaching. When a product has not yet been approved by an appropriate regulatory body for the use described in your paper, please specify this, or that the product is still investigational.

**Submitting your paper**

This journal uses Taylor & Francis' Submission Portal to manage the submission process. The Submission Portal allows you to see your submissions across Taylor & Francis' journal portfolio in one place. To submit your manuscript please click here.

By submitting your paper to *Disability and Rehabilitation* you are agreeing to originality checks during the peer-review and production processes.

The Editor of *Disability and Rehabilitation* will respond to appeals from authors relating to papers which have been rejected. The author(s) should email the Editor outlining their concerns and making a case for why their paper should not have been rejected. The Editor may choose to accept the appeal and secure a further review, or to not uphold the appeal. In case of the latter, the Editor of *Disability and Rehabilitation: Assistive Technology* will be consulted.

On acceptance, we recommend that you keep a copy of your Accepted Manuscript. Find out more about sharing your work.

**Data Sharing Policy**

This journal applies the Taylor & Francis Basic Data Sharing Policy. Authors are encouraged to share or make open the data supporting the results or analyses presented in their paper where this does not violate the protection of human subjects or other valid privacy or security concerns.

Authors are encouraged to deposit the dataset(s) in a recognized data repository that can mint a persistent digital identifier, preferably a digital object identifier (DOI) and recognizes a long-term preservation plan. If you are uncertain about where to deposit your data, please see this information regarding repositories.

Authors are further encouraged to cite any data sets referenced in the article and provide a Data Availability Statement.

At the point of submission, you will be asked if there is a data set associated with the paper. If you reply yes, you will be asked to provide the DOI, pre-registered DOI, hyperlink, or other persistent identifier associated with the data set(s). If you have selected to provide a pre-registered DOI, please be prepared to share the reviewer URL associated with your data deposit, upon request by reviewers.

Where one or multiple data sets are associated with a manuscript, these are not formally peer reviewed as a part of the journal submission process. It is the author's responsibility to ensure the soundness of data. Any errors in the data rest solely with the producers of the data set(s).

**Publication charges**

There are no submission fees, publication fees or page charges for this journal.

Color figures will be reproduced in color in your online article free of charge.

**Copyright options**

Copyright allows you to protect your original material, and stop others from using your work without your permission. Taylor & Francis offers a number of different license and reuse options, including Creative Commons licenses when publishing open access. Read more on publishing agreements.

**Complying with funding agencies**

We will deposit all National Institutes of Health or Wellcome Trust-funded papers into PubMedCentral on behalf of authors, meeting the requirements of their respective open access (OA) policies. If this applies to you, please tell our production team when you receive your article proofs, so we can do this for you. Check funders' OA policy mandates here. Find out more about sharing your work.

**My Authored Works**

On publication, you will be able to view, download and check your article's metrics (downloads, citations and Altmetric data) via My Authored Works on Taylor & Francis Online. This is where you can access every article you have published with us, as well as your free eprints link, so you can quickly and easily share your work with friends and colleagues.

We are committed to promoting and increasing the visibility of your article. Here are some tips and ideas on how you can work with us to promote your research.

# Section Three Critical Appraisal

Word count (excluding references, tables and appendices): 3744

Derval McCormack

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

**All correspondence should be sent to:**

Derval McCormack
Doctorate in Clinical Psychology
Lancaster University
Furness College
United Kingdom
LA1 4GY

Email: d.mccormack@lancaster.ac.uk

Tel: 01524 59297

# Critical Appraisal

The critical appraisal will begin with a brief summary of the preceding sections, highlighting the novel contributions to multiple sclerosis (MS) and dystonia literature. The appraisal will then reflect on key decisions and challenges encountered throughout various stages of the thesis. In doing so, the strengths and limitations will be considered, along with any professional, ethical, and personal issues experienced. The critical appraisal provides space for further discussion of the main findings and implications for future research.

## Summary of Systematic Literature Review

The systematic literature review examined whether there was a relationship between body image and psychological outcome in MS. The searching process highlighted the limited amount of research in this area, with 13 quantitative studies being included in the final review. For the purpose of this study, the concept of body image encompassed individuals' perception or appraisal of external and internal aspects of their bodies. The psychological outcomes that were examined included depression, anxiety, self-esteem, and quality of life (QoL). The relationship between body image and psychological outcome in MS was explicitly examined in ten of the studies, using correlation or regression analysis. Given the limited amount of research available in this area, the review also included three intervention studies, which examined body image and psychological outcome measures both pre and post a given intervention. The findings from this review suggested that there is a relationship between body image and psychological outcome in MS, identifying that a more positive body image is associated with improved mood, reduced anxiety, increased self-esteem, and greater QoL. However, the causal nature of these associations was unclear, and further research is needed to explore these relationships using more detailed analysis. A longitudinal study may be particularly beneficial in providing insight into cause and effect (Caruana et al., 2015). The review indicated that body image could be a useful target of psychological intervention in MS. Randomised controlled trials investigating the effectiveness of such interventions in improving body image and psychological outcome are recommended.

## Summary of Empirical Paper

The empirical paper used qualitative methods to examine the lived experiences of psychological distress in people with neck dystonia (ND), using interpretative phenomenological analysis (IPA). The purpose of this study was to gain greater insight into people’s experiences and understandings of distress. The study included 11 participants, who were interviewed using a semi-structured schedule. Four overarching themes emerged from the findings: 1) Uncertainty in the lead up to diagnosis; “I was so full of fear”, 2) The isolating nature of the condition; “you don’t know who to turn to”, 3) Mind and body relationship; “is my head turning because I’m thinking about it?”, and 4) A challenging new way of life; “living life sideways”. The encompassing subthemes identified that uncertainty, abandonment, loss, stigma, and isolation were central to participants’ experiences of distress. The majority of individuals believed that psychological distress came from living with the debilitating nature of ND. Some participants offered the tentative belief that psychological distress also developed prior to onset of ND. This study offers a valuable contribution to the literature, providing a person perspective on experiences of psychological distress in ND. The findings highlighted the need for greater psychological input for individuals with this condition, to support a person-centred and holistic approach to care. The most prominent limitation of this study was the difficulty in setting inclusion criteria, given the complexities of ND presentations. Further research should explore lived experiences of psychological distress in other groups within ND that were not captured within the scope of the current project. This may include older populations or individuals presenting with other health conditions. Interestingly, several participants in the present study spoke about experiences of physical or emotional trauma prior to onset of ND. A future study may explore people's belief about aetiology in idiopathic dystonia.

## Reflections on the Systematic Literature Review

### *Selecting a Topic*

The process of selecting a topic for the systematic literature review occurred after the empirical topic had been decided upon, and was one of the most challenging aspects of the review. Previous studies had demonstrated that body image was an important factor in predicting psychological outcome for people with ND (Jahanshahi, 1991; Jahanshahi and Marsden, 1990; Lewis et al., 2008), and I was interested in examining this further. However, my initial scoping searches revealed that there had been very few studies investigating the relationship between body image and psychological outcome in ND, and therefore this search was broadened for feasibility purposes. The Cochrane guidance for conducting systematic reviews (Higgins & Thomas, 2023) was particularly helpful at this stage of the process, as it encouraged me to refer to the FINER criteria (Cummings et al., 2007) when developing my research question. These criteria emphasise the importance of feasibility, and suggest that scoping methods are used to ensure that sufficient evidence exists to answer the research question, without yielding unmanageable amounts of studies. To broaden the search, I considered examining the relationship between body image and psychological distress across other movement disorders, however research in this area was also limited. I noticed during this initial scoping process that I was being pulled toward MS literature, with my interests in MS arising from both professional and personal (close friend) experience of this condition. The FINER criteria highlight the importance of authors being interested in their review topics, to support with commitment and engagement throughout the task (Cummings et al., 2007). Considering these factors, I decided to focus this review on the relationship between body image and psychological outcome in people with MS. This decision was made in collaboration with research supervisors to ensure that there was sufficient knowledge and experience of MS within the research team.

While MS is not a movement disorder, there are similarities between this condition and ND, in the sense that both are chronic neurological illnesses. Both MS and ND are associated with debilitating and challenging symptoms, loss of function, and changes to postural appearance (Ghasemi et al., 2017; Papathanasiou et al., 2001; Williams et al., 2017). There has been previous evidence to suggest the presence of increased psychological and cognitive difficulties in both MS (Bobholz & Gremley, 2011; Boeschoten et al., 2017) and ND (Costanzo et al., 2021; O'Connor et al., 2023). Therefore, I felt that such findings may be relevant to clinical psychologists working within neurological environments, to which people with both MS and ND present.

### ***Search Strategy***

I made the decision to use a broad search strategy for this research, using terms related to MS and body image only, to ensure sensitivity of the search (Lee et al., 2012). I did not want to limit psychological outcome by including specific terms in the search string, and therefore manually searched the returned papers to capture this field. This supported me to exclude articles that were not relevant to the question, thus maintaining a balance between sensitivity and specificity (Lee et al., 2012). The process of manually searching for studies that included a psychological outcome was manageable within the scope of the present review, given the limited amount of research in this area. This may not have been feasible if the initial search string yielded a high number of studies.

## Inconsistent Outcome Measures

A limitation of this review was the heterogeneity across measures of body image in the included studies. The measures examined different dimensions of body image such as body satisfaction, attractiveness, sexual difficulties, body-esteem, body comfort, and interoceptive awareness. Given the limited number of studies that had examined the relationship between body image and psychological outcome, I made the decision to include all measures of body image. However, this did create challenges in terms of meaningfully synthesising the data. The inconsistencies across body image measures suggested the lack of a gold standard when assessing body image in clinical settings. A previous systematic review supported the reliability and validity of most body image measures, however reported that suitability varied across populations (Kling et al., 2019). Their findings were based on the examination of eight body image measures, four of which were used in the present review. However, given the lack of consideration to ecological validity, it is unclear how well these measures reflect a person's daily experiences (Heron et al., 2015). It would be helpful for future research to examine the suitability of body image measures in people with long-term health conditions, such as MS.

### *Data Synthesis*

I decided upon a narrative approach to synthesising the findings from the returned studies. This type of approach is recommended where studies are insufficiently similar, meaning that statistical methods such as meta-analysis are not feasible (Popay et al.,2006). This approach relies on words and storytelling to bring information from studies together, and I initially felt worried about 'doing it right'. Following published guidelines provided me with a sense of structure to ensure that the process remained systematic, despite not being grounded in rigorous statistics. Tabulation was particularly helpful for me during this process, and involved inputting extracted data into tables to allow patterns to be identified (Popay et al., 2006).

### *Personal Reflections on Working with Distinct Topics*

Throughout this thesis, I documented my thoughts and feelings about the process in a reflective journal, with the aim of supporting personal learning and development, and offering transparency for the reader (Ortlipp, 2008). I used this journal to reflect upon the continual challenges of examining two distinct conditions: MS and ND. I was aware that being organised would help me to manage my learning, and minimise feelings of stress, which are common in trainee clinical psychologists (Pakenham & Stafford-Brown, 2012). I found it helpful to set specific reading periods aside for each condition, to allow me to balance my learning of both conditions equally.

Studying these two distinct conditions allowed me to foster my own interests within clinical neuropsychology and health psychology. The literature highlighted the current gaps in psychological input for both ND (Comella & Bhatia, 2015) and MS (Thomas et al., 2006), however it became evident that these gaps differed significantly. For example, a larger body of research currently exists regarding the effectiveness of psychological interventions in MS. Thomas et al. (2006) identified 16 randomised trials, and provided some evidence to suggest the benefits of CBT in supporting with adjustment and coping in MS. A further review (Di Cara et al., 2022) demonstrated the effectiveness of mindfulness in improving psychological outcome in MS. Conversely, just one systematic review has been conducted in dystonia populations, with results being inconsistent due to poor methodologies across studies (Bernstein et al. 2016). I was curious about these discrepancies, and wondered whether less research had been carried out in ND due to it being less common than MS. The global prevalence of ND is 1.8 per 100,000 (LaHue et al., 2020), compared with 35.9 per 100,000 in MS (Walton et al., 2020). This current gap in research evidenced the importance of the present study, and I noticed myself becoming more engaged with the process as a result.

## Reflections on the Empirical Paper

### *Selecting the Focus for Study*

Selecting a topic and method for this research was an important first step, and was carried out in collaboration with my supervisors to ensure feasibility. I had developed an interest in ND through previous research employing quantitative methods (Monaghan et al., 2021), and was keen to conduct further study using a qualitative design. I was particularly drawn to the varying perspectives in the literature regarding the reasons why people with ND experience high levels of psychological distress (Moriarty et al., 2022), and wanted to learn more about the perspectives of people living with the condition.

It was important to choose a method of qualitative analysis that was consistent with the epistemological position of the research. IPA tends to focus on people's experiences and understandings, and therefore was deemed an appropriate fit regarding the research aims (Smith et al., 2022). Supervision supported me to hold this epistemological stance in mind when formulating the research questions, with the assumption that the data would provide insight into people's experiences and sense-making (Smith et al., 2022).

### *Service User Involvement*

I consulted an expert by experience throughout the process, in line with published guidelines for conducting psychological research (Sheldon & Harding, 2010). One of their recommendations was to use accessible language, and so I used the term "neck dystonia" rather than "cervical dystonia" throughout this thesis. I noticed variation in terminology used by participants at interview stage, with some referring to their condition as "neck dystonia", and others as "cervical dystonia". On reflection, including more experts by experience would have allowed me to facilitate a discussion around this. It may also have been helpful to ask participants' for their preferred language, for use in the report.

### *Inclusion Criteria*

Deciding upon inclusion criteria that ensured homogeneity of the sample (Smith et al., 2022) was one of the most challenging aspects of this research. The initial inclusion criteria did not set an upper age limit or exclude those with concurrent health conditions. However, when I started data collection, I recognised that the criteria were not robust enough to ensure a sufficiently homogenous sample, in line with IPA requirements. The first person I spoke with was experiencing age-related health difficulties that were affecting them on a daily basis. There were also significant barriers to conducting the interview due to the participant not speaking English fluently. Supervision provided me with a space to discuss the possible confounding effect that other conditions may have, in terms of their impact on psychological distress. I considered ways to refine the original inclusion criteria, to ensure both homogeneity, and a representative sample (Connelly, 2020). I made the pragmatic decision to exclude individuals over 70, those with significant concurrent health conditions, and individuals who were not fluent in English. Despite this feeling like a sensible research decision, it felt difficult explaining this to individuals who had expressed an interest in participating. The initial response from recruitment had been positive, demonstrating people's willingness to have their voices heard.  I noticed an internal conflict within myself, where I was feeling guilty for excluding individuals from participating, but also having an understanding that such decisions would lead to better quality research. I managed these feelings through use of supervision to discuss research challenges, and by contacting people individually to explain the reasons for changing the inclusion criteria. I noticed these feelings of guilt resurfacing later in the research, when themes of abandonment and isolation emerged as salient experiences within the findings.

The challenges of the inclusion criteria remained throughout data collection, despite the aforementioned amendments. As the research progressed, it was evident that a large number of participants were living with other health conditions. This resulted in complex case-by-case research decisions regarding eligibility, which involved discussions with the wider research team. I interviewed one participant, for example, who was recovering from breast cancer. I needed to make a decision regarding her eligibility to participate based on whether her distress was arising from cancer or dystonia, a decision that was not straightforward. It became evident from listening to the recording of this interview that the participant's distress was the result of living with ND. She spoke of her experiences of breast cancer solely as a way of describing the impact that dystonia was having upon her life. This experience was therefore relevant to the research question, as it powerfully and emotively described the challenges of living with ND. Conversely, I made the difficult decision to exclude other individuals from analysis due to complexities in their presentation suggesting non-focal dystonia or the presence of other conditions. For example, one individual who had received a diagnosis of focal ND reported that symptoms of hand tremor were causing him significant psychological distress. I discussed the atypical nature of this presentation in supervision, and given the importance of homogeneity, made the difficult decision to exclude the participant from this research. Another individual reported symptoms of pain resulting from another neck condition, alongside symptoms for ND. It was felt that this participant may have been experiencing distress differently due to the former symptoms, and therefore the decision was made to exclude him from the study. Despite not meeting the inclusion criteria for this present study, these individuals provided valuable insights into the complexities in ND, and the difficulties in setting research parameters. This was emphasised further by individuals who believed the onset of ND was connected to earlier trauma, highlighting the difficulties in defining idiopathic dystonia, both clinically and in research. Future research is needed to further understand these complexities, and their impact on psychological distress, perhaps using a collective case design (Stake, 2000).

### *Data Collection*

The process of data collection developed my professional interests in working with this population, through direct contact with people who live with ND. It was a privilege to carry out this research, and to have the opportunity to listen to people's stories and experiences first-hand. Due to the nature of this research, the participants spoke about emotive and distressing times in their lives, and this created a personal conflict regarding my dual role as a researcher and a trainee clinical psychologist. At times, I noticed myself wanting to step into the therapist role by providing validation for distress, or offering psychoeducation and reassurance. I reflected on the similarities between qualitative research and therapy, in the sense that both seek to empower the individuals involved (Bourdeau, 2000). As the interviews progressed, I used supervision to consider ways to support empowerment within the context of the current study. This allowed me to develop a safe and validating space that was conducive to rich and detailed data collection.

Participants in this study had the opportunity to be interviewed by video or telephone. There is evidence to suggest the effectiveness of telephone (Drabble et al., 2016) and video interviews (Archibald et al., 2019; Irani, 2019) in qualitative research, in terms of accessibility, convenience and cost-effectiveness. The use of such methods allowed me to reach participants throughout the UK, rather than being limited to a specific geographical region. Despite these advantages, such methods have received criticism in terms of difficulties reading body language or providing comfort if participants become distressed (Irani, 2019). I initially considered using video methods only as it would allow me to observe non-verbal communication, such as facial expressions and body language (Saarijärvi & Bratt, 2021). However, I was aware that not all participants would have access to such technology, or be comfortable using it (Irani, 2019), and so made the decision to offer telephone interviews as an accessible alternative. At times, I experienced some technological delays, which for the most part were easily resolved. However, the recording of one telephone interview did not capture the sound, and therefore was excluded. This was ethically challenging because this individual had given up their time to participate, and I was unable to use their data in the analysis. I approached this issue by being transparent with the individual about what had happened, and we considered how their voice might be heard in dystonia research going forward, as a participant or expert by experience. I confirmed that I would share the results of the study's finding with them upon completion. On reflection, it may have been helpful to offer this person service user involvement in the present study given that their data was no longer usable. Going forward, I have learned the importance of anticipating technological barriers and employing strategies to manage this, for example, testing equipment prior to each interview.

### *Data Analysis*

I approached this thesis as a novice IPA researcher, and therefore was nervous about 'getting it right'. As such, I worked closely with the recommended steps involved in IPA analysis, published by Smith et al. (2022). This helped me to develop my confidence with this methodology, and become more comfortable with the process of analysing and interpreting. At first, I found it challenging to move from descriptive to interpretative, and noticed I was being quite tentative in this process. The guidelines helped me to engage with the experiential significance of what participants were saying, and to focus particularly on the sense they were making of these experiences (Nizza et al., 2021). I initially noticed that I was finding patterns of convergence within the data, however the guidance supported me to look for divergence also, to support a quality IPA study that acknowledged individual idiosyncrasies within this population (Nizza et al., 2021).

There were challenges to analysing data from a relatively large sample by IPA standards, in particular choosing extracts for the narrative. The iterative nature of IPA helped me to manage my selection of data, refine themes, and maintain a continual process of analysis. In line with guidance, my aim was to ensure that the narrative was engaging and persuasive to the reader, supported by carefully selected participant extracts (Nizza et al., 2021). It was challenging selecting appropriate extracts from the transcripts to support the narrative account, while concurrently attempting to give a voice to all participants. In my efforts to create an engaging and compelling narrative, I noticed how I was initially selecting extracts from participants who were particularly articulate. In line with guidance by Smith et al. (2022), the iterative process of writing and rewriting supported me to ensure that extracts from all participants were included. This ensured that the final analysis was representative of the whole sample (Smith, 2011).

## Conclusion

Overall, this thesis includes two distinct papers that offer novel findings, relevant to the field of clinical psychology. This thesis has provided greater understanding of the psychological difficulties experienced by two distinct neurological conditions: MS and ND. The findings are particularly relevant to psychologists working within neuropsychological and health settings. The literature review indicated a relationship between body image and psychological outcome in MS, thus suggesting that clinical interventions that target body image could be useful. Future research examining the effectiveness of such interventions is recommended. The empirical paper demonstrated the psychological difficulties that people with ND experience, and provided an insight into people's beliefs regarding these difficulties. Future research is recommended to address complexities in ND presentations, and to develop psychological interventions to support individuals further.

There were a number of challenges encountered across different stages of the thesis process. Consultation with my supervisory team, service user involvement, and published guidelines supported me when making such decisions. Overall, I feel confident in the decisions that I made during this process. I believe I have developed my skills as a researcher, and look forward to carrying out further studies.

# References

Archibald, M. M., Ambagtsheer, R. C., Casey, M. G., & Lawless, M. (2019). Using zoom videoconferencing for qualitative data collection: perceptions and experiences of researchers and participants. *International journal of qualitative methods*, *18*, 1609406919874596. https://doi.org/10.1177/160940691987459

Bernstein, C. J., Ellard, D. R., Davies, G., Hertenstein, E., Tang, N. K., Underwood, M., Sandhu, H. (2016). Behavioural interventions for people living with adult-onset primary dystonia: a systematic review. *BMC neurology*, *16*, 1-14. https://doi.org/10.1186/s12883-016-0562-y

Bobholz, J. A., & Gremley, S. (2011). Multiple Sclerosis and Other Demyelinating Disorders. In *The Little Black Book of Neuropsychology. A Syndrome Based Approach* (pp. 647–662). Springer. https://doi.org/10.1007/978-0-387-76978-3

Boeschoten, R. E., Braamse, A. M. J., Beekman, A. T. F., Cuijpers, P., van Oppen, P., Dekker, J., & Uitdehaag, B. M. J. (2017). Prevalence of depression and anxiety in Multiple Sclerosis: A systematic review and meta-analysis. *Journal of the Neurological Sciences*, *372*, 331–341. https://doi.org/10.1016/j.jns.2016.11.067

Bourdeau, B. (2000). Dual relationships in qualitative research. *The Qualitative Report*, *4*(3), 1-6. https://doi.org/10.46743/2160-3715/2000.2081

Caruana, E. J., Roman, M., Hernández-Sánchez, J., & Solli, P. (2015). Longitudinal studies. *Journal of thoracic disease*, *7*(11), E537. 10.3978/j.issn.2072-1439.2015.10.63 https://doi.org/10.3978/j.issn.2072-1439.2015.10.63

Comella, C., & Bhatia, K. (2015). An international survey of patients with cervical dystonia. *Journal of neurology*, *262*, 837-848. https://doi.org/10.1007/s00415-014-7586-2

Connelly, L. M. (2020). Inclusion and Exclusion Criteria. *Medsurg nursing*, *29*(2).

Costanzo, M., Belvisi, D., Berardelli, I., Maraone, A., D'Antonio, F., Baione, V., ... & Berardelli, A. (2021). Motor and non-motor subtypes of cervical dystonia. *Parkinsonism & Related Disorders*, *88*, 108-113. https://doi.org/10.1016/j.parkreldis.2021.06.008

Cummings, S., Browner, W., & Hulley, S. (2007). Conceiving the research question and developing the study plan. In S. Hulley, S. Cummings, & W. Browner (Eds.), *Designing Clinical Research: An Epidemiological Approach* (pp. 14–22). Lippincott Williams & Wilkins.

Di Cara, M., Grezzo, D., Palmeri, R., Lo Buono, V., Cartella, E., Micchia., K., …& Corallo, F. (2022). Psychological well-being in people with multiple sclerosis: a descriptive review of the effects obtained with mindfulness interventions. *Neurological Sciences*, 1-7. https://doi.org/10.1007/s10072-021-05686-1

Drabble, L., Trocki, K. F., Salcedo, B., Walker, P. C., & Korcha, R. A. (2016). Conducting qualitative interviews by telephone: Lessons learned from a study of alcohol use among sexual minority and heterosexual women. *Qualitative Social Work*, *15*(1), 118-133. https://doi.org/10.1177/147332501558561

Ghasemi, N., Razavi, S., & Nikzad, E. (2017). Multiple sclerosis: pathogenesis, symptoms, diagnoses and cell-based therapy. *Cell Journal (Yakhteh)*, *19*(1), 1. https://doi.org/ 10.22074/cellj.2016.4867

Heron, K. E., Mason, T. B., Sutton, T. G., & Myers, T. A. (2015). Evaluating the real-world predictive validity of the body image quality of life inventory using ecological momentary assessment. *Body Image*, *15*, 105-108. https://doi.org/10.1016/j.bodyim.2015.07.004

Higgins, J., & Thomas, J. (2023). *Cochrane Handbook For Systematic Reviews Of Interventions.* (2nd ed.). Wiley-Blackwell.

Irani, E. (2019). The use of videoconferencing for qualitative interviewing: Opportunities, challenges, and considerations. *Clinical Nursing Research*, *28*(1), 3-8. https://doi.org/10.1177/105477381880317

Jahanshahi, M. (1991). Psychosocial factors and depression in torticollis. *Journal of psychosomatic research*, *35*(4-5), 493-507. https://doi.org/10.1016/0022-3999(91)90044-O

Jahanshahi, M., & Marsden, C. D. (1990). Body concept, disability, and depression in patients with spasmodic torticollis. *Behavioural neurology*, *3*(2), 117-131. https://doi.org/10.3233/BEN-1990-3206

Kling, J., Kwakkenbos, L., Diedrichs, P. C., Rumsey, N., Frisen, A., Brandao, M. P., ... & Fitzgerald, A. (2019). Systematic review of body image measures. *Body Image*, *30*, 170-211. https://doi.org/10.1016/j.bodyim.2019.06.006

LaHue, S. C., Albers, K., Goldman, S., Lo, R. Y., Gu, Z., Leimpeter, A., ... & Tanner, C. M. (2020). Cervical dystonia incidence and diagnostic delay in a multiethnic population. *Movement Disorders*, *35*(3), 450-456.https://doi.org/10.1002/mds.27927

Lee, E., Dobbins, M., DeCorby, K., McRae, L., Tirilis, D., & Husson, H. (2012). An optimal search filter for retrieving systematic reviews and meta-analyses. *BMC medical research methodology*, *12*, 1-11. https://doi.org/10.1186/1471-2288-12-51

Lewis, L., Butler, A., & Jahanshahi, M. (2008). Depression in focal, segmental and generalized dystonia. *Journal of neurology*, *255*(11), 1750-1755. doi: 10.1007/s00415-008-0020-x https://doi.org/10.1007/s00415-008-0020-x

Monaghan, R., Cogley, C., Burke, T., McCormack, D., O'Riordan, S., Ndukwe, I., ... & O'Keeffe, F. (2021). Non-motor features of cervical dystonia: Cognition, social cognition, psychological distress and quality of life. *Clinical Parkinsonism & Related Disorders*, *4*, 100084. https://doi.org/10.1016/j.prdoa.2020.100084

Moriarty, A., Rafee, S., Ndukwe, I., O'Riordan, S., & Hutchinson, M. (2022). Longitudinal follow-up of mood in cervical dystonia and influence on age at onset. *Movement Disorders Clinical Practice*, *9*(5), 614-618. https://doi.org/10.1002/mdc3.13457

Nizza, I. E., Farr, J., & Smith, J. A. (2021). Achieving excellence in interpretative phenomenological analysis (IPA): Four markers of high quality. *Qualitative Research in Psychology*, *18*(3), 369-386. https://doi.org/10.1080/14780887.2020.1854404

O'Connor, S., Hevey, D., Burke, T., Rafee, S., Pender, N., & O'Keeffe, F. (2023). A Systematic Review of Cognition in Cervical Dystonia. *Neuropsychology Review*, 1-21. https://doi.org/10.1007/s11065-022-09558-z

Ortlipp, M. (2008). Keeping and using reflective journals in the qualitative research process. *The qualitative report*, *13*(4), 695-705. https://doi.org/10.46743/2160-3715/2008.1579

Pakenham, K. I., & Stafford-Brown, J. (2012). Stress in clinical psychology trainees: Current research status and future directions. *Australian Psychologist*, *47*(3), 147-155. https://doi.org/10.1111/j.1742-9544.2012.00070.x

Papathanasiou, I., MacDonald, L., Whurr, R., & Jahanshahi, M. (2001). Perceived stigma in Spasmoic Torticollis. *Movement Disorders: Official Journal of the Movement Disorder Society*, *16*(2), 280-285. https://doi.org/10.1002/mds.1055

Popay, J., Roberts, H. M., Sowden, A., Petticrew, M., Arai, L., Rodgers, M., & Britten, N. (2006). Guidance on the conduct of narrative synthesis in systematic reviews.

Saarijärvi, M., & Bratt, E. L. (2021). When face-to-face interviews are not possible: tips and tricks for video, telephone, online chat, and email interviews in qualitative research. https://doi.org/10.1093/eurjcn/zvab038

Sheldon, K., & Harding, E. (2010). Good practice guidelines to support the involvement of service users and carers in clinical psychology services. British Psychological Society.

Smith, J. A. (2011). Evaluating the contribution of interpretative phenomenological analysis. *Health psychology review*, *5*(1), 9-27. https://doi.org/10.1080/17437199.2010.510659

Smith, J. A., Flowers, P., & Larkin, M. (2022). *Interpretative phenomenological analysis: Theory, method and research* (2nd ed.). Sage Publications.

Stake, R. E. Case studies (2000) Handbook of Qualitative Research. *NK Denzin and YS Lincoln (Eds.)*, 435-454.

Thomas, P. W., Thomas, S., Hillier, C., Galvin, K., & Baker, R. (2006). Psychological interventions for multiple sclerosis. *Cochrane database of systematic reviews*, (1). https://doi.org/10.1002/14651858.CD004431.pub2

Walton, C., King, R., Rechtman, L., Kaye, W., Leray, E., Marrie, R. A., Robertson, N., La Rocca, N., Uitdehaag, B., van der Mei, I., Wallin, M., Helme, A., Angood Napier, C., Rijke, N., & Baneke, P. (2020). Rising prevalence of multiple sclerosis worldwide: Insights from the Atlas of MS, third edition. *Multiple Sclerosis Journal*, *26*(14), 1816–1821. https://doi.org/10.1177/1352458520970841

Williams, L., McGovern, E., Kimmich, O., Molloy, A., Beiser, I., Butler, J. S., ... & Hutchinson, M. (2017). Epidemiological, clinical and genetic aspects of adult onset isolated focal dystonia in Ireland. *European journal of neurology*, *24*(1), 73-81. https://doi.org/10.1111/ene.13133

# Section Four: Ethics Section

Derval McCormack

Doctorate in Clinical Psychology

Division of Health Research, Lancaster University

**All correspondence should be sent to:**

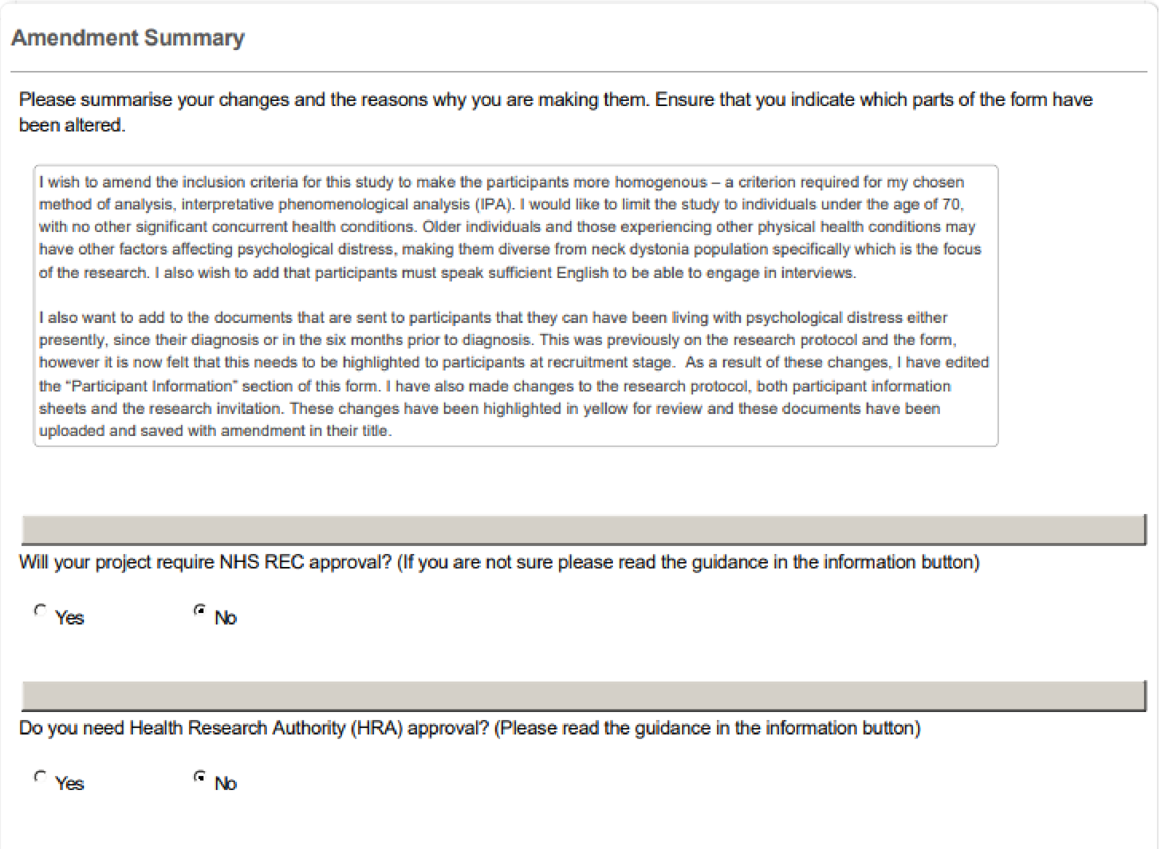
Derval McCormack
Doctorate in Clinical Psychology
Lancaster University
Furness College
United Kingdom
LA1 4GY

Email: d.mccormack@lancaster.ac.uk

Tel: 01524 59297

## 1. Approved Application to Ethics

## Lived experiences of psychological distress in neck dystonia



**Amendment Summary**

Please summarise your changes and the reasons why you are making them. Ensure that you indicate which parts of the form have been altered.

I wish to amend the inclusion criteria for this study to make the participants more homogenous – a criterion required for my chosen method of analysis, interpretative phenomenological analysis (IPA). I would like to limit the study to individuals under the age of 70, with no other significant concurrent health conditions. Older individuals and those experiencing other physical health conditions may have other factors affecting psychological distress, making them diverse from neck dystonia population specifically which is the focus of the research. I also wish to add that participants must speak sufficient English to be able to engage in interviews.

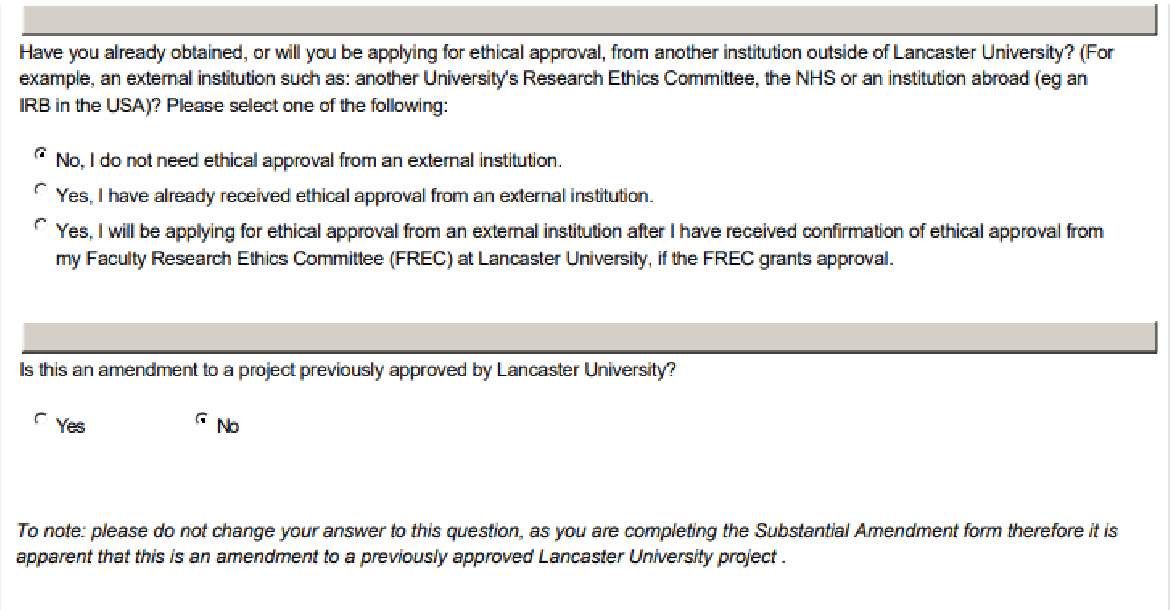
I also want to add to the documents that are sent to participants that they can have been living with psychological distress either presently, since their diagnosis or in the six months prior to diagnosis. This was previously on the research protocol and the form, however it is now felt that this needs to be highlighted to participants at recruitment stage. As a result of these changes, I have edited the "Participant Information" section of this form. I have also made changes to the research protocol, both participant information sheets and the research invitation. These changes have been highlighted in yellow for review and these documents have been uploaded and saved with amendment in their title.

Will your project require NHS REC approval? (If you are not sure please read the guidance in the information button)

○ Yes ● No

Do you need Health Research Authority (HRA) approval? (Please read the guidance in the information button)

○ Yes ● No



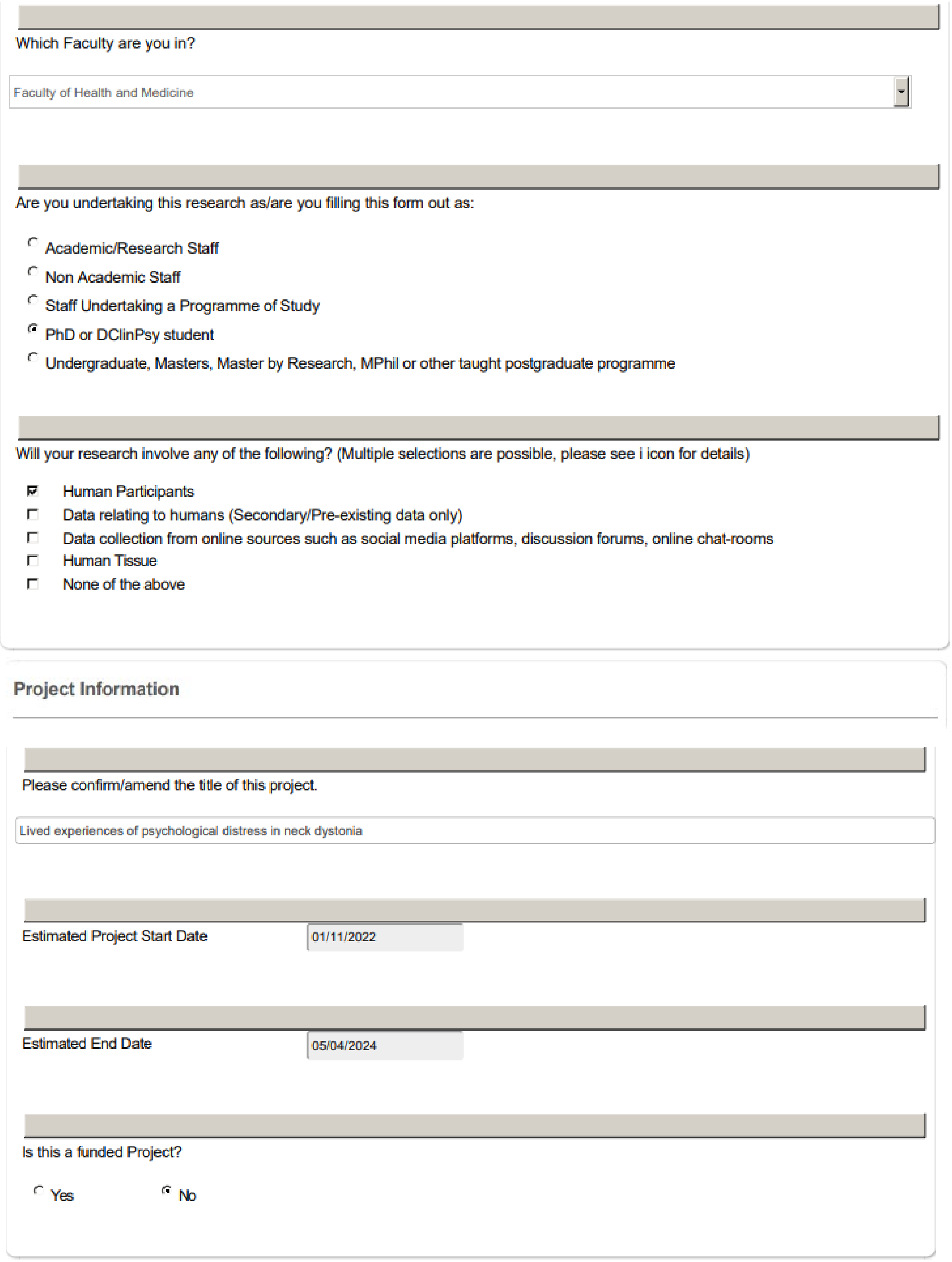
Have you already obtained, or will you be applying for ethical approval, from another institution outside of Lancaster University? (For example, an external institution such as: another University's Research Ethics Committee, the NHS or an institution abroad (eg an IRB in the USA)? Please select one of the following:

● No, I do not need ethical approval from an external institution.
○ Yes, I have already received ethical approval from an external institution.
○ Yes, I will be applying for ethical approval from an external institution after I have received confirmation of ethical approval from my Faculty Research Ethics Committee (FREC) at Lancaster University, if the FREC grants approval.

Is this an amendment to a project previously approved by Lancaster University?

○ Yes ● No

*To note: please do not change your answer to this question, as you are completing the Substantial Amendment form therefore it is apparent that this is an amendment to a previously approved Lancaster University project .*



Which Faculty are you in?

Faculty of Health and Medicine

Are you undertaking this research as/are you filling this form out as:

○ Academic/Research Staff
○ Non Academic Staff
○ Staff Undertaking a Programme of Study
● PhD or DClinPsy student
○ Undergraduate, Masters, Master by Research, MPhil or other taught postgraduate programme

Will your research involve any of the following? (Multiple selections are possible, please see i icon for details)

☑ Human Participants
☐ Data relating to humans (Secondary/Pre-existing data only)
☐ Data collection from online sources such as social media platforms, discussion forums, online chat-rooms
☐ Human Tissue
☐ None of the above

## Project Information

Please confirm/amend the title of this project.

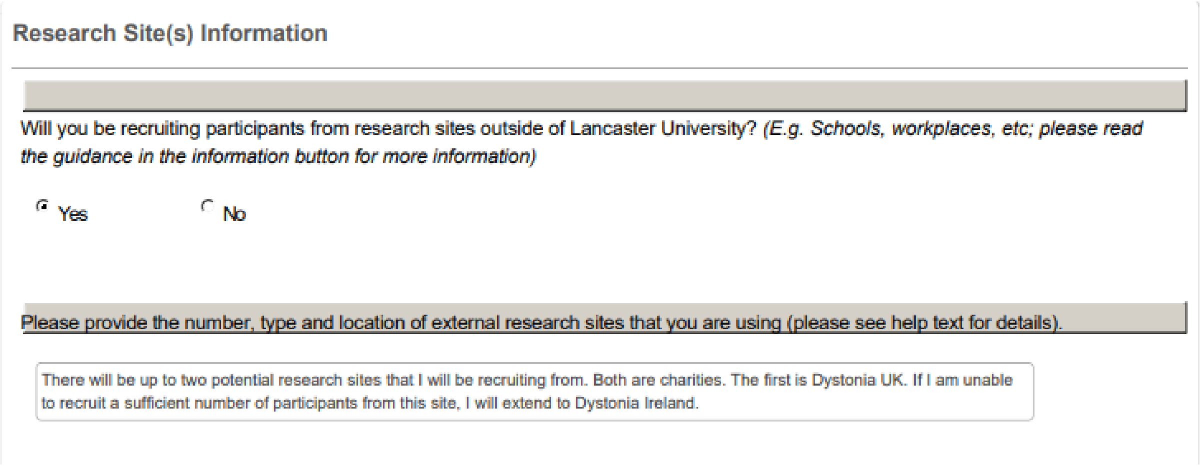
Lived experiences of psychological distress in neck dystonia

Estimated Project Start Date 01/11/2022

Estimated End Date 05/04/2024

Is this a funded Project?

○ Yes ● No



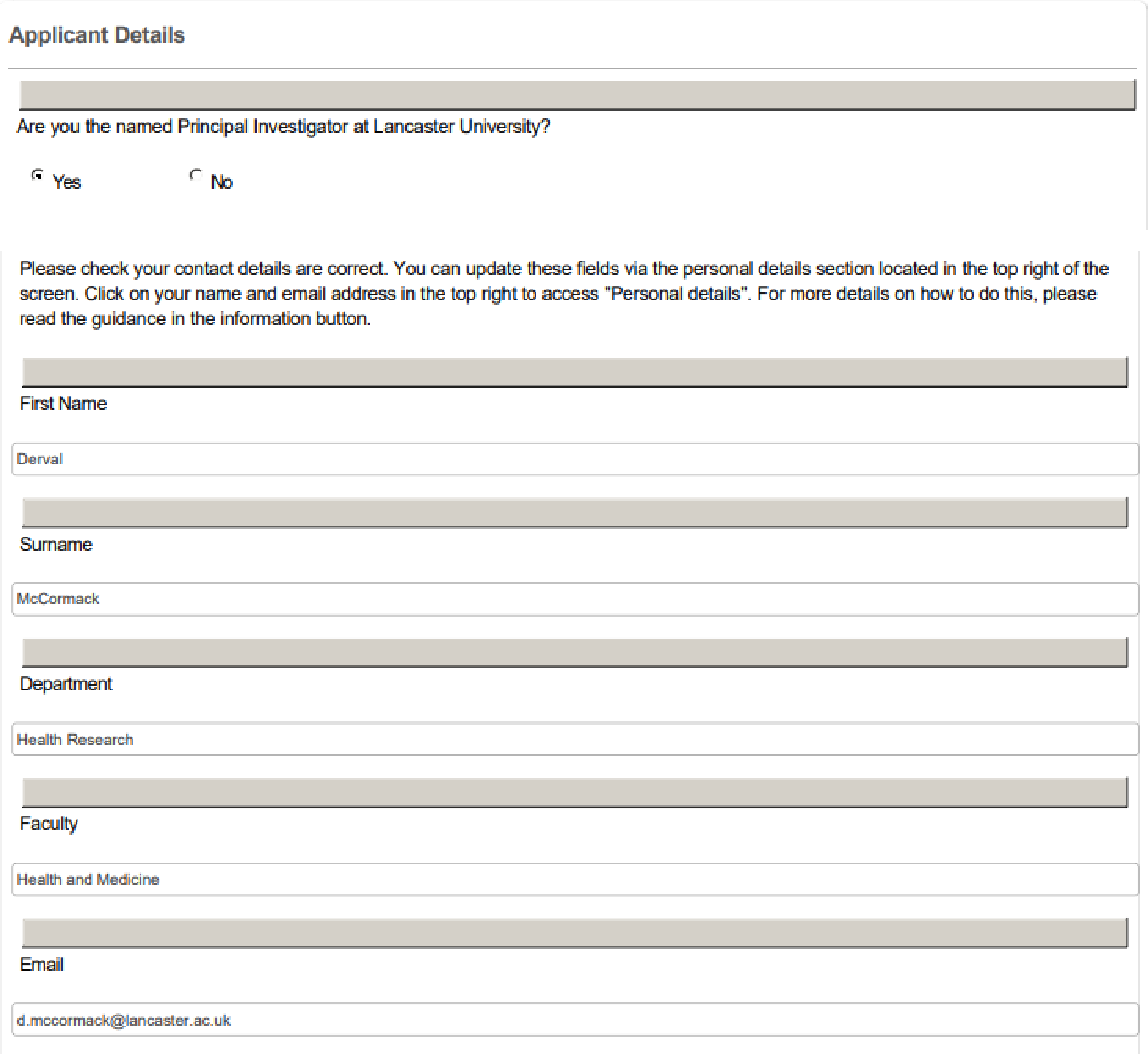
### Research Site(s) Information

Will you be recruiting participants from research sites outside of Lancaster University? *(E.g. Schools, workplaces, etc; please read the guidance in the information button for more information)*

● Yes ○ No

Please provide the number, type and location of external research sites that you are using (please see help text for details).

There will be up to two potential research sites that I will be recruiting from. Both are charities. The first is Dystonia UK. If I am unable to recruit a sufficient number of participants from this site, I will extend to Dystonia Ireland.



### Applicant Details

Are you the named Principal Investigator at Lancaster University?

● Yes ○ No

Please check your contact details are correct. You can update these fields via the personal details section located in the top right of the screen. Click on your name and email address in the top right to access "Personal details". For more details on how to do this, please read the guidance in the information button.

First Name

Derval

Surname

McCormack

Department

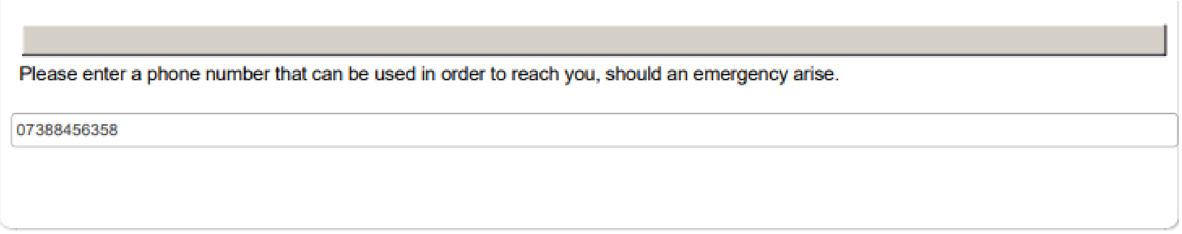
Health Research

Faculty

Health and Medicine

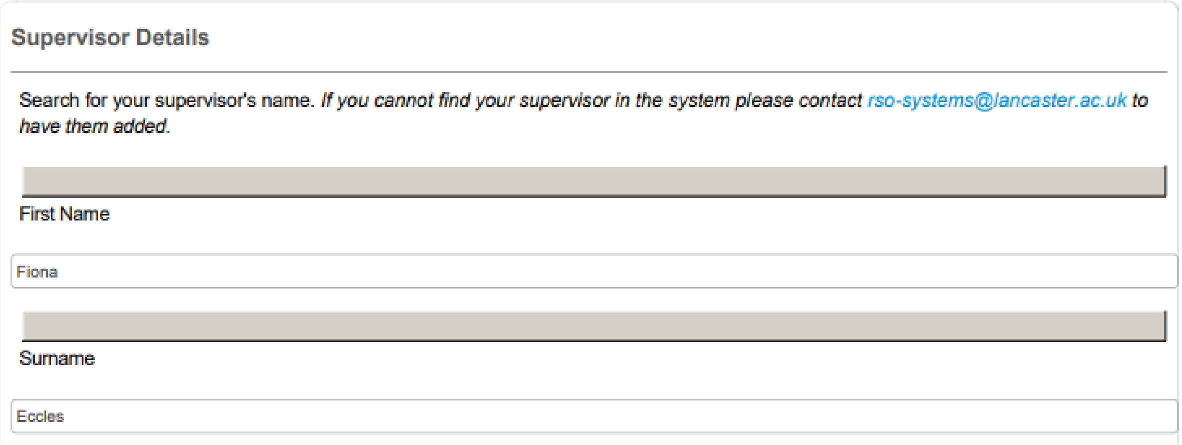
Email

d.mccormack@lancaster.ac.uk



Please enter a phone number that can be used in order to reach you, should an emergency arise.

07388456358



## Supervisor Details

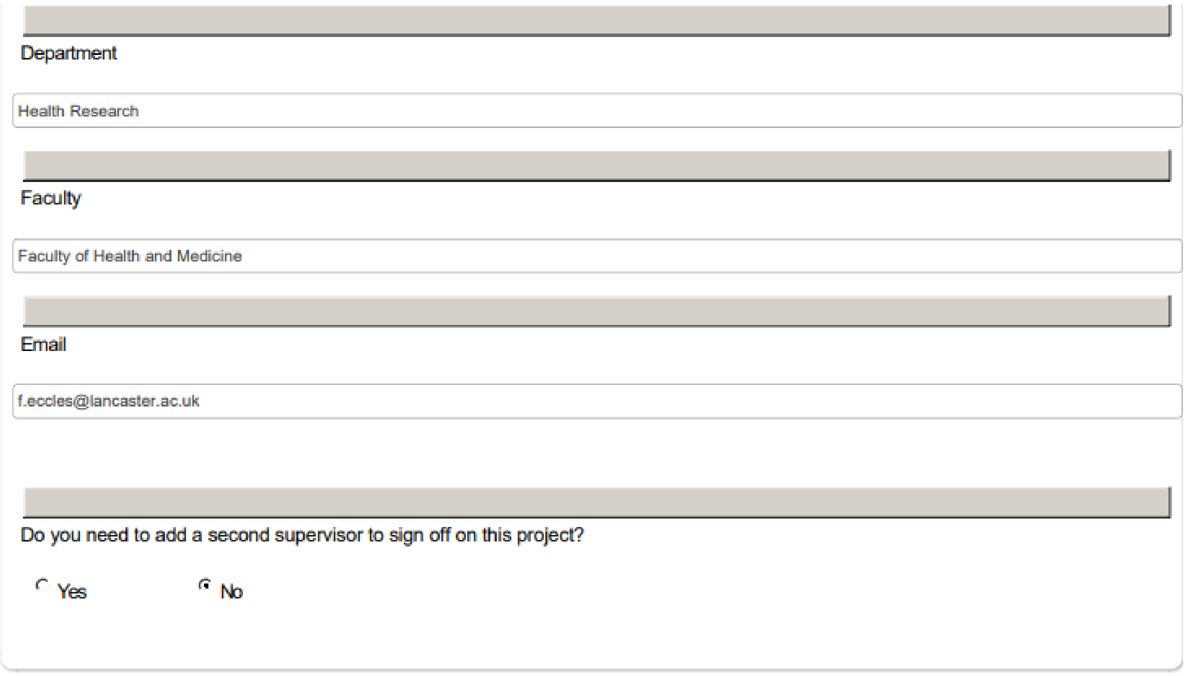
Search for your supervisor's name. *If you cannot find your supervisor in the system please contact* *rso-systems@lancaster.ac.uk* *to have them added.*

First Name

Fiona

Surname

Eccles



Department

Health Research

Faculty

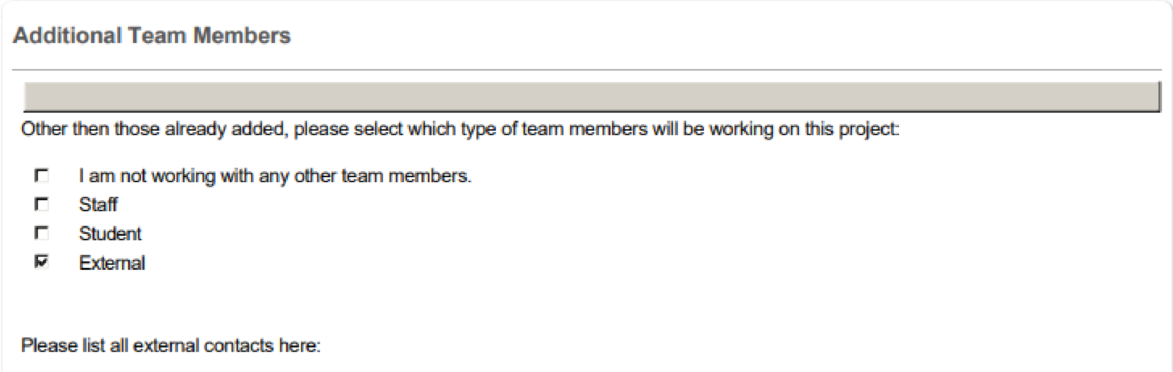
Faculty of Health and Medicine

Email

f.eccles@lancaster.ac.uk

Do you need to add a second supervisor to sign off on this project?

○ Yes ● No

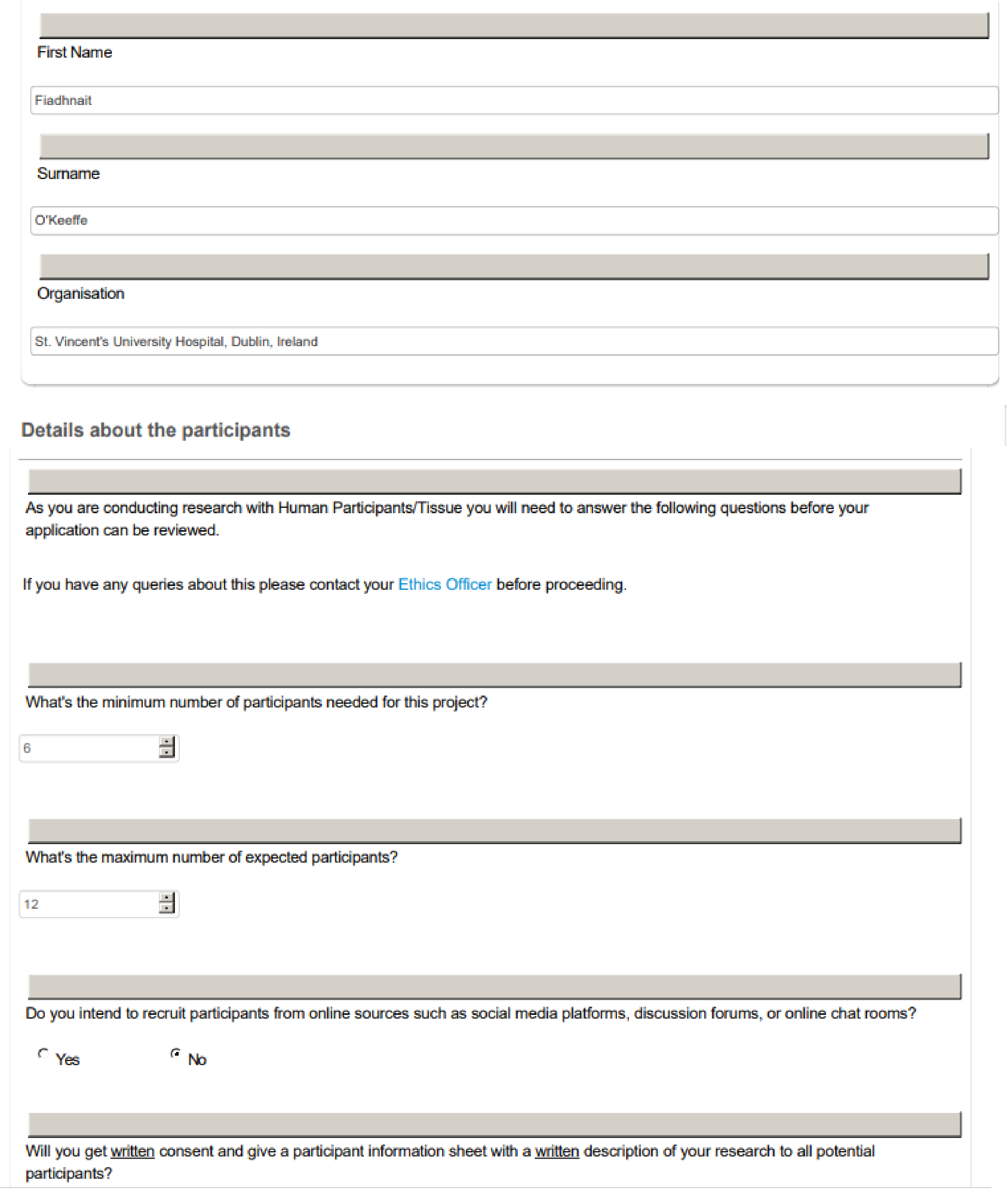


## Additional Team Members

Other then those already added, please select which type of team members will be working on this project:

☐ I am not working with any other team members.
☐ Staff
☐ Student
☑ External

Please list all external contacts here:



First Name

Fiadhnait

Surname

O'Keeffe

Organisation

St. Vincent's University Hospital, Dublin, Ireland

**Details about the participants**

As you are conducting research with Human Participants/Tissue you will need to answer the following questions before your application can be reviewed.

If you have any queries about this please contact your Ethics Officer before proceeding.

What's the minimum number of participants needed for this project?

6

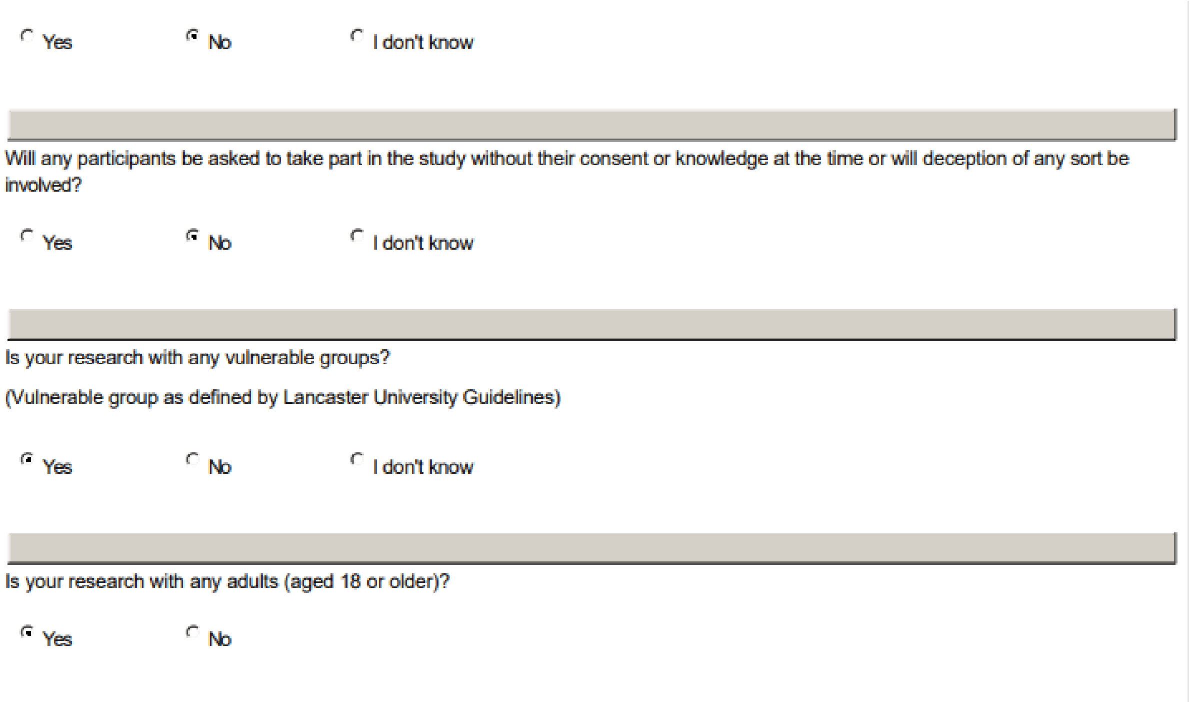
What's the maximum number of expected participants?

12

Do you intend to recruit participants from online sources such as social media platforms, discussion forums, or online chat rooms?

○ Yes ● No

Will you get written consent and give a participant information sheet with a written description of your research to all potential participants?



○ Yes ● No ○ I don't know

Will any participants be asked to take part in the study without their consent or knowledge at the time or will deception of any sort be involved?

○ Yes ● No ○ I don't know

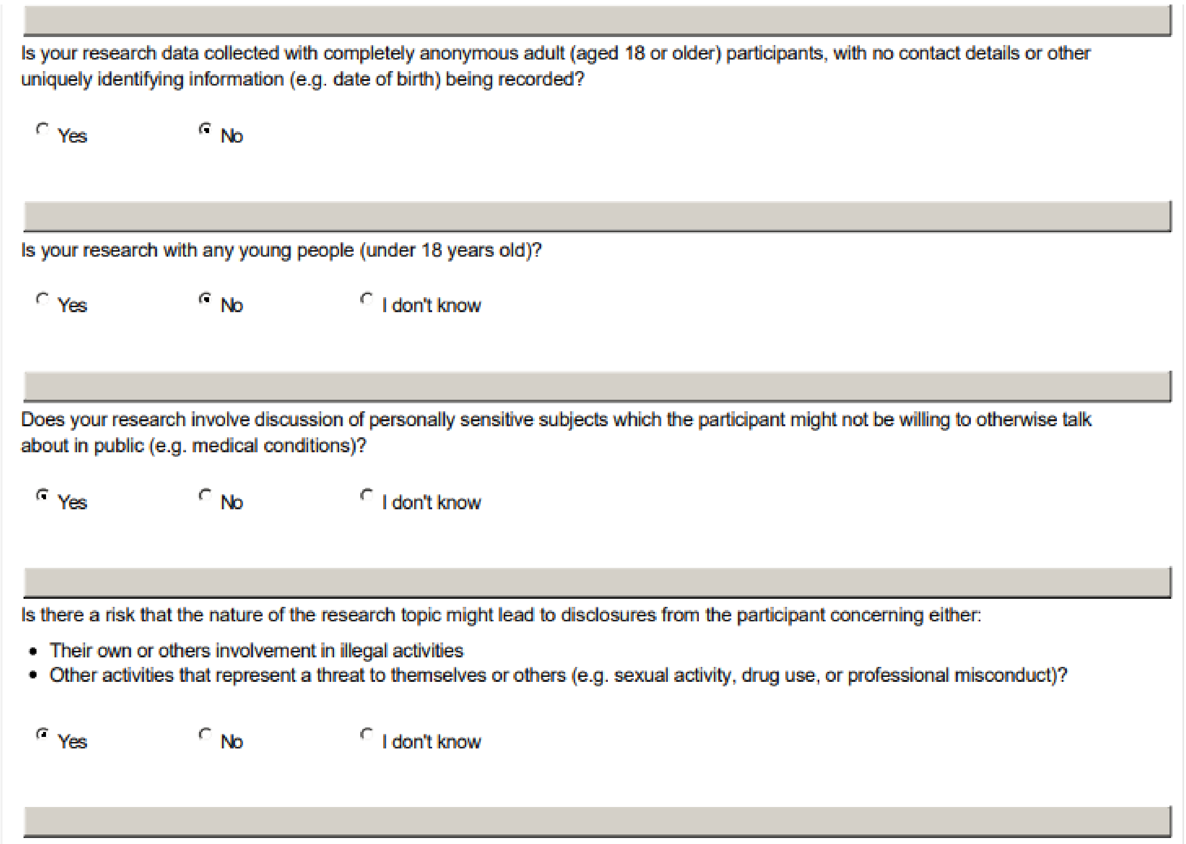
Is your research with any vulnerable groups?

(Vulnerable group as defined by Lancaster University Guidelines)

● Yes ○ No ○ I don't know

Is your research with any adults (aged 18 or older)?

● Yes ○ No



Is your research data collected with completely anonymous adult (aged 18 or older) participants, with no contact details or other uniquely identifying information (e.g. date of birth) being recorded?

○ Yes ● No

Is your research with any young people (under 18 years old)?

○ Yes ● No ○ I don't know

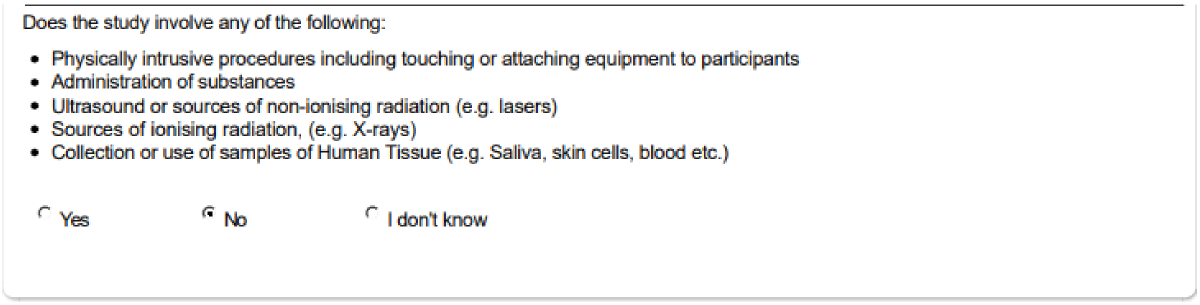
Does your research involve discussion of personally sensitive subjects which the participant might not be willing to otherwise talk about in public (e.g. medical conditions)?

● Yes ○ No ○ I don't know

Is there a risk that the nature of the research topic might lead to disclosures from the participant concerning either:

- Their own or others involvement in illegal activities
- Other activities that represent a threat to themselves or others (e.g. sexual activity, drug use, or professional misconduct)?

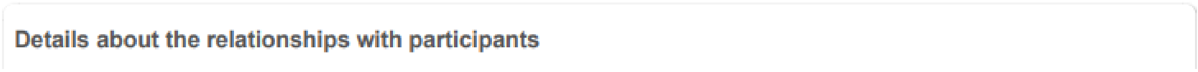
● Yes ○ No ○ I don't know



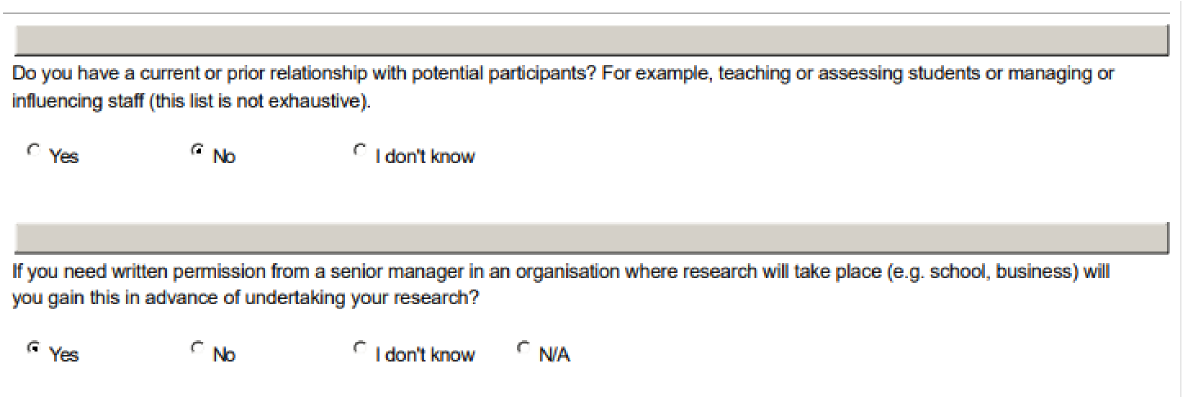
Does the study involve any of the following:

- Physically intrusive procedures including touching or attaching equipment to participants
- Administration of substances
- Ultrasound or sources of non-ionising radiation (e.g. lasers)
- Sources of ionising radiation, (e.g. X-rays)
- Collection or use of samples of Human Tissue (e.g. Saliva, skin cells, blood etc.)

○ Yes ● No ○ I don't know



**Details about the relationships with participants**

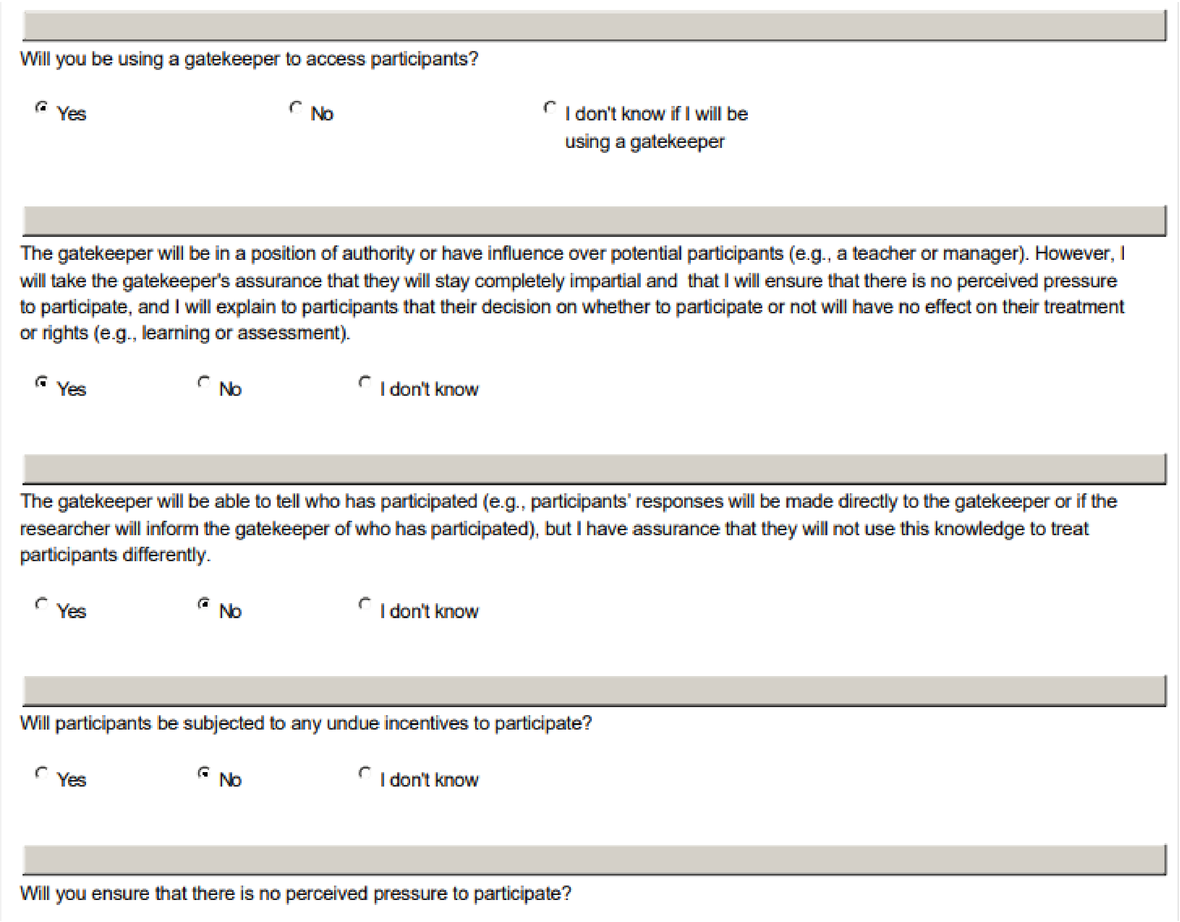


Do you have a current or prior relationship with potential participants? For example, teaching or assessing students or managing or influencing staff (this list is not exhaustive).

○ Yes ● No ○ I don't know

If you need written permission from a senior manager in an organisation where research will take place (e.g. school, business) will you gain this in advance of undertaking your research?

● Yes ○ No ○ I don't know ○ N/A



Will you be using a gatekeeper to access participants?

● Yes ○ No ○ I don't know if I will be using a gatekeeper

The gatekeeper will be in a position of authority or have influence over potential participants (e.g., a teacher or manager). However, I will take the gatekeeper's assurance that they will stay completely impartial and that I will ensure that there is no perceived pressure to participate, and I will explain to participants that their decision on whether to participate or not will have no effect on their treatment or rights (e.g., learning or assessment).

● Yes ○ No ○ I don't know

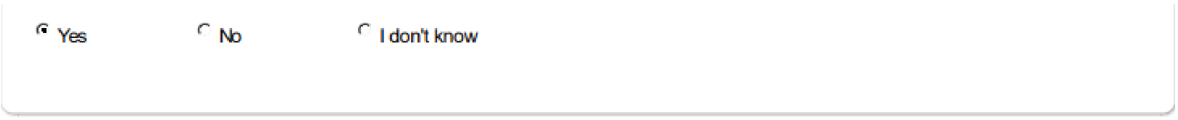
The gatekeeper will be able to tell who has participated (e.g., participants' responses will be made directly to the gatekeeper or if the researcher will inform the gatekeeper of who has participated), but I have assurance that they will not use this knowledge to treat participants differently.

○ Yes ● No ○ I don't know

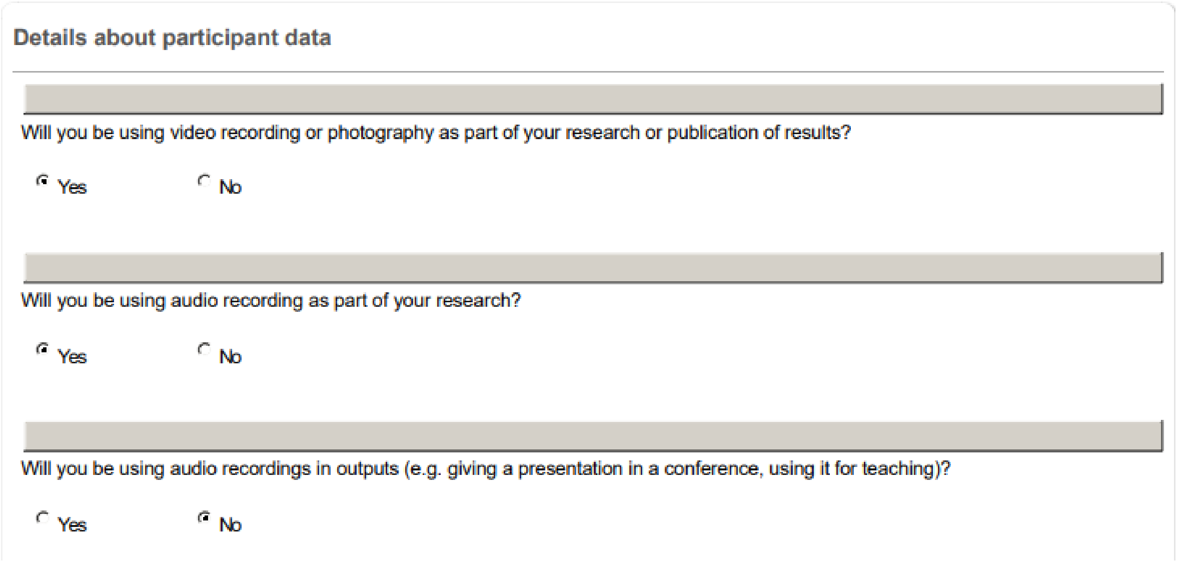
Will participants be subjected to any undue incentives to participate?

○ Yes ● No ○ I don't know

Will you ensure that there is no perceived pressure to participate?



● Yes ○ No ○ I don't know



**Details about participant data**

Will you be using video recording or photography as part of your research or publication of results?

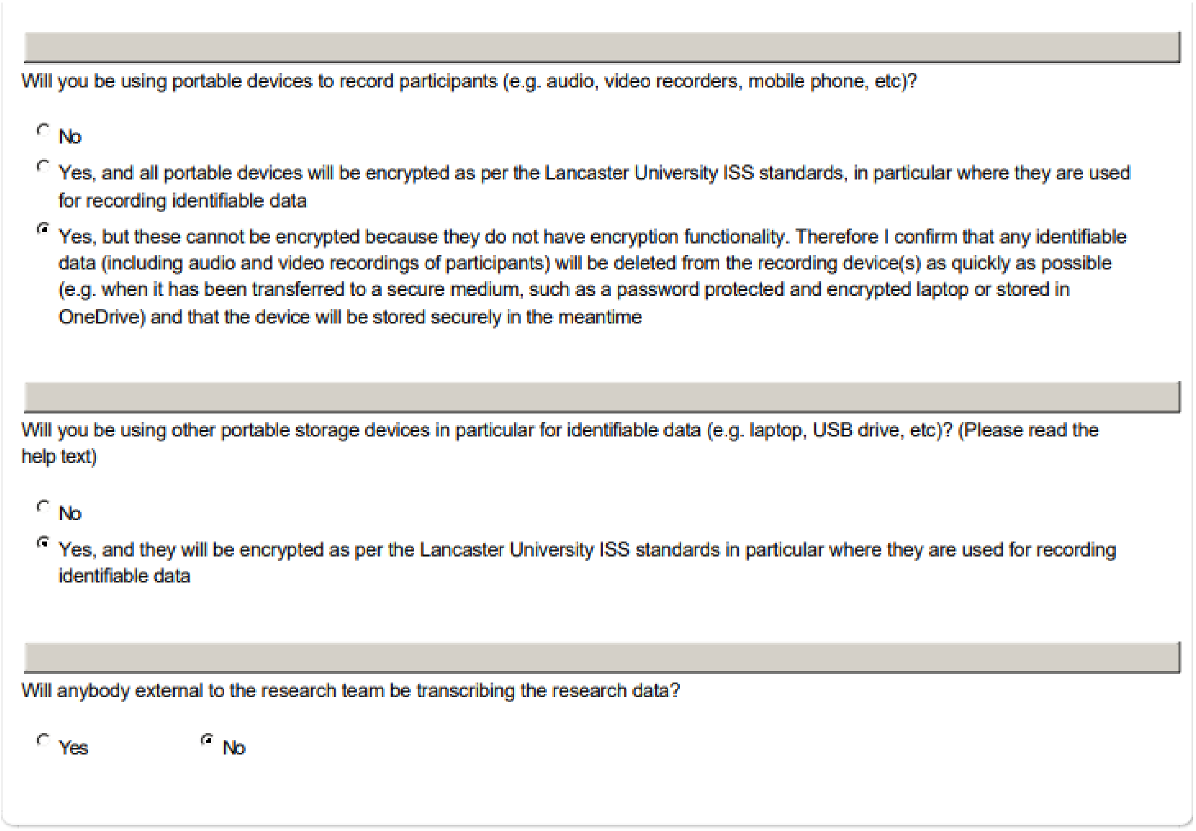
● Yes ○ No

Will you be using audio recording as part of your research?

● Yes ○ No

Will you be using audio recordings in outputs (e.g. giving a presentation in a conference, using it for teaching)?

○ Yes ● No



Will you be using portable devices to record participants (e.g. audio, video recorders, mobile phone, etc)?

○ No

○ Yes, and all portable devices will be encrypted as per the Lancaster University ISS standards, in particular where they are used for recording identifiable data

● Yes, but these cannot be encrypted because they do not have encryption functionality. Therefore I confirm that any identifiable data (including audio and video recordings of participants) will be deleted from the recording device(s) as quickly as possible (e.g. when it has been transferred to a secure medium, such as a password protected and encrypted laptop or stored in OneDrive) and that the device will be stored securely in the meantime

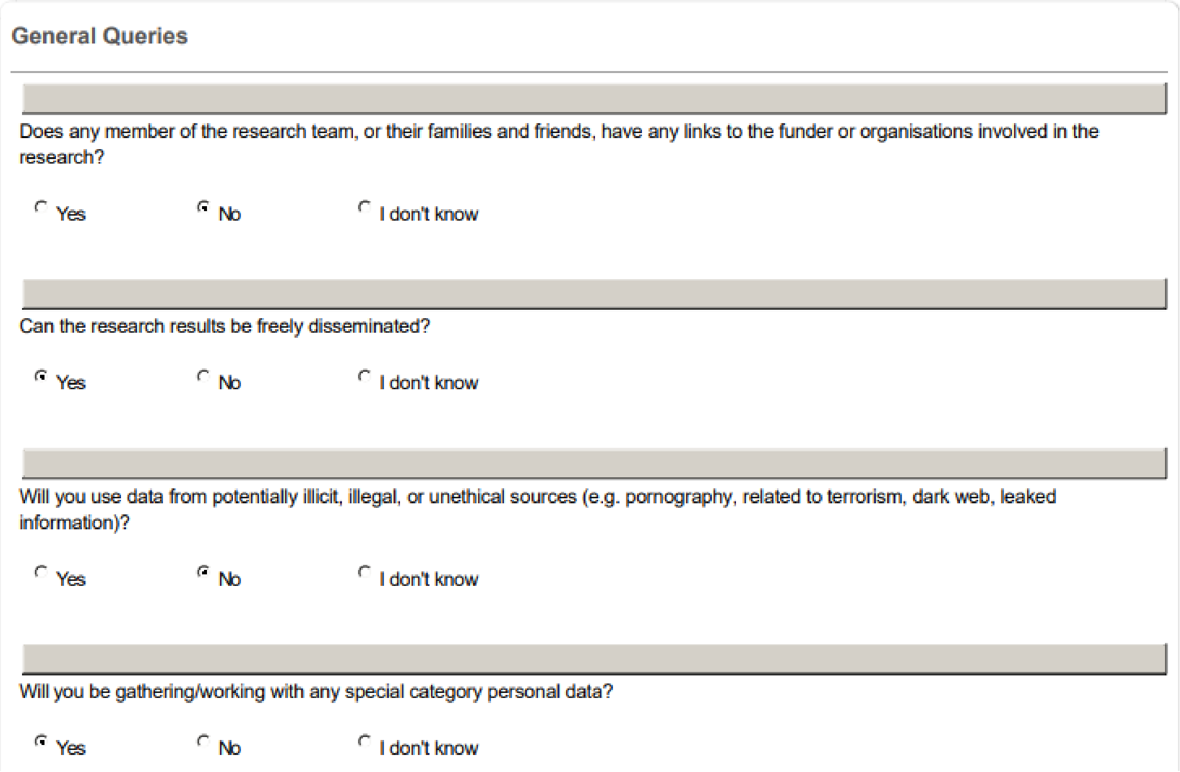
Will you be using other portable storage devices in particular for identifiable data (e.g. laptop, USB drive, etc)? (Please read the help text)

○ No

● Yes, and they will be encrypted as per the Lancaster University ISS standards in particular where they are used for recording identifiable data

Will anybody external to the research team be transcribing the research data?

○ Yes ● No



**General Queries**

Does any member of the research team, or their families and friends, have any links to the funder or organisations involved in the research?

○ Yes ● No ○ I don't know

Can the research results be freely disseminated?

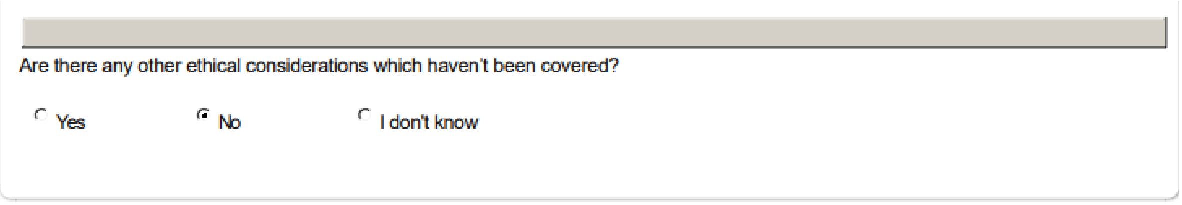
● Yes ○ No ○ I don't know

Will you use data from potentially illicit, illegal, or unethical sources (e.g. pornography, related to terrorism, dark web, leaked information)?

○ Yes ● No ○ I don't know

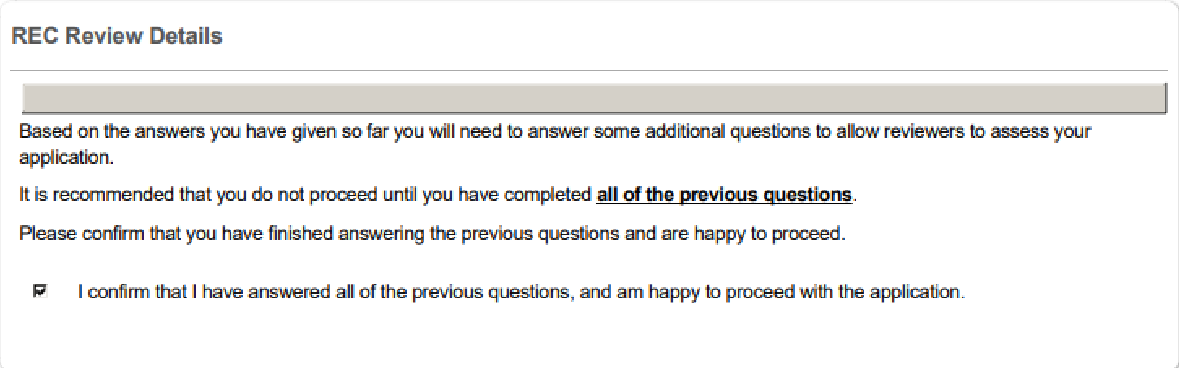
Will you be gathering/working with any special category personal data?

● Yes ○ No ○ I don't know



Are there any other ethical considerations which haven't been covered?

○ Yes ● No ○ I don't know



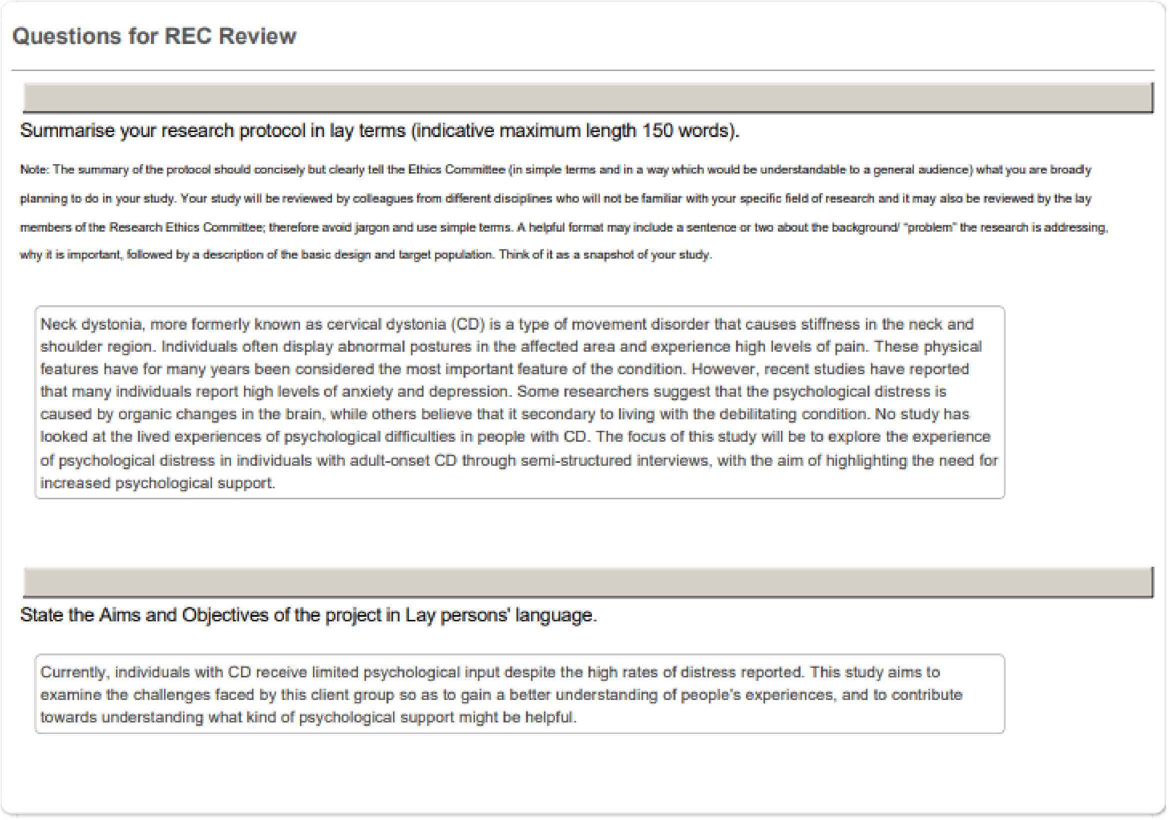
**REC Review Details**

Based on the answers you have given so far you will need to answer some additional questions to allow reviewers to assess your application.

It is recommended that you do not proceed until you have completed **all of the previous questions**.

Please confirm that you have finished answering the previous questions and are happy to proceed.

☑ I confirm that I have answered all of the previous questions, and am happy to proceed with the application.



## Questions for REC Review

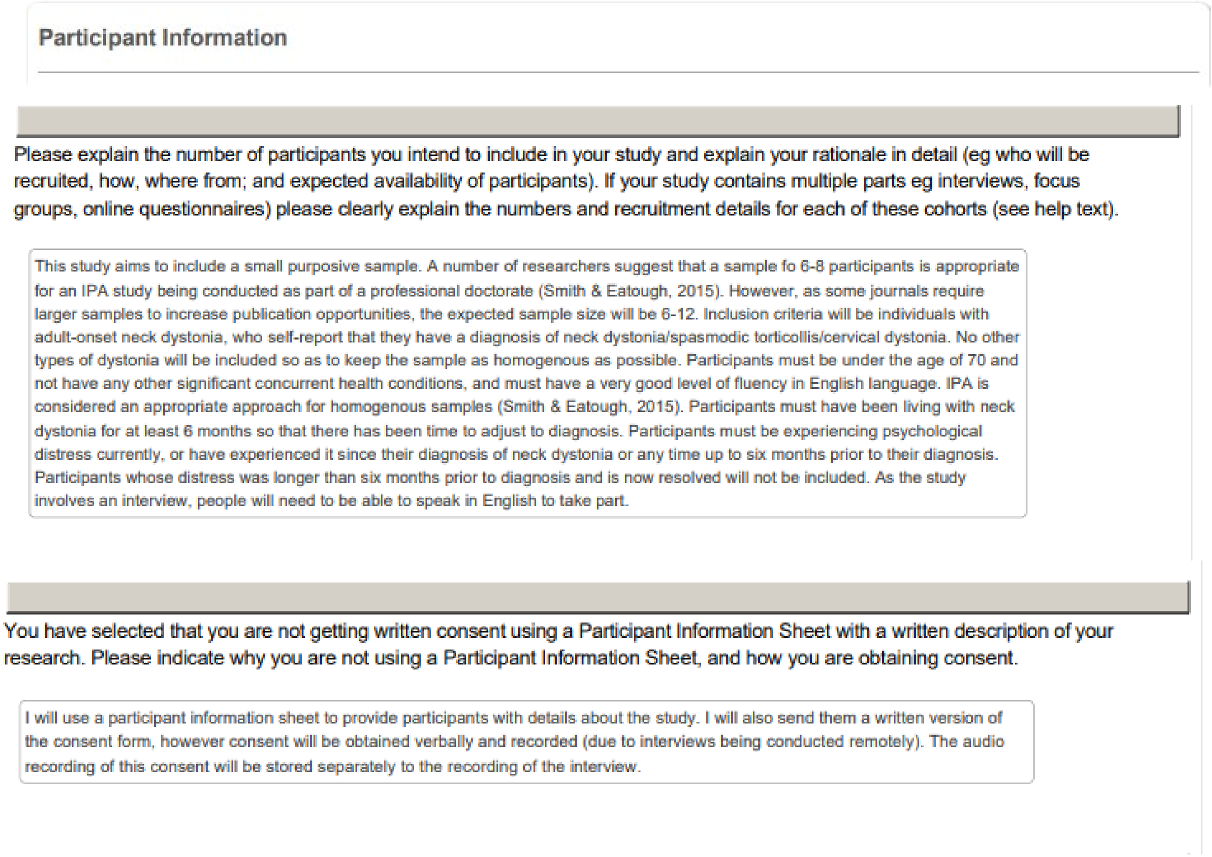
Summarise your research protocol in lay terms (indicative maximum length 150 words).

Note: The summary of the protocol should concisely but clearly tell the Ethics Committee (in simple terms and in a way which would be understandable to a general audience) what you are broadly planning to do in your study. Your study will be reviewed by colleagues from different disciplines who will not be familiar with your specific field of research and it may also be reviewed by the lay members of the Research Ethics Committee; therefore avoid jargon and use simple terms. A helpful format may include a sentence or two about the background/ "problem" the research is addressing, why it is important, followed by a description of the basic design and target population. Think of it as a snapshot of your study.

Neck dystonia, more formerly known as cervical dystonia (CD) is a type of movement disorder that causes stiffness in the neck and shoulder region. Individuals often display abnormal postures in the affected area and experience high levels of pain. These physical features have for many years been considered the most important feature of the condition. However, recent studies have reported that many individuals report high levels of anxiety and depression. Some researchers suggest that the psychological distress is caused by organic changes in the brain, while others believe that it secondary to living with the debilitating condition. No study has looked at the lived experiences of psychological difficulties in people with CD. The focus of this study will be to explore the experience of psychological distress in individuals with adult-onset CD through semi-structured interviews, with the aim of highlighting the need for increased psychological support.

State the Aims and Objectives of the project in Lay persons' language.

Currently, individuals with CD receive limited psychological input despite the high rates of distress reported. This study aims to examine the challenges faced by this client group so as to gain a better understanding of people's experiences, and to contribute towards understanding what kind of psychological support might be helpful.



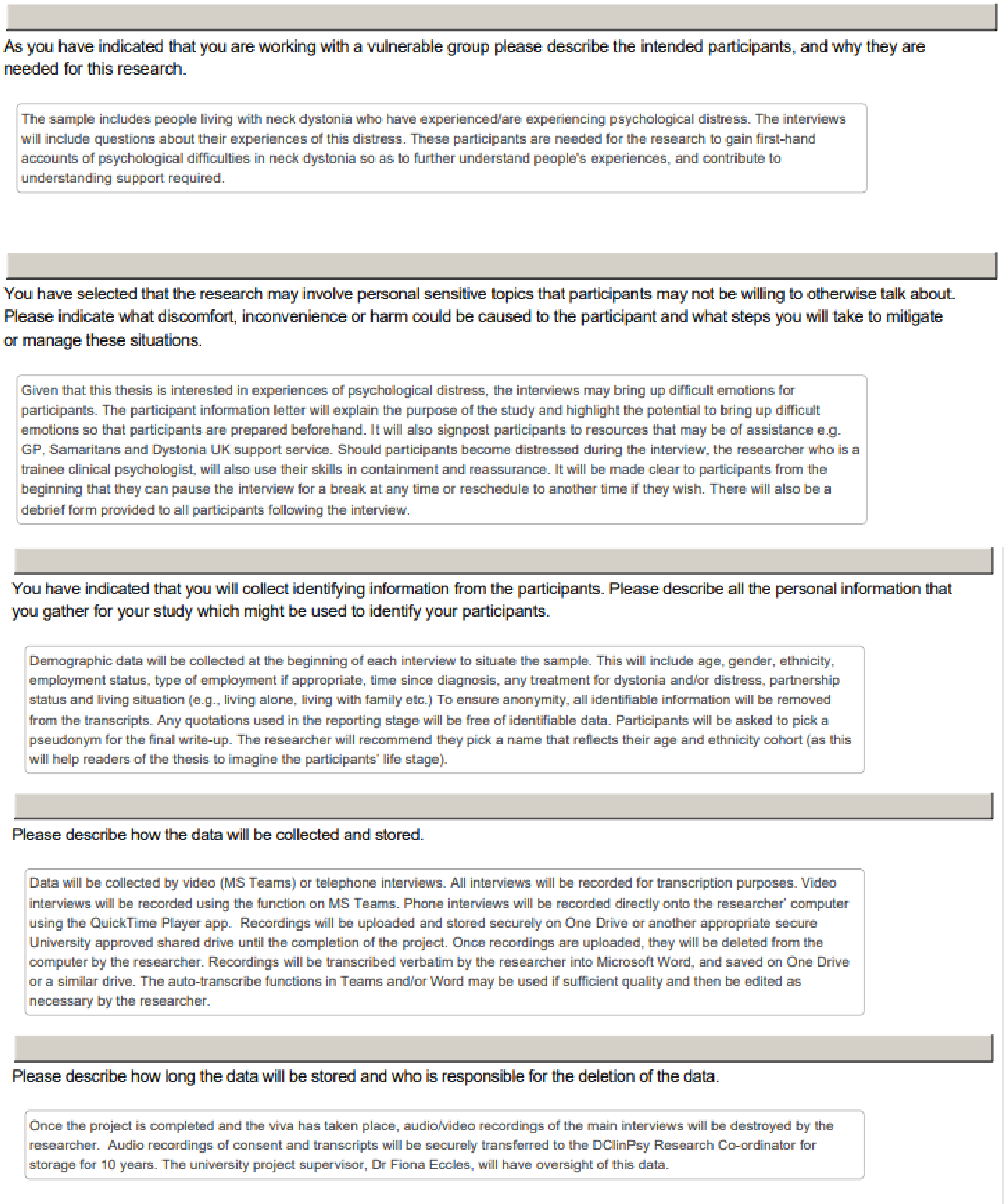
## Participant Information

Please explain the number of participants you intend to include in your study and explain your rationale in detail (eg who will be recruited, how, where from; and expected availability of participants). If your study contains multiple parts eg interviews, focus groups, online questionnaires) please clearly explain the numbers and recruitment details for each of these cohorts (see help text).

This study aims to include a small purposive sample. A number of researchers suggest that a sample fo 6-8 participants is appropriate for an IPA study being conducted as part of a professional doctorate (Smith & Eatough, 2015). However, as some journals require larger samples to increase publication opportunities, the expected sample size will be 6-12. Inclusion criteria will be individuals with adult-onset neck dystonia, who self-report that they have a diagnosis of neck dystonia/spasmodic torticollis/cervical dystonia. No other types of dystonia will be included so as to keep the sample as homogenous as possible. Participants must be under the age of 70 and not have any other significant concurrent health conditions, and must have a very good level of fluency in English language. IPA is considered an appropriate approach for homogenous samples (Smith & Eatough, 2015). Participants must have been living with neck dystonia for at least 6 months so that there has been time to adjust to diagnosis. Participants must be experiencing psychological distress currently, or have experienced it since their diagnosis of neck dystonia or any time up to six months prior to their diagnosis. Participants whose distress was longer than six months prior to diagnosis and is now resolved will not be included. As the study involves an interview, people will need to be able to speak in English to take part.

You have selected that you are not getting written consent using a Participant Information Sheet with a written description of your research. Please indicate why you are not using a Participant Information Sheet, and how you are obtaining consent.

I will use a participant information sheet to provide participants with details about the study. I will also send them a written version of the consent form, however consent will be obtained verbally and recorded (due to interviews being conducted remotely). The audio recording of this consent will be stored separately to the recording of the interview.



As you have indicated that you are working with a vulnerable group please describe the intended participants, and why they are needed for this research.

The sample includes people living with neck dystonia who have experienced/are experiencing psychological distress. The interviews will include questions about their experiences of this distress. These participants are needed for the research to gain first-hand accounts of psychological difficulties in neck dystonia so as to further understand people's experiences, and contribute to understanding support required.

You have selected that the research may involve personal sensitive topics that participants may not be willing to otherwise talk about. Please indicate what discomfort, inconvenience or harm could be caused to the participant and what steps you will take to mitigate or manage these situations.

Given that this thesis is interested in experiences of psychological distress, the interviews may bring up difficult emotions for participants. The participant information letter will explain the purpose of the study and highlight the potential to bring up difficult emotions so that participants are prepared beforehand. It will also signpost participants to resources that may be of assistance e.g. GP, Samaritans and Dystonia UK support service. Should participants become distressed during the interview, the researcher who is a trainee clinical psychologist, will also use their skills in containment and reassurance. It will be made clear to participants from the beginning that they can pause the interview for a break at any time or reschedule to another time if they wish. There will also be a debrief form provided to all participants following the interview.

You have indicated that you will collect identifying information from the participants. Please describe all the personal information that you gather for your study which might be used to identify your participants.

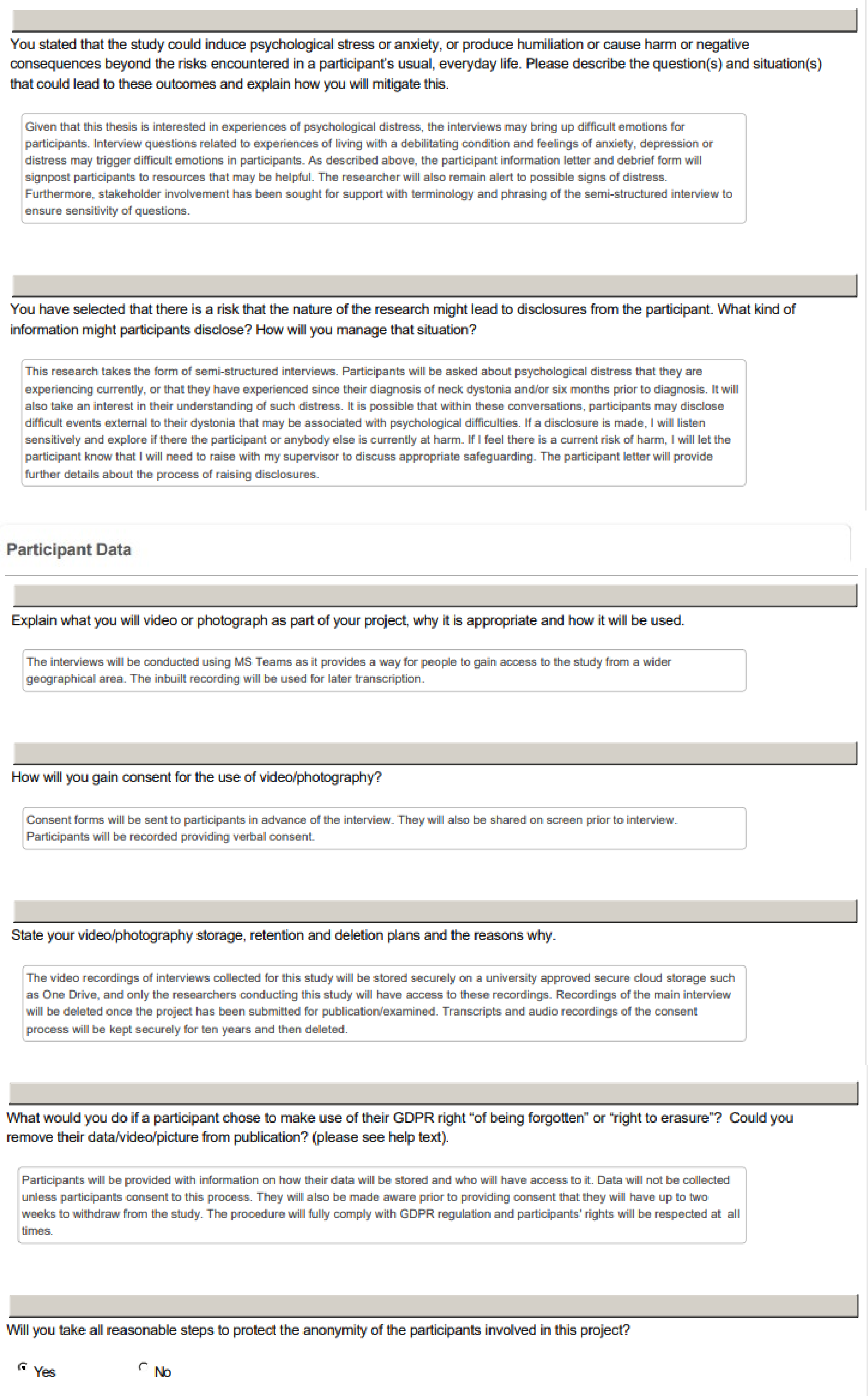
Demographic data will be collected at the beginning of each interview to situate the sample. This will include age, gender, ethnicity, employment status, type of employment if appropriate, time since diagnosis, any treatment for dystonia and/or distress, partnership status and living situation (e.g., living alone, living with family etc.) To ensure anonymity, all identifiable information will be removed from the transcripts. Any quotations used in the reporting stage will be free of identifiable data. Participants will be asked to pick a pseudonym for the final write-up. The researcher will recommend they pick a name that reflects their age and ethnicity cohort (as this will help readers of the thesis to imagine the participants' life stage).

Please describe how the data will be collected and stored.

Data will be collected by video (MS Teams) or telephone interviews. All interviews will be recorded for transcription purposes. Video interviews will be recorded using the function on MS Teams. Phone interviews will be recorded directly onto the researcher' computer using the QuickTime Player app. Recordings will be uploaded and stored securely on One Drive or another appropriate secure University approved shared drive until the completion of the project. Once recordings are uploaded, they will be deleted from the computer by the researcher. Recordings will be transcribed verbatim by the researcher into Microsoft Word, and saved on One Drive or a similar drive. The auto-transcribe functions in Teams and/or Word may be used if sufficient quality and then be edited as necessary by the researcher.

Please describe how long the data will be stored and who is responsible for the deletion of the data.

Once the project is completed and the viva has taken place, audio/video recordings of the main interviews will be destroyed by the researcher. Audio recordings of consent and transcripts will be securely transferred to the DClinPsy Research Co-ordinator for storage for 10 years. The university project supervisor, Dr Fiona Eccles, will have oversight of this data.



You stated that the study could induce psychological stress or anxiety, or produce humiliation or cause harm or negative consequences beyond the risks encountered in a participant's usual, everyday life. Please describe the question(s) and situation(s) that could lead to these outcomes and explain how you will mitigate this.

Given that this thesis is interested in experiences of psychological distress, the interviews may bring up difficult emotions for participants. Interview questions related to experiences of living with a debilitating condition and feelings of anxiety, depression or distress may trigger difficult emotions in participants. As described above, the participant information letter and debrief form will signpost participants to resources that may be helpful. The researcher will also remain alert to possible signs of distress. Furthermore, stakeholder involvement has been sought for support with terminology and phrasing of the semi-structured interview to ensure sensitivity of questions.

You have selected that there is a risk that the nature of the research might lead to disclosures from the participant. What kind of information might participants disclose? How will you manage that situation?

This research takes the form of semi-structured interviews. Participants will be asked about psychological distress that they are experiencing currently, or that they have experienced since their diagnosis of neck dystonia and/or six months prior to diagnosis. It will also take an interest in their understanding of such distress. It is possible that within these conversations, participants may disclose difficult events external to their dystonia that may be associated with psychological difficulties. If a disclosure is made, I will listen sensitively and explore if there the participant or anybody else is currently at harm. If I feel there is a current risk of harm, I will let the participant know that I will need to raise with my supervisor to discuss appropriate safeguarding. The participant letter will provide further details about the process of raising disclosures.

## Participant Data

Explain what you will video or photograph as part of your project, why it is appropriate and how it will be used.

The interviews will be conducted using MS Teams as it provides a way for people to gain access to the study from a wider geographical area. The inbuilt recording will be used for later transcription.

How will you gain consent for the use of video/photography?

Consent forms will be sent to participants in advance of the interview. They will also be shared on screen prior to interview. Participants will be recorded providing verbal consent.

State your video/photography storage, retention and deletion plans and the reasons why.

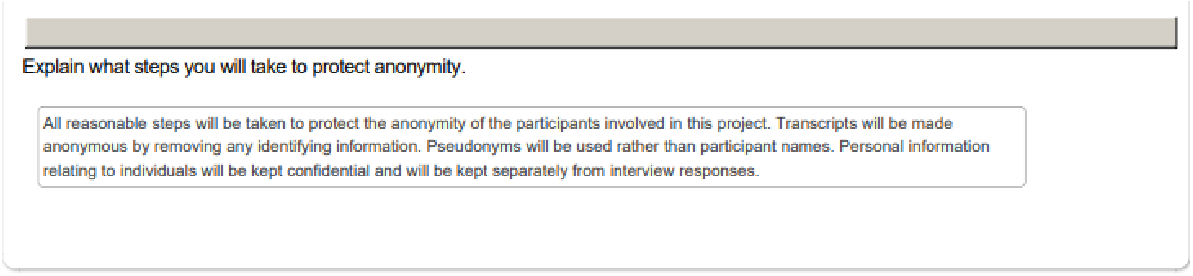
The video recordings of interviews collected for this study will be stored securely on a university approved secure cloud storage such as One Drive, and only the researchers conducting this study will have access to these recordings. Recordings of the main interview will be deleted once the project has been submitted for publication/examined. Transcripts and audio recordings of the consent process will be kept securely for ten years and then deleted.

What would you do if a participant chose to make use of their GDPR right "of being forgotten" or "right to erasure"? Could you remove their data/video/picture from publication? (please see help text).

Participants will be provided with information on how their data will be stored and who will have access to it. Data will not be collected unless participants consent to this process. They will also be made aware prior to providing consent that they will have up to two weeks to withdraw from the study. The procedure will fully comply with GDPR regulation and participants' rights will be respected at all times.

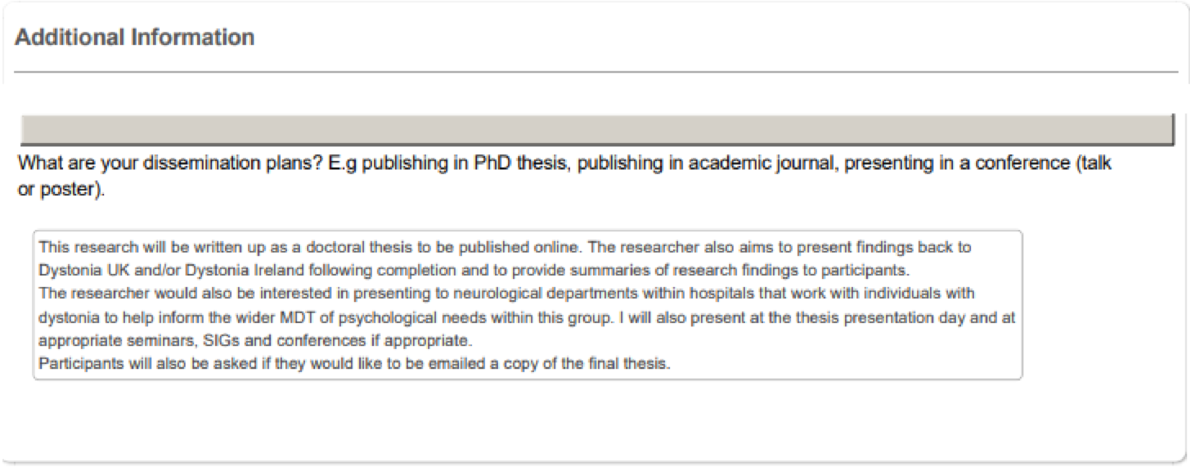
Will you take all reasonable steps to protect the anonymity of the participants involved in this project?

● Yes ○ No



Explain what steps you will take to protect anonymity.

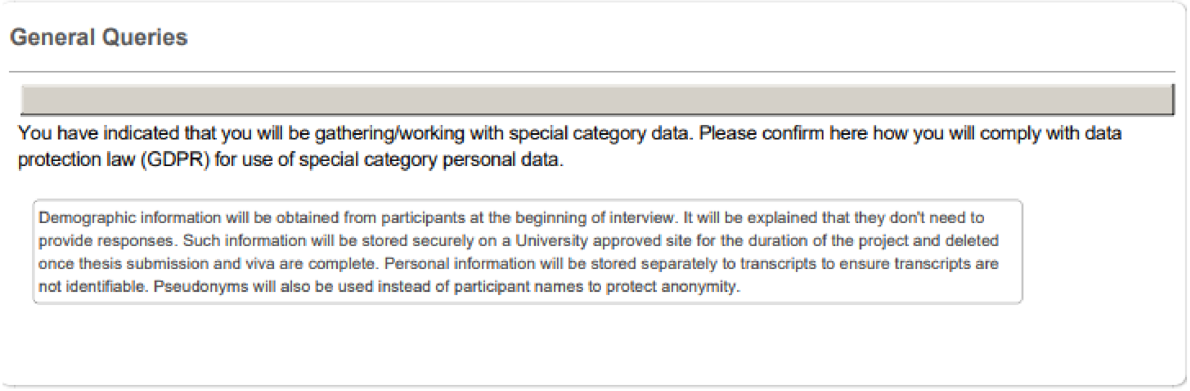
All reasonable steps will be taken to protect the anonymity of the participants involved in this project. Transcripts will be made anonymous by removing any identifying information. Pseudonyms will be used rather than participant names. Personal information relating to individuals will be kept confidential and will be kept separately from interview responses.



### Additional Information

What are your dissemination plans? E.g publishing in PhD thesis, publishing in academic journal, presenting in a conference (talk or poster).

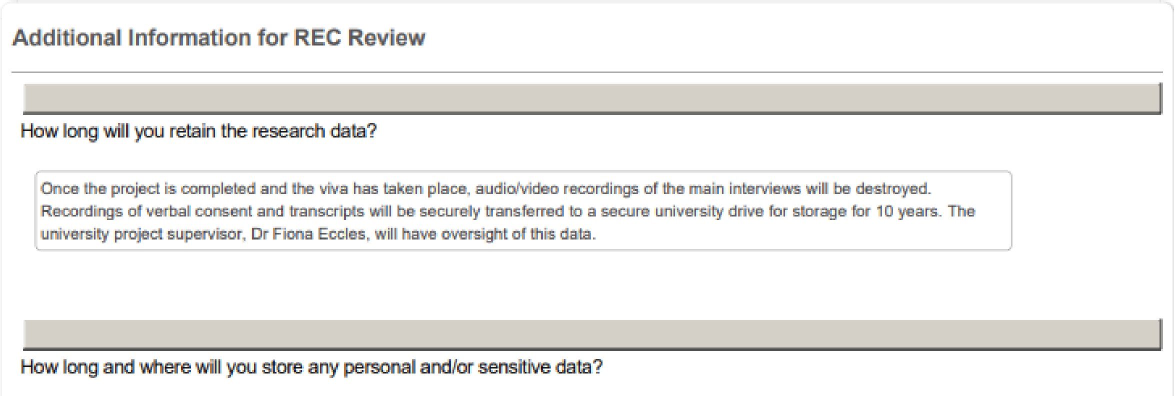
This research will be written up as a doctoral thesis to be published online. The researcher also aims to present findings back to Dystonia UK and/or Dystonia Ireland following completion and to provide summaries of research findings to participants.
The researcher would also be interested in presenting to neurological departments within hospitals that work with individuals with dystonia to help inform the wider MDT of psychological needs within this group. I will also present at the thesis presentation day and at appropriate seminars, SIGs and conferences if appropriate.
Participants will also be asked if they would like to be emailed a copy of the final thesis.



### General Queries

You have indicated that you will be gathering/working with special category data. Please confirm here how you will comply with data protection law (GDPR) for use of special category personal data.

Demographic information will be obtained from participants at the beginning of interview. It will be explained that they don't need to provide responses. Such information will be stored securely on a University approved site for the duration of the project and deleted once thesis submission and viva are complete. Personal information will be stored separately to transcripts to ensure transcripts are not identifiable. Pseudonyms will also be used instead of participant names to protect anonymity.

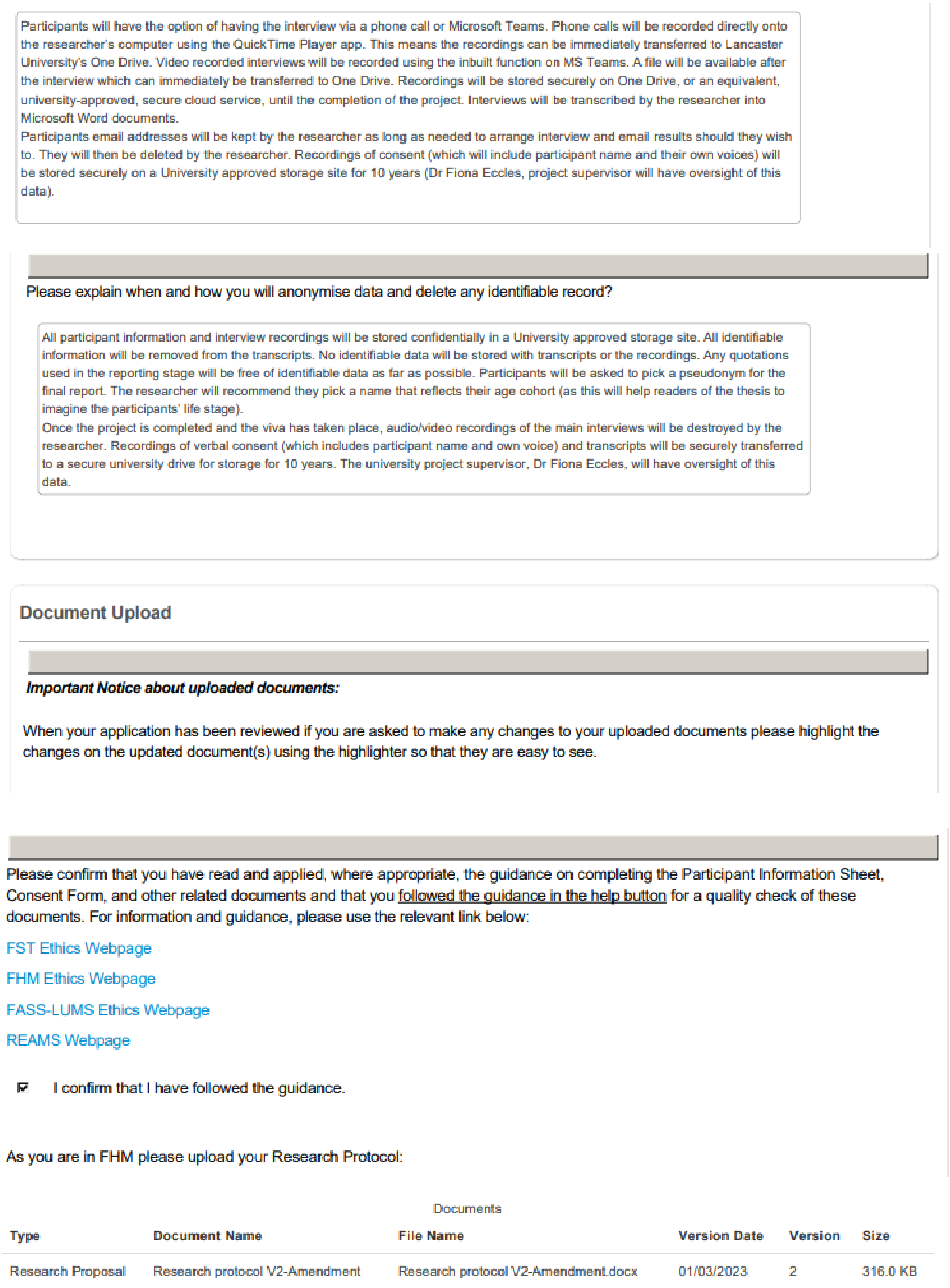


### Additional Information for REC Review

How long will you retain the research data?

Once the project is completed and the viva has taken place, audio/video recordings of the main interviews will be destroyed. Recordings of verbal consent and transcripts will be securely transferred to a secure university drive for storage for 10 years. The university project supervisor, Dr Fiona Eccles, will have oversight of this data.

How long and where will you store any personal and/or sensitive data?



Participants will have the option of having the interview via a phone call or Microsoft Teams. Phone calls will be recorded directly onto the researcher's computer using the QuickTime Player app. This means the recordings can be immediately transferred to Lancaster University's One Drive. Video recorded interviews will be recorded using the inbuilt function on MS Teams. A file will be available after the interview which can immediately be transferred to One Drive. Recordings will be stored securely on One Drive, or an equivalent, university-approved, secure cloud service, until the completion of the project. Interviews will be transcribed by the researcher into Microsoft Word documents.

Participants email addresses will be kept by the researcher as long as needed to arrange interview and email results should they wish to. They will then be deleted by the researcher. Recordings of consent (which will include participant name and their own voices) will be stored securely on a University approved storage site for 10 years (Dr Fiona Eccles, project supervisor will have oversight of this data).

Please explain when and how you will anonymise data and delete any identifiable record?

All participant information and interview recordings will be stored confidentially in a University approved storage site. All identifiable information will be removed from the transcripts. No identifiable data will be stored with transcripts or the recordings. Any quotations used in the reporting stage will be free of identifiable data as far as possible. Participants will be asked to pick a pseudonym for the final report. The researcher will recommend they pick a name that reflects their age cohort (as this will help readers of the thesis to imagine the participants' life stage).

Once the project is completed and the viva has taken place, audio/video recordings of the main interviews will be destroyed by the researcher. Recordings of verbal consent (which includes participant name and own voice) and transcripts will be securely transferred to a secure university drive for storage for 10 years. The university project supervisor, Dr Fiona Eccles, will have oversight of this data.

## Document Upload

***Important Notice about uploaded documents:***

When your application has been reviewed if you are asked to make any changes to your uploaded documents please highlight the changes on the updated document(s) using the highlighter so that they are easy to see.

Please confirm that you have read and applied, where appropriate, the guidance on completing the Participant Information Sheet, Consent Form, and other related documents and that you followed the guidance in the help button for a quality check of these documents. For information and guidance, please use the relevant link below:

FST Ethics Webpage

FHM Ethics Webpage

FASS-LUMS Ethics Webpage

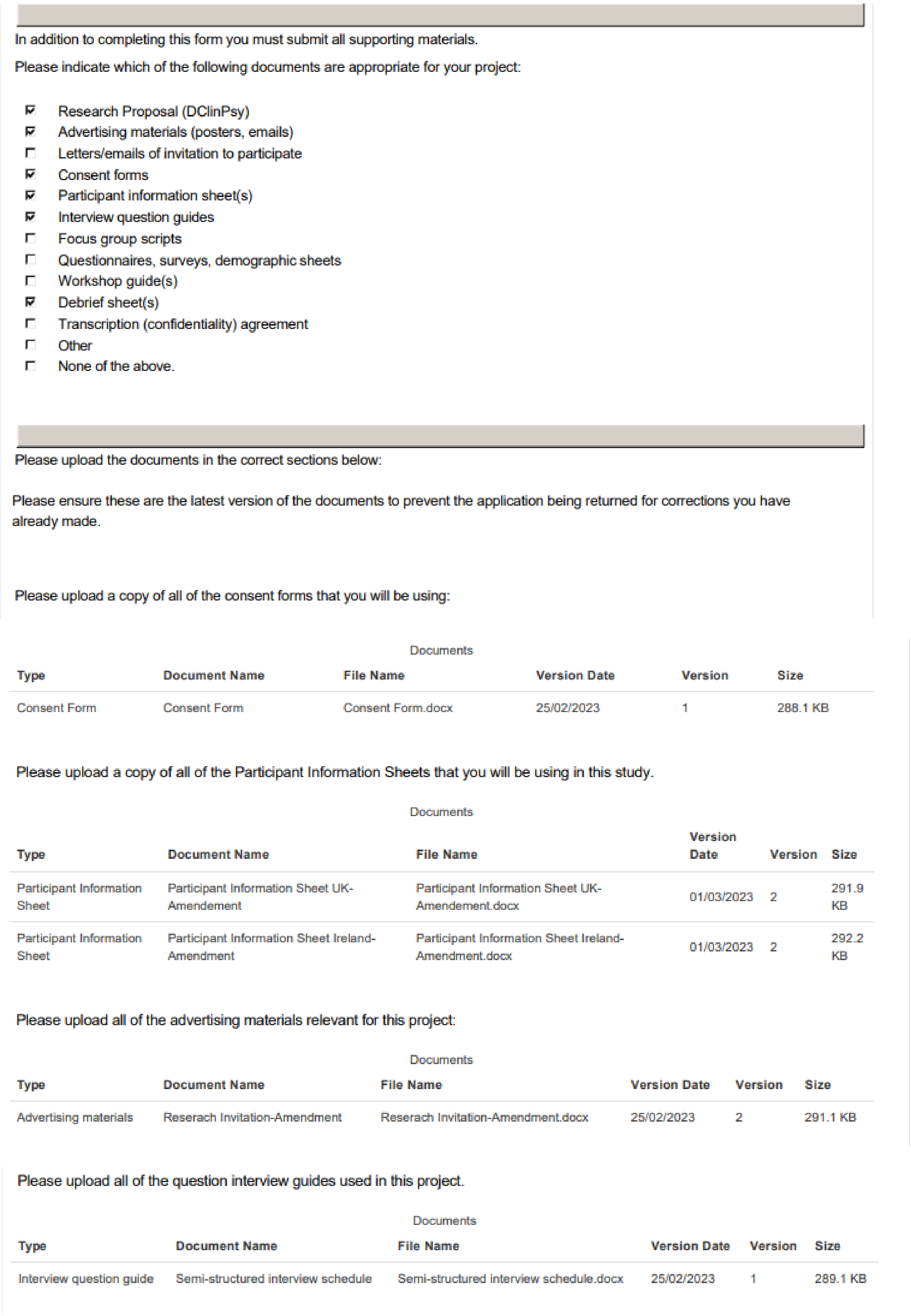
REAMS Webpage

☑ I confirm that I have followed the guidance.

As you are in FHM please upload your Research Protocol:

Documents

| Type | Document Name | File Name | Version Date | Version | Size |
|---|---|---|---|---|---|
| Research Proposal | Research protocol V2-Amendment | Research protocol V2-Amendment.docx | 01/03/2023 | 2 | 316.0 KB |



In addition to completing this form you must submit all supporting materials.

Please indicate which of the following documents are appropriate for your project:

- ☑ Research Proposal (DClinPsy)
- ☑ Advertising materials (posters, emails)
- ☐ Letters/emails of invitation to participate
- ☑ Consent forms
- ☑ Participant information sheet(s)
- ☑ Interview question guides
- ☐ Focus group scripts
- ☐ Questionnaires, surveys, demographic sheets
- ☐ Workshop guide(s)
- ☑ Debrief sheet(s)
- ☐ Transcription (confidentiality) agreement
- ☐ Other
- ☐ None of the above.

Please upload the documents in the correct sections below:

Please ensure these are the latest version of the documents to prevent the application being returned for corrections you have already made.

Please upload a copy of all of the consent forms that you will be using:

Documents

| **Type** | **Document Name** | **File Name** | **Version Date** | **Version** | **Size** |
|---|---|---|---|---|---|
| Consent Form | Consent Form | Consent Form.docx | 25/02/2023 | 1 | 288.1 KB |

Please upload a copy of all of the Participant Information Sheets that you will be using in this study.

Documents

| **Type** | **Document Name** | **File Name** | **Version Date** | **Version** | **Size** |
|---|---|---|---|---|---|
| Participant Information Sheet | Participant Information Sheet UK-Amendement | Participant Information Sheet UK-Amendement.docx | 01/03/2023 | 2 | 291.9 KB |
| Participant Information Sheet | Participant Information Sheet Ireland-Amendment | Participant Information Sheet Ireland-Amendment.docx | 01/03/2023 | 2 | 292.2 KB |

Please upload all of the advertising materials relevant for this project:

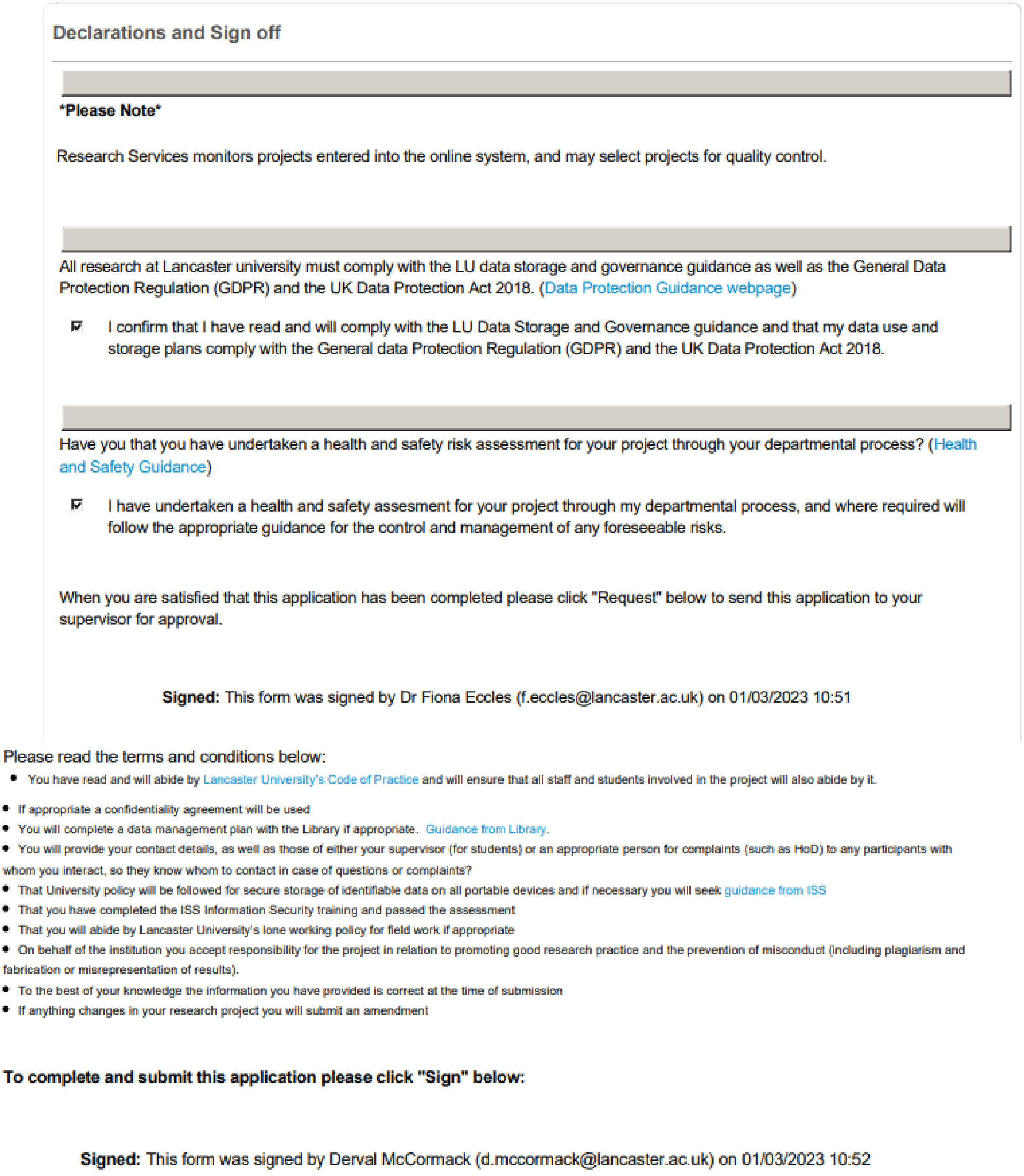
Documents

| **Type** | **Document Name** | **File Name** | **Version Date** | **Version** | **Size** |
|---|---|---|---|---|---|
| Advertising materials | Reserach Invitation-Amendment | Reserach Invitation-Amendment.docx | 25/02/2023 | 2 | 291.1 KB |

Please upload all of the question interview guides used in this project.

Documents

| **Type** | **Document Name** | **File Name** | **Version Date** | **Version** | **Size** |
|---|---|---|---|---|---|
| Interview question guide | Semi-structured interview schedule | Semi-structured interview schedule.docx | 25/02/2023 | 1 | 289.1 KB |



Declarations and Sign off

***Please Note***

Research Services monitors projects entered into the online system, and may select projects for quality control.

All research at Lancaster university must comply with the LU data storage and governance guidance as well as the General Data Protection Regulation (GDPR) and the UK Data Protection Act 2018. (Data Protection Guidance webpage)

☑ I confirm that I have read and will comply with the LU Data Storage and Governance guidance and that my data use and storage plans comply with the General data Protection Regulation (GDPR) and the UK Data Protection Act 2018.

Have you that you have undertaken a health and safety risk assessment for your project through your departmental process? (Health and Safety Guidance)

☑ I have undertaken a health and safety assesment for your project through my departmental process, and where required will follow the appropriate guidance for the control and management of any foreseeable risks.

When you are satisfied that this application has been completed please click "Request" below to send this application to your supervisor for approval.

**Signed:** This form was signed by Dr Fiona Eccles (f.eccles@lancaster.ac.uk) on 01/03/2023 10:51

Please read the terms and conditions below:

- You have read and will abide by Lancaster University's Code of Practice and will ensure that all staff and students involved in the project will also abide by it.
- If appropriate a confidentiality agreement will be used
- You will complete a data management plan with the Library if appropriate. Guidance from Library.
- You will provide your contact details, as well as those of either your supervisor (for students) or an appropriate person for complaints (such as HoD) to any participants with whom you interact, so they know whom to contact in case of questions or complaints?
- That University policy will be followed for secure storage of identifiable data on all portable devices and if necessary you will seek guidance from ISS
- That you have completed the ISS Information Security training and passed the assessment
- That you will abide by Lancaster University's lone working policy for field work if appropriate
- On behalf of the institution you accept responsibility for the project in relation to promoting good research practice and the prevention of misconduct (including plagiarism and fabrication or misrepresentation of results).
- To the best of your knowledge the information you have provided is correct at the time of submission
- If anything changes in your research project you will submit an amendment

**To complete and submit this application please click "Sign" below:**

**Signed:** This form was signed by Derval McCormack (d.mccormack@lancaster.ac.uk) on 01/03/2023 10:52

## 2. Research Protocol

**Title:** Lived experiences of psychological distress in neck dystonia

**Applicant:** Derval McCormack

**Supervisors:** Dr Fiona Eccles (Lecturer, Doctorate in Clinical Psychology, Lancaster University)

Dr Fiadhnait O'Keeffe (Principal Specialist Neuropsychologist, St Vincent's University Hospital, Dublin).

# Introduction

## Background

Dystonia is a hyperkinetic movement disorder characterised by sustained muscle contractions resulting in abnormal, repetitive and often painful postures (Albanese et al., 2013). Adult-onset isolated neck dystonia, also known as cervical dystonia or spasmodic torticollis, is the most common form of focal dystonia marked by involuntary muscle contractions affecting the head and neck (Ben-Shlomo et al., 2002). Symptoms tend to develop slowly and include head tremor, twisting of the head and pulling of the neck (Ben-Shlomo et al., 2002). Previous epidemiology studies have provided inconsistent results for the prevalence of the condition, therefore it remains difficult to determine, with estimates ranging from 20 to 4,100 cases per million (Defazio et al., 2013). A recent review reported 42 as the mean age of onset and a 2:1 female to male ratio (Defazio et al., 2013). While there is no cure for the condition, the physical spasmodic symptoms can be alleviated by botulinum toxin injection (Monaghan et al., 2021).

The physical features of neck dystonia have long been considered the most distinct feature of the condition (Costanzo et al., 2021), however there has been increased interest over recent years in the prevalence of psychological distress within this client group. A number of studies have reported high rates of mood difficulties, in particular anxiety and depression in neck dystonia. A study by Tomic et al. (2016) found depression to be present in 42.1% and anxiety present in 57.9% of their 19 participants. A similar study with 30 participants found depression to be present in 46.3% and anxiety present in 26.6% of neck dystonia participants (Ceylan et al., 2019). A recent study by Monaghan et al. (2021) also reported high levels of psychological distress in their cohort. The condition has also been reported to have a significant impact on people's quality of life and their ability to engage in activities of daily living. Pain, fatigue and disease severity have all been found to be associated with reduced quality of life (Werle et al., 2014; Smit et al., 2017 ). Some studies have also reported high levels of stigma experienced by people with neck dystonia, arising mostly from the postural abnormalities that affect physical appearance (Papathanasiou et al., 2001). A qualitative study by Williams (2005) found that neck dystonia impacted individuals' ability to engage in activities of daily living due to social embarrassment, pain and physical limitations.

There is currently a debate within the literature as to whether psychological distress in neck dystonia is caused by neurobiological processes or if it is secondary to living with the condition itself, or indeed some combination of the two. Berardelli et al. (2015) for example support the argument that mood disorder is a primary symptom of the condition caused by neurobiological mechanisms. They found that over the course of five years, psychological difficulties within individuals with neck dystonia remained stable despite an improvement in disease severity. Ndukwe et al. (2021) also support the view that mood disorder is reflected by disordered neurobiological processes, suggesting that anxiety and depression are primary premotor symptoms of neck dystonia. This belief may explain the tendency for some neurologists to treat symptoms of psychological distress in neck dystonia solely from a medical perspective. There are other studies however that report these difficulties in mood are secondary to the physical symptoms experienced as part of the condition. For example, pain and disease severity have been found to be associated with increased psychological distress in this client group (Müller et al. 2002, Tomic et al., 2016). It is likely that a biopsychosocial framework may be helpful to more fully understand distress in this population.

A number of quantitative studies suggest the interaction of physical, psychological and social factors in understanding distress in neck dystonia. Negative body concept has been found to be associated with reduced quality of life and higher levels of depression (Paracka et al., 2020; Jahanshahi & Marsden, 1990; Lewis & Jahansahi, 2008). Changes in physical appearance have also been found to be associated with lower levels of self-esteem in neck dystonia (Jahanshahi, 1991). Research also demonstrates the impact of socioeconomic factors, with unemployment and lower education level being associated with reduced quality of life (Camfield et al., 2002; Zetterberg et al., 2008; Queiroz et al., 2011). Symptoms associated with neck dystonia can make it difficult for people to sustain employment (Ortiz et al., 2019), which can negatively impact a person's identity and social networks (Johnson et al., 2004). Furthermore, recent literature demonstrates that illness perceptions are also associated with psychological outcome in neck dystonia (O'Connor et al., 2022b).

While these studies suggest that a number of factors may be associated with psychological difficulties for individuals with neck dystonia, further research would be helpful to look more psychologically at the underlying processes behind some of these findings. To date, qualitative literature into this area has been limited, however Caldwell (2011) did strengthen some of the findings above using qualitative methods, indicating an association between disfigurement in neck dystonia and feelings of rejection and hopelessness.

## Clinical Implications

Understanding the psychological impact of neck dystonia has particular relevance to clinical psychologists working with this population. Treatment plans in the past have primarily focused on the medical needs of this client group while the psychological needs have tended to be neglected (Burke et al., 2020). A systematic review by Bernstein et al. (2016) examined the effectiveness of behavioural interventions for people with dystonia, however they were unable to draw robust conclusions due to poor methodologies across studies. In the UK, individuals with neck dystonia who are experiencing psychological difficulties are usually referred by their GP for interventions such as Cognitive Behavioural Therapy (CBT) or mindfulness (Dystonia UK, 2021). However, such treatment options are based on recommendations developed for non-dystonic populations and therefore lack a person-centred approach (Zurowski et al., 2013). Given the high prevalence rates of anxiety and depression within this patient population, it appears that a greater understanding of their psychological needs is important in tailoring appropriate psychological intervention.

### Aim of Research

The majority of research into psychological distress in neck dystonia has used quantitative methods. This thesis aims to investigate individuals' lived experiences of psychological distress using a qualitative design. It is hoped that this study will allow for detailed accounts of patients' experience of anxiety and depression that will increase understanding of the psychological needs of this client group. The study aims to answer two main research questions: what are the experiences of psychological distress in neck dystonia, and how do people with neck dystonia understand their psychological difficulties.

## Method

### Design

Given that this thesis project is interested in participants' experiences, the study will be qualitative in design. The methodological approach that will be used is interpretative phenomenological analysis (IPA). IPA is a model rooted in phenomenology and is interested in how people make sense of their personal experiences. It aims to explore lived experiences of individuals and to examine how people make sense of their personal world (Smith & Eatough, 2015). It is particularly helpful in understanding how people perceive significant events in their lives and is often used as a methodological approach within health and clinical psychology (Smith & Eatough, 2015). As this study is interested in how individuals with neck dystonia understand experiences of psychological distress, this method is deemed most appropriate. People with lived experiences of neck dystonia have been involved and consulted throughout the design process. They provided input regarding terminology of the project title, semi-structured interview schedule, participant information sheet and debrief form.

A semi-structured interview schedule will be used to collect data to guide questioning. The schedule consisted of the following sections: experiences of neck dystonia, psychological distress, impact of neck dystonia on quality of life and support for distress. The interview has been designed to take place over 45-60 minutes.

Demographic data will be collected at the beginning of each interview to situate the sample. This will include age, gender, ethnicity, employment status, type of employment if appropriate, time since diagnosis, any treatment for dystonia and/or distress, partnership status and living situation (e.g., living alone, living with family etc.)

### Participants

This study aims to include a small purposive sample. A number of researchers suggest that a sample size of 6-8 is appropriate for an IPA study being conducted as part of a professional doctorate (Smith & Eatough, 2015). However, as some journals require larger samples to increase publication opportunities, the expected sample size will be 6-12. Inclusion criteria will be individuals with adult-onset neck dystonia, who self-report that they have a diagnosis of neck dystonia/spasmodic torticollis/cervical dystonia. No other types of dystonia will be included so as to keep the sample as homogenous as possible. Participants will be aged 18 to 69 to target a working age sample where participants are less likely to be also experiencing additional health problems due to ageing. Participants aged 70 and above and those experiencing other serious concurrent health problems which affect their day-to-day functioning will be excluded. Participants must have been living with neck dystonia for at least 6 months so that there has been time to adjust to the diagnosis. Participants must be experiencing psychological distress currently, or have experienced it since their diagnosis of neck dystonia or any time up to six months prior to their diagnosis. Participants whose distress was longer than six months prior to diagnosis and is now resolved will not be included. IPA is considered an appropriate approach for homogeneous samples (Smith & Eatough, 2015). Participants must speak a level of English such that they can take part in an interview.

**Procedure**

Following expression of interest in the study, participants will be provided with a participant information sheet and consent form. They will be encouraged to read the materials and ask any questions before providing consent to take part. Consent will be obtained verbally prior to the interview taking place and recorded separately. The study will be advertised via the Dystonia UK website. The research invitation will also be included in the charity's newsletter which is emailed to members and may also be advertised through their social media channels. The advertisement will include a short description of the study and will invite those who are interested to make direct contact with the researcher by email. If the study does not yield sufficient participants, it will then be extended to Dystonia Ireland using similar methods. If sufficient number of participants has still not been reached, the researcher will advertise on a project specific Twitter account. The direct messaging function will be turned off to avoid people sharing personal information. Following participants making contact with the researcher, they will be sent a participant information sheet and available time slots for interview. The interviews will be conducted by video using MS Teams so as to reach a wide geographical network and support inclusivity. Participants will be offered the option of a telephone interview should they prefer that to video. The consent form will be sent to their email prior to the interview, and the consent will be taken by reading out the questions at the beginning of the interview. Participants will be audio-recorded giving this verbal consent. This will be a separate recording to the main interview for long-term storage purposes. All interviews will be recorded for transcription purposes. Video interviews will be recorded using the function on MS Teams. Phone interviews will be recorded directly onto the researcher' computer using the QuickTime Player app. Recordings will be uploaded and stored securely on One Drive or another appropriate secure University approved shared drive until the completion of the project. Once recordings are uploaded, they will be deleted from the computer. Recordings will be transcribed verbatim by the researcher into Microsoft Word, and saved on One Drive or a similar drive. The auto-transcribe functions in Teams and/or Word may be used if sufficient quality and then be edited as necessary by the researcher. Following the interview, participants will be emailed a debrief form. This will include information about dissemination of the research, as well as support services should the interviews trigger any psychological distress. Once the project is completed and the viva has taken place, audio/video recordings of the main interviews will be destroyed. Audio recordings of consent and transcripts will be securely transferred to the DClinPsy Research Co-ordinator for storage for 10 years. The university project supervisor, Dr Fiona Eccles, will have oversight of this data.

**Proposed analysis**

The data will be analysed using IPA. In line with guidelines by Smith & Eatough (2015), the following steps will be used when analysing the data.

- Transcribing the data verbatim. A record of any thoughts or points that appear significant at this stage will be kept.
- In line with IPA methodology, transcripts will be worked through one by one. Once transcribed, data will be read several times to ensure that interpretation later remains grounded in the participants' own accounts. Notes will be made at this point with anything that seems important.
- These notes will be used to identify more specific themes.

- Each transcript will be analysed individually to identify super-ordinate themes, before the researcher connects common themes across transcripts.
- Finally a narrative account of the interplay between researcher interpretation and participants' accounts will be written up formally. Quotes from transcripts will be used at this point.

**Anonymity**

To ensure anonymity, all identifiable information will be removed from the transcripts. Any quotations used in the reporting stage will be free of identifiable data. Participants will be asked to pick a pseudonym for the final write-up. The researcher will recommend they pick a name that reflects their age and ethnicity cohort (as this will help readers of the thesis to imagine the participants' life stage).

**Managing Distress**

Given that this thesis is interested in experiences of psychological distress, the interviews may bring up difficult emotions for participants. The participant information sheet will explain the purpose of the study and highlight the potential to bring up difficult emotions so that participants are prepared beforehand. It will also signpost participants to resources that may be of assistance e.g. GP, Samaritans and Dystonia UK support service. Should participants become distressed during the interview, the researcher who is a trainee clinical psychologist, will also use their skills in containment and reassurance. It will be made clear to participants from the beginning that they can pause the interview for a break at any time or reschedule to another time if they wish. There will also be a debrief form provided to all participants following the interview.

The process of confidentiality will be outlined in the participant information sheet. Interview recordings and any information relating to participants will be stored securely and confidentially. If the researcher is concerned of any harm coming to the participant or others, confidentially may need to be broken. In this case, the researcher will speak with project supervisor(s) as soon as possible and take appropriate action (for example, contact the police or social services).

**Withdrawal of consent**

Following interview, there will be a two week window in which participants can withdraw their data should they want to. This information will be provided in the consent form.

**Timescale**

Sept/Oct 2022-Submit ethics

Dec 2022-Begin recruitment through Dystonia UK

Jan 2023-Write introduction

Spring to summer 2023- Schedule interviews with participants and write methodology

June 2023-Complete interviews and transcriptions

July-September 2023-Analysis

October 2023-Write up findings

December 2023-Write up discussion

March 2024-Final submission

## References

Albanese, A., Bhatia, K., Bressman, S. B., DeLong, M. R., Fahn, S., Fung, V. S., ... & Teller, J. K. (2013). Phenomenology and classification of dystonia: a consensus update. *Movement disorders*, 28(7), 863-873. https://doi.org/10.1002/mds.25475

Berardelli, I., Ferrazzano, G., Pasquini, M., Biondi, M., Berardelli, A., & Fabbrini, G. (2015). Clinical course of psychiatric disorders in patients with cervical dystonia. *Psychiatry research*, *229*(1-2), 583-585. https://doi.org/10.1016/j.psychres.2015.07.076

Bernstein, C. J., Ellard, D. R., Davies, G., Hertenstein, E., Tang, N. K. Y., Underwood, M., & Sandhu, H. (2016). Behavioural interventions for people living with adult-onset primary dystonia: a systematic review. *BMC neurology*, *16*(1), 1-14. https://bmcneurol.biomedcentral.com/articles/10.1186/s12883-016-0562-y

Ben-Shlomo, Y., Camfield, L., & Warner, T. (2002). What are the determinants of quality of life in people with cervical dystonia?. *Journal of Neurology, Neurosurgery & Psychiatry*, *72*(5), 608-614. http://dx.doi.org/10.1136/jnnp.72.5.608

Burke, T., Monaghan, R., McCormack, D., Cogley, C., Pinto-Grau, M., O'Connor, S., ... & O'Keeffe, F. (2020). Social cognition in cervical dystonia: A case-control study. *Clinical Parkinsonism & Related Disorders*, *3*, 100072. https://doi.org/10.1016/j.prdoa.2020.100072

Caldwell, D. M. M. (2001). Without a normal face: Grief and loss experiences of those afflicted with spasmodic torticollis. Gonzaga University.

Camfield, L., Ben-Shlomo, Y., Warner, T. T., & Epidemiological Study of Dystonia in Europe (ESDE) Collaborative Group. (2002). Impact of cervical dystonia on quality of life. *Movement disorders*, *17*(4), 838-841. https://doi.org/10.1002/mds.10127

Ceylan, D., Erer, S., Zarifoğlu, M., Türkeş, N., & Özkaya, G. (2019). Evaluation of anxiety and depression scales and quality of LIFE in cervical dystonia patients on botulinum toxin therapy and their relatives. *Neurological Sciences*, *40*(4), 725-731. https://link.springer.com/article/10.1007/s10072-019-3719-9

Costanzo, M., Belvisi, D., Berardelli, I., Maraone, A., Baione, V., Ferrazzano, G., ... & Berardelli, A. (2021). Effect of Botulinum Toxin on Non-Motor Symptoms in Cervical Dystonia. *Toxins*, *13*(9), 647. https://doi.org/10.3390/toxins13090647

Defazio, G., Jankovic, J., Giel, J. L., & Papapetropoulos, S. (2013). Descriptive epidemiology of cervical dystonia. *Tremor and Other Hyperkinetic Movements*, *3*.

*I feel I'm struggling with my dystonia. Where can I get psychological support?* Dystonia UK. https://www.dystonia.org.uk/FAQs/professional-psychological-support-faq

Jahanshahi, M. (1991). Psychosocial factors and depression in torticollis. *Journal of psychosomatic research*, *35*(4-5), 493-507. https://doi.org/10.1016/0022-3999(91)90044-O

Jahanshahi, M., & Marsden, C. D. (1990). Body concept, disability, and depression in patients with spasmodic torticollis. *Behavioural neurology*, *3*(2), 117-131. https://doi.org/10.3233/BEN-1990-3206

Johnson, K. L., Amtmann, D., Yorkston, K. M., Klasner, E. R., &amp; Kuehn, C. M. (2004). Medical, Psychological, Social, and Programmatic Barriers to Employment for People with Multiple Sclerosis. Journal of Rehabilitation, 70(1).

Lewis, L., Butler, A., & Jahanshahi, M. (2008). Depression in focal, segmental and generalized dystonia. *Journal of neurology*, *255*(11), 1750-1755. doi: 10.1007/s00415-008-0020-x

Monaghan, R., Cogley, C., Burke, T., McCormack, D., O'Riordan, S., Ndukwe, I., ... & O'Keeffe, F. (2021). Non-motor features of cervical dystonia: Cognition, social cognition, psychological

Müller, J., Kemmler, G., Wissel, J., Schneider, A., Voller, B., Grossmann, J., ... & Poewe, W. (2002). The impact of blepharospasm and cervical dystonia on health-related quality of life and depression. *Journal of neurology*, *249*(7), 842-846. https://link.springer.com/article/10.1007/s00415-002-0733-1

Ndukwe, I., O'Riordan, S., Walsh, C. B., & Hutchinson, M. (2020). Mood disorder affects age at onset of adult-onset cervical dystonia. *Clinical Parkinsonism & Related Disorders*, *3*, 100049. https://doi.org/10.1016/j.prdoa.2020.100049

O'Connor, S., Hevey, D., & O'Keeffe, F. (2022). Illness Perceptions, Coping, Health-Related Quality of Life and Psychological Outcomes in Cervical Dystonia. *Journal of Clinical Psychology in Medical Settings,* 1-14. doi: 10.1007/s10880-022-09851-2

Ortiz, R. M., Scheperjans, F., Mertsalmi, T., & Pekkonen, E. (2019). Comorbidity and retirement in cervical dystonia. *Journal of neurology*, *266*(9), 2216-2223. https://link.springer.com/article/10.1007/s00415-019-09402-0

Papathanasiou, I., MacDonald, L., Whurr, R., & Jahanshahi, M. (2001). Perceived stigma in Spasmoic Torticollis. *Movement disorders: official journal of the Movement Disorder Society*, *16*(2), 280-285. https://link.springer.com/article/10.1007/s00415-019-09402-0

Paracka, L., Wegner, F., Escher, C., Klietz, M., de Zwaan, M., Abdallat, M., ... & Krauss, J. K. (2020). Body concept and quality of life in patients with idiopathic dystonia. *Brain Sciences*, *10*(8), 488. https://doi.org/10.3390/brainsci10080488

Queiroz, M. R., Chien, H. F., & Barbosa, E. R. (2011). Quality of life in individuals with cervical dystonia before botulinum toxin injection in a Brazilian tertiary care hospital. *Arquivos de neuro-psiquiatria*, *69*, 900-904. doi: 10.1590/s0004-282x2011000700010

Smit, M., Kamphuis, A. S., Bartels, A. L., Han, V., Stewart, R. E., Zijdewind, I., & Tijssen, M. A. (2017). Fatigue, sleep disturbances, and their influence on quality of life in cervical dystonia patients. *Movement disorders clinical practice*, *4*(4), 517-523. https://doi.org/10.1002/mdc3.12459

Smith, A. & Eatough, V. (2015). Interpretative Phenomenological Analysis. In Lyons, E. & Coyle, A. (Eds.), *Analysing Qualitative Data in Psychology* (pp. 50-67). SAGE.

Tomic, S., Petkovic, I., Pucic, T., Resan, B., Juric, S., & Rotim, T. (2016). Cervical dystonia and quality of life. *Acta Neurologica Belgica*, *116*(4), 589-592. https://link.springer.com/article/10.1007/s13760-016-0634-1

Tong, A., Sainsbury, P., & Craig, J. (2007). Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International journal for quality in health care*, *19*(6), 349-357. https://doi.org/10.1093/intqhc/mzm042

Werle, R. W., Takeda, S. Y. M., Zonta, M. B., Guimarães, A. T. B., & Teive, H. A. G. (2014). The physical, social and emotional aspects are the most affected in the quality of life of the patients with cervical dystonia. *Arquivos de neuro-psiquiatria*, *72*, 405-410. https://doi.org/10.1590/0004-282X20140044

Williams, J. N. (2005). *The Everyday Lived Experiences of People with Cervical Dystonia: A Qualitative Approach* (Doctoral dissertation, Mount Saint Vincent University). http://hdl.handle.net/10587/2034

Zetterberg, L., Aquilonius, S. M., & Lindmark, B. (2009). Impact of dystonia on quality of life and health in a Swedish population. *Acta neurologica scandinavica*, *119*(6), 376-382. https://doi.org/10.1111/j.1600-0404.2008.01111.x

Zurowski, M., McDonald, W. M., Fox, S., & Marsh, L. (2013). Psychiatric comorbidities in dystonia: emerging concepts. *Movement Disorders*, *28*(7), 914-920. https://doi.org/10.1002/mds.25501

# Appendices

## Appendix 4-A

## Ethics Review Feedback (22nd September 2022)

FHM-2022-1010-RECR-1 Lived experiences of psychological distress in neck dystonia

Dear Derval McCormack,

Thank you for submitting your ethics application in REAMS, Lancaster University's online review system for research. Your application has been reviewed by member(s) of your Faculty Research Ethics Committee (FREC). You will find their comments in the table below.

Please read the comments carefully, provide the information requested and revise your application and the attachments accordingly. If you replace an attachment please remember to delete the old version and upload the new documents as Version V2 (V3 etc.). Once you resubmit your application it will be reviewed again by member(s) of the FHM.

Yours sincerely,

Faculty Research Ethics Officer on behalf of FHM

Link to application: https://reams.lancaster-university.uk/Project/Index/1079

| Title | Comment |
|---|---|
| REC - Advertising Materials | Suggest that the advert is made more 'attractive' to assist recruitment. Perhaps we haven't seen what will be presented on the Dystopia UK website, a word document was submitted. |

## Appendix 4-B

## Approval Letter from Ethics Board (6th March 2023)

**Name:** Derval McCormack
**Supervisor:** Fiona Eccles
**Department:** Health Research
FHM **REC Reference:** FHM-2023-1010-SA-1
**Title:** Lived experiences of psychological distress in neck dystonia

Dear Derval McCormack,

Thank you for submitting your ethics application in REAMS, Lancaster University's online ethics review system for research. The application was recommended for approval by the FHM Research Ethics Committee, and on behalf of the Committee, I can confirm that approval has been granted for this application.

As Principal Investigator/Co-Investigator your responsibilities include:

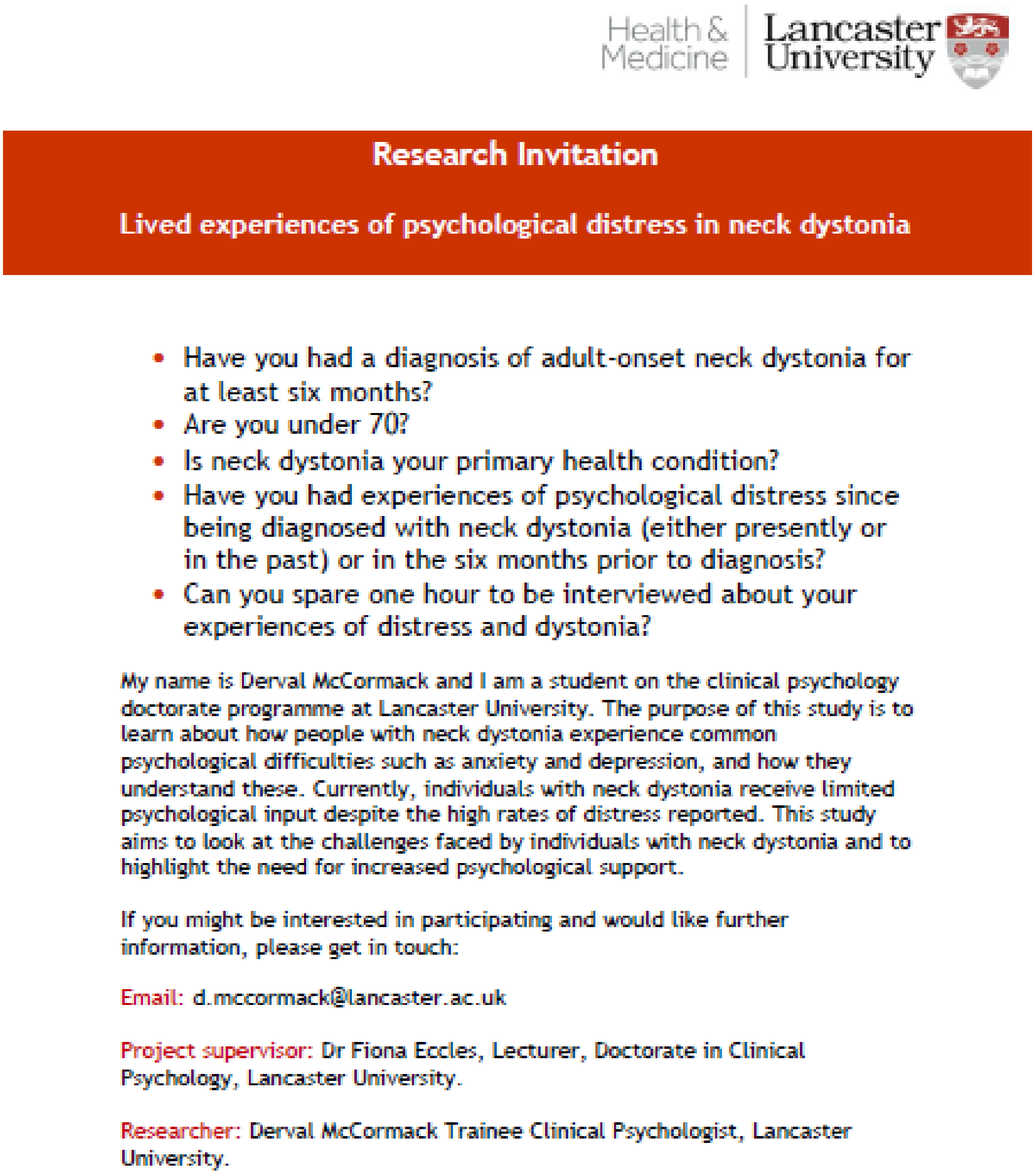
- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licences and approvals have been obtained.
- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress).
- submitting any changes to your application, including in your participant facing materials (see attached amendment guidance).

Please keep a copy of this email for your records. Please contact me if you have any queries or require further information.

Yours sincerely,
Dr Laura Machin
Chair of the Faculty of Health and Medicine Research Ethics Committee
fhmresearchsupport@lancaster.ac.uk

## Appendix 4-C

## Research Invitation used in Recruitment Process



Health & Medicine

Lancaster University

**Research Invitation**

**Lived experiences of psychological distress in neck dystonia**

- Have you had a diagnosis of adult-onset neck dystonia for at least six months?
- Are you under 70?
- Is neck dystonia your primary health condition?
- Have you had experiences of psychological distress since being diagnosed with neck dystonia (either presently or in the past) or in the six months prior to diagnosis?
- Can you spare one hour to be interviewed about your experiences of distress and dystonia?

My name is Derval McCormack and I am a student on the clinical psychology doctorate programme at Lancaster University. The purpose of this study is to learn about how people with neck dystonia experience common psychological difficulties such as anxiety and depression, and how they understand these. Currently, individuals with neck dystonia receive limited psychological input despite the high rates of distress reported. This study aims to look at the challenges faced by individuals with neck dystonia and to highlight the need for increased psychological support.

If you might be interested in participating and would like further information, please get in touch:

Email: d.mccormack@lancaster.ac.uk

Project supervisor: Dr Fiona Eccles, Lecturer, Doctorate in Clinical Psychology, Lancaster University.

Researcher: Derval McCormack Trainee Clinical Psychologist, Lancaster University.

## Appendix 4-D

## Participant Information Sheet

### Participant Information Sheet

### Lived experiences of psychological distress in neck dystonia

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

Thank you for your interest in this research project. My name is Derval McCormack and I am conducting this research as a student on the Clinical Psychology Doctorate programme at Lancaster University, Lancaster, United Kingdom.

**What is the study about?**

The purpose of the study is to gain insight into how people with neck dystonia experience common psychological difficulties such as anxiety and depression and how they understand these. Currently, individuals with neck dystonia receive limited psychological input despite the high rates of distress reported. This study aims to examine the challenges faced by this group and to highlight the need for increased psychological support.

**Why have I been approached?**

This study is interested in individuals with adult-onset neck dystonia. We are interested in the experience of people who have been living with the condition for at least 6 months. Individuals must have experienced distress (e.g., anxiety, depression, or stress) either currently or since their neck dystonia diagnosis (or within the six months prior to their diagnosis) and you can still be experiencing distress now. It does not matter whether you have seen a doctor about the distress or not. This study is interested in the experiences of individuals under the age of seventy where neck dystonia is their primary health condition. Those with other significant concurrent health conditions and other types of dystonia will not be able to take part. As the study involves an interview, people will need to be able to speak in English to take part.

**Do I have to take part?**

No. Participating in this research study is completely voluntary and you can withdraw at any time before, during or up to two weeks post-interview.

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

If you decide to take part in the research, please reply by email to the researcher to arrange an interview at a time convenient for you. The interview will take up to an hour and will be conducted via MS Teams. If preferable a phone call can also be arranged. The interviewer will ask about your experiences of living with neck dystonia, as well as how it has impacted upon you psychologically.

**Will my data be identifiable?**

The recordings of interviews collected for this study will be stored securely on a university approved secure cloud storage, and only the researchers conducting this study will have access to these recordings:

- Audio/ video recordings of the main interview will be deleted once the project has been submitted for publication/examined.
- The files on the computer will be encrypted (that is no-one other than the researcher will be able to access them) and the computer itself will be 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.
- All your personal data will be confidential and will be kept separately from your interview responses.
- Transcripts and audio recordings of the consent process will be kept securely for ten years and then deleted.

There are some limits to confidentiality. If what is said in the interview suggests 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 have to do this.

**What will happen to the results?**

The results will be summarised and reported in a thesis and may be submitted for publication in an academic journal. A report of the findings will also be shared with you and the other interviewees (should you want to see it) and with Dystonia UK.

**Are there any risks?**

There are minimal risks anticipated with participating in this study. However, if you experience any distress following participation you are encouraged to inform the researcher. There are also some helpful resources provided at the end of this sheet of services that you may wish to contact for further support.

### Are there any benefits to taking part?

Although you may find participating interesting, there are no direct benefits in taking part. There may be indirect benefits to taking part, as the research aims to increase understanding of neck dystonia and to contribute to better treatment and management of the condition.

### Who has reviewed the project?

This study has been reviewed and approved by the Faculty of Health and Medicine Research Ethics Committee at Lancaster University.

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

If you have any questions about the study, please contact the main researcher:

Derval McCormack

Email: d.mccormack@lancaster.ac.uk

You can also contact the research supervisor:
Dr Fiona Eccles

Email: f.eccles@lancaster.ac.uk

Tel: 01524 592 807

### 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: +44 (0)7507 857069
Research Director for Clinical Psychology Email: I.smith@lancaster.ac.uk
Division of Health Research, Faculty of Health and Medicine
Lancaster University
B31 Floor, Health Innovation One
Sir John Fisher Drive
Lancaster University
Lancaster
LA1 4AT

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

Dr Laura Machin Tel: +44 (0)1524 594973

Chair of Faculty of Health and Medicine Research Ethics Committee

Email: l.machin@lancaster.ac.uk

Faculty of Health and Medicine

(Lancaster Medical School)

Lancaster University

Lancaster

LA1 4YG

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

**Resources in the event of distress**

Should you feel distressed either as a result of taking part, or in the future, the following resources may be of assistance.

**Your GP or consultant:** we recommend speaking to your own hospital consultant or GP for support.

**Samaritans**

www.samaritans.org

Tel: 116 123

Email: jo@samaritans.org

**Dystonia UK**

www.dysonia.org.uk

Tel: 020 7793 3651

Email: support@dystonia.org.uk

## Appendix 4-E

## Semi-Structured Interview Schedule

**Interview Schedule with Prompt Questions**

**Topic 1-Experiences of Neck Dystonia**

Can you describe your journey to getting a diagnosis of neck dystonia?

-How long have you been living with neck dystonia?

-Can you describe for me the symptoms of neck dystonia?

-What symptoms are most prominent for you?

**Topic 2-Impact on Quality of Life**

-How does neck dystonia impact your daily life (e.g., family, friends, work/not being able to work)

-What has been most difficult for you about this condition/what are the main challenges?

-Has neck dystonia stopped you from doing anything that you would like to do?

**Topic 3: Experiences and Understanding of Psychological Distress**

-When we talk about psychological distress we are referring to things like anxiety, depression and stress. Are these things that you recognise you experience?

-Can you tell me about any experiences of psychological distress?

Did you have any psychological difficulties prior to diagnosis?

-What do you think has contributed to distress for you?

-Some researchers say that mood difficulties are as much a symptom of neck dystonia as motor symptoms. Other researchers believe that mood difficulties happen as a result of living with the movement symptoms. How do you understand psychological distress in neck dystonia?

**Topic 4: Support for Psychological Distress**

-Have you ever received psychological support for anxiety, depression or stress?

-Have you talked to your neurologist about how your mood is affected? How have these conversations gone? What support or treatment was recommended for you?

-Have you ever been prescribed medication for your mood? How do you find the medication?

-What do you find is most helpful to you in managing any psychological difficulties that you have?

-How have family/friends/work responded to psychological distress you have experienced?

-Do you think you receive sufficient psychological support or do you think there is a need for more?

**Anything else?**

- Is there anything I might have missed that you think might be relevant or helpful for me to know in the context of this research?

# Appendix 4-F

## Consent Form

### Consent Form

**NO NEED TO SIGN THIS FORM. WE WILL GO THROUGH TOEGETHER PRIOR TO INTERVIEW AND VERBAL CONSENT WILL BE RECORDED.**

### Consent information

**Study Title:** Lived experiences of psychological distress in neck dystonia

**Please note you do not need to fill in this form or post it back. It is just for information.** The researcher will go through each point during the interview and you will be able to give verbal consent.

If you have any questions or queries regarding this information before the interview before please speak to the researcher, Derval McCormack.

1. I confirm that I have read the information sheet and fully understand what is expected of me within this study.
2. I confirm that I have had the opportunity to ask any questions and to have them answered.
3. I understand that my interview will be video (or audio) recorded and then made into an anonymised written transcript.
4. I understand that video (or audio) recordings will be kept until the research project has been examined.
5. I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my medical care or legal rights being affected.
6. I understand that after the interview I have up to two weeks to change my mind and withdraw consent for my data to be used.

7. I understand that the information from my interview will be combined 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. I consent to information and quotations from my interview being used in reports, conferences and training events. These will also be anonymous.

9. I understand that the researcher will discuss data with their supervisor as needed.

10. I understand that any information I give will remain confidential and anonymous unless it is thought that there is a risk of harm to myself or others, in which case the researcher may need to share this information with their research supervisor.

11. I consent to Lancaster University keeping written transcriptions of the interview for 10 years after the study has finished.

## Appendix 4G

## Debrief Form

### Debrief Form

Thank you for participating in this study and sparing your time to contribute to research.

The purpose of the study is to learn about how people with neck dystonia experience common psychological difficulties such as anxiety and depression, and how they understand these. Currently, individuals with neck dystonia receive limited psychological input despite the high rates of distress reported. This study aims to examine the challenges faced by individuals with neck dystonia and to highlight the need for increased psychological support.

The interview will be transcribed anonymously into a word document. The researcher will look for common themes across all interview transcripts. These themes will be written up in a report and published by Lancaster University.  The researcher hopes to publish the final report in an academic journal.

If you would like, the researcher will email or post a copy of the final report to your address. If you have any questions about the research or the process, please contact d.mccormack@lancaster.ac.uk

If you have experienced any distress from things discussed during the interview, or at any point in the future, then the following resources may be helpful:

**Your GP:** we recommend speaking to your own hospital consultant or GP for support.

**Samaritans**
www.samaritans.org
Tel: 116 123
Email: jo@samaritans.org

**Dystonia UK**
www.dysonia.org.uk
Tel: 020 7793 3651
Email: support@dystonia.org.uk