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Doctoral Thesis:
Exploring the impact and experience of living with dystonia

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Statement of Total Word Count

Section	Main Text	Appendices (including tables, figures and references)	Total
Thesis Abstract	263	-	263
Literature Review	8,826	8,096	16,922
Research Paper	9,509	7,693	17,202
Critical Appraisal	4,022	889	4,911
Ethics Section	2,395	4,958	7,353
Total	25,015	21,636	46,651

Thesis Abstract

This thesis is a discussion of the impact and experience of living with dystonia. The literature review investigated factors relating to health related quality of life (HRQOL) for people living with dystonia. The main research paper used interpretative phenomenological analysis to explore the experiences of people living with the condition. The thesis concludes with a critical appraisal of the two papers and the process of working with two different research paradigms.

The literature review found that depression and anxiety had a significant, negative impact on HRQOL. Dystonia severity and psychosocial variables such as body concept and self-esteem were also implicated. A series of pervasive issues had a negative impact on the methodological quality of reviewed studies. These include a lack of engagement with key concepts such as HRQOL, depression and anxiety. Suggestions were made for future research designs.

The main research paper produced three themes: (1) Dealing with ignorance and uncertainty: navigating health services with a rare, poorly understood condition; (2) The challenge of social isolation: overcoming barriers to positive social identity; and (3) Fear of psychological explanations: the impact of stigmatised attitudes towards psychological explanations for dystonia symptoms. Participants described their experiences of isolation and alienation which resulted from having a rare, visible and chronic health condition. Academic and clinical implications were discussed.

Finally, the critical appraisal reflected on the process of writing using two different research paradigms. Adherence to the philosophical assumptions of each approach was considered a strength of the thesis. The limitations of the two approaches were discussed as well as recommendations for further research on dystonia.

Declaration

This thesis records work undertaken for the Doctorate in Clinical Psychology at the Division of Health Research at Lancaster University from June 2014 to July 2018. The work presented here is the author's own, except where due reference is made. The work has not been submitted for the award of a higher degree elsewhere.

Name: Andrew Morgan

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Acknowledgements

I would like to thank the people who took part in my study. Hearing their stories was a privilege and had a significant impact on me. I hope that my writing has done them justice and that it helps them to reach their goals of raising awareness of dystonia and improving the experiences of people with the condition.

I would also like to thank my supervisors Dr Fiona Eccles and Dr Pete Greasley for making this research possible, not only through their academic advice but also their patience and understanding. Thanks also to Dr Anna Daiches and the other people on the course who believed that I could finish this thesis.

Finally, I would like to thank Keith, my father, for listening to me talk about my ideas and opinions about these topics for the past five years.

Contents

Word Count	-
Thesis Abstract	-
Declaration	-
Acknowledgments	-
Section One: Literature Review	
Title page	1-1
Abstract	1-2
Introduction	1-3
Methods	1-8
Results	1-10
Discussion	1-16
References	1-33
Table 1	1-47
Table 2	1-50
Table 3	1-56
Figure 1	1-65
Appendix 1-A: Author Guidelines for <i>Disability and Rehabilitation</i>	1-66
Appendix 1-B: Implications for rehabilitation	1-75
Appendix 1-C: Quality appraisal checklist	1-76
Appendix 1-D: Data extraction form	1-77

Section Two: Research Paper

Title page	2-1
Abstract	2-2
Introduction	2-3
Methods	2-5
Results	2-10
Discussion	2-22
References	2-36
Table 1	2-48
Table 2	2-50
Table 3	2-52
Appendix 2-A: Author Guidelines for <i>Disability and Rehabilitation</i>	2-53
Appendix 2-B: Implications for rehabilitation	2-61
Appendix 2-C: Excerpt from annotated transcript	2-62
Appendix 2-D: Table of emergent themes for one participant	2-65
Appendix 2-E: Excerpt from table of group level themes	2-67

Section Three: Critical Appraisal

Title page	3-1
Main body	3-2
References	3-15

Section Four: Ethics Section

Title page	4-1
Ethics proposal	4-2

Appendix 4-A: Thesis protocol	4-13
Appendix 4-B: Consent form for research study	4-26
Appendix 4-C: Participant information sheet for research study	4-28

Section One: Literature Review

**Factors influencing health-related quality of life for people with dystonia: A
systematic review**

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Factors influencing health-related quality of life for people with dystonia: a systematic review

Abstract

Purpose: To conduct a systematic review of research into factors which influence health related quality of life (HRQOL) for people with dystonia.

Method: PsycINFO, PubMed, MEDLINE, AMED, CINAHL, EMBASE, IBSS and Web of Science databases were systematically searched for eligible studies, using keywords related to “quality of life” and “dystonia”. Quantitative studies which analysed the relationship of demographic, clinical and psychosocial variables to HRQOL were included. Treatment studies were excluded. Quality assessment was conducted on all eligible studies.

Results: 1788 citations were identified, of which 18 were eligible. Disease severity, depression and anxiety were consistently found to have a negative relationship with HRQOL scores. Several psychosocial variables such as body concept and self-esteem were positively related to HRQOL. A series of pervasive issues had a negative impact on the methodological quality of reviewed studies. A lack of engagement with key concepts such as HRQOL and depression may have contributed to the consistency of results and reduced the clinical applicability of the findings. The excessive use of subscales and multiple tests on the same datasets without adjustment of significance threshold may also have produced statistically significant results with limited clinical significance.

Conclusion: Future research examining associations between clinical and psychosocial variables and HRQOL should engage with key concepts and established theoretical frameworks to identify candidate variables for further study. The use of complementary research methods such as cluster analysis or qualitative approaches can also be productive in this regard. Clinicians should assess people with dystonia for non-motor symptoms and offer multidisciplinary interventions according to the idiosyncratic impact of these symptoms on individuals' quality of life.

Keywords: dystonia; dystonic disorders; health status; movement disorders; quality of life

Introduction

Dystonia is defined as a movement disorder characterised by uncontrollable, sustained or repetitive movements and postures [1]. It is the third most commonly treated movement disorder after Parkinson's disease (PD) and essential tremor. A survey of eight European countries estimated that primary dystonia (dystonia with no known exogenous cause) occurs in between 15 and 30 people in every 100,000 [2]. In practice, the term dystonia refers to two broad concepts. It describes both a motor phenomenology comprised of dystonic symptoms, and a group of neurological disorders in which the symptoms occur [3]. According to the predominant neurological model of dystonia, the disorder is caused by dysfunction of the basal ganglia involving abnormal dopaminergic activity, resulting in a lack of inhibitory control over movement [4].

Dystonic symptoms can, however, be produced by a range of causes including neurological degeneration, brain injury, substance use and infection [3].

This heterogeneity has led to a complex system of categorisation and diagnosis. Dystonia diagnoses have historically been categorised in terms of aetiology, age of onset and/or the location of symptoms in the body [5, 6, 7]. When categorisation is based upon aetiology, diagnoses are divided into idiopathic, inherited (genetic) and acquired types. The most common of these is idiopathic, although as knowledge of genetics increases, previously unknown causes are revealed to be inherited. In cases where there is an obvious exogenous cause of dystonia symptoms such as infection or head injury, the disorder is referred to as acquired, secondary or non-primary dystonia [8].

Generalised dystonia is diagnosed when spasms occur in multiple areas of the body. These spasms can continue to occur when the person is at rest, and can lead to visibly distorted posture. Historically the term dystonia has been used colloquially to

describe this general form, whereas focal disorders have been given specific diagnostic labels [9]. Focal dystonia is diagnosed when effects are mainly localised to a particular body part or area. It is the most common type of primary dystonia: a US study found that approximately ten times as many people were diagnosed with focal dystonia compared to generalised dystonia [10]. Cervical dystonia, or torticollis, is the most common subtype of focal dystonia and is characterized by the twisting of the neck and head [11]. Another major subtype of focal dystonia is blepharospasm which involves repetitive contractions of the eyelids and surrounding muscles, often resulting in visual disability [12]. Segmental dystonia is diagnosed when spasms occur in multiple, adjoining locations. Cranial dystonia, for example, may be diagnosed if multiple areas of the head are affected. Dystonia which begins in childhood is more likely to have a discoverable cause, and to progress from focal to generalized symptoms. Adults are more likely to develop long term focal dystonia, although the age at which specific subtypes are most common varies. Cervical dystonia, for example, is most likely to occur in people over the age of 40, whereas blepharospasm is most likely over the age of 50 [3, 13, 14].

Available treatment options are most commonly aimed at ameliorating the distinctive motor symptoms of dystonia [15]. It is difficult for clinicians to make recommendations on an individual basis because the efficacy of treatments varies significantly based upon manifestation of symptoms and aetiology. Botulinum toxin (BTX) injections are a common choice and lead to a reduction in muscle spasms in a large proportion of patients with focal dystonia, with only minor side effects [16]. Research suggests that the benefits from an injection last for approximately three months, and continue for approximately six injections [17]. For people with enduring symptoms for whom BTX is not a viable option, deep brain stimulation of the globus

pallidus interna (DBS) can be considered [18]. Evidence suggests it can reduce symptoms for between three to ten years and is particularly effective at treating generalised and segmental dystonia [16]. In some people however, DBS can actually exacerbate symptoms [19]. Alternative pharmaceutical treatments include anticholinergics, benzodiazepines and antispasmodics, although these are ineffective for many and can lead to unpleasant side effects [20]. Research into physical and psychological therapies shows that, although participants are positive about the impact of these interventions, there is currently little evidence of their long term effectiveness [21].

The neurological model of dystonia has been dominant in both research and clinical practice since the 1970s, expanding in more recent times to include genetic components [22]. In medical settings the ability of this model to inform differential diagnosis has been limited by the multitude of possible causes for dystonic symptoms and the range of different disorders which manifest in similar ways [23]. Misdiagnoses resulting from an inability to comprehensively describe the condition or its causal elements has a negative impact upon both clinicians and people with dystonia [24]. Discussion of the issue of categorisation led to a consensus statement from researchers, wherein the phenomenology of dystonia was given equal weighting alongside aetiology in a new system of categorisation [3]. In practice, this model encourages clinicians to focus on the symptoms experienced by their patients as well as putative neurological or genetic causes when considering treatment and diagnostic categorisation.

This change in emphasis has coincided with the increasing popularity of health related quality of life (HRQOL) as a measure of the impact of health conditions [25]. HRQOL aims to assess the extent to which health influences an individual's ability to function, as well as their subjective perceptions of health across the three broad areas of

physical, psychological, and social wellbeing [26, 27]. The concept of health in this context includes the idea of optimal living and not just the absence of disease [28]. The subjective aspect of HRQOL entails that the individual's evaluation and interpretation of their quality of life in their particular context is given priority over objective ratings of wellbeing derived from techniques such as observation [29]. HRQOL represents a pragmatic, person-centred alternative to traditional ways of measuring the severity of a disorder because it takes the experience of the individual as an end goal, and not simply as a means of identifying underlying biological issues [30]. As such, the HRQOL concept involves recognition that therapeutic interventions should not only be aimed at ameliorating symptoms, but also at enabling the individual to live as optimally as possible [31]. HRQOL assessment can complement a phenomenological conception of dystonia by determining the severity of symptoms and priorities for treatment in terms of the impact it has on the individual's life, according to their own priorities and personal context [32]. This recognition of wider context makes HRQOL assessment well suited to dealing with the complexity inherent in what the research refers to as "non-motor symptoms", such as depression or social isolation.

There are other advantages which have contributed to an increase in the popularity of HRQOL assessment as a means of measuring not only service user experience but also clinical effectiveness and value [33, 34]. Self-report questionnaires of the kind used in HRQOL assessment are more effective than clinical interviews in assessing individuals' perceptions of their own wellbeing [35]. They are also more reliable than clinical interviews due to their predetermined structure and fit more readily into the dominant quantitative paradigm for healthcare research [36]. The most commonly used measure in HRQOL studies is the 36-item short-form health survey (SF-36) [37]. This is a self-administrated questionnaire consisting of eight multi-item

dimensions, covering emotional wellbeing, functional status and overall evaluation of health. Physical (PCS) and mental component scores (MCS) are generated by calculating the mean of relevant items. The ubiquity of the SF-36 is attributable in part to the large number of translations and adaptations available for different populations [28]. The popularity of the measure makes it an attractive choice for research because of the ease of comparison with other studies. Although the SF-36 has well established norms, its factor structures have not yet been tested with a dystonia population [38].

Disease specific questionnaires such as the Cervical Dystonia Impact Profile (CDIP-58) [39] and Craniocervical Dystonia Questionnaire (CDQ-24) [38] have become viable options for assessing HRQOL in the past decade. The structures used in these questionnaires are predicated on interviews with people with dystonia. The application of the HRQOL concept to people with disabilities is particularly complex due to the growing recognition of the social aspects of disability, in addition to any physical or functional impairment [40]. Bespoke questionnaires designed with this population in mind are more likely to be responsive to the holistic impact of ill health and disability on HRQOL [27]. In the case of dystonia, the growing recognition of the importance of non-motor symptoms demands that the conceptual model of the illness underlying the formulation of HRQOL questionnaires be one which is sensitive to this wider impact [39].

Both the CDIP-58 and the CDQ-24 have good levels of reliability, validity and sensitivity [38, 41]. The CDIP-58 is specifically for use with people with cervical dystonia and has eight subscales measuring head and neck symptoms, pain and discomfort, upper limb activities, walking, sleep, annoyance, mood and psychosocial functioning. These subscales correlate well with comparable subscales of the SF-36. The CDQ-24 is for use with people with either cervical dystonia or blepharospasm and

is comprised of five subscales measuring stigma, emotional well-being, pain, activities of daily living, and social/ family life. Its subscales also correlate well with the SF-36. Unlike the SF-36, the CDIP-58 and CDQ-24 both include questions about sleep and stigma, as well as ratings of specific dystonia related physical functioning and pain [38, 42]. The CDQ-24 employs a stigma subscale and includes a sleep item in the pain subscale, whereas the CDIP-58 employs a sleep subscale and includes several stigma related items as part of the psychosocial functioning subscale.

This review aims to gather research into factors related to HRQOL for people with dystonia, to present an overview of key findings and to assess methodological rigour. It will discuss ways in which existing research designs can be improved and how complementary methods can enhance understanding of this topic. It will also discuss how this information can inform assessments and interventions aimed at improving HRQOL and rehabilitation for people with dystonia.

Method

Search strategy

A systematic search of eight databases was conducted from their inception to the 16th of August 2017: PsycINFO, PubMed, MEDLINE, AMED, CINAHL, EMBASE, IBSS and Web of Science. Google Scholar and the reference lists of relevant studies were also searched. Database thesauruses and Medical Search History (MeSH) terms were searched for key words relating to quality of life and dystonia. Search strings were then developed for each database, in consultation with an academic librarian. The following is an example search string used with the MEDLINE database: ("Dystonia"[Mesh] OR "Dystonic Disorders"[Mesh] OR "Blepharospasm"[Mesh]) AND ("Quality of

Life"[Mesh] OR "Sickness Impact Profile"[Mesh] OR "Health Status"[Mesh]). Full details of search strings are given in table 1.

[Insert table 1 around here]

Selection criteria

Quantitative studies investigating demographic, clinical and/or psychosocial variables and their relationship with HRQOL were included. There were no exclusion criteria concerning dystonia characteristics, co-morbidities, age or gender. Treatment studies which measured HRQOL pre and post intervention were excluded. This was because either the expectation or experience of change relative to previous health perceptions could confound the post-treatment assessment of HRQOL. People who were receiving ongoing treatment were not excluded as this is a common experience for people with dystonia. Only English language publications were considered.

Data extraction

Quality assessment was conducted using a bespoke tool developed by Soh et al. [32] for use in their review of factors related to HRQOL for people with PD (appendix 1-C). It is informed by the cross-sectional variant of the Strengthening the Reporting of Observational studies in Epidemiology (STROBE) guidance [43] and Hayden et al.'s guide to quality assessment in systematic reviews [44]. This tool facilitates a descriptive appraisal of several areas of study methodology related to risk of bias, internal and external validity and reliability of results. Study characteristics were extracted using a checklist adopted from the Soh et al. [32] review (appendix 1-D). Two supervisors also

performed a quality assessment of a sample of papers to check for bias.

Results

Study selection

The initial literature search returned 1788 articles; after duplicates were removed 987 remained (figure 1). The titles and abstracts of these articles were then examined, and those clearly ineligible for inclusion were removed. Full-text examination of the remaining 45 papers resulted in 18 eligible studies for review.

[Insert figure 1 around here]

Study characteristics and quality assessment

Table 2 presents the results of the quality assessment. Table 3 presents detailed information about the characteristics of each study including key findings.

[Insert tables 2 and 3 around here]

All the studies featured in this review used a cross-sectional design and investigated the relationship of a selection of demographic, clinical and psychosocial variables to self-reported assessment of HRQOL. Six studies justified their choice of variables with reference to established theoretical frameworks [45-47, 49, 54, 55]. Other studies justified their choice as either seeking to confirm results from similar research

[58, 61], or as a speculative exploration of possible candidate variables [31, 50-52, 56, 57, 59, 60]. Research objectives and study designs were generally well described across the studies.

Twelve studies were conducted in Europe [31, 45-51, 54, 55, 57, 60] two in the USA [56, 58], two in Brazil [52, 59] and two in Asia [54, 61]. The mean age of participants in studies ranged from 39.5 to 60 years old. Focal dystonia was the sole type of dystonia in ten samples [45, 48, 49, 51-55, 57, 59] and the most common overall. One study included only people with segmental dystonia [31], one only people with dopa responsive dystonia [46], and the rest used a mixed sample.

The majority of studies recruited their participants from hospitals or clinics [31, 45, 47-49, 52-54, 57, 59, 61]. Two studies recruited via patient association contact lists [55, 60] and one study used an existing epidemiological sample [50]. Two studies did not specify where their sample was drawn from [51, 56]. The majority of studies used consecutive or convenience sampling [31, 45, 43, 50-55, 57-61]. Of the six studies which employed control groups [46-49, 54, 55], one of them used random sampling [49]. Three studies did not specify the sampling technique used [46, 48, 56]. Sample sizes ranged from 19 to 351, with a median of 82. Two studies justified their sample size [55, 59]. One conducted an a posteriori analysis of two similar studies and estimated that a sample of 50 participants would have a power of 0.85 [55]. The other study conducted an a priori power analysis which found that a sample of 70 participants would generate an analytic power of 0.98, with an effect size of 0.5 [59]. Three studies had sample sizes in the range of 19 to 28 participants [31, 46, 57], which could undermine their ability to detect a significant effect. Characteristics of participants were thoroughly described in 13 of the studies [31, 45-47, 49, 50, 52, 53, 56-60]. In the five studies where questionnaires were sent to potential participants [45, 49, 50, 54, 60], the

characteristics of non-responders were not reported. Questionnaire return rates ranged from 46% [49] to 90% [54], with a median of 75%. One study with a return rate of 68.3% suggested that this could introduce a selection bias and affect the generalisability of their findings [60].

All of the studies utilised some combination of correlation and regression models for their statistical analysis. Papers which performed a large number of statistical tests using subscales of clinical and HRQOL measures were most vulnerable to the possibility of type 1 error [45-48, 51, 55, 57, 60, 61]. Simpler designs involving fewer tests were less susceptible to this issue [31, 49, 52, 53, 58, 59]. One study adjusted their significance threshold to account for multiple tests, using a p-value of <0.005 [50], while two studies utilised the Bonferroni correction [54, 56]. Assessment of the appropriateness of statistical methods and sample size was complicated by studies not reporting the number of tests conducted or non-significant results [46, 48, 52, 56, 59-61]. The issue of minimising type 1 and type 2 error is particularly relevant to these exploratory studies of dystonia due to the rarity of the condition and the desire to investigate as many candidate variables as possible. This puts the onus on researchers to be conservative with their use of subscales and to have a strong justification for their choice of candidate variables. Although there is debate about the most effective way to mitigate such problems [62], readers should exercise caution when interpreting the results of studies which did not attend to this issue.

The concept of HRQOL was given a limited definition in one of the studies [31]. Fifteen studies used generic HRQOL measures [31, 45-57, 61], the most popular of which was the SF-36. This choice was most commonly justified on the basis that it was well established in the broader area of health research, although often no explicit justification was given for its use. In some cases a specific measure was not available

for a given type of dystonia. Three studies used a specific, dystonia related measure [57, 59, 60], either the CDIP-58, CDQ-24 or the Thai version of the HFS-30 [63]. The use of these measures was justified on the basis of their increased sensitivity, although one of these studies had a mix of dystonia types in their sample, which may negate this benefit [60].

Disease related variables

Greater severity of motor symptoms was typically measured with the Toronto Western Spasmodic Torticollis Rating Scale (TWSTRS) [64] which includes subscales for severity of motor symptoms, disability and pain. Worse severity scores were consistently found to be related to worse scores on the physical subscales of HRQOL measures [31, 50-52, 54, 55, 57, 59, 61]. Three studies conducted separate tests using the pain subscale of disease severity measures [55, 57, 59]. All studies found that worse scores on the pain subscale of the TWSTRS were associated with worse scores on pain subscales of HRQOL measures. One study found that the TWSTRS subscale was correlated with worse total SF-36 scores [59]. One study created their own pain rating scale and found it was a non-significant predictor of HRQOL [31]. Another defined the variable categorically as presence of pain in the affected area. Analysis found that it explained SF-36 variance for patients with cervical dystonia and blepharospasm, but not writer's cramp [51].

The association between disease duration and HRQOL was investigated in nine studies [31, 45, 47, 49, 51, 52, 54, 56, 60] and produced inconsistent results. While most studies found no significant association [31, 49, 52, 54, 56, 60], two studies found that longer duration was linked to poorer scores on the bodily pain subscale of the SF-36 [47, 51]. In one case a similar association was found with the social functioning

subscale as well [47]. One study found that longer duration was linked to better MCS and PCS scores on the SF-36 [45].

Type of dystonia was not consistently found to have a significant relationship with HRQOL outcomes. One study found that people with cervical dystonia and blepharospasm scored significantly lower on the physical subscales of the SF-36 than those with focal hand dystonia, although there was no significant difference on the mental subscales [51]. Two other studies found no significant relationship between type of dystonia and HRQOL outcomes [50, 60]. In contrast, the one study which compared primary and secondary dystonia found that mean SF-36 scores for all but two subscales were significantly lower for secondary as opposed to primary [47]. It should be noted that this study conducted a between groups comparison on a sample of only 43 participants and so further investigation is required. With regards to treatment, no systematic differences in results were observable when comparing the findings of studies which required participants to be receiving BTX injections [48, 54, 57] with those which did not [31, 45-47, 49-53, 55, 56, 57-61].

Psychosocial variables

Depression was the most commonly investigated psychosocial variable and in each study was found to have a significant relationship with HRQOL outcomes: higher levels of depression were associated with lower levels of HRQOL [31, 45-47, 50, 51, 53-56, 61]. One study found that higher levels of self-reported psychopathology were associated with lower levels of HRQOL [48]. Anxiety was also commonly investigated, although the results were slightly more inconsistent. Five studies found a significant inverse relationship between anxiety and HRQOL outcomes [31, 45, 51, 55, 56, 61]. Two studies found a significant inverse relationship between anxiety and the mental subscales on the SF-36, but not the physical [47, 54]. One study found no significant

result [45], but it should be noted that this study had a sample size of only 23 and therefore its ability to detect a significant effect may be compromised.

Negative body concept was found to be related to poorer HRQOL outcomes. It was the most powerful explanatory variable in a regression model using PCS scores explaining 44% of variance, the second most powerful explanatory variable being functional disability at 5%. Along with depression scores, negative body concept explained 34% of variance in a model using MCS scores [50]. Negative body concept was also the second most powerful explanatory variable in a model using the EuroQol EQ-5D HRQOL questionnaire; together with functional disability it explained 42% of variance [50]. One study found a significant relationship between increased PCS and MCS scores and both higher self-esteem and lower sense of stigma [45]. Another study found no significant relationship between stigma and HRQOL but it had a relatively small sample size of only 28 participants [31].

Results showed that higher levels of physical and leisure activity were related to better HRQOL outcomes. One study found that level of physical activity explained a high proportion of variance in HRQOL scores in a regression model alongside a range of demographic variables and satisfaction with treatment [60]. One study found that increased engagement in leisure activities was correlated with higher HRQOL scores [31] and another found the same for higher levels of social participation [45]. Employment status was a significant part of regression models in three studies [50, 51, 60]. Studies found that higher levels of fatigue and sleeplessness were linked to lower HRQOL scores on a range of scales in correlation and regression analysis [46, 56, 58].

Demographic variables

Regarding gender differences, females were found to have worse HRQOL outcomes than males in two studies [60,61], although five studies found no significant difference

between genders [31, 50, 51, 54, 56]. The only Chinese study in the review [61] suggested that perhaps cultural differences could explain their result as norms were taken from a rural Chinese population. The European study which found a similar result [60] speculated that perhaps the stigmatising effect of dystonia was more powerful for women than men. It should be noted that this study was the only one which used specialised cervical dystonia measures with multiple other dystonia types and therefore this result should be interpreted with caution. Non-significant results were not consistently reported, but the relationship between age [31, 51, 53, 54, 56] and marital status [31, 51, 54] and HRQOL was commonly found to be non-significant.

Discussion

Disease characteristics

Severity of motor symptoms was the one disease characteristic which was consistently found to have a significant negative relationship with HRQOL. Investigation of the disease duration variable produced contradictory findings which may be due to a number of factors. In degenerative movement disorders such as PD, symptoms are expected to become more severe over time and thus reduce HRQOL. In dystonia however, symptoms typically do not worsen to the same extent. Some researchers have suggested that in dystonia increased duration of disease may facilitate psychological adjustment [65, 66]. For people experiencing life changing levels of functional disability, Lee et al. [49] hypothesised that a shift in demand may occur, and over time an individual may readjust their self-concept and goals accordingly. Lee et al. invoked the theory of the hedonic treadmill to explain how people may adjust to adverse circumstances in a relatively short amount of time and then refocus their concerns on

meeting their ongoing existential needs, given the appropriate levels of support or resilience. As factors such as resilience were not commonly included in the statistical analysis of reviewed studies, it is difficult to speculate about their impact on HRQOL outcomes. The two studies which found that longer duration was linked to reduced HRQOL did so on a maximum of two subscales of the eight tested, and no composite or global scores were measured. This is a complex issue, and further research into specific psychosocial factors which potentially mediate the impact of disease duration on HRQOL is needed before firm conclusions are drawn.

Type of dystonia was not consistently found to have a significant relationship with HRQOL outcomes. This was the variable most affected by the size and composition of the samples used: in cases where the sample was large enough to justify such analysis, the balance of dystonia type subgroups was often insufficient to allow for meaningful comparisons. This could explain the variety in results and future research must account for this. The current lack of clarity surrounding this issue lends support to the idea that, as per the phenomenological model of dystonia [3], clinicians should focus on the presence of specific symptoms, rather than using the particular diagnosis of dystonia to direct their interventions [60].

Psychosocial variables

Both depression and anxiety were found to have a negative relationship with HRQOL for people with dystonia. This finding is in keeping with similar studies of people with PD [32]. Commonly the studies in this review recommended that psychological therapy and in some cases, pharmaceutical intervention, should be considered for people with dystonia who experience significant depression and anxiety. The ability to make causal attributions is limited however, due to the cross-sectional nature of the research.

There was a lack of engagement with the concepts of depression and anxiety in reviewed studies. None of the studies defined depression and anxiety, except in terms of scores on psychometric tests, and it was common for studies to use measures for which dystonia specific norms are not well established [22]. Leentjens et al. [67] advise that researchers exercise caution when making inferences based on the norms of measures such as the Beck Depression Inventory (BDI) [68] and Beck Anxiety Inventory (BAI) [69] as studies of PD found that symptoms of the condition confounded the measures significantly. They warn that the use of non-specific norms can result in an unacceptably high rate of over-attribution of depression. This represents a potential risk because there is a lack of evidence for the safety and efficacy of psychotropic medication for people with dystonia [70].

It was typical for studies to speculate that depression and anxiety may play a causal role in the impairment of HRQOL in subjects, as “co-morbidities”. This view is in keeping with a medical model perspective, however a biopsychosocial model of depression and anxiety may conceptualise them as being outcomes caused by the interaction of negative experiences and intermediary psychosocial variables [71], and therefore as inappropriate for use as predictor variables in regression models. Researchers have also suggested that the negative bias inherent in depression may confound the use of self-report questionnaires in general [15]. In some cases, the ability for depression and anxiety to explain variance in models of regression may actually hinder the aim of studies to develop an understanding of a variety of variables related to HRQOL. One study attempted to develop this understanding by testing a range of candidate variables such as self-esteem, social support and social participation, and HRQOL scores, finding numerous significant correlations [45]. However, when these variables were entered into a regression model with depression and anxiety as co-

predictors, these additional psychosocial variables either had a reduced effect or lost significance. Conceptual overlap between variables can reduce sensitivity in results and mask useful information. There is the possibility of collinearity in such designs, which could decrease statistical power and lead to the exclusion of relevant predictor variables when generating models [72, 73]. Only one study reported checking for collinearity [55], and this should be considered in future research.

One study in this review concluded that the overall power of their explanatory models was limited, even with the inclusion of depression and anxiety, and that this suggests a range of factors not yet investigated [60]. Similarly, a treatment study of people with dystonia found that, although depression was correlated with HRQOL before BTX injections, neither it nor any other independent variable predicted improvement after treatment. They concluded that this may reflect a complex, multimodal interaction of variables [25]. In order for more nuanced theoretical models of psychosocial processes to be developed it is crucial that the impact of specific psychosocial factors such as self-esteem are not hidden by overarching concepts such as depression. Developing knowledge of more specific variables can help clinicians to target and tailor person-centred interventions.

Studies found that higher levels of physical, social and leisure activity lead to better HRQOL outcomes. The negative functional effect of motor symptoms on levels of physical activity is well established, for example leading to reduced activity due to a fear of falling or exacerbating pain [60]. The impact of non-motor symptoms on reduced physical and social activity related to dystonia is less well established and requires further examination [74]. One relevant concept to investigate further is that of stigma, which entails that the fear of negative evaluation by others and can lead to avoidance of social contact [75]. One study in this review found a significant

relationship between stigma and HRQOL outcomes [45]. Research has found that social phobia is one of the most commonly diagnosed psychiatric conditions for people with dystonia [22]. This suggests that psychosocial variables may help to explain why some people with dystonia become less physically and socially active. Better understanding of these variables can help to inform decisions about intervention aimed at increasing levels of activity.

Some researchers have hypothesised that depression and dystonia may co-occur because of a shared casual mechanism, namely dysfunction of the basal ganglia [55]. Even if one accepts the premise that depression is a disease with a biological aetiology, verification of this hypothesis requires significant advances in neuroscience and understanding of the basal ganglia. Until then it remains somewhat speculative.

Methodological quality

The methodological quality of reviewed studies was negatively affected by consistent issues such as the frequent use of multiple tests without adjusting significance thresholds and a lack of engagement with core concepts such as HRQOL and depression. Although key findings were generally consistent across studies, it is possible that conceptual issues such as the overlap of clinical and psychosocial measures with HRQOL questionnaires may explain some of this consistency.

Implications

This review confirms that non-motor symptoms such as depression and anxiety have a significant negative relationship with HRQOL for people with dystonia, and that clinicians should direct their assessments and interventions to ameliorate these symptoms. Enacting the recommendations of the reviewed studies on an individual basis requires more specific information about the factors influencing the impact of

non-motor symptoms [76]. Further research using the conventional cross-sectional design should focus on engaging with established theoretical bases and eliciting from these a range of prospective variables for further investigation. Several reviewed studies demonstrated how these research designs could use existing theoretical frameworks to inform their selection of variables [45, 49, 56]. Further engagement with core concepts can also improve the applicability of results. One example of this would be rethinking the use of HRQOL measures and their overlap with other clinical and psychometric tests.

HRQOL

Engagement with the concept of HRQOL was lacking in reviewed studies, an issue which is common in health research [77]. A more thorough examination of HRQOL can help researchers to apply and analyse the concept more effectively [36]. One of the most consistent issues which was not discussed by reviewed studies is the conflation of Health status (HS) and HRQOL in questionnaires such as the SF-36 [77]. Unlike the concept of HS, the concept of HRQOL is inherently individual and subjective [78]. HS measures the health perceptions of individuals as they relate to symptomatology, functionality and social conditions, rather than internal experiences. Objective questions such as how many times someone engages in a particular activity overlap with functional disability questionnaires, for example, and could confound results. Oudsten et al. [77] argue that HRQOL questionnaires should ask participants about their attitudes regarding reduced levels of physical activity, for example. These insights can help clinicians to prioritise interventions on the basis of the importance of symptoms to people with dystonia.

The SF-36 remains the most popular HRQOL measure, partly for the ease of comparisons with other clinical populations. There is evidence to suggest however, that

specific assessments may be more sensitive to particular features of dystonia [20], something which is particularly important when working with smaller samples, as is often the case with rarer conditions. For example, disfigurement, negative body image, and poor self-esteem are commonly experienced by people with dystonia [78] and are not covered in the SF-36 [38]. The impact of these features is thought to contribute to the floor and ceiling effects which are present when using the SF-36 with a dystonia population [79]. These effects are most commonly observed with the subscales which assess physical and social functioning. Researchers have argued that it is a fundamental misconception to equate the loss of capacity to engage in functional activities to HRQOL, and that through environmental modifications and a change in psychological relationship to remaining functional ability, people can mitigate the effect on their HRQOL [27].

The adoption of HRQOL measures for specific dystonia types such as the CDQ-24 and CDIP-58 is relatively recent and validated measures do not yet exist for many types of dystonia. One study in this review that used these specific measures did so with subtypes of dystonia for which they were not intended [60], which may negate any advantage in sensitivity [42]. A wider range of validated, dystonia specific measures are needed. Besides using dystonia specific measures, future studies could also further explore the concept of HRQOL by using more person-centred measures such as the Measure Yourself Medical Outcome Profile (MYMOP) [80]. The MYMOP asks respondents to rate symptoms and activities which are most important to them, as well as their wider psychosocial resources. Mallinson [81] argues that the use and validation of standardised questionnaires such as the SF-36 has become ritualistic and ceased to engage with the key issue of meaning, leading to misleading self-report scores and

distorted communication. The MYMOP is more sensitive to change than the SF-36 and its use was found to improve communication between clinicians and their patients [80].

Other designs

Different research designs can complement those used by studies in this review and help to identify potential variables for further investigation. One example is provided by Graham et al. [82], who conducted a cluster analysis to determine which groups of interrelated illness perceptions coalesced into schemata for people with various muscle diseases. This is a useful complimentary method as it has the potential to investigate patterns of psychosocial variables within individuals, which can facilitate discussions of differences between individuals. Graham found three common patterns of illness perceptions amongst participants. The “realistic” schema involved people with more severe symptoms who had appropriately lower expectations for treatment and personal control, and a stronger emotional representation of their illness. These people had the lowest HRQOL and mood. The other two groups shared similar, lower levels of illness severity, and were labelled the “helpful” and “unhelpful” schemata. The “helpful” group had a greater sense of self-control compared to the “unhelpful group”. They showed a better understanding of the illness, identified with it less and gave it a weaker emotional representation. According to Leventhal’s Self-Regulation Model [83], illness perceptions influence the coping strategies that people adopt and the emotional impact of their illness, and therefore can provide vital information to direct psychological intervention. Investigating the relationship between illness perceptions and HRQOL for people with dystonia could be a useful avenue to pursue in future research.

Qualitative methods can also play an important role in developing knowledge of potential key variables, as it can emphasise the subjective experience of individuals and

work with the complexity inherent in discussions of individual differences [84].

Papathanasiou et al. [74] provide an example of how qualitative methods can enhance quantitative approaches, by using a two stage research design wherein interviews were used to inform the development of a bespoke stigma questionnaire that was responsive to the concerns of participants. The qualitative data they collected allowed them to compare specific aspects of the experience of stigma with studies of other populations and led them to conclude that there are significant differences, for example, people with dystonia were more likely to feel apologetic for their condition. They concluded that for psychosocial variables such as stigma, there may be a complexity and individualised impact which requires nuanced analysis. As with person-centred HRQOL measures, allowing participants to articulate which areas are more significant to them can produce results which are more reflective of the idiosyncratic impact of non-motor symptoms.

Clinical implications

Research into other movement disorders such as PD can provide direction for dystonia specific interventions and research. Research evaluating pain interventions for people with PD recommends a multidisciplinary approach combining physical therapy, education and consultation with appropriate specialists [85]. In the area of sleep, studies of people with PD also found that the majority of people with PD experience significant sleep disturbance [86] and that this has a negative impact on HRQOL [87]. Research is still developing in this area but there is preliminary evidence that pharmaceutical and therapeutic interventions such as cognitive behaviour therapy (CBT) may be helpful, depending on appropriate person-centred assessment [88]. People with dystonia should be assessed for sleep problems and, if appropriate, their feedback used to inform sleep hygiene interventions [89]. This review also recommends that health services consider

helping people with dystonia to engage in social activities. More dystonia specific research needs to be conducted on the impact of such interventions, but studies of people with PD suggest they could be beneficial [90]. Considering the role of identity and stigma, alongside the impact of functional disability and pain in reduced levels of activity, holistic, multidisciplinary intervention is appropriate [15].

Studies which investigated depression and anxiety were virtually unanimous in finding that this has a significant negative relationship with HRQOL for people with dystonia. People who have been diagnosed with dystonia should be offered a psychological assessment to determine if they would benefit from psychological intervention. Given the complexity of the impact of dystonia, it is appropriate for this assessment to be conducted by a clinical psychologist [91]. Other psychosocial variables identified by this review such as self-esteem and social engagement could also be investigated as part of a broader psychological formulation. Psychological formulation is a process which aims to construct a working model of an individual's presenting issue. This model is co-constructed by the clinician and the service user and takes into account social and psychological factors as well as the individual's wider context [92]. The use of formulation in complex cases has been shown to improve the efficiency of treatment as part of integrated, proactive care and to minimise unhelpful or inappropriate intervention [91]. The HRQOL concept could be utilised to facilitate discussion and reflection on the subjective impact of dystonia and the relative importance of different aspects of health to the service user. These topics could then be given priority as part of a holistic care plan and used to inform multidisciplinary intervention targeting, for example, sleep issues or pain management. Clinical psychologists could work directly with service users involved in such interventions to help them make decisions about their treatment and to adhere to treatment plans, or to

provide specific therapeutic intervention aimed at ameliorating experiences of depression or anxiety [93].

Psychological formulation and case consultation can assist multidisciplinary teams (MDTs) by increasing understanding of a specific service user and facilitating person centred care [94]. Indirect work such as this can also help to ensure that intervention is consistent and goal directed across different parts of the MDT [91]. Generally speaking, MDTs could also benefit from training provided by a clinical psychologist aimed at enhancing their ability to work with a client group who often have complex psychosocial needs, as is the case with people with dystonia [95]. Additionally, complex cases such as these can be emotionally challenging for health professionals due to frustrations with providing effective intervention and subsequent feelings of uncertainty and inadequacy [96]. Through supervision and consultation clinical psychologists can provide health professionals with the opportunity to reflect on these challenges in an emotionally supportive environment [95].

Enacting the common recommendation of psychological intervention aimed at ameliorating distress is problematic because of a lack of research into these interventions with this specific population. A systematic review of behavioural interventions for people with dystonia found the evidence in favour to be limited, largely due to poor methodological quality [97]. They said that the lack of evidence may also be attributed to adult onset dystonia often being misdiagnosed as an emotional illness, for which behavioural interventions are considered inappropriate. Relaxation was the technique with the strongest evidential support. A case study for CBT found a reduction in both BDI and BAI scores, and in self-reported motor symptoms [98]. The final follow up assessment was after six months so it is unclear how enduring the effect is, or how generalisable the findings are. Sandhu et al. [99] conducted a pilot study for a

combined program of CBT and mindfulness. The study provided no inferential statistics, but there was a general trend of reduction in non-motor symptoms and increased wellbeing. Only nine of the 28 original participants completed the treatment, suggesting a possible self-selection bias.

Another potential intervention which warrants further investigation is Acceptance and Commitment Therapy (ACT), particularly as it relates to increased psychological flexibility. Graham et al. [100], conducted a study which found that higher levels of psychological flexibility, being present in the here-and-now, were related to higher levels of HRQOL for people with muscular dystrophy. The researchers recommended ACT as an intervention which can increase psychological flexibility due to its emphasis on staying in the present moment and accepting challenging thoughts as transient events separate from one's role as a detached thinker. A case study of ACT for functional movement disorders found that it improved symptom intensity and mood, and enabled the participant to engage more fully in personally meaningful activity [101]. The authors suggested that these positive effects may have been partly attributable to an improvement in psychological flexibility. A systematic review of ACT for chronic disease concluded that although there was a lack of high quality research, it was a potentially productive area for exploration [102].

The cluster analysis conducted by Graham et al. [82] of illness schemata for people with muscle disease highlights three main targets which could help to reduce depression and anxiety in people with dystonia. Firstly, intervention could address beliefs about the nature of the disease, the impact of treatment, time course and expectations of the future. This can be accomplished through education about dystonia and related psychological processes, something which has already proven to be effective in helping people to manage the emotional impact of other chronic health

conditions [103]. Secondly, it could reduce the strength of emotional representation of the disease. One way to achieve this could be to use CBT to challenge negative thought patterns and their impact on emotion, something which has been successfully utilised by people with PD [104]. Another approach would be to teach skills such as mindfulness to help people with dystonia to manage their emotional relationship with the illness, which has also been successfully utilised in many health conditions [105,106] and which has preliminary evidence for its efficacy in the context of dystonia [99]. Thirdly, intervention could target the impact of illness perceptions on identity and values. The concept of a “spoiled identity” is useful in understanding the impact of chronic ill health on the psychological wellbeing and HRQOL of people with dystonia [74]. When diagnosed with a health condition, people can come to associate themselves with negative stereotypes related to the condition. Research into people coping with various chronic health conditions has suggested that intervention aimed at adjusting these perceptions of illness and identity can have a positive impact on psychological wellbeing [107]. Psychological intervention can potentially help individuals to integrate their experiences into their identity concept, to respond to changing demands and to adjust their values and aims accordingly [84]. Replicating this research with people with dystonia is a necessary next step.

Regardless of what specific interventions are found to be useful for people with dystonia, these methods should be applied in response to a person-centred assessment and formulation [108]. This approach has been shown to improve clinical outcomes and satisfaction for service users [92, 109]. It can also increase understanding about individuals’ values and goals, and the way these interact to comprise perceptions related to HRQOL. HRQOL assessment can itself be a clinical tool to inform psychological formulation, as it creates opportunity for discussion of factors such as resilience and the

impact of early experiences and culture [35]. As well as emphasising the experience of the service user, a HRQOL approach to the assessment of dystonia focuses on a holistic sense of wellbeing involving factors such as autonomy, social support and self-efficacy. It therefore requires that the clinician be responsive to the concerns of service users when determining the most helpful course of action, rather than using the service user's experiences as a means to make a diagnosis and then prescribe an intervention in a top-down manner [34]. Service user feedback indicates that they are primarily interested, not in classification or diagnosis, but rather in the wider impact of chronic illness on their ability to live the lives as they wish [36]. The HRQOL concept provides psychologists, and clinicians generally, with the opportunity to listen and be responsive to concerns of the service user. This knowledge should form the basis of further intervention.

Limitations

Similarities between HRQOL outcome measures and measures of predictor variables represent a confound in reviewed designs. Some questions on depression and anxiety psychometrics such as the BDI and BAI are practically identical to those found in the mental component subscales of the SF-36 and the emotional wellbeing and social life subscales of the CDQ-24. The CDIP is less affected by this issue due to questions generally being presented in a disease specific context, for example, 'during the past 2 weeks, how often has spasmodic torticollis caused you to feel depressed?' This issue of confounded questionnaires extends to other predictor variables such as functional disability, which overlaps with the physical functioning subscales of HRQOL questionnaires, and pain which has directly analogous subscales in each of the three main HRQOL questionnaires. These variables were amongst those most consistently

found to be related to HRQOL in reviewed studies. This overlap of measures may explain some of this consistency and results should be interpreted with this in mind.

Some studies were vulnerable to high risk of type 1 error due to excessive testing of their samples. The use of multiple outcome measures, testing of subscales, component scores and total scores on HRQOL measures, and the use of a large amount of predictor variables all contributed to this issue. The problem of multiple testing was exacerbated by a lack of a priori justification for selected variables, predictions about regression models and hypotheses [110]. The lack of reporting of non-significant results also contributes to the issue of type 1 error rates in this area. Only three studies aimed to mitigate risk of type 1 error through adjustment of significance thresholds. Failure to respond to these concerns makes interpretation of the data more difficult and undermines the process of null hypothesis testing as a whole [111]. Simmons [112] suggests that in an exploratory research area these issues might be difficult to fully avoid, for example the use of multiple outcome measures is in keeping with recommendations [28], and because of this researchers should interpret and report their results with explicit reference to these issues. Reporting should feature clear description of the design and analysis, explicit justification for methodological choices and the self-identification of studies as exploratory [113]. This is something future studies in this area can improve upon.

Type 2 error was also an issue for some reviewed studies, as is often the case in rare disease research due to a lack of potential participants [114]. The methods of correlation and regression are suitable for these research questions, but their operation assumes relatively large sample sizes. In order to mitigate this requirement in rare disease studies, it is imperative that hypotheses and power calculations are made a priori, that reporting is clear and comprehensive, and that results are cautiously

interpreted in the context of a potentially under powered study [115]. Reviewed studies with small sample sizes did typically report this as a limitation, although only one study reported conducting an a priori power calculation, with another conducting a post hoc calculation. There is evidence to suggest that a priori power analyses are preferable to post hoc ones as the latter are often biased and ignore the confidence intervals of the observed effect size [116]. A priori calculation is recommended for future research in this area, particularly when working with restricted sample sizes.

All the studies in this review were cross-sectional in design and therefore the ability to make causal attributions is limited. Evidence suggests that generic HRQOL measures are not optimal for studying the dystonia population [117], and their popularity in reviewed studies could introduce a systematic bias in results. There was inconsistency in the use of these measures, with studies alternatively reporting subscale, composite and global scores, making direct comparison between studies more difficult. Some studies investigated the relationship of HRQOL and key variables as a secondary objective, and so did not report or analyse results in full detail. This contributed to a lack of clarity around the reporting on non-significant results and of effect sizes. Although grey literature collections were searched, publication bias could be considered as a limitation.

Conclusion

Disease severity, depression and anxiety were consistently found to have a negative relationship with HRQOL. Several psychosocial variables such as body concept and self-esteem were also implicated. Future research should seek to confirm these findings and to expand the range of psychosocial variables being investigated. These variables should be drawn from theoretical frameworks which aim to explain how they mediate

the impact of negative life experiences on HRQOL. Engagement with core concepts such as HRQOL, depression and anxiety can better inform the use of appropriate measures and research designs. Complementary research methods such as qualitative analysis can provide additional insight into potential candidate variables and their idiosyncratic relationship with HRQOL. Person-centred assessment of non-motor symptoms should be considered a priority for clinicians. These assessments should be concerned with and informed by the experiences and priorities of the person with dystonia. Multidisciplinary interventions should target the factors which are found to be most impactful.

Declaration of interest

The author reports no conflicts of interest.

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Table 1. Database search matrices.

Search matrix (PubMed)

Focus 1	Focus 2
Dystonia	Health Related Quality of Life
<ul style="list-style-type: none"> • “Dystonia”[Mesh] • “Dystonic Disorders”[Mesh] • “Blepharospasm”[Mesh] 	<ul style="list-style-type: none"> • “Quality of Life”[Mesh] • “Sickness Impact Profile”[Mesh] • “Health Status”[Mesh]

Search matrix (MEDLINE)

Focus 1	Focus 2
Dystonia	Health Related Quality of Life
<ul style="list-style-type: none"> • MH “Dystonic Disorders” • MH “Dystonia” • MH “Blepharospasm” 	<ul style="list-style-type: none"> • MH “Quality of Life” • MH “Sickness Impact Profile” • MH “Health Status”

Search matrix (CINAHL)

Focus 1	Focus 2
Dystonia	Health Related Quality of Life
<ul style="list-style-type: none"> • MH “Dystonic Disorders+” • MH “Blepharospasm” • MH “Torticollis” 	<ul style="list-style-type: none"> • MH “Quality of Life+”

Search matrix (PsycINFO)

Focus 1	Focus 2
Dystonia	Health Related Quality of Life
<ul style="list-style-type: none"> • DE “Muscular Disorders” • DE “Movement Disorders” • DE “Torticollis” • Blepharospasm 	<ul style="list-style-type: none"> • DE “Quality of Life” • DE “Life Satisfaction”

Search matrix (AMED)

Focus 1	Focus 2
Dystonia	Health Related Quality of Life
<ul style="list-style-type: none"> • DE “Muscular Disorders” • DE “Torticollis” • Blepharospasm 	<ul style="list-style-type: none"> • DE “Quality of Life” • DE “Life Satisfaction”

Search matrix (EMBASE)

Focus 1	Focus 2
Dystonia	Health Related Quality of Life
<ul style="list-style-type: none"> • *dystonia/ 	<ul style="list-style-type: none"> • *"quality of life"/

Search matrix (IBSS)

Focus 1	Focus 2
Dystonia	Health Related Quality of Life
<ul style="list-style-type: none"> • Dystonia • Blepharospasm 	<ul style="list-style-type: none"> • “Quality of life” • “Life satisfaction”

Search matrix (Web of Science)

Focus 1	Focus 2
Dystonia <ul style="list-style-type: none">• Dystonia• Blepharospasm• Torticollis	Health Related Quality of Life <ul style="list-style-type: none">• “Quality of life”• “Life satisfaction”

Within focus terms are combined with OR. Focus 1 and 2 are combined with AND

Table 2. Quality assessment.

Study	Research objectives	Study design	Participants	Non-responders	Sampling method
Basurovic, et al., 2012 [31]	Stated	Described	Described	N/A	Described
Ben-Shlomo, et al., 2002 [45]	Stated	Described	Described	Not Described	Described
Brüggemann, et al., 2014 [46]	Stated	Described	Described	N/A	Limited
Degirmenci, et al., 2013 [47]	Stated	Described	Described	N/A	Described
Gündel, et al., 2007 [48]	Stated	Described	Limited	N/A	Limited
Lee, et al., 2015 [49]	Stated	Described	Described	Not Described	Described
Page, et al., 2007 [50]	Stated	Described	Described	Not Described	Described
Pekmezovic, et al., 2009 [51]	Stated	Described	Limited	N/A	Limited
Queiroz, et al., 2011 [52]	Stated	Described	Described	N/A	Described

Setthawatchara- wanich, et al., 2011 [53]	Limited	Described	Described	N/A	Described
Skogseid, et al., 2007 [54]	Stated	Described	Limited	Not Described	Limited
Smit, et al., 2016 [55]	Stated	Described	Not Described	N/A	Limited
Soeder, et al., 2009 [56]	Stated	Described	Described	N/A	Described
Tomic, et al., 2016 [57]	Limited	Described	Described	N/A	Described
Wagle Shukla, et al., 2015 [58]	Stated	Described	Described	N/A	Described
Werle, et al., 2014 [59]	Stated	Described	Described	N/A	Described
Zetterberg et al., 2009 [60]	Stated	Described	Described	Not Described	Described
Zhang, et al., 2010 [61]	Limited	Limited	Limited	N/A	Described
	Stated	Described	Described	Described	Described
	Limited	Limited	Limited	Limited	Limited
	Not Stated	Not Described	Not Described	Not Described	Not Described

Study	Sample size	HRQOL definition	HRQOL instrument	Clinimetrics	Variables description
Basurovic, et al., 2012 [31]	Not Justified	Limited	Limited	Insufficient	Described
Ben-Shlomo, et al., 2002 [45]	Not Justified	Not Defined	Not Justified	Insufficient	Described
Brüggemann, et al., 2014 [46]	Not Justified	Not Defined	Not Justified	Insufficient	Described
Degirmenci, et al., 2013 [47]	Not Justified	Not Defined	Not Justified	Insufficient	Described
Gündel, et al., 2007 [48]	Not Justified	Not Defined	Not Justified	Insufficient	Not Described
Lee, et al., 2015 [49]	Not Justified	Not Defined	Limited	Insufficient	Described
Page, et al., 2007 [50]	Not Justified	Not Defined	Limited	Adequate	Limited
Pekmezovic, et al., 2009 [51]	Not Justified	Not Defined	Not Justified	Insufficient	Limited
Queiroz, et al., 2011 [52]	Not Justified	Not Defined	Not Justified	Insufficient	Described

Setthawatchara- wanich, et al., 2011 [53]	Not Justified	Not Defined	Not Justified	Insufficient	Described
Skogseid, et al., 2007 [54]	Not Justified	Not Defined	Limited	Insufficient	Described
Smit, et al., 2016 [55]	Justified	Not Defined	Not Justified	Insufficient	Described
Soeder, et al., 2009 [56]	Not Justified	Not Defined	Limited	Adequate	Described
Tomic, et al., 2016 [57]	Not Justified	Not Defined	Limited	Insufficient	Described
Wagle Shukla, et al., 2015 [58]	Not Justified	Not Defined	Limited	Insufficient	Described
Werle, et al., 2014 [59]	Justified	Not Defined	Justified	Insufficient	Described
Zetterberg et al., 2009 [60]	Not Justified	Not Defined	Limited	Adequate	Limited
Zhang, et al., 2010 [61]	Not Justified	Not Defined	Limited	Insufficient	Not Described
	Justified	Defined	Justified	Adequate	Described
	Limited	Limited	Limited	Limited	Limited
	Not Justified	Not Defined	Not Justified	Insufficient	Not Described

Study	Variables selection	Statistical analysis	Key results	Limitations
Basurovic, et al., 2012 [31]	Limited	Limited	Stated	Not Described
Ben-Shlomo, et al., 2002 [45]	Justified	Appropriate	Stated	Described
Brüggemann, et al., 2014 [46]	Justified	Limited	Stated	Limited
Degirmenci, et al., 2013 [47]	Justified	Limited	Stated	Limited
Gündel, et al., 2007 [48]	Not Justified	Unable to Determine	Stated	Limited
Lee, et al., 2015 [49]	Justified	Appropriate	Stated	Described
Page, et al., 2007 [50]	Limited	Appropriate	Stated	Described
Pekmezovic, et al., 2009 [51]	Limited	Appropriate	Stated	Limited
Queiroz, et al., 2011 [52]	Limited	Appropriate	Stated	Described

Sethawatchara- wanich, et al., 2011 [53]	Not Justified	Appropriate	Stated	Not Described
Skogseid, et al., 2007 [54]	Justified	Limited	Stated	Not Described
Smit, et al., 2016 [55]	Justified	Limited	Stated	Described
Soeder, et al., 2009 [56]	Limited	Appropriate	Stated	Described
Tomic, et al., 2016 [57]	Limited	Limited	Stated	Limited
Wagle Shukla, et al., 2015 [58]	Justified	Appropriate	Stated	Described
Werle, et al., 2014 [59]	Limited	Appropriate	Stated	Described
Zetterberg et al., 2009 [60]	Limited	Appropriate	Stated	Described
Zhang, et al., 2010 [61]	Justified	Unable to Determine	Stated	Described
	Justified	Appropriate	Stated	Described
	Limited	Limited	Limited	Limited
	Not Justified	Unable to Determine	Not Stated	Not Described

Table 3. Study characteristics.

Study*	Country	Sample	HRQOL measure	Key findings
Basurovic, et al., 2012 [31]	Serbia	<i>n</i> = 28, neurology clinic. All segmental dystonia. Mean age 53.1 ± 15.8 years, 39% female. Mean disease duration 9.9 ± 7.9 years.	SF-36	Higher values of depression (B = -0.498, p=0.0007) and disease severity (B=-0.526, p=0.0004) predicted lower HRQOL scores. Higher amounts of leisure activity was a predictor of higher HRQOL scores (B=0.611, p=0.001).
Ben-Shlomo, et al., 2002 [45]	Europe	<i>n</i> = 289, movement disorder clinic. All focal dystonia. Mean age 55.3 ± 13.2 years, 65% female. Mean disease duration 11.9 ± 8.9 years.	SF-36	Longer disease duration and lower levels of disease severity, depression and anxiety were linked to higher PCS scores. The same variables were also linked to higher MCS scores, along with living alone and self-deprecation.

Brüggemann, et al., 2014 [46]	Germany	<p>$n = 23$, hospital. All dopa-responsive dystonia. Mean age 39.2 ± 20.1 years, 70% female. Mean disease duration 30 ± 19.4 years.</p>	WHO-Qol BREF	<p>Lower HRQOL scores were linked to higher levels of depression and impaired sleep quality, collectively explaining a variance of 66.4% in a regression model.</p>
Degirmenci, et al., 2013 [47]	Turkey	<p>$n = 43$, neurology clinic. 23 focal, 5 segmental and 15 secondary dystonia. Mean age 55 ± 15.7 years, 67% female. Mean disease duration 8.31 ± 8.5 years.</p>	SF-36	<p>Dystonia type (secondary dystonia had worse HRQOL outcomes) and lower levels of depression and anxiety were predictors of higher HRQOL scores in multiple regressions across a range of SF-36 subscales.</p>

Gündel, et al., 2007 [48]	Germany	<p><i>n</i> = 50, hospital. All focal dystonia. Mean age 57.6 years, 56% female. Mean disease duration 11.5 ± 9.5 years.</p>	FLZ	<p>Lower levels of ‘self-rated psychopathology’ (<i>p</i><0.001) and the presence of a current close relationship (<i>p</i>=0.028) predicted higher HRQOL scores.</p>
Lee, et al., 2015 [49]	Germany	<p><i>n</i> = 243, neurology clinic. All focal dystonia. Mean age 48 ± 10 years, 33% female. Mean disease duration 7.6 ± 4.2 years.</p>	FLZ	<p>Investigated disease duration and disease course and found no significant results.</p>

Page, et al., 2007 [50]	UK	<p><i>n</i> = 276, sample from epidemiological study. 188 focal, 32 segmental, 21 hemi, 10 multifocal, 25 generalised. Mean age 55 years, 66% female. Mean disease duration 14 ± 11.3 years.</p>	SF-36, EuroQol EQ-5D	<p>Body concept and function disability together accounted for 42% of variance in EuroQOL scores. For PCS scores, the most important predictors were body concept (44%) and functional disability (5%). For MCS scores, the most important predictors were body concept and depression, which combined to explain 34% of variance.</p>
Pekmezovic, et al., 2009 [51]	Serbia	<p><i>n</i> = 157, source not stated. All focal dystonia. Mean age 50.5 years, 59% female. Mean disease duration 7.3 years,</p>	SF-36	<p>Depression, anxiety and pain were the most significant factors explaining HRQOL in multiple regression.</p>

Queiroz, et al., 2011 [52]	Brazil	<p>$n = 65$, movement disorder clinic. All focal dystonia. Mean age 49.9 ± 14.60 years, 57% female. Median disease duration 11 years.</p>	SF-36	<p>Disease severity was moderately correlated with two subscales of the SF-36 (role-physical and body pain). Women scored lower than men on two subscales (vitality and mental health).</p>
Setthawatcharawanich, et al., 2011 [53]	Thailand	<p>$n = 85$, hospital. All focal dystonia. Mean age 58 years, 71% female. Mean disease duration unknown.</p>	Thai HFS-30	<p>Depression was positively correlated with HFS-30 scores ($r = 0.55$, $p = 0.000$; $r = 0.74$, $p = 0.000$) in participants with hemifacial spasm and blepharospasm respectively.</p>

Skogseid, et al., 2007 [54]	Norway	<p><i>n</i> = 70, hospital. All focal dystonia. Mean age 53 years, 64% female. Mean disease duration 11.5 years.</p>	SF-36	<p>Higher levels of disease severity (<i>p</i> < 0.001) and depression (<i>p</i> = 0.001) were linked with lower SF-36 scores on a range of SF-36 subscales in a multivariate regression. Anxiety was linked with the vitality, role limitation: emotional and mental health subscales.</p>
Smit, et al., 2016 [55]	Netherlands	<p><i>n</i> = 50, outpatient clinic, patient association. All focal dystonia. Mean age 54 years, unknown % female. Mean disease duration 13.2 years.</p>	RAND-36	<p>Depression, anxiety and disease severity were the most important predictors of HRQOL scores on a range of SF-36 subscales.</p>

Soeder, et al., 2009 [56]	USA	<p><i>n</i> = 73, unknown source. 46 focal, 17 segmental, 1 hemi, 7 generalised dystonia. Mean age 55.4 ± 16 years, 60% female. Mean disease duration 11.2 ± 13 years.</p>	SF-36	<p>PCS scores on the SF-36 were correlated with depression scores (<i>r</i> = -0.37; <i>p</i> = 0.002) and higher ratings of tiredness. MCS scores were correlated with depression scores (<i>r</i> = -0.63, <i>p</i><0.001), state anxiety (<i>r</i> = -0.50, <i>p</i><0.001) and trait anxiety (<i>r</i> = -0.62, <i>p</i><0.001) and higher ratings of tiredness.</p>
Tomic, et al., 2016 [57]	Croatia	<p><i>n</i> = 19, hospital. All focal dystonia. Mean age 59.4 ± 13 years, 58% female. Mean disease duration 9 ± 6.5years.</p>	SF-36, CDQ-24	<p>Level of disability was significantly correlated with all scales of the CDQ-24. Disability and pain were positively correlated with the physical function, disability and pain subscales of the SF-36.</p>

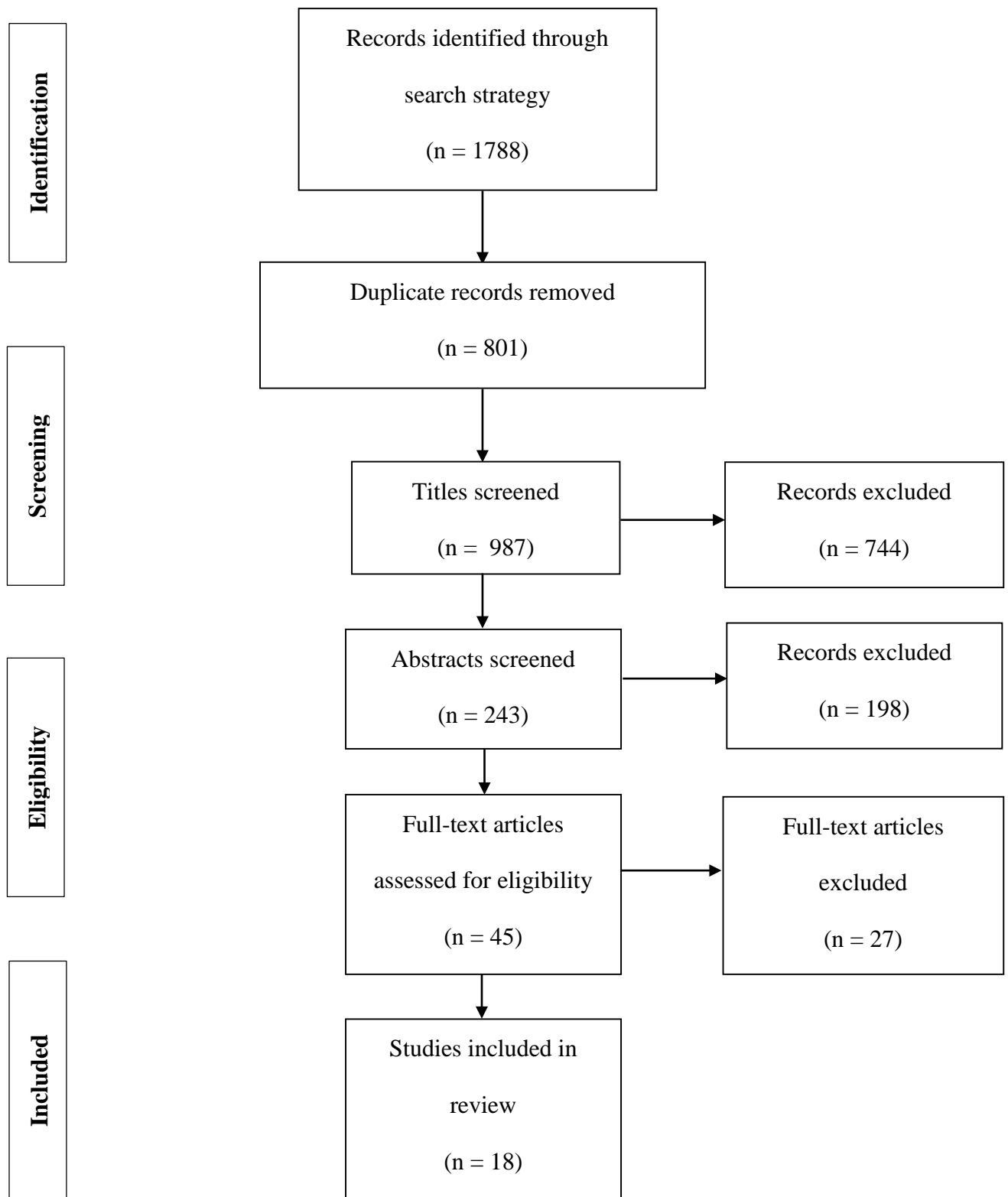
Wagle	USA	<p><i>n</i> = 91, movement disorder clinic. 64 focal, 18 segmental, 9 generalised dystonia. Mean age 60 ± 17 years, 73% female. Mean disease duration 7.5 ± 8 years.</p>	SF-36	<p>Worse fatigue and sleep scores on multiple measures correlated significantly with HRQOL.</p>
Werle, et al., 2014 [59]	Brazil	<p><i>n</i> = 70, hospital. All focal dystonia. Median age 50 years, 63% female. Mean disease duration unknown.</p>	CDQ-24	<p>Higher levels of dystonia severity were correlated with worse HRQOL scores.</p>

Zetterberg et al., 2009 [60]	Sweden	<i>n</i> = 351, patient association. 284 focal, 35 segmental, 20 multifocal, 7 generalised dystonia. Mean age 59 ± 11 years, 73% female. Mean disease duration 14 years.	CDQ-24, CDIP-58	21% of variance in scores in the symptom domain of the CDIP-58 were explained by high levels of physical activity and satisfaction with treatment. 35% of variance in the impact on daily activities domain was explained by level of physical activity and employment. Satisfaction with treatment and disability pension explained 16% of variance in scores on the psychosocial sequelae domain.
Zhang, et al., 2010 [61]	China	<i>n</i> = 76**, neurology clinic.	SF-36	Linear regression found four predictive factors: gender (b = 5.704, P = 0.007), anxiety (b = 1.605, p = 0.003), depression (b = 1.939, p < 0.001) and disease severity (b = 0.469, p < 0.001).

*all studies used cross-sectional designs

**only part of the sample completed HRQOL measures, therefore reported demographics for full sample are unrepresentative

Figure 1. Study search flow chart.



Appendix 1-A

Author guidelines

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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.

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 - 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.

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 - 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.
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Appendix 1-B

Implications for rehabilitation

- Dystonia has a pervasive, negative impact on the lives of people with the condition.
- Physical and psychosocial factors such as disease severity, depression, anxiety and fatigue have been found to be related to HRQOL for people with dystonia.
- Multidisciplinary interventions, including psychology, should aim to mitigate the negative impact of these factors.

Appendix 1-C**Quality appraisal checklist**

1. Were the objectives of the study clearly stated?
2. Was the design of the study clearly described?
3. Were characteristics of participants clearly described?
4. Were characteristics of non-responders adequately described?
5. Was the sampling method for recruitment of participants appropriately described?
6. Was the sample size used appropriately justified?
7. Was the concept of HRQOL clearly defined?
8. Was a reason provided to justify the HRQOL instrument selected?
9. Was adequate evidence provided regarding the clinimetric properties of the HRQOL instrument selected?
10. Were all demographic and/or clinical variables examined clearly described?
11. Was a reason provided to justify why the demographic and/or clinical variables selected were examined in relation to HRQOL?
12. Were appropriate statistical methods used?
13. Were the key findings of the study clearly stated?
14. Were limitations of the study clearly described?

Appendix 1-D

Data extraction form

1. Study design

What were the primary research aims?

What is the study design?

2. Participants

Participant details (sample size, age, disease duration, sex, country)

Recruitment method and source

Inclusion and exclusion criteria

3. Measurement tools

What was the HRQOL instrument used?

4. Statistical analysis

What statistical analyses were used?

5. Clinical variables

What clinical variables were included in the analyses?

What results were found?

6. Discussion and conclusion

What limitations were identified in the study by the authors?

What recommendations were made?

Section Two: Research Paper

Experiences of living with dystonia

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Word Count: 9,509 (excluding references, tables, figures, and appendices)

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Prepared for submission to *Disability and Rehabilitation* (see appendix 2-A)

Experiences of living with dystonia

Abstract

Purpose: Dystonia is a chronic and incurable movement disorder. This qualitative study aimed to enhance understanding of the condition by exploring the experience of living with dystonia.

Method: Interpretative Phenomenological Analysis (IPA) was used to analyse data gathered through semi-structured interviews. Eight participants were recruited via a UK based dystonia charity.

Results: Three superordinate themes emerged from the data: (1) Dealing with ignorance and uncertainty: navigating health services with a rare, poorly understood condition; (2) The challenge of social isolation: overcoming barriers to positive social identity; and (3) Fear of psychological explanations: the impact of stigmatised attitudes towards psychological explanations for dystonia symptoms.

Conclusion: Coping with a rare and chronic condition led to participants feeling isolated and stigmatised by health care services and their communities. These experiences represented a challenge to participant's identities, which they were able to overcome in part through the use of social support, particularly from other people with dystonia. Practical recommendations for reducing the stigmatising experiences of people with dystonia can help to ease the process of adjustment to the illness and enable people to pursue meaningful lives and positive identities. Recommendations for research are aimed at increasing knowledge about these processes.

Keywords: dystonia; identity; interpretative phenomenological analysis; rare diseases; social isolation

Introduction

The term dystonia refers to a group of heterogeneous, chronic movement disorders characterised by uncontrollable muscle spasms which can lead to contorted posture or repetitive movements. Types of dystonia have typically been categorised according to age of onset, location of symptoms and aetiology [1, 2, 3]. When dystonia is presumed to be the sole neurological cause of symptoms it is labelled primary dystonia, as opposed to secondary dystonia which has a known exogenous cause. The prevalence of dystonia is difficult to estimate due to problems accurately diagnosing such a heterogeneous disorder. A meta-analysis conducted in 2012 found a rate of 16.43 per 100,000 people for primary dystonia, but the authors believe this to be an underestimation [4].

Traditionally the impact of dystonia has been conceptualised in terms of the severity of motor symptoms [5]. Spasms and contortions can cause significant pain and discomfort, reducing people's ability to work and access the community [6]. As such, dystonia also has a significant social impact and represents an economic health burden [7]. Treatment for dystonia is typically focused on relieving motor symptoms rather than targeting pathogenesis. Treatments include pharmacological intervention, botulinum toxin injections (BTX) and deep brain stimulation of the globus pallidus interna (DBS). Dystonia is difficult to treat and diagnose effectively due to individual variation in symptoms and reactivity to treatment. This means that people with the condition often have to undergo a frustrating process of trial and error, enduring side effects of treatment with no improvement in their symptoms [8]. This has a negative emotional impact on both patients and health professionals [9].

Although most research and clinical intervention is aimed at ameliorating the physical symptoms of dystonia, attention is increasingly being paid to its psychological

and social impact [10]. Research has found that people with dystonia have reduced health related quality of life (HRQOL), scoring lower on all domains in comparison to healthy controls [11]. There is also an above average prevalence of psychiatric diagnoses for people with dystonia, with one study finding that 57% of people with dystonia have a psychiatric diagnosis [12]. Severity of motor symptoms do not adequately explain the psychological impact of the condition; for example, severity of depressive symptoms is not significantly correlated with severity of motor symptoms [10]. Neither are differences in HRQOL outcomes fully explained by severity of motor symptoms [13]. The experience of chronic illness itself can be argued to be inherently linked to psychological distress, representing a critical life event which demands a process of adjustment, the success of which depends on the availability of psychological and social resources [14]. The psychological and social aspects of dystonia require further investigation in order to more fully understand the impact of the disorder [15].

The research which has been conducted into the links between psychosocial variables and HRQOL outcomes thus far has found that depression and anxiety have a negative relationship with HRQOL [6, 16, 17]. Psychosocial variables such as self-esteem and body concept, and cognitive variables such as memory and attention, have also been found to be associated with HRQOL [13, 18]. Although this body of research has been successful in establishing some fundamental knowledge about the impact of the condition, thus far the majority of recommendations for research and practice are typically generic and make little reference to the specific nature of dystonia [19, 20].

Despite recent trends towards qualitative research into chronic neurological conditions, dystonia remains one of the conditions with the fewest studies. This underrepresentation is typical of rarer conditions because they are less visible and considered less of a public burden. They also have less of an established research

foundation and potential participants are more scarce [21]. Qualitative research methods are well positioned to both develop understanding of the condition and elucidate the existing quantitative literature [22]. A phenomenological study of the impact of dystonia on quality of life found that participants faced significant barriers to social interaction and that this negatively affected their sense of self [23]. Another study explored life changes for patients with primary dystonia after DBS [24]. Thematic analysis found that, although participants viewed their treatment positively, surgery sparked a challenging process of adjustment. Participants wanted more support from health professionals during this process.

This qualitative study aims to increase knowledge about the experience of living with dystonia and in doing so provide insight for professionals working to support people with dystonia, helping them to understand what support might be helpful, and how and when it should be delivered. It also aims to aid researchers of both quantitative and qualitative approaches in their choice of topics of interest.

Method

Design

IPA was chosen to explore the experience of living with dystonia. As an idiographic, in-depth approach it is capable of dealing with the complex interactions of psychological, social and contextual factors inherent in the experience of living with chronic illness [25]. By providing a thorough analysis of individual cases and trying to capture as much personal nuance and context as possible, IPA aims to generate information which is specific [17], and which can supplement the general understanding currently established by quantitative research. As a hermeneutic method, IPA is well suited for understanding

the experience of chronic ill health because diagnosis often inspires a search for meaning and a period of adjustment and reflection [14]. There is an increasing acceptance that ill health is, in part, personally constructed and that people's perceptions and interpretations of their embodied experience are of paramount importance [25]. The phenomenological emphasis of IPA lends itself to investigating these perceptions. Finally, an inductive method such as IPA is useful when the knowledge at stake may be outside the perceptual field of researchers [26], as is the case with the specific needs and values of people with dystonia.

This study used semi-structured interviews to encourage participants to choose the topics which were most personally salient and to describe them in their own words. This approach benefits the hermeneutic aspect of IPA by facilitating a rich, first person account of experience whilst also ensuring that a specific range of topics are covered across interviews. It is also a collaborative approach suitable for phenomenological analysis because it emphasises that participants are the primary experts of their experience [25]. Ethical approval for this study was received from the Lancaster University Faculty of Health and Medicine Research Ethics Committee.

Participant recruitment

Potential participants were required to have first-hand experience of living with dystonia, to be 18 years old or over and be able to speak English. Participants needed to be available for approximately one hour and be able to use a telephone, computer or meet in person.

Participants were recruited via the Dystonia Society, a UK based dystonia charity. In the initial recruitment phase 50 information packs were sent to people on the Dystonia Society's mailing list. Two participants replied and were recruited for the study. Contact was made with the remaining participants via advertising on the

Society's social media accounts. Twenty people responded to the advert and the first six who met the criteria were chosen to take part. A total of eight interviews were conducted: this number was considered to represent the optimal balance between facilitating idiographic and between-participant analysis [17]. All participants provided written consent.

Participant characteristics are presented in table 1. Participants were given pseudonyms to protect their anonymity and identifying information in the transcripts was changed.

[table 1 around here]

Data collection

Semi-structured interviews ranging in length from 46 to 91 minutes were conducted by the researcher either in person, via telephone or via online video conferencing. The choice of modality was at the discretion of participants to maximise accessibility and comfort. Interviews were recorded with a digital recording device and transcribed verbatim. The interview schedule (table 2) was developed according to the recommendations of Smith et al. [17]. Questions were made general to encourage participants to choose topics which were salient to them. Prompts were used as necessary to facilitate reflection [27], although participants were encouraged to lead the conversation [25].

[table 2 around here]

Data analysis

Analysis was informed by Smith et al. [17] and based upon an interpretivist perspective [28]. The researcher engaged in a hermeneutic cycle which began with repeated, line by line readings of each transcript and the taking of analytic notes. In keeping with an idiographic approach, each transcript was analysed as completely as possible before moving on to the next [29]. After potential themes were identified within each transcript, comparisons between transcripts were conducted. This was a cyclical process with interpretation moving between each individual transcript and the data as a whole. Superordinate themes were produced from emergent themes via processes of subsumption and abstraction. Care was taken to avoid over interpretation and repeated readings of transcripts ensured themes were grounded in the data. The analysis was considered complete when a set of superordinate and subordinate themes were produced which represented an appropriate balance between salience for participants and representativeness of participants [17].

To ensure legitimate and rigorous analysis it is imperative that the interpretivist philosophical foundation of IPA is articulated consistently throughout the research process [30]. As an inherently interpretive method it was important that the researcher continually evaluated their own bias to ensure interpretations were a legitimate reflection of the data. As such this study was conducted with reference to principles discussed by Yardley [31] and De Witt and Ploeg [30], with the aim of ensuring transparency and legitimacy. Transparency and a critical reflective stance were maintained through the keeping of a detailed audit trail [32] and reflective diary [33] at each step of the process. The researcher used these tools to analyse their bias and orientation towards the data. As an intrinsic feature of a hermeneutic method, bias cannot be avoided [34], but rather should be articulated as an essential part of the

context of interpretation [35, 36]. Legitimacy of interpretation during the process was facilitated by having a selection of participants check initial analysis of their data [37]. Two supervisors also checked the legitimacy of the analysis. Sufficient quotations from transcripts are included to ensure interpretation was grounded in the data and to allow the reader to draw their own conclusions about the legitimacy of interpretation [32].

Personal statement

At the time that I was designing this study I had no personal experience of chronic ill health as an adult, although I had clinical experience of working with people who had. Prior to conducting the analysis I had to use the NHS for such a health problem and encountered a number of the issues that participants would later discuss. This gave me increased empathy for the frustration and hopelessness of struggling to receive a diagnosis for a chronic condition, as well as the systematic, negative impact such a situation can have on one's life. These aspects of participants' accounts became more salient to me as a result and therefore my experience of ill health influenced the analysis as a whole.

My clinical experience of working with people who had endured psychological suffering and seeing them cope in extreme circumstances also influenced my position as researcher. I felt myself wishing for the best possible outcome for participants and being moved by their stories. I was particularly sensitive to the influence of health service power dynamics and the importance of person centred understanding due to the impact I have seen these have on service user experience. Finally, the sense of 'rooting for the underdog' was enhanced by my personal construal of people who overcome adverse circumstances as being in some sense heroic or exemplary. These aspects imbued the analysis with emotional vigour. My clinical

training helped me to manage my emotions and direct the flow of the semi-structured interview whilst still being responsive to the participant.

Results

Three superordinate themes emerged from the data: (1) Dealing with ignorance and uncertainty: navigating health services with a rare, poorly understood condition; (2) The challenge of social isolation: overcoming barriers to positive social identity; and (3) Fear of psychological explanations: the impact of stigmatised attitudes towards psychological explanations for dystonia symptoms. Superordinate and supporting subordinate themes are presented in table 3.

[table 3 around here]

Dealing with ignorance and uncertainty

The first superordinate theme related to the experience of having a rare, poorly understood and unpredictable condition. Participants were forced to cope with their own and others' ignorance and a subsequent sense of uncertainty about their condition and the future. All participants contributed to this theme.

Struggling to escape the darkness

Several participants described the experience of searching for a diagnosis as akin to being in the dark. Even in cases where an accurate diagnosis was given relatively quickly, this process was experienced as a "struggle", which for some participants

would last for years. The rarity of the condition meant that neither they nor the health professionals involved understood the cause of their distressing experiences. This, combined with the oppositional attitude of some health professionals, left participants feeling isolated:

It was very stressful, very traumatic (...) ¹ when nobody believes you in the medical world, you just sort of think, well I'm on my own here, how, how am I going to resolve this? So, ermm, really it was very difficult seeing some of these [health professionals] ². I'm not saying they were all awful people, but I did have a few along the way that, that did make me feel pretty bad. [Jennifer]

The participants who did not receive an early diagnosis experienced a chain of unhelpful referrals, diagnostic testing and treatment. As avenues for referral were exhausted, contact with their General Practitioner (GP) lessened and participants felt obliged to take control of their care. The internet was a powerful mechanism for education, making contact with third party support services and seeking out private treatment:

I'd managed to do a bit of research myself on the computer and found an organization. [I] got the information from them, and then I, you know, I sort of self-diagnosed really, but the local hospital still weren't having it. They refused to believe that blepharospasm was a form of dystonia (...) the consultant wasn't having it, so (...) I had to arrange a

¹ (...) indicates removed text.

² [text in square brackets] indicates additional information.

private appointment initially. I basically walked into the consultant's room and within five minutes he'd diagnosed me, which was a huge relief. [Hailey]

The experience of taking control was empowering for participants but was accompanied by feelings of resentment at a perceived lack of support from the National Health Service (NHS) and at having to pay large sums of money for the answers they needed. The moment of diagnosis itself was transformative for participants, so much so that it created a surreal tension between delight at having their experiences validated and the reality of being diagnosed with an incurable movement disorder. Sheryl described receiving a diagnosis of dystonia as a gift:

[A] friend had gone with me, and when I came out I went (...) "Hey, I've got dystonia!" [laughing] I was just so relieved that at long last someone was saying to me, I've got a name for these symptoms that I've got and that are visible to other people (...) I said "that's the best Christmas present I've ever had", being told I've got dystonia [laughing] cos I was just so relieved that at long last I had an answer. [Sheryl]

Walking on the treadmill of dystonia

After the struggle to receive a diagnosis was over, participants faced living with an unpredictable, poorly understood movement disorder, treatment of which has variable impact and significant side effects [8]. Participants felt initially optimistic due to the possible range of treatment options and their belief in the expertise of health professionals. Over time, as options were exhausted, this optimism gave way to an acceptance of dystonia as a pervasive and chronic condition:

I felt at first, quite optimistic, that (...) a way of managing it could be found because you know, they'd say "we'll try this drug and we'll try that drug" and as time went on and I went from one to another and Botox didn't work, then I think it dawned on me that, you know, hold on, this thing isn't going to be managed. [Matthew]

Multiple participants invoked a treadmill metaphor to describe the experience of receiving trial and error treatment with no significant improvement in their symptoms. This lack of predictability had an emotional impact on participants who were worn down by the cycle of hope and disappointment, and who felt trapped by an inability to plan for the future:

You build everything up and (...) think, "oh great, this is going to be it", and then as I say, you do get knocked off so it's just very, as I say, sad I suppose (...) It just plays with your emotions I suppose really and, and plays with your life because you don't know where, where you're going (...) one minute you think you can and then the next minute you think, "oh well, that's not going to happen now", so it's a very, very, sort of, wavy road that you (...) follow. [Jennifer]

Participants spoke about the emotionally draining nature of dystonia as a "twenty-four-seven" pervasive disorder. Maria described how the temporary respite afforded by treatment dictated the rhythm of her life. She was not alone in worrying about the sustainability of this cycle. Gradual physical decline due to age, unmanaged symptoms and adaptation to treatment left participants facing an uncertain future:

I've had very mixed results from [treatment] as well. I mean, when I first started going I did, once the Botox kicks in (...) you get about four weeks where you're quite stable and

then obviously it wears off so you're back to, sort of, square one again, so I did feel like I was living in a constant two monthly cycle. (...) Obviously I'm getting older and I've had the injections for over seven years now (...) They don't seem to be working anymore. [Maria]

Acceptance and working for a positive future

Coping with a lack of hope for improvement and a sense of gradual decline posed a challenge for participants. Those with the most positive outlook on the future focused on taking responsibility for their interpretation of experiences in light of a lack of control over their health. Participants emphasised the importance of living day by day:

It's an acceptance, I've got it, err, you know, unless there's some sort of (...) silver bullet, that's going to magically appear, it's sort of, acceptance that I've got this for the rest of my life, so let's make the best of it, let's make the best of everyday. [Jacob]

Some participants sought to replace meaningful aspects of their lives that had been lost, such as work, by committing to new goals. Hailey saw the limitations of dystonia as a "challenge" to be overcome and in committing to doing so, developed a positive, efficacious aspect of her identity:

I probably wouldn't have done quite as much as, as I have (...) I think my character has changed really because I do, I do see various things in life as a bit of a challenge. I think before, ermm, I was, you know, getting on with my job (...) Part of me thinks, "well I have to do it now because I don't know what my eyes will be like in five years' time" (...) At the back of my mind, you have to go out, you have to go and do it now, none of

us know what's round the corner anyway do we? (...) I do take life more head on really than I ever did. [Hailey]

Participants struggled initially to integrate dystonia into their concept of a meaningful life and identity, a process made more difficult by the rarity of the disorder and the uncertainty of the future. Meeting other people with dystonia, particularly those who had had it for a longer time, proved a compelling demonstration of the possibility of this integration. Ethan suffered from severe panic attacks and described himself as a “recluse” until he attended his first support group meeting:

I plucked up the courage to go along one day and it was the best thing that ever happened. It really, really helped, actually seeing people that were in the same boat as you. People understood what you were going through (...) Some of them have had it for twenty five year, thirty year, so it was good to see them and speak to them about their experiences and how they coped (...) I go to it like, religiously, never miss it (...) I wouldn't say it saved my life but it really, really helped because at that point I was not even leaving the house. [Ethan]

The challenge of social isolation

The second superordinate theme related to the experience of social isolation as a consequence of having dystonia and associated visible differences. Participants felt excluded from their communities and the emotional impact was manifold. This was an experience shared across all participants.

Isolation as a threat to identity

Participants mourned identities lost as a result of their disability, which resulted in increased feelings of isolation. Ethan was still in touch with his former colleagues, “the taxi boys”, through social media but he spoke with a real despondency and sense of loss about his inability to take part in the activities which to him were a crucial part of group membership:

I used to go to the golf outings with the boys, the taxi boys, that’s all had to stop. I cannot do any of that now (...) Aye, I loved it (...) I don’t do any of the things anymore, it’s too much for me (...) I hate it, I would rather go out to the pub on a Sunday and watch the football with the boys (...) or going on the golf outings. That’s the parts I really, really miss. [Ethan]

The lack of understanding and acknowledgment from friends led some participants to intentionally divide their social circles and focus on people with whom they were closer. In doing so they sacrificed part of their identity. For Anna this was her sense of self as a fun, spontaneous, party-goer. She was typical in experiencing a sense of guilt for engaging in this self-protective behaviour. This guilt was balanced by feelings of alienation and bitterness towards friends who did not make an effort to accommodate her or acknowledge her negative experiences:

I find it very hard to swallow because they’re like, “well why aren’t you coming out? Why aren’t you drinking?” And then it was kind of a case of, I realised it was easier to shrink my friendship group and have people who cared, who understood [that] I could leave myself with maybe four weeks of being in agony because I pushed myself to go to

something that I really shouldn't have done and I just, it was easier to, it sounds awful, but just cut people like that out who just didn't understand. [Anna]

Visible difference impacting social experiences

Participants described having a constant, tiring awareness of difference due to their visible contortions and tremors. The negative responses of strangers were experienced by participants as judgmental and were damaging to their self-esteem, making them feel socially isolated. Ethan described how previous aspects of his life which he enjoyed like socialising and talking with customers of his taxi firm were disrupted and came to represent burdens. He was typical of participants in coping with the experience of negative evaluation by isolating himself from others, compounding the barriers to social interaction resulting from pain, fatigue and functional disability:

It was just a living nightmare (...) it just basically stopped my life in track and ermm, just basically made me a prisoner in my own home. I didn't like going out because people were staring, ermm, it affected my balance, it knocked my balance off so people were assuming you were either drunk or drugged up because you were staggering.

Ermm, I became basically a recluse, I wouldn't leave the house, I hated going out.

[Ethan]

Changing social identity in response to isolation

The barriers to social engagement presented by dystonia forced participants to challenge themselves in order to find new groups and identities which could satisfy their need for emotional and practical support. Support groups, either online or in person, allowed

participants to make contact with other people with dystonia. This provided an avenue for sharing experiences and receiving validation, which in turn reduced feelings of isolation. Engagement with support groups also gave participants the opportunity to support others with the condition, which they found added a meaningful aspect to their lives. For Hailey meeting someone with similar symptoms after years of believing she was alone was a relief and a delight:

People were describing symptoms and treatments and what have you, and (...) I thought, “oh my goodness! It’s not just me!” (laughing) “There’s other people!” (...) [I met] this person locally (...) She was the first person I’d seen with it, and I said, “your eyes are exactly like mine, I can’t believe it!” (...) Meeting other people that share the same symptoms, it’s so helpful. [Hailey]

Meeting people with similar experiences allowed participants to let go of the psychological tension resulting from fear of judgement. They described moving experiences of mutual recognition and shared compassionate understanding:

One of the ladies that I [volunteer] with, she’s got a benign tremor (...) so I’ve really connected with her (...) It feels quite, err, a relief to be honest, because I don’t need to say too much to her. I see what she’s going through (...) It just feels more relaxed, more (...) it’s just like a kindred spirit I suppose. She, she’s got the same thing as me and we, I just, yeah, I just feel compassionate towards her and she does to me I suppose. [Maria]

For some, learning to accept support was a challenge to their independence and identity. For others, trying to access support was a difficult task which required them to adjust identity in a different way. Several participants described a process of “hardening” in the face of rejection or criticism, particularly from health professionals.

Jennifer spoke about feeling forced to become more demanding and aggressive with a mixed sense of sadness and pride:

I didn't ask for [dystonia] I, I've got [it], I've got to deal with it and you've got to believe that I'm, that I've got those problems. So, to be told by consultants, "pull yourself together, don't be so stupid", be sworn at in consulting rooms and everything, I don't have to put up with that, that's just not right. So, ermm, I, I've had to learn to stand my ground a bit shall we say, with, with how I've been treated over the course of time with these people. So, it's made me a stronger person I think, definitely. [Jennifer]

Fear of psychological explanations

The third superordinate theme related to participants' perception that psychological explanations for their symptoms were stigmatising. This theme was articulated by six of the eight participants.

It was common for participants to express their relief at receiving a physical explanation for their symptoms. There was a collective sense that psychological factors are "all in your head", with connotations that they are untreatable, are unworthy of medical resources and indicative of personal weakness. The discrepancy between the intensity of their experiences and the message that there was not anything "really" wrong with them made participants such as Jennifer feel alienated and angry. For her, receiving a physical label after a long struggle meant validation and recognition of her experiences:

To actually finally have a doctor or a professor actually say to me, "this is a proper condition", ermm, was a massive relief (...) Just, knowing that I had a condition so that if anybody else asked me I can say, "right ok, I've got this, it is a real condition, it's not

all in my head, I'm not making it up", ermm, was a massive relief. So, ermm, I was so grateful to that guy for, for sort of telling me that yes, I have got a condition. I'm not, not, just making it up. [Jennifer]

Some health professionals' attitudes towards a rare condition which they could not diagnose and treat as being the fault of patients led participants to internalise doubt and blame:

Nothing was helping and the way [the physiotherapist] spoke to me was really, really hard because she was like, "but you're just doing this, you're making this happen, you're not trying", but I would go home and (...) do everything they told me to every day (...) I was thinking, "well, why's this happening if they say that (...) it will all just go away?" And at that point that's when I thought, "hang on I can't have somebody just telling me this and it's all in my head and you're making this happen" (...) and I thought, "oh god am I going crazy?" (...) I didn't ever have to see her again, but it was just, it was horrible being treated like that. [Anna]

Some participants felt responsible for their symptoms and so struggled against them, resulting in frustration, exhaustion and a feeling of failure. Sheryl eventually had to take extended leave from work due to exhaustion after living with undiagnosed dystonia for three years. The unpredictable nature of her symptoms led both her and health professionals to believe that she could control her symptoms if she was committed enough:

I used to try and practise walking (...) and sort of say to myself, "come on, you know, it's just you, it's just", I'd began to question myself, "is it psychological?" and I used to practise walking and sometimes it would be better than others and if I had a good

moment I'd be, you know, "of course it must be psychological because you weren't too bad then", and the next day I'd be bad again and I'd think, "oh!" [Sheryl]

The negative connotations of psychological explanations caused some participants to feel embarrassed and ashamed. Sheryl described how she lived in fear of having to tell people that her struggles were "her fault". Her greatest moment of relief was the first time that she heard from a health professional that there was a physical explanation for her symptoms, even though they did not know what it was or how to treat it:

[The doctor] said (...) "As far as I can see, and we're all agreeing, including the professor, that it's definitely physical, you do know that don't you?" I was almost in tears and she said, "what's the matter?", and I said, "well, in the end I was absolutely convinced it was psychological and I'd been so worried thinking, how am I going to tell everyone that it's all in my head? I'd be so mortified." [Sheryl]

Participants' own stigmatised attitudes towards psychological explanations contributed to their unwillingness to engage with psychological services when feeling distressed about their situation. Ethan was one of three who previously experienced suicidal ideation and he was typical of participants in preferring to speak informally to trusted people about his distress, rather than accepting a referral to psychological services:

I would rather just [talk to my GP], I've always kind of just done it that, that way. I think it's cos I know him it's good to get it out in the open to him. I would feel quite, I don't know, ermm, nervous, apprehensive about, with a stranger. [Ethan]

Discussion

The most salient experiences for participants of this study were those relating to isolation and its impact on their sense of self. This is consistent with the evidence that positive group membership and social identity have a significant impact on wellbeing and quality of life, operating as part of a positive feedback cycle between perceptions of the self and others [38]. Equally, stigmatised identities and group memberships have been found to produce negative feedback and reduce wellbeing [39]. With regards to stigma, people with dystonia are in a particularly vulnerable position. The disorder represents an intersection of rare, visible, chronic and often unexplained illness, all of which have associated stigma [40, 41, 42]. In order to increase social connectedness and wellbeing for people with dystonia, it is important to understand and mitigate experiences of stigma and isolation.

Navigating health services with a rare condition

Participants reacted with anger at the struggle they faced for a diagnosis and the antagonistic relationship that developed with health professionals. Until this point they conceived of health professionals as typically supportive and efficacious. Rare or unexplained conditions can represent a challenge to traditional patient-doctor relationships and cause conflict, demanding that both parties adjust their role perceptions in order to achieve progress [41]. Typically doctors are less likely to have knowledge of the specific condition and patients are obliged to assume more control, as was the case for participants in this study. A study of GP attitudes towards people with medically unexplained symptoms found that GPs felt uneasy giving up their traditional authority and felt powerless due to their inability to help patients [41]. A similar study found that some GPs resent the amount of time and emotional investment taken up by

these difficult cases, and fear making costly errors in judgement [43]. Stigmatisation of patients seen to be upsetting established processes and balance of roles can lead to GPs providing a less person-centred service, shorter consultations and operating with impaired clinical judgment [41, 44]. Although participants were not explicitly labelled as having ‘medically unexplained’ or purely psychogenic symptoms, their experiences echo these findings. Bodenheimer et al. [45] argue from a systemic perspective that the neglect which participants experienced can be attributed in part to an acute-focused care system where emergencies are a priority and chronic cases are often left to self-management.

When participants found their care in a state of inertia they turned to the internet as a source of knowledge and empowerment. Although participants spoke about this empowerment in positive terms, they were consistent with other groups in expressing a preference for health professionals taking control of their care [46]. Although the internet represents a useful resource for accessing other perspectives and adding context to consultations, participants felt exposed to misinformation and exploitation. In order to facilitate self-management for people with rare and undiagnosed conditions, GPs should receive training aimed at developing a facilitator role, wherein they explain and contextualise information for patients, rather than competing with them for authority [46, 47, 48]. The ability for GPs to move flexibly between facilitator and expert roles in a way that is responsive to the needs of patients allows them to provide a more effective service [48].

The most common request of participants was that the NHS improve GPs’ awareness of dystonia to enable accurate and timely diagnosis and treatment. Rare diseases are generally associated with delayed diagnosis, misdiagnosis and harmful and unnecessary intervention [49]. GPs often fall victim to the availability heuristic and

premature closure during diagnosis, identifying a common explanation for symptoms and then stopping the search for alternatives [50]. However, it is unrealistic to expect GPs to be able to consistently and accurately identify rare diseases due to a lack of specialist training and consultation and research time. It is necessary therefore, for GPs to be able to utilise the expertise of specialist health professionals in these situations. Grounded theory research into the relationship between GPs and consultants has found that both parties lack the time to confer due to excessive workload and desynchronised workflow [51]. The subsequent lack of information sharing and its impact on holistic care was an issue highlighted by participants of this study. Sampson et al. [51] recommend the instigation of a “universal pause”, a protected time for information sharing between GPs and specialists. The more efficient use of information already in the system would complement the work of third sector organisations and rare disease centres in increasing awareness through education aimed at both the public and health professionals [52]. Helping GPs to more effectively manage the care of people with rare or undiagnosed conditions may reduce the associated emotional burden and improve the experience of patients.

Finding social support

Participants described experiencing stigmatising reactions to their ambiguous, visible symptoms. These reactions caused feelings of shame and participants subsequently limited their interaction with their communities. This experience echoes the findings of IPA studies of stroke survivors and people with Parkinson’s disease [53, 54]. This self-imposed social isolation can have a snowball effect whereby people begin to feel depressed and, as Ethan put it, become recluses [55].

Chronic illness was experienced as a biographical disruption which threatened the identity of participants [56]. The identity restoration hypothesis posits that people

with threatened identities will often feel compelled to develop or repair their social identity [38], a prediction which is consistent with the experience of participants. This challenging process is particularly salient for people who are members of a group which they cannot leave, for example those with incurable illnesses [57]. The experience of participants and related research suggests that support groups are one of the most effective ways to facilitate this restoration [38, 58, 59]. Support groups can promote positive social comparisons and cultivate a sense that overcoming the challenges of ill health makes the person more virtuous than before. When participants saw their peers coping effectively they were inspired and spoke about the inner strength this engendered in themselves. This process of creative redefinition [60] can form part of a positive feedback cycle wherein positive conceptions of one's peers enhance personal identity [38]. In keeping with the testimony of participants, research shows that peer interaction provides perspective and information for people who are otherwise isolated in their experiences [61, 62]. Group meetings also offer a supportive venue for expressing negative emotions, which helps people feel acknowledged and validated [59, 63]. Participants in this and other IPA studies reported that they valued the opportunity to give, as well as receive, support of this kind [64].

Members of support groups are more likely than non-members to have accepted their condition and incorporated it into a positive identity [38, 65]. The challenge of coming to terms with a life changing illness is compounded by the anxiety provoked by initial engagement with face to face support groups [58]. These barriers mean that, as with three participants in this study, some people prefer one to one peer interaction or online support groups. A content analysis of online support groups showed that they tend towards a biomedical emphasis and the sharing of technical information, rather than emotional support [66]. As such, they represent an alternative for people who want

specific, pragmatic, experiential knowledge from their peers but for whom face to face groups are unsuitable [67].

Dystonia support groups are typically operated by non-profit organisations or group members themselves. They represent a cost-effective means of ameliorating deficits in support provided by the NHS [66, 67]. Participants in this study complained about health professionals' lack of awareness of support groups, which they felt was indicative of a lack of holistic support in general. Bodenheimer et al. [45] claim that connecting patients with chronic illness to community resources should be a priority for acute-focused care services, given the inability of these services to deal with the long term needs of patients. Participants requested that GPs inform people of available support provided by non-profit dystonia services and support groups to help with the process of adjustment and ongoing self-management. An IPA study of people with acquired brain injuries also recommended this as part of effective person-centred care and emphasised the key role of social identity in wellbeing for people with neurological conditions [64]. As well as providing information about support groups, GPs are also well placed to use their psychological training to mitigate patient anxiety about initial engagement [58].

Dealing with self-doubt and alienation

Participants were almost unanimous in their perception of psychological explanations for their symptoms as dismissive and stigmatising. Health professionals' negative attitudes towards psychological explanations were perceived as reinforcing the pre-existing, stigmatised views of participants. Although all participants reported significant psychological distress, with two attempting suicide, none had received specific, psychological support before or after their diagnosis. Some participants expressed negative attitudes towards utilising mental health services and were wary of being

further stigmatised by health professionals. It is possible that their negative experiences during the struggle for diagnosis contributed to these attitudes. Due to the common experience of severe psychological distress, enabling the option of psychological intervention for people with dystonia should be a priority for health services. In order to achieve this, stigma around psychological explanations in general must be reduced on the part of both patients and health professionals.

That health professionals often have a stigmatised attitude towards psychological explanations is well established in the literature. A review of nurses' attitudes echoed the experience of participants, finding that respondents believed psychological symptoms were attributable to personal weakness, laziness and lack of self-control [68]. This pattern is present across different types of health professionals, including medical specialists [69]. When dealing with patients with unexplained symptoms, health professionals can feel obliged to convince them of psychological explanations for their experiences and perceive their resistance itself as evidence of psychological issues [41]. Crucially for people with dystonia, people who are thought to be suffering from psychogenic symptoms are liable to have their physical symptoms taken less seriously [68].

Participants' own stigmatised views of psychological distress and mental health also represented a barrier to engagement. A mixed methods review of the impact of mental health related stigma on help seeking found that psychoeducation can be an effective way of reducing such stigma [70]. Interviews with recipients of psychiatric care found that psychoeducation is less stigmatising when offered pre-emptively [71]. As the initial point of contact for people experiencing symptoms of dystonia, GPs are well placed to introduce patients to the possibility of psychological distress as a consequence of having a rare, undiagnosed or chronic disorder.

In terms of ongoing management of distress, thematic analysis of people with multiple sclerosis found that patients wanted more emotional support from their GPs [69, 72]. An interview study of GPs found that they are likely to rate the quality of their emotional support more highly than patients [73]. A review of psychosocial interventions provided by GPs found that there is significant variability in quality of support depending on the individual GP [74]. The researchers concluded that this was due in part to a lack of focused training, which emphasises GPs pre-existing attitudes and skills regarding emotional support. Controlled trials of effective psychological management in consultations suggest that even a small amount of focused training can raise standards of emotional support provided by GPs [75]. Training of this sort can be compatible with budgetary and time constraints, taking as little as eight hours [76], and therefore represents an effective way for health services to enable GPs to provide a consistent quality of support to patients.

Clinical implications

Every participant in this study struggled at some point with psychosocial issues related to dystonia, although none of them had undergone any subsequent psychological assessment or therapy. Several participants expressed an unwillingness to do so, in part due to stigmatised attitudes concerning psychological issues. In these cases clinical psychologists can work indirectly to help service users, for example as part of multidisciplinary teams (MDTs) or through staff training and consultation [77]. This study suggests that a range of health professionals can benefit from increased sensitivity to the psychological challenges facing service users surrounding diagnosis and treatment of a complex illness. Training and consultation for MDTs is particularly pertinent for teams working with people whose health conditions are ostensibly physical, but which have a manifold psychosocial impact that is often not directly

treated [78]. As discussed, complex, rare illnesses like dystonia which are difficult to diagnose and treat can also represent a significant emotional burden for health professionals. Clinical psychologists can provide supervision for GPs and other professionals to allow them to discuss their own concerns in an emotionally supportive environment [78].

Clinical psychologists can also provide formulations and case consultation for MDTs to assist them in better understanding the unique needs of individuals and the idiosyncratic impact of a heterogeneous disorder such as dystonia. Increased understanding of the individual can better enable teams to deliver care in a consistent and proactive manner [79]. Formulation can also assist differential diagnosis by elucidating the relative contribution of physical and psychosocial factors to a given clinical presentation [78]. If formulations were offered to service users presenting with complex, medically unexplained symptoms prior to diagnosis, it could reduce the financial and emotional cost of unhelpful intervention and diagnostic testing.

If barriers to engagement were reduced to the extent that patients felt able to engage in psychological intervention, this could assist with a range of issues raised in this study such as the process of psychological adjustment, decision making about treatment and adherence to care plans [80]. In terms of therapy, Acceptance and Commitment Therapy (ACT) represents a potentially effective model. In the context of chronic ill health, ACT aims to facilitate the process of acceptance of illness and commitment to meaningful action intended to enhance wellbeing [81]. A review of ACT for chronic illness concluded that the evidence base is currently small and of low quality, but that there are suggestions that the approach could potentially be useful for this population [82]. A pilot study found preliminary evidence that ACT can be useful for reducing self-stigma for people with substance abuse issues [83]. A case study of

ACT for people with functional movement disorders found that the participant experienced improved mood and reduced interference of symptoms with meaningful activity [84]. A treatment study of people with chronic pain found that acceptance of pain and an increase in values-based action correlated with a reduction in anxiety, depression and disability after a three-month follow up [85]. A review of behavioural interventions for adult onset primary dystonia [86] similarly concluded that more quality research is needed, but that there is reason to believe that behavioural interventions could assist self-management of dystonia outside clinical settings. An example of such a programme was piloted by Sandhu et al. [87], who concluded that a combined cognitive behavioural and mindfulness programme could assist self-management and an integrated approach to managing the psychosocial impact of dystonia.

HRQOL

The emphasis that participants placed on experiences of stigma and social isolation is in keeping with the conceptual model of the condition employed by established dystonia specific HRQOL questionnaires such as the Cervical Dystonia Impact Profile (CDIP-58) [88] and Craniocervical Dystonia Questionnaire (CDQ-24) [89]. This study therefore supports the use of these disease specific measures in place of, or in addition to, generic measures such as the 36-item short-form health survey (SF-36) [90] which does not contain subscales or items related to stigma and social isolation to the same extent. This study also showed however that the concepts of stigma and social isolation are nuanced and manifest in different ways for different people. For example, some participants were severely impacted by the perceived negative evaluations of other people, whereas others were more impacted by their own negative perceptions of ill health, manifesting in self-blame and criticism. In terms of social isolation, participants

felt both isolated from their communities and isolated from medical expertise, feeling forced to take control of their own care. The former sense of isolation is represented in the HRQOL questionnaires, but not the latter. Future measures may therefore benefit from discrete social isolation and stigma subscales, allowing for the nuance of these factors to be more fully explored. This study also recommends that measures take account of levels of available support for dealing with the burden of self-care experienced by people with dystonia.

Perhaps the most significant issue with existing HRQOL measures is a lack of subjective rating of importance of items and subscales. Item and subscale scores are currently given equal weighting in the calculation of total scores which implies that, for example, physical and psychological wellbeing have an equal impact on HRQOL. Evidence shows that this is often not the case and that through a process of psychological and environmental adjustment, people with physical disabilities can adjust their values based on remaining functional ability and the pursuit of optimal psychological wellbeing [91]. Subjective perception is a core aspect of the HRQOL construct [92] and as such should be reflected in HRQOL measures through either a weighting system, wherein participants highlight subscales or items which are most important and subsequently contribute more to summed scores, or through the use of a semi-structured questionnaire like the Measure Yourself Medical Outcome Profile (MYMOP) [93] which allows participants to choose which specific issues to rate based on their subjective importance. Using this salience based IPA study as an example, participants did not choose to discuss physical issues to nearly the same extent as psychosocial ones, and yet on existing HRQOL these areas would be weighted equally in score calculations. On a subjectively weighted questionnaire, it would be clear that it was social isolation and stigma which were the most pressing concerns and targets for

intervention. It should be noted that semi-structured questionnaires such as the MYMOP are more suited for clinical use rather than quantitative research due to the difficulty of comparison between participants' ratings.

Another insight from this study is that the heterogeneity of the physical manifestation of dystonia did not prevent the emergence of salient, group-wide themes related to psychosocial issues. Future dystonia specific measures could benefit from addressing salient psychosocial aspects of HRQOL such as stigma, without focusing on specific dystonia types, as the CDIP-58 and CDQ-24 do. This would facilitate the use of disease specific measures for a much wider selection of people with dystonia. Due to the clinical importance of pain and body part specific physical symptoms, perhaps these scales could include a supplemental subscale for use with types such as cervical dystonia.

Strengths and limitations

Despite dystonia being a heterogeneous condition, the sample for this study was sufficiently homogenous for effective IPA analysis. The consistency of experiences between people with different types of dystonia suggests that the most salient aspects of experience are not related to the particular manifestation of motor symptoms, but the more general psychological and social impact of the condition. The complexity of the issues described by participants justifies the use of IPA and warrants further analysis using similar methods [23]. This study was able to identify a range of topics which have thus far received little attention in dystonia research.

Participants in this study represent a self-selecting sample as they had experiential knowledge of the specific phenomenon of interest, were motivated enough to participate and were in contact with the Dystonia Society. The majority of people who were aware of the study chose not to participate. It is possible that participants

were motivated to tell their stories because of how adverse their experiences were and that this gives a particularly negative impression of the experience of living with dystonia. Additionally, one of the main functions of the Dystonia Society is advocacy so it is possible that people who were engaged with the Society's social media accounts were more likely to want to raise awareness of the plight of people with the condition. By comparison, a survey of patients with cervical dystonia found that 53% were able to return to a normal routine and 70% were happy with their relationship with their doctor, although this was after a difficult period surrounding diagnosis [94]. The nature of IPA data collection may explain this apparent discrepancy however. As an open and inductive method, often utilising semi-structured interviews, IPA asks participants to select and describe their most salient experiences. This includes recalling their emotional reaction at the time. As such, discussion of 'a difficult period surrounding diagnosis' could include significant negative emotion and evocative description, as it did in this study. Given that most of the participants had in some sense adjusted to the presence of dystonia in their lives, they may have responded as the patients in the survey did when asked to make a contemporary judgement of their relationship to health professionals and their illness in general.

Another aspect of recruitment which bears consideration is that advertising on social media networks requires that potential participants have internet access, be computer literate to some degree and be relatively active on social media. The sampling could therefore have excluded a set of people who, for example, were from lower socioeconomic groups or had greater levels of disability. Studies of internet sampling have concluded however that online sampling methods reach a relatively diverse socioeconomic range [95]. This study sought to minimise the impact of disability on accessibility by offering participants the choice of telephone, video conferencing or face

to face interviews. This also ensured that it was possible to recruit a sufficient sample. Although telephone interviews lack some information communicated through non-verbal behaviour, comparisons of telephone and face to face interviews found no significant difference in quality of data [96, 97].

The philosophical assumptions of IPA dictate that the interpretation of a given data set cannot be generalised to a wider population due to IPA's ideographic perspective and the irreducibility of context [98]. The methodological concept of generalisability, or external validity, is replaced by the idea of theoretical transferability: the extent to which lessons learned from one set of experiences may inform the interpretation of another set [29]. This principle means that rigour in IPA is not achieved through representative sampling but through the articulation of potential bias and the justification of methodological decisions with reference to the philosophical assumptions of the method [99].

Further research

Longitudinal studies are one of the most neglected areas of qualitative research [21]. An IPA study with multiple interviews per participant, beginning shortly after diagnosis, could shed light on the process of adjustment and meaning making. Recording the experiences of participants whilst they are still recent may produce different results than in studies such as this, where participants are often reflecting on experiences which took place years earlier [14]. In terms of quantitative research, more high quality studies of the efficacy of ACT and behavioural interventions for people with dystonia and related conditions are needed, particularly in the form of randomised controlled trials with long term follow up assessments [81].

This study has identified a range of psychological processes and potential variables of interest that warrant further attention. Future research should investigate the

impact of stigma, availability of social resources and patient-health professional dynamics on the wellbeing of people with dystonia. Although this study was not designed to investigate differences between participants, another issue which demands attention is the effect of pre-morbid personality and attachment on how people are impacted by these factors. Research with other populations suggests it could be an important part of the complexity of this topic [99, 100, 101].

Conclusion

Participants in this study described a struggle for explanation, recognition and validation of their experiences. This left participants feeling isolated from health professionals and their community, and alienated from themselves. The mutual exchange of support with peers helped participants to integrate the experience of dystonia with a positive sense of identity. Although people with dystonia represent a particularly vulnerable group in terms of exposure to stigma, the experiences described by participants were typically in keeping with those of people with other, similar health conditions. The recommendations for practice and research could help shorten the process of acceptance and the development of a positive identity for people with dystonia.

Declaration of interest

The author reports no conflicts of interest.

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Table 1. Participant characteristics.

Pseudonym	Gender	Age	Age at first symptoms	Dystonia type*	Interview type	Ethnicity	Employment status
Matthew	Male	61	53	Oromandibular	Face to face	White British	Retired
Hailey	Female	66	43	Blepharospasm	Telephone	White British	Retired
Jennifer	Female	50	35	Fixed	Telephone	White British	Retired
Anna	Female	31	23	Segmental	Video conferencing	White Indo-Caribbean	Employed
Sheryl	Female	55	34	Generalised	Telephone	White British	Retired
Jacob	Male	56	54	Cervical	Telephone	White British	Unemployed
Maria	Female	56	26	Cervical	Telephone	White British	Retired
Ethan	Male	42	36	Cervical	Telephone	White British	Retired

*Description of dystonia types:

Oromandibular- Dystonia of the mouth, tongue or jaw

Blepharospasm- Dystonia of the eyelids

Fixed- Fixed limb postures after peripheral injury

Segmental- Dystonia affecting two contiguous areas of the body

Generalised- Dystonia affecting multiple areas of the body

Cervical- Dystonia of the neck

Table 2. Interview schedule.

Topic area 1: Background and diagnosis

- Can you tell me about when you first experienced symptoms of dystonia?
- Can you tell me about any medical treatment you have received for dystonia?

Topic area 2: The physical impact of symptoms

- What kind of symptoms do you experience?
- What impact, if any, do your symptoms have on your day to day life?

Topic area 3: The psychological impact of symptoms

- Has the experience of having dystonia had any impact on how you feel/your state of mind?
- How do you feel about having dystonia?

Topic area 4: Support and coping

- Is there anything that helps you to manage dystonia?

- Are there any specific people or groups that help you to manage having dystonia?

Topic area 5: General issues

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

Table 3. Summary of themes.

Superordinate themes	Subordinate themes
Dealing with ignorance and uncertainty	Struggling to escape the darkness
	Walking on the treadmill of dystonia
	Acceptance and working for a positive future
The challenge of social isolation	Isolation as a threat to identity
	Visible difference impacting social experience
	Changing social identity in response to isolation
Fear of psychological explanations	

Appendix 2-A

Author guidelines

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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.

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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 .
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Appendix 2-B

Implications for rehabilitation

- Dystonia has a pervasive, negative impact on the lives of people with the condition.
- The struggle for diagnosis marks the beginning of a difficult period of psychological adjustment. The challenge of adjustment is compounded by social isolation and stigma.
- Support groups and peer interaction help people to integrate dystonia into their concept of a meaningful life and identity.
- Health professionals should play a pivotal role in assisting patients during the process of adjustment and on-going self-management through sensitive communication and signposting to wider support services.

Appendix 2-C

Excerpt from annotated transcript (Jennifer)

Notes	LINE NO.	TEXT	Themes
	96	P: Unless you actually put in	
	97	fixed dystonia my one for some	
	98	reason doesn't seem to be in the,	
	99	in the normal band of them, you've	
<u>'Soul searching', 'grief' implying loss and bereavement period.</u> First major mention of emotional impact beyond 'quite annoying'	100	got to, sort of, actually put the	
	101	word 'fixed' in, ermm, to sort of	
	102	know what my one, one is about	
	103	so, ermm, and it was four and a	
First mention of hope	104	half years of a lot of ... soul	Grief for past life
	105	searching I suppose, and sort of,	Soul searching
	106	ermm, grief, not knowing what I,	
<i>Emphatic- 'completely' 'Changed my life' it didn't change back, permanent.</i> Grief as mourning for old life.	107	what was up with my ankle, hoping	
	108	that obviously it was going to	
	109	improve and get better. It	
	110	completely changed my life in the	
	111	fact that I couldn't do lots of things	Permanent change
Repeated mention of how important work was for her	112	that I, I did before hand, ermm, I	
Panic attacks- she believes they're connected	113	also started suffering badly with	
	114	panic attacks and I later learned	Two problems
	115	that there is a linkup between	
<i>Never- journey still ongoing after diagnosis</i>	116	dystonia and panic attacks, ermm,	
<i>Right- life changed completely, for the worse/wrong</i>	117	so I had to give up work because	
	118	the panic attacks got so bad,	
	119	ermm, and obviously with the	
	120	ankle problem as well, ermm, so	
<i>'Finally', 'lots more things'- emphasis on time and inaccuracy of clinical activity</i>	[5]	it's basically, it's, it's, never been	
	121	right. They've finally come to the	
<u>My brain- physical attribution</u>	122	conclusion after having a lot more	
	123	things done after they discovered	
	124	it was fixed dystonia, ermm, that	
Hopeless to make it 'right'	125	that's it basically. My brain has	
	126	shut that particular thing down	
	127	that operates the, the usage of	
	128	that ankle, ermm, and there's	
	129	nothing now that will, that will	Trial and error
<i>'Hard struggle'- fighting</i>	130	open it up again. The only way it	
<u>Come to terms with- acceptance</u>	131	will work is if I'm under a general	
<i>Completely- totalising</i>	132	anaesthetic, ermm, and then my	
	133	ankle moves perfectly normally but	Physical attribution
<u>Coming to terms- soul searching. Emotional aspect</u>	134	as soon as I wake up again then	
<u>journey, flip side of clinical</u>	135	that's it, it's back to being	

<p><u>aspect</u>. She thinks their view is lacking in empathy?</p> <p>Positive- learning about mechanics. <u>Removing ignorance makes people feel better.</u></p> <p><u>'Everybody'- us vs them</u></p> <p>Their emotions are negative, dismissive ones. Complete opposite to what she wanted</p> <p>No point- hopeless</p> <p><u>They actively instilled fear of individual fault in her. You're different and it's in you (vs your unusual diagnosis)</u></p> <p>Stuck- refers directly to cyclical nature of journey and feeling of lack of progress/positive development</p> <p><u>Live with- new life after complete change.</u> Sounds like someone who is in the middle of an ongoing thing, not backwards looking/forwards looking like some</p> <p><i>Pauses, emphatic- this is a negative and final outcome</i></p> <p><i>Repeatedly describes new info as 'interesting', search for understanding. knowledge is valuable/ignorance scary</i></p> <p>'relief'</p> <p>Repetition of 'actually'- like success was a mirage</p> <p><u>In the dark- mystery, ignorance</u></p>	<p>136</p> <p>137</p> <p>138</p> <p>139</p> <p>140</p> <p>141</p> <p>142</p> <p>143</p> <p>144</p> <p>145</p> <p>146</p> <p>147</p> <p>148</p> <p>149</p> <p>150</p> <p>151</p> <p>152</p> <p>153</p> <p>154</p> <p>155</p> <p>156</p> <p>157</p> <p>158</p> <p>159</p> <p>160</p> <p>161</p> <p>162</p> <p>163</p> <p>164</p> <p>165</p> <p>166</p> <p>167</p> <p>168</p> <p>169</p> <p>170</p> <p>171</p> <p>172</p> <p>173</p> <p>174</p> <p>175</p> <p>176</p> <p>177</p> <p>178</p> <p>179</p> <p>180</p> <p>181</p>	<p>completely fixed. So, it's been a, a hard struggle and a lot to sort of, come to terms with, and ... it was interesting I suppose as well to sort of realise that there is a link, because I've had so much treatment for the panic attacks as well, ermm, it was nice to actually learn that the two were linked up because everybody that I was going to see, ermm, was sort of getting fed up basically, sort of saying, 'well it's not working, this works with everybody else, it's not working with you, there's no point in me seeing you any more', ermm, and I was beginning to sort of think, well what the hell is wrong with me, sort of thing. But then when they, sort of said to me that there is a link between the two, it sort of made me realise that I'm sort of stuck in this vicious cycle basically, ermm, and that the panic attacks and this was something that I've basically now got to live with ...</p> <p>I: You mentioned visiting with a doctor who, err, was able to give you some useful information and link the different things together-</p> <p>P: Yeah</p> <p>I: What was that like?</p> <p>P: Ermm, that, that was really interesting, it was, as I say it was a neurological hospital in [location], ermm, and it was a, a relief to actually go in and actually go in and see him, and actually be told you have got this condition, ermm, because prior to that obviously everybody was just completely in</p>	<p>Struggle</p> <p>Acceptance</p> <p>Emotional journey, not just physical</p> <p>Knowledge as a reward</p> <p>Us vs them</p> <p>Undervalued</p> <p>Hopeless</p> <p>Abandoned</p> <p>Internalising negative reaction of medics</p> <p>Blame/responsibility for difference</p> <p>Forced compromise</p> <p>Knowledge as a reward</p> <p>Shared ignorance</p>
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<u>Ignorance of professionals=ignorance of patient</u>	182	the dark as to why my ankle wouldn't move, ermm, and I did	
--	-----	--	--

Appendix 2-D

Table of emergent themes for one participant (Jennifer)

Themes	Line	Key Words/Phrases
<i>Solving the mystery of a rare condition</i>		
Shared ignorance	180	'Everybody was just completely in the dark'
Feeling blamed by medics	63/148/245	'making it all up' 'this works with everyone else' 'you're hard wired'
Doubting self	209/454	'why am I so different?' 'He's cured everybody, why not me?'
Relief of validation	189	'massive relief, I was so grateful to that guy for telling me that I'm not just making it up'
<i>Living with an unwanted guest</i>		
Unexpected arrival	978/264	'Suddenly' 'taken over' life
Personified problems	385	'The ankle' 'the panic' 'working together'
Lost life	220/387	'Very traumatic' 'no end of grief on every aspect'
Forced compromise	522/857	'Stuck with the condition', 'juggle what's best for each', doing 'the best I can' to 'live with it'
<i>The up and down cycle of medical intervention</i>		
Chain of engagements	38	'going to various hospitals, having various treatments done, operations, MRI scans etc. etc.'

Highs and lows	674/692	'very up and down ride', 'following a wavy road'
Cost of waiting and cost of treating	561	'I'm not going through that again'
Loss of hope	770/623	'I've hit a brick wall' 'I'm not ever going to get this fixed'
<i>Becoming stronger through adversity</i>		
Taking responsibility and feeling proud	1166/1174	'very proud' 'proving to myself that I could do it'
Toughened by negative experiences	1098	'nobodies got the right to tell me you haven't got fixed dystonia because I have'
Dystonia not the worst thing	785	'the panic attacks have more, ermm, been a hindrance in my life'
Kinder to self, less kind to others	1001/1008	'kinder to myself', 'selfish thing'
<i>Adjusting to being different</i>		
Visible impact of problem is permanent reminder of difference	875	'I'm constantly aware that I'm the odd one out, I'm the different one'
Emotional impact of others' behaviour	892	'Just hurts... People look at you strange'
Coping by masking problem	897	'I wear an ankle support on my ankle, not because I need the ankle support'
Distance from friends	963	'Some friends not prepared to listen, there's no point'
<i>Separating the body and the mind</i>		
Physical is more real than mental	254/175	'Genuine problem' not 'all in my head'
Externalising body parts	834	'The ankle'
Shame of mental health issues	369	'I didn't want anybody to know'
Experience of mental health issues increasing understanding	803	'with any sort of mental problem, I have a lot more patience and tolerance'

Appendix 2-E

Excerpt from table of group level themes

Dealing with ignorance and uncertainty	Line
<i>Struggling to escape the darkness</i>	
Jennifer- It was very stressful, very traumatic (...) when nobody believes you in the medical world, you just sort of think, well I'm on my own here, how, how am I going to resolve this?	280
Hailey- I sort of self-diagnosed really but the local hospital still weren't having it	133
Sheryl- I thought, well if she says I'm making it up, I know I'm not making it up (...) I'll just leave it and see what happens and we'll see if it is anything and it does get worse I may get somewhere	373
<i>Walking on the treadmill of dystonia</i>	
Matthew- I felt at first, quite optimistic, that a way of managing it could be found because you know, they'd say 'we'll try this drug and we'll try that drug' and as time went on and I went from one to another and Botox didn't work then I think it dawned on me that, you know, hold on, this thing isn't going to be managed	262
Sheryl- I found it hard in the end because I didn't know what to expect... he said they'd try to turn [DBS] up more, I got even worse still... And every time they'd try to go a bit higher to try and get things to improve I got worse. If anything I thought, oh things are much harder now than before the op	1016
Maria- I just think living with a movement disorder constantly, twenty-four-seven is just really tiring, it's just exhausting. You just think, I just want to be able to walk normally or sit normally somewhere and relax and just enjoy it but your, it's like you're trapped in your own body like, ermm, yeah, you're trapped within the dystonia if that makes sense	777

Acceptance and working for a positive future

- Mark- It's an acceptance, I've got it, err, you know, unless there's some sort (...) a silver bullet, that's going to magically appear, 1184
it's sort of, acceptance that I've got this for the rest of my life, so let's make the best of it, let's make the best of everyday
- Ethan- [It was] a relief to be, ermm, to know that it was not all in your head basically. It, it was all real. Ermm, there was people out 702
there, because with it being such a rare, obviously, disorder. Just good to meet people that actually knew what you were going
through. Ermm, they could understand the, obviously the pain and discomfort that you were, you were going through.
- Anna- I've had people messaging me saying, "it's really empowering to hear what you do and how you still carry on and how 486
you've adapted your life and get on with things" and I'm like, "ah actually I'm not doing that bad", (...) so it's been a boost for me
as well.
-

Section Three: Critical Appraisal

Reflections on the process of working with two different research paradigms

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Reflections on the process of working with two different research paradigms

This paper is a critical appraisal of the literature review and the main research paper of this thesis. In this paper I will reflect on the strengths and limitations of the thesis as a whole, as well as the process of working with two different research paradigms.

Implications for further research are also considered.

Research findings

In the literature review I found that disease severity and a range of “non-motor symptoms” of dystonia were linked with health related quality of life (HRQOL) in correlation and regression models. Higher levels of depression and anxiety were consistently found to be predictors of poorer HRQOL, whilst psychosocial factors such as body concept and self-esteem were found to have a positive relationship with HRQOL scores. Although a biopsychosocial model would consider depression and anxiety to be outcomes arising from the interaction between negative experiences and mediating psychosocial factors such as resilience [1], the reviewed papers employed these concepts as explanatory variables. As such, the models used in reviewed papers generally do not tell us much about *why* people with dystonia may become depressed or anxious. This understanding is particularly important for clinicians such as psychologists who target interventions towards psychosocial processes, and who create formulations to enhance understanding of these processes.

My interpretive phenomenological analysis (IPA) study of people’s experiences of living with dystonia helps to shed some light on this question. Participants described

a series of negative experiences which adversely affected their sense of identity, their relationships with health professionals and their attitude towards the future. One way of conceptualising the experiences recounted in the three themes of the IPA study is as different manifestations of isolation. The first theme described how difficult relationships with health professionals left participants feeling abandoned and forced to cope on their own. This caused feelings of uncertainty and fear, and ultimately led to participants empowering themselves and taking control of their care. The second theme revolved around the idea of social isolation caused by damaged self-esteem and practical barriers to social engagement. Peer interaction helped participants to reconstruct a positive sense of self and to commit to new, meaningful life goals. The third theme described experiences of alienation arising from stigmatised attitudes expressed by both participants and health care professionals. These attitudes informed conceptions of psychological distress as illusory and caused by a deficit of moral character in the form of a lack of determination or self-control. This conception was in contrast to the intensity and experiential “reality” of dystonia symptoms and related distress. This juxtaposition left some participants doubting their own judgement and even their sanity.

A range of psychosocial processes are therefore highlighted in the IPA study which could help to explain the impact of dystonia on HRQOL and the role it plays in experiences of depression and anxiety. This information echoes the findings of the papers from the literature review which attempted to investigate psychosocial variables [2, 3, 4] and together these insights provide a foundation from which researchers working with quantitative methods can choose their variables of interest, informed by psychological theories which are responsive to the complexity of the topic. Examples highlighted in the thesis include social identity theory [5], chronic illness as

biographical disruption [6], and the work of Graham et al. on illness representation and psychological flexibility [7, 8]. The findings of the IPA study also offer a new, complementary dimension to the findings discussed in the literature review, one of adjustment and change over time.

Reflections on the strengths and limitations of IPA

IPA was an appropriate choice of method for the main research paper. The approach was chosen in the hope that the idiographic, inductive and hermeneutic aspects of IPA would enable an open investigation of the topic, unburdened by the presumptions of researchers which characterise the quantitative literature. In this way, IPA facilitated one of the main aims of the paper, to provide a baseline of understanding that could inform future research and clinical intervention. As someone who works in the applied field of clinical psychology, the emphasis that IPA places on the relationship of the individual to their experiences, in the form of personal meaning [9], is of paramount importance. The majority of my clinical work is on a one to one basis and my main aim as a clinician is to help people to construe their experiences in a way which is productive and satisfying to them. The idiographic and phenomenological emphases of IPA facilitate this in a way which some other qualitative approaches such as discourse analysis or grounded theory do not. These approaches often focus on the level of systems and higher order theories, whilst IPA focuses on the experiential perspective of the individual [10, 11].

As with any research method, IPA is limited by its philosophical assumptions. However, unlike some more flexible approaches, IPA is explicit about these assumptions and the subsequent demands on the researcher. These demands include recognition that an individual's relationship to knowledge is necessarily mediated

through their subjective perspective, and that understanding of experience is an act of interpretation which occurs within a given context. The flexibility of the method is primarily in terms of choosing which features of this context to emphasise [12]. The extent to which I was responsive to these demands throughout the main paper is, I think, a strength of the thesis. The essential nature of perspective means that IPA exists in opposition to many scientific methods, for example those analysed in the literature review, which are predicated on a positivist epistemology. I would also argue that the ontological assumptions of IPA which necessitate the rejection of claims about a knowable, objective reality, position it as a quasi-scientific research method. Since IPA employs a hermeneutic cycle which layers interpretation on top of interpretation, I believe IPA is better understood as a creative work than a scientific one. The specific epistemological and ontological stances of IPA entail a series of limitations and potential pitfalls which are not typically applicable to quantitative approaches. I believe it is a more productive use of IPA to emphasise its strengths whilst articulating and critically engaging with its limitations, rather than trying to mitigate those limitations through adoption of popular conceptions of what constitutes rigour or “good science”. Attempts to codify IPA to make it compatible with these conceptions may actively minimise the potential strengths of the approach [13]. I will now illustrate how the main research paper of this thesis is in line with this position, whilst discussing some of the common alternative positions represented in the literature review.

Perhaps the most divisive issue in the IPA literature is the concept of rigour. Much has been written in favour of the position that the concepts of reliability and validity are inappropriate for IPA research, and this was the stance I took when designing and writing the main research paper. The writings which had the most influence on me personally were from De Witt and Ploeg [14] and Lincoln and Guba

[15]. It is not uncommon, however, to find IPA research which employs these concepts without addressing this debate. The argument against the use of reliability and validity centres on the claim that they have a foundation in positivism and realism [16]. In this context, the concept of reliability implies that the ability of the method to produce an identical outcome if performed again is a key aspect of its integrity. In this way the method can be seen as a kind of recipe with a consistent outcome. IPA depends on the hermeneutic cycle which assumes that any given interpretation is the product of a complex interaction of personal and contextual factors and will change indefinitely as these factors vary [17]. As a creative work, there is no definitive point at which one can say an IPA analysis is complete. In my case, I commented in my reflexive journal about the influence that my own personal experience of using the National Health Service (NHS) with an undiagnosed health condition had on how I looked at transcripts, even months after I thought I had completed my analysis. My experience gave personal salience to different aspects of the accounts of participants and made me see the work as a whole in a more sympathetic light. Although another researcher may produce a similar analysis given the same transcripts, it would be impossible to reproduce the exact conditions under which this analysis was written.

Regarding validity, the term is often used in a flexible manner and even in IPA research can be synonymous with the concept of trustworthiness [18]. Here I am referring to the common assumption of quantitative research, the idea that there exists external criteria against which knowledge claims can be judged [19]. The reification of concepts typically involved in this process of abstraction and deduction is not justified by the philosophical assumptions of IPA. Rather, IPA sees the experiences of individuals as a whole, irreducible from its context and relationships, both in terms of the information provided by participant testimonies and the interpretation of the

researcher [20]. As such, drawing comparisons between interpretations is a somewhat inductive process wherein the researcher or reader selectively attends to the aspects of the accounts which are most salient to them. In my journal I wondered if perhaps the very process of comparing accounts and crystallising perceived similarities into themes is in contradiction to the holistic aspects of IPA. I did my best to remain cognisant of this during analysis and to be cautious when making generalisations which effectively involve the idea of an “average” participant who emerges from the combination and reduction of accounts.

IPA analysis still benefits from rigour however, both in terms of assisting the researcher to work in a systematic way which is consistent across all aspects of the study, and in terms of assisting the reader in judging the quality of the work [21]. In my study I favoured the approach of legitimacy through transparency and resonance, informed by De Witt and Ploeg [14]. I employed these concepts in two ways, firstly as aspirational goals to which I would refer throughout the process to inform my decision making. Secondly, these concepts informed my understanding of the work as a communication to the reader and acted as a way for me to anticipate and affect their experience. I aimed to write a study that clearly communicated the reasoning for my decisions and that would be resonant for the reader. In service of transparency, I addressed the philosophical justification for my choices wherever possible and made quotations from participants the central focus of the paper. With regard to resonance, I believe the concept is central to IPA. The approach seeks to highlight aspects of experience that are most resonant for participants and the researcher creates from this an interpretation which is resonant for them [9]. It is on the basis of resonance therefore, that the researcher can choose between an effectively infinite number of possible interpretations. Similarly, the reader has to make a decision about the value of the work

without utilising a positivist rubric which makes reference to external truth. As a creative work the quality of IPA cannot be comprehensively judged according to a set of external standards, it is from the perspective of the reader that a work is either “good” or “bad”, useful or not [22]. If they feel the interpretation is meaningful to them and hence feels “true” from their perspective, then in that individual instance, the work can be considered a success [23]. Due to the necessity of interpretation from the reader, I believe they should be considered as the third part of the hermeneutic cycle. During the writing process I showed my work to someone with a research background and experience of chronic health issues. When they were moved by the recounting of participants’ experiences and the story being told, I felt confident that I had managed to transfer the emotional vigour that I experienced during interviews into the writing and I felt proud that I had done justice to the experiences of participants.

Bias is another controversial issue for IPA. Typically in positivist models, bias is seen as detrimental to good quality research. The concept of bracketing, the idea that through reflection one could identify and remove bias from interpretation, was originally integral to phenomenology and is still represented in the literature [11]. I believe that interpretation, bias and perspective are synonymous in this context. Bias is an irreducible part of experience and therefore from a phenomenological perspective, is part of “reality” [24] and thus bracketing is impossible [23]. Indeed, if researchers believe it is possible, this could create complacency and actively harm their ability to take a critical stance. Researchers should not aim to remove bias, rather they should highlight it where possible and engage with it critically. Throughout my work I asked myself, to what extent is my own perspective represented in the analysis as opposed to that of participants? The key issue I wanted to avoid was putting words in the mouths of participants, particularly with regard to the aforementioned “averaging” and abstraction

involved in generating themes. I wanted to have the most resonant and robust themes possible and at times the best way to do this would have been to interpret the testimony of participants to fit this end. Having the principles of rigour at hand helped me to adhere to interpretations which aimed at legitimacy and not expediency.

One of the fundamental limitations of IPA is that, due to the fact that interpretations are irreducible from their context, conclusions from IPA research cannot be generalised to a wider population [25]. There is no justification for assuming that what is true for one set of experiences will also be true for other people in ostensibly similar circumstances (i.e. external validity). Rather the concept of transferability should be used to judge the extent to which lessons learned from a set of experiences may inform the interpretation of another set [26]. Again, the reader plays a key role; it is their responsibility to make this assessment in light of their own experiences and interests. As such this puts the onus on the researcher to make the process of interpretation clear, to justify it with reference to participant testimonies and to embed discussions in the context of theory.

Reflections on the process of working with two different research paradigms

The first comment I made in my reflexive journal was about the challenge of simultaneously writing a literature review and a research paper, each with fundamentally different philosophical assumptions. Due to my stance as an epistemological pluralist, I was able to reconcile this tension. This position claims that in any given context there are a plurality of useful perspectives on knowledge [27] and that utilising a multiplicity of perspectives recognises and reflects the complexity and multifaceted ontology of any given phenomenon [28]. Gallivan writes about the moral dimension of pluralism versus methodological dogmatism with regards to information

systems research [29]. His claim is that a narrow focus on testing to the exclusion of the generation of ideas and exploration of theoretical blind spots is dangerous. I believe this issue is particularly pertinent with regards to psychology where research can have a direct impact on the wellbeing of individuals who are not equally represented in the process, for example service users, in the form of potentially harmful clinical interventions [30, 31, 32].

Although I felt justified in moving between perspectives the question remained, to what extent can the two papers can be considered commensurate and compatible? I favour a model of levels of analysis as proposed by Kuhn, whereby the two papers represent fundamentally different ways of looking at a similar phenomenon, and as such are incommensurate [33]. This position entails a rejection of the possibility of triangulation, the claim that the combination of different levels of analysis increases confidence of measurement [28]. For example, the triangulation of internal cognitive states, environmental factors and presumed biological bases as is often employed in psychiatric diagnosis [34]. The two levels of analysis cannot have simple correspondence, but could both be potentially useful in different contexts. I do not accept the position that choosing one perspective in a given context necessitates the rejection of the other in all contexts [35]. One of the claims of the theory of pragmatism is that researchers do not primarily make methodological choices on the basis of the philosophical assumptions of a given approach: practical concerns such as social and institutional agreements about values, disciplines and interests are more influential [36]. Although I find this a convincing position, I do not believe that this relieves the researcher of the responsibility to engage with the assumptions of their method. Philosophical choices must be justified and articulated, both for the integrity of the research and to aid the critical judgement of the reader.

There are multiple ways in which the two paradigms represented in this thesis could be considered compatible [28]. One example is provided by Papathanasiou et al. [37] who demonstrated how different paradigms could be used to inform one another by generating a structured questionnaire from qualitative interviews. Bager-Charleson et al. [38] constructed a qualitative interview schedule on the basis of quantitative questionnaires, providing an alternative approach to a sequential design [39]. In the case of this thesis, my choice of topics for the IPA study was influenced by findings from the literature review, whilst findings from the IPA study provide researchers with a number of ideas for potential variables which could be investigated using quantitative analysis.

Punch claims that one of the main consequences of choosing a particular methodological position is the impact on the way in which concepts are defined [40]. The difference between how psychological distress is conceptualised by papers in the literature review and participants in the research paper provides an effective example of this. The research analysed in the literature review operationalised depression and anxiety as scores on a psychometric test and conceptualised them in the context of a medical model of mental health, often describing them as “co-morbid” with dystonia. Within that paradigm I felt it was appropriate to engage with these concepts in this way, albeit from a critical perspective. Participants in the IPA study often made no reference to these medical concepts. Rather they spoke at length about distressing experiences such as isolation, pessimism about the future and damaged self-esteem, experiences which have their parallels in quantitative psychometric testing. Due to the phenomenological nature of IPA I felt it was appropriate to engage with this grounded, experiential conception of psychological distress. This illustrates another approach to the compatibility of paradigms, one which recognises the ontological complexity of phenomena by addressing both social and individual levels of analysis (i.e. structure and

agent) [28]. It could be argued that in this context, depression and anxiety are socially mediated medical constructs and represent in part the concept of “abnormal psychology” as social judgement [41].

As an applied psychologist, the reductionist process of abstraction and averaging of participants experiences through psychometric testing and inferential statistics is limited in what it can tell me about how best to help a given individual or how they might construe their experiences [42]. Rather, as a reader I must reflect on the given information and then examine my personal experience for insights that might be transferable to the individual in question. However, for organisations such as the NHS which operate on utilitarian premises, the concept of an abstract, average person is far more useful when making decisions about funding or best practice. In this way, although they are not commensurate, both levels of analysis are valuable to the practice of psychology.

Reflections on data collection and implications for future research

One aspect of the main research paper which illustrates the importance of methodological choices is the data collection, specifically the nature of the sampling and the interviews. It could be argued that the sample was self-selecting in that, by advertising to people who were engaged with the social media accounts of a charity which participates in political activism, I may have selected people who were particularly motivated to communicate the negative aspects of their experience. Several participants were explicit in their desire to raise awareness of dystonia and the plight of people with the condition. This may have led to the main paper giving a particularly negative impression of the experience of living with dystonia. Survey research found that 53% of people with dystonia said they were able to return to a normal routine at

some point and 70% said they had a positive relationship with their doctor, after a difficult process of diagnosis [43]. Although these findings may appear to contradict those of the main researcher paper, I would argue that the nature of the methodologies may explain the difference. An open and inductive IPA study such as this explicitly asks people to choose their most salient experiences and to speak about the emotional resonance of these experiences. In this sense I was asking participants to recount some of their most difficult experiences and to relive the emotion involved. I suspect that if participants were asked to make a contemporary judgement on their quality of life, most of them would have responded as the people in the survey study did. They had undergone a process of adjustment which helped them come to terms with their negative experiences but which did not remove their salience. Both approaches then offer a perspective on the phenomenon of having dystonia and provide insight into different aspects. Both are limited and potentially misleading if not interpreted in light of the methodology upon which they are based.

A more practical limitation of IPA that I encountered was related to the ubiquity of certain types of discourse, those which are predominantly associated with positivist and quantitative approaches to research and understanding. During interviews participants often spoke in descriptive, narrative terms about their experiences and required constant prompting to explicate *why* they had chosen to speak about the given experience, what it meant to them and how it made them feel. Their focus was on the “facts” of the situation, the order of events and who was involved. This was the case even after a pre-interview briefing about the style of the interview and its aims, and it took a significant amount of time for participants to adjust to the specific demands of the interview. It occurred to me that in an hour long interview, the rate and extent to which participants adapt to these demands is likely to have an impact on the nature of

the resulting transcript. That proficiency impacts upon one's capacity for phenomenological reflection was one of the original insights of Husserl [9].

Due to the impact of proficiency and the fact that IPA often asks people to make meaning of events in a post hoc fashion, often years later, I would like to see further research involving repeated interviews with the same participants. A study could interview people with dystonia at several different points, starting after diagnosis, and then at intervals thereafter. This design would be responsive to the idea that meaning making changes over time and that coping with chronic health issues is a process, as was reported by participants in my study. Participants would have a chance to develop their own understanding of phenomenological inquiry and questions could be focused more on recent experiences and involve less post hoc reflection.

Conclusion

In this thesis I attempted to maintain a critical awareness of the limitations of two different paradigms, whilst maximising the strengths of each approach. In this critical appraisal I have made an argument for the compatibility of the paradigms, whilst highlighting the limits of commensurability. This critical awareness was made possible through reflection and engagement with the philosophical assumptions of each approach. The process of philosophical engagement and the maintenance of a critical stance is important to help researchers to produce rigorous work and communicate it to the reader in a way which facilitates their own interpretation and application of the work.

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Section Four: Ethics Section

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Faculty of Health and Medicine Research Ethics Committee (FHMREC) Lancaster University

Application for Ethical Approval for Research involving direct contact with human participants

1. Apply to the committee by submitting:
 - a. The University's Stage 1 Self Assessment (part A only) and the Project Questionnaire. These are available on the Research Support Office website: [LU Ethics](#)
 - b. The completed application **FHMREC form**
 - c. Your full research proposal (background, literature review, methodology/methods, ethical considerations)
 - d. All accompanying research materials such as, but not limited to,
 - 1) Advertising materials (posters, e-mails)
 - 2) Letters/emails of invitation to participate
 - 3) Participant information sheets
 - 4) Consent forms
 - 5) Questionnaires, surveys, demographic sheets
 - 6) Interview schedules, interview question guides, focus group scripts
 - 7) Debriefing sheets, resource lists

Please note that you DO NOT need to submit pre-existing handbooks or measures, which support your work, but which cannot be amended following ethical review. These should simply be referred to in your application form.

2. Submit all the materials electronically as a **SINGLE email attachment in PDF format** by the deadline date.
3. Submit one collated and signed paper copy of the full application materials

in time for the FHMREC meeting. If the applicant is a student, the paper copy of the application form must be signed by the Academic Supervisor.

4. Committee meeting dates and application submission dates are listed on the [FHMREC website](#). Applications must be submitted by the deadline date, to:

Dr Diane Hopkins

B14, Furness College

Lancaster University,

LA1 4YG

d.hopkins@lancaster.ac.uk

5. Prior to the FHMREC meeting you may be contacted by the lead reviewer for further clarification of your application.
6. Attend the committee meeting on the day that the application is considered, if required to do so.

1. **Title of Project:** Experiences of living with dystonia

2. **Name of applicant/researcher:** Andrew Morgan

3. **Type of study**

Includes *direct* involvement by human subjects.

Involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants. Please complete the University Stage 1 Self Assessment part B. This is available on the Research Support Office website: [LU Ethics](#).

Submit this, along with all project documentation, to Diane Hopkins.

4. If this is a student project, please indicate what type of project by marking the relevant box: (please note that UG and taught PG projects should complete **FHMREC form UG-tPG**, following the procedures set out on the [FHMREC website](#))

PG Diploma	<input type="checkbox"/>	Masters dissertation	<input type="checkbox"/>	DClinPsy SRP	<input type="checkbox"/>	PhD Thesis	<input type="checkbox"/>	PhD Pall. Care	<input type="checkbox"/>
PhD Pub. Health	<input type="checkbox"/>	PhD Org. Health & Well Being	<input type="checkbox"/>	PhD Mental Health	<input type="checkbox"/>	M D	<input type="checkbox"/>		
DClinPsy Thesis	<input checked="" type="checkbox"/>								

Applicant Information

5. Appointment/position held by applicant and Division within FHM Trainee
Clinical Psychologist

6. Contact information for applicant:

E-mail:
a.morgan2@lancaster.ac.uk

Telephone: OMITTED (please give a number on which you can be contacted at short notice)

Address: Faculty of Health and Medicine, C12 Furness College, Lancaster University, Lancaster, LA1 4YG

7. Project supervisor(s), if different from applicant: Dr Pete Greasley, Dr Fiona Eccles

8. Appointment held by supervisor(s) and institution(s) where based (if applicable):
Dr Greasley: Teaching Fellow, Department of Clinical Psychology, Lancaster University;
Dr Eccles: Lecturer in Research Methods, Department of Clinical Psychology, Lancaster University

9. Names and appointments of all members of the research team (including degree where applicable)

Andrew Morgan, BA (Hons), Graduate

Diploma Psychology Pete Greasley

BSc(Hons), PhD

Fiona Eccles MPhys, DPhil, Graduate Diploma Psychology, DClInPsy

The Project

NOTE: In addition to completing this form you must submit a detailed research protocol and all supporting materials.

10. Summary of research protocol in lay terms (indicative maximum length 150 words):

This is a qualitative study looking at the experiences of people living with a neurological movement disorder called dystonia. Dystonia is characterised by involuntary, repetitive movements and abnormal postures. Individual interviews with approximately eight participants will be conducted to gain an insight into the psychological issues experienced by those living with the condition. The interview will ask them open ended questions to enable them to talk about the things that are most important to them about living with this condition. The principal investigator will examine participants' responses and elicit themes that connect people within this group. These themes will be discussed in the write up with reference to what they can tell researchers and health professionals about the best way to enhance the psychological wellbeing of people living with dystonia.

11. Anticipated project dates (month and year only)

Start date: December 2014

End date May 2018

12. Please describe the sample of participants to be studied (including maximum & minimum number, age, gender):

Approximately 6-12, aged 18 and over

13. How will participants be recruited and from where? Be as specific as possible

The Dystonia Society has agreed to publicise this research through their national support group meetings and the distribution of participant information sheets to members on their mailing list, pending ethical approval. Due to the geographical location of the researcher, recruitment will focus upon the northwest, particularly groups meeting in Manchester and Liverpool. The principal investigator will attend Manchester and Liverpool group meetings, with the consent of group members, to discuss the study and distribute information to group members. If group members are interested in taking part, they will be able to contact the principal investigator at their convenience to arrange an interview. If it is not possible to arrange a mutually appropriate location for a face-to-face interview, a telephone or skype interview may be conducted. In this case, a consent form will be posted to the participant with a stamped, addressed envelope which they will return prior to the meeting. Participants will be made aware prior to any skype/internet interview that these interviews cannot be totally secure and if they wish to withdraw at this point then they may.

As a contingency plan, if it is not possible to recruit sufficient participants via support group

meetings, social media networks, such as Twitter, attached to the Dystonia Society will be contacted. The study will be advertised through these networks to boost recruitment numbers to the desired range.

14. It is intended that information about the study will be distributed to the support group meetings or through the Dystonia Society mailing list, so that people can consider their involvement in their own time and not feel pressured to take part. If they decide they would like to take part, they can make contact with the researcher to arrange an interview. They will be asked to sign a consent form at that interview. If a telephone or skype interview is to be conducted, consent forms will be sent to potential participants via post for them to return prior to interview. Potential participants will be judged to be capable of giving informed consent unless there is reason to think otherwise. Examples of reasons why informed consent may not be considered applicable would include presentation indicative of intellectual disability, at which point, if they wanted to take part, consent would be sought from their carer in keeping with the standards set out by the Mental Capacity Act 2005.

15. What discomfort (including psychological eg distressing or sensitive topics), inconvenience or danger could be caused by participation in the project? Please indicate plans to address these potential risks.

There is the potential for distressing material to arise during interviews. The researcher will also be mindful of the wellbeing of the participant throughout the interview. Participants will be given time to think about their answers and will be encouraged to take a break if the need arises. Participants will also be able to stop the interview at any time and be offered the opportunity to reschedule if they so wish. Participants will be encouraged to raise appropriate issues within the context of their Dystonia society support group if necessary. Participants will be given the contact information of relevant local support services as part of their participant information sheet.

16. What potential risks may exist for the researcher(s)? Please indicate plans to address such risks (for example, noting the support available to you; counselling considerations arising from the sensitive or distressing nature of the research/topic; details of the lone worker plan you will follow, and the steps you will take).

The investigator will be working alone and as such will do so in accordance with the Lancashire Care Lone Worker Policy. In accordance with this policy, the environment of interview locations will be assessed with regards to any hazards. The investigator will familiarise themselves with the emergency procedures of the facility, such as evacuation routes. As interviews will be conducted in public buildings rather than participants homes, it is anticipated that there will be people in the area. The researcher will log details of the timing and location of the meeting with staff working at the premises. The researcher will abide by the facilities' signing in procedures and make themselves known to staff. The researcher will also have a mobile phone with them in case of emergency.

17. Whilst we do not generally expect direct benefits to participants as a result of this research, please state here any that result from completion of the study.

This study will increase understanding of the experience of living with dystonia, to be shared with relevant health care professionals via reports and publication. Participants may also experience psychological benefit from discussing their experiences.

18. Details of any incentives/payments (including out-of-pocket expenses) made to participants:

Travel costs can be reimbursed up to the value of £20, via the Lancaster University Clinical Psychology department expenses system.

19. Briefly describe your data collection and analysis methods, and the rationale for their use. Please include details of how the confidentiality and anonymity of participants will be ensured, and the limits to confidentiality.

This study will use individual, semi-structured interviews adopting an interpretive phenomenological approach and analysis (IPA). Participants will be encouraged to talk about the issues that are most salient to them, so an open format should promote this. This approach is intended to put the expertise of participants at the forefront of the project. IPA was chosen because of the importance of understanding the personal experience of the participants.

As the possibility exists that sensitive material may be disclosed during participant interview, standard duty of care/confidentiality procedures will be in place whereby the investigator is obliged to report information, which leads them to believe that the participant or someone else may be at significant risk of harm, to the relevant service.

20. If relevant, describe the involvement of your target participant group in the design and conduct of your research.

Advice regarding study design and methodological decisions has been gained from the Dystonia Society, via their information telephone line and correspondence with their board members. Discussions about the nature of the condition and the experience of living with it have informed the choice of IPA analysis. The Dystonia Society have also offered advice regarding making contact with potential participants and the importance of respecting the boundaries of group meetings, i.e. not making contact without prior approval from support group members. The Lancaster University Public Involvement Network (LUPIN) were also involved in the initial stages of development.

21. What plan is in place for the storage of data (electronic, digital, paper, etc.)?

Please ensure that your plans comply with the Data Protection Act 1998.

Interviews will be recorded on a digital audio recorder and then transferred as soon as possible after each interview to password protected file space on the Lancaster University server where they will be saved as encrypted files and then deleted from the audio recorder. They will then be transcribed by the investigator.

Participant names will be replaced with pseudonyms and any identifiable details removed. The initial audio recordings will be deleted after the study has been assessed.

Once interview recordings are transcribed, a key will be created to convert participant names into ID codes or pseudonyms and stored in encrypted files on the university server, separate from the study data. This identifying information will be deleted once the project is assessed. It is important that participants are able to withdraw their consent at any point, and ask for their data to be withdrawn up until the project is complete.

Participants will be made aware of this through the consent form. After the study is complete, anonymised versions of the research data will be transferred to the university system and stored for 10 years. Consent forms will be scanned and stored electronically, in a separate location from other documents. All personal data stored by at the university is in the custody of Dr Bill Sellwood, programme director for the clinical psychology doctorate.

22. Will audio or video recording take place? no audio video
If yes, what arrangements have been made for audio/video data storage? At what point in the research will tapes/digital recordings/files be destroyed?

Interviews will be recorded on a digital audio recorder and then transferred as soon as possible after each interview to password protected file space on the Lancaster University server where they will be saved as encrypted files and then deleted from the audio recorder. They will then be transcribed by the investigator.

Participant names will be replaced with pseudonyms and any identifiable details removed. The initial audio recordings will be deleted after the study has been assessed. Audio files will be uploaded from the recorder to secure servers at the earliest possible opportunity as the recorders themselves cannot be encrypted.

23. What are the plans for dissemination of findings from the research? If you are a student, include here your thesis.

The finding will be written up into a thesis as part of the doctorate in clinical psychology. The findings will be presented to the Dystonia Society for their own information. They could also be presented to individual local group meetings if they should choose. It is intended that the research will be submitted to a peer reviewed journal. If the opportunity arises, this information would also be presented at academic conferences.

24. What particular ethical considerations, not previously noted on this application, do you think there are in the proposed study? Are there any matters about which you wish to seek guidance from the FHMREC?

None

Signatures: Applicant: Andrew Morgan

Date: 18/08/2017

*Project Supervisor (if applicable): Pete Greasley



Date: 18/08/2017

*I have reviewed this application, and discussed it with the applicant. I confirm that the project methodology is appropriate. I am happy for this application to proceed to ethical review.



Applicant: Andrew Morgan
Supervisor: Pete Greasley
Department: Health Research
FHMREC Reference: FHMREC15140

05 September 2016

Dear Andrew

Re: Experiences of living with dystonia.

Thank you for submitting your research ethics amendment application for the above project for review by the **Faculty of Health and Medicine Research Ethics Committee (FHMREC)**. The application was recommended for approval by FHMREC, and on behalf of the Chair of the Committee, I can confirm that approval has been granted for this research project.

As principal 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 licenses 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 details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information.

Tel:- [REDACTED]

Email:- [REDACTED]

Yours sincerely,

[REDACTED]



**Faculty of Health and Medicine Research Ethics Committee (FHMREC) Lancaster University
Application for Amendment to Previously Approved Research**

1. Name of applicant: **Andrew Morgan**
2. E-mail address and phone number of applicant: **a.morgan2@lancaster.ac.uk 07970529372**
3. Title of project: **Experiences of living with dystonia**
4. FHMREC project reference number: **FHMREC14024**
5. Date of original project approval as indicated on the official approval letter (month/year): **01/2015**
6. Please outline the requested amendment(s)
Note that where the amendment relates to a change of researcher, and the new researcher is a student, a full application must be made to FHMREC
Extend the expected completion date to 05/2018
7. Please explain your reason(s) for requesting the above amendment(s):
I am returning to the project after several months of ill health so the original timescales are not applicable

Guidance:

- a) Resubmit your research ethics documents (the entire version which received final approval, including all participant materials, your application form and research protocol), with all additions highlighted in yellow, and any deletions simply 'struck through', so that it is possible to see what was there previously.
- b) This should be submitted as a single PDF to [Diane Hopkins](#). There is no need to resubmit the Governance Checklist

Applicant electronic signature: **Andrew Morgan** Date **19/08/17**

Student applicants: please tick to confirm that you have discussed this amendment application with your supervisor, and that they are happy for the application to proceed to ethical review

Project Supervisor name (if applicable): **Pete Greasley** Date application discussed **20/08/17**

You must submit this application from your Lancaster University email address, and copy your supervisor in to the email in which you submit this application

July 2016

Appendix 4-A

Thesis protocol (v1 25/10/2014) for study title:

Experiences of living with dystonia

Principal Investigator: Andrew Morgan, Trainee Clinical Psychologist, Lancaster University

University Project Supervisor: Dr Pete Greasley, Teaching Fellow, Lancaster University Field

Supervisor: Dr Fiona Eccles, Lecturer in Research Methods, Lancaster University

Background

Dystonia is a neurological condition affecting control of movement and typically features involuntary, repetitive and sustained muscle contractions and postures (Lewis, Butler, & Jahanshahi, 2008). People with dystonia are commonly categorised either in terms of the location of symptoms in the body, the age of onset or aetiology (Degirmenci, Oyekcin, Bakar, & Kurklu, 2013). Generalised dystonia is diagnosed when spasms occur in a variety of areas of the body; focal dystonia is diagnosed when spasms tend to stay localised to a particular body part. Segmental dystonia involves spasms in multiple, adjoining locations. When dystonia is categorised according to aetiology, it is typically divided into idiopathic, inherited (genetic) and acquired categories.

Dystonia is the third most commonly treated movement disorder after Parkinson's disease and essential tremor, and is estimated to occur in between 15 and 30 people in every 100,000 (Phukan, Albanese, Gasser, & Warner, 2011). From a neurological perspective, dystonia is most commonly explained as being a dysfunction of the basal ganglia involving abnormal dopaminergic activity which results in a lack of inhibitory control over movement (Degirmenci et al., 2013), although understanding of the condition is still developing. The symptoms comprising dystonia can be produced by a range of causes including neurological degeneration, brain injury, genetics and substance use; the Dystonia Society website (Dystonia Society, 2014) lists more than 50 known causes.

Although it is defined as a movement disorder, the most significant negative impacts of dystonia are pain resulting from uncomfortable posture, and psychological distress (Lim, 2007; Miguel, Tiago, & Ferreira, 2009). Pekmezovic et al. (2009) found that, along with diagnosis of depression and anxiety, pain was the most effective predictor of poor quality of life in people with dystonia.

The causes of psychological distress are multiple. Evidence has been found that poor body and self-concept, social isolation and stigma, and disruption of activities of daily living are amongst the best predictors of impaired quality of life (Basurovic, Svetel, Pekmezovic, & Kostic, 2012; Lewis et al., 2008). Studies have found that, for people experiencing symptoms of dystonia, quality of life is impaired across all domains of the SF-36 assessment (Ware Jr & Gandek, 1998)

when compared with a non-clinical sample. Quality of life was particularly impaired with regards to physical and social functioning (Camfield, Ben-Shlomo, & Warner, 2002; Page, Butler, & Jahanshahi, 2007).

There is an association between the experience of dystonia symptoms and receiving a diagnosis of psychological disorder, the most common example of which is major depressive disorder. Some estimates claim a 25% prevalence of depression in people suffering with dystonia (Jahanshahi, 2005). There is also evidence of an overrepresentation of anxiety and obsessive compulsive disorder diagnoses amongst people with dystonia.

Sleep problems are common and stress and fatigue have been found to negatively impact on physical, social and psychological functioning in people with the condition (Avanzino et al., 2010; Miguel et al., 2009). The implication of this is that, without adequate psychological support, people experiencing the symptoms of dystonia could be at risk of entering into a negative cycle of distress. Intervention is predominately aimed at relieving symptoms and most commonly takes the form of medication. When this proves ineffective then invasive surgery involving deep brain stimulation can be employed (Ben-Shlomo, Camfield, & Warner, 2002).

There have been increasing calls to develop our understanding of the psychological impact that dystonia has so that candidates for therapeutic intervention can be properly identified and person centred interventions tailored to individuals' specific needs (Reckess, Zahodne, Fennell, & Bowers, 2009). As a life-long condition, the sooner that people in need of intervention can be identified and helped, the more the burden of the condition can potentially be reduced (Degirmenci et al., 2013). Furthermore, given the chronicity of psychological difficulties including depression, providing support early could potentially reduce the need for future intervention from services.

The vast majority of research into the impact of the condition and its interventions has been quantitative studies using self-report quality of life measures (Diamond & Jankovic, 2005; Kuyper, Parra, Aerts, Okun, & Kluger, 2011). These studies have provided estimates of prevalence of psychological diagnoses, and often utilised regression analysis to isolate relevant variables thought to contribute to effects on quality of life. This is both in terms of

negative impacts of the condition and the mediating influence of psychological variables such as locus of control on extent of negative impact.

A search of the literature found one qualitative study on psychological aspects of dystonia, which looked at the impact of deep brain stimulation on participants' experiences (Hariz, Limousin, Tisch, Jahanshahi, & Fjellman-Wiklund, 2011). Interviews were analysed using grounded theory and focused in large part on the changes brought about in participants' lives as a result of treatment. The researchers commented on the ability of qualitative methods to illicit new information about the needs of patients and to chart the development of their struggle with symptoms over an extended period of time.

Existing knowledge about the experience of living with symptoms of dystonia and the impact of symptoms on psychological wellbeing is limited. The information that has been gathered is limited predominately to that derived from quantitative designs, as described above. The development of the topic area would be promoted therefore, by engaging with the experiences of people living with the condition through qualitative methods. Understanding of key aspects of the disorder and its psychological impact could be increased and this could lead to better targeting and development of interventions for people with dystonia experiencing psychological distress.

This study aims to contribute to this development by conducting a qualitative analysis of interviews with people about their personal experiences of living with the condition. Due to the current state of research in the field, this study is intended to be exploratory, and as such will explore fundamental areas including the impact of symptoms on work, home life and psychological wellbeing, the use of external and internal sources of support, and the experience of coping with symptoms. Interviews will be analysed using interpretative phenomenological analysis (IPA).

Design

Data Collection:

Since this is a qualitative IPA study, which aims to produce an in-depth analysis of participants' experiences, a small sample of 6-12 people is required. Individual semi-structure interviews will be used; it is expected that interviews will take between 60-90 minutes. Semi- structured interviews were chosen as they allow for participants' experiences to guide the investigation. Interviews will be structured around existing theory and issues raised through quantitative and qualitative research into dystonia and similar related conditions such as Parkinson's Disease. Members of the Dystonia Society have also been consulted. Topic areas will focus on the experience of being diagnosed, the experience of receiving treatment, the nature of symptoms and how they impact upon home and working life, and methods of coping with psychological distress. There will also be the opportunity for participants to discuss issues that they feel are important.

This approach to interviewing is appropriate for an IPA study as it facilitates discussions of participants' personal experiences and the points that they feel are most pertinent.

At the start of each interview written consent will be obtained from the participant. Interviews will be recorded using a digital audio recorder. The interview schedule will be used to initiate and guide interviews, although the line of questioning will be responsive to the participant's answers and direction. The researcher's academic supervisor will listen to an early interview recording to check the researcher's interview style and line of questioning.

Recruitment:

The Dystonia Society have given permission to make contact with their regional support groups for people living with dystonia, pending ethical approval and their subsequent approval of the project.

Assuming the consent of group members is given, the principal investigator will attend group meetings and discuss the study in person. Should this not be possible, information will be sent to the groups via post or email.

Groups will be given information sheets to distribute amongst their attendees or through the mailing list, which inform members about the study and provide contact details for the principal

investigator. Group members will be asked to email or write to the principal investigator with their own contact details. Upon receipt of contact details, the researcher will contact participants to arrange a convenient interview time in their local area. Local facilities such as libraries and community centres will be used as a neutral venue for interviews. Participants must be 18 years old or over. Due to the geographical location of the principal investigator, recruitment will focus initially on groups located in the north west of England.

If it is not possible to recruit the intended amount of people from the North West, then the contingency plan is to utilise the Dystonia Society's social networks, such as Twitter, and website to advertise the study to a wider audience. Where it is not feasible to conduct a face-to-face interview, a telephone or skype interview may be conducted. In this case, a consent form will be posted to the participant with a stamped, addressed envelope which they will return prior to the meeting. Participants will be made aware prior to any skype/internet interview that these interviews cannot be totally secure and if they wish to withdraw at this point then they may.

Data Analysis:

Data from the interviews will be analysed through interpretative phenomenological analysis. This approach is intended to access accounts of the personal experience of participants from a particular group, and the understanding they have made of their experiences (Brocki & Wearden, 2006). It seeks to avoid making general claims about the experience of individuals outside this group (Smith & Osborn, 2008).

IPA is a dynamic process wherein the researcher attempts to engage with the personal meaning of the participant and interpret their accounts in the context of existing theory. This process is also necessarily influenced by the researcher's own assumptions and way of making meaning (Brocki & Wearden, 2006). The analysis extends beyond a list of participants' experiences and involves the researcher's critical interpretation of them.

The vast majority of IPA use is in health psychology (Smith, 1999). This is partly due to the developing perception of illness as more than a biomedical pathology and seeing it also as a social construction (Brocki & Wearden, 2006). IPA may be a complementary approach to quantitative analysis because it can handle potentially more complex explanations involving many different variables or ideas of interest, and it can challenge assumptions built into

theoretical models by researchers (Johnson, Burrows, & Williamson, 2004). IPA deals with individuals' narratives and given the long-term nature of many health conditions, including dystonia, this ability to account for the passage of time is considered a strength of the approach. This study will follow the guidance of Smith and Osborn (2008). The research question has been intentionally left broad to allow flexible investigation of the area and not the testing of a hypothesis. The estimate of 6-12 participants is based upon the ability of a single researcher to dedicate the requisite amount of time to a detailed analysis of interviews and is in accordance with standard use of IPA (Reid, Flowers, & Larkin, 2005).

Transcripts will be read repeatedly by the researcher and annotated according to information of interest. This process is deliberately open ended to allow meaningful information to come forth from the text (Smith & Osborn, 2008). After a transcript has been re-read, it will be annotated again with emerging theme titles. This represents a process of abstraction where more psychological constructs are invoked to understand the data. Emergent themes are then collected and the researcher groups them according to superordinate themes. Final themes are then written up in the form of a narrative account which seeks to interpret personal testimonies into a structure which relates to psychological theory. Throughout this process, the researcher will refer back to the original transcripts to ensure interpretation is grounded in the data; academic supervisors will be checking this process to help the researcher monitor his own influences on the information.

Dissemination:

Results of the research will be written up as a research report as part of the thesis of the principal investigator. This work is a requirement for completion of a doctorate in clinical psychology. It will also be disseminated via the Dystonia Society who has expressed interest in the findings. Upon invitation, the investigator will attend relevant local group meetings or provide them with written information to disseminate findings. It is intended that the research be submitted to a peer reviewed journal. If the opportunity arises, this information would also be presented at academic conferences

Ethical considerations:

There is the potential for sensitive material to arise during interview. Participants will be encouraged to raise appropriate issues within the context of their Dystonia society support group. Participants will be given the contact information of relevant local support services as part of their participant information sheet. The researcher will also be mindful of the wellbeing of the participant throughout the interview. Participants will be given time to think about their answers and will be encouraged to take a break if the need arises. Participants will also be able to stop the interview at any time and offered the opportunity to reschedule if they so wish.

Confidentiality will be respected, although with recognition of the duty of the investigator to report information that has implications for the safety of themselves or others this information to health services or the police as appropriate. Efforts will be made to communicate with the participant in such cases.

It is hoped that the opportunity to contribute to the development of health services for people with dystonia and to register their views and experience will justify any cost or risk of harm to participants. Based on this principle, the findings of the study will be made available for feedback to participants, if they wish. There is the potential that the findings of this study could improve provision of psychological intervention for this population.

During the publication process, care will be taken in to avoid any combination of incidental details which could lead to individuals being identifiable.

Data storage:

Interviews will be recorded on an electronic audio recorder and then transferred as soon as possible after each interview to password protected file space on the Lancaster University server where they will be saved as encrypted files and then deleted from the audio recorder. They will then be transcribed by the investigator at which point participant names will be replaced with pseudonyms and any identifiable details removed. The initial audio recordings will be deleted once the study has been submitted for assessment. The initial audio recordings will be deleted after the study has been assessed. Audio files will be uploaded from the recorder to secure servers at the earliest possible opportunity as the recorders themselves cannot be encrypted.

Once transcribed, a key will be created to convert participant names into ID codes or pseudonyms and stored in encrypted files on the university server, separate from the study data. This identifying information will be deleted once the project is assessed. It is important that participants are able to withdraw their consent at any point, and ask for their data to be withdrawn up until the project is complete. Participants will be made aware of this through the consent form. After the study is complete, anonymised versions of the research data will be transferred to the university system and stored for 10 years. Consent forms will be scanned and stored electronically, in a separate location from other documents. All personal data stored by at the university is in the custody of Dr Bill Sellwood, programme director for the clinical psychology doctorate. Potential participants will be judged to be capable of giving informed consent unless there is reason to think otherwise. Examples of reasons why informed consent may not be considered applicable would include presentation indicative of intellectual disability, at which point, if they wanted to take part, consent would be sought from their carer in keeping with the standards set out by the Mental Capacity Act 2005.

Practicalities:

Participants may be asked to travel within their local area to attend the interview. Local facilities such as libraries and community centres will be contacted to arrange a location for interview. This decision was made in collaboration with the Dystonia Society who agreed that, given that support group members cannot be contacted regarding their preferences before ethical approval is sought, this represented the best compromise available.

Participants will have their travel costs reimbursed up to the value of £20, via the Lancaster University expenses system. As funding is not available for a translator, only participants who speak English will be considered eligible to participate. Further to this, the process of interpretation could prove problematic for in depth, IPA analysis.

The investigator will be working alone and as such will do so in accordance with the Lancashire Care Lone Worker Policy. In accordance with this policy, the environment of interview locations will be assessed with regards to any hazards. The investigator will familiarise themselves with the emergency procedures of the facility, such as evacuation routes. As interviews will be conducted in public buildings rather than participants homes, it is anticipated that there will be people in the area. The researcher will log details of the timing and location of the meeting with

staff working at the premises. The researcher will abide by the facilities' signing in procedures and make themselves known to staff. The researcher will also have a mobile phone with them in case of emergency.

Service user involvement:

The Dystonia Society is involved in the development of the project. They have helped to facilitate access to participants and to address theoretical assumptions drawn from the literature by the researcher. The Lancaster University Public Involvement Network (LUPIN) were also involved in the initial stages of development.

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Appendix 4-B

Consent form (v1 25/10/2014) for:

Experiences of living with dystonia

We are asking if you would like to take part in a research project looking at people's experiences of living with dystonia. Before you consent to participate in the study we ask that you read the participant information sheet and mark each box below with your initials if you agree. If you have any questions before signing the consent form please speak to the principal investigator, Andrew Morgan.

- | | Please initial box
after statement |
|---|---------------------------------------|
| 1. I confirm that I have read the information sheet and fully understand what is expected of me within this study | <input type="checkbox"/> |
| 2. I confirm that I have had the opportunity to ask any questions and to have them answered. | <input type="checkbox"/> |
| 3. I understand that my interview will be audio recorded and then made into an anonymised, written transcript. | <input type="checkbox"/> |
| 4. I understand that audio recordings will be kept until the research project has been examined. | <input type="checkbox"/> |
| 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. I can request that my data be removed from the study and destroyed up to the point of publication. | <input type="checkbox"/> |
| 6. I understand that the information from my interview will be pooled with other participants' responses, anonymised and may be published | <input type="checkbox"/> |
| 7. I consent to anonymised information and quotations from my interview being used in reports, conferences and training events. | <input type="checkbox"/> |

- 8. I understand that any information I give will remain strictly confidential and anonymous unless it is thought that there is a risk of harm to myself or others, in which case the principal investigator may need to share this information with their research supervisor.

- 9. I consent to Lancaster University keeping written transcriptions of the interview for 10 years after the study has finished.

- 10. I consent to take part in the above study.

Name of Participant _____ Signature _____ Date _____

Name of Researcher _____ Signature _____ Date _____

Appendix 4-C

Participant Information Sheet (v1 24/10/2014) for:

Experiences of living with dystonia

My name is Andrew Morgan and I am conducting this research as part of my doctoral training on the Clinical Psychology programme at Lancaster University.

What is the study about?

The purpose of this study is to investigate the experiences of people living with dystonia. It is hoped that by doing this we can learn more about dystonia and the best way to offer support.

Why have I been approached?

You have been approached because the study is looking at the experiences of people who are living with dystonia. Hearing about your experiences could be helpful in improving our understanding of dystonia and how best to help people who live with it.

Do I have to take part?

No. It's completely up to you to decide whether or not you take part and your decision will not affect the care you receive or your participation within any groups or organisations.

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

If you decide you would like to take part, you will be asked to provide your email address or telephone number so that I can arrange an interview which may last for about an hour. The interview will be quite informal asking you about what living with dystonia is like for you, what effects it has on your life and how you cope with symptoms. You will be encouraged to talk about the issues that are important to you. People taking part in the study will be given the opportunity to take part in a second interview if they would like to. If it is not possible to meet in person, the interview could be conducted over the telephone or via internet telephone programs such as Skype.

Will my data be confidential?

The information you provide is confidential. The data collected for this study will be

stored securely and only the researchers conducting this study will have access to this data.

- Audio recordings will be deleted after the work has been assessed
- The files on the computer will be encrypted (that is no-one other than the researchers will be able to access them) and the computer itself is 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.
- At the end of the study, anonymised versions of the data will be kept on secure computers for up to ten years and only be accessible to the researchers.

There are some limits to confidentiality: if what is said in the interview makes me think that you, or someone else, is at significant risk of harm, I will have to break confidentiality and tell somebody. I will inform you before doing so, whenever possible.

What will happen to the results?

The results will be summarised and written up in a report as part of my doctorate and may be submitted for publication in an academic or professional journal. The Dystonia Society will also receive a copy of the report.

Are there any risks?

There are no risks anticipated with participating in this study. However, if you experience any distress following your interview you are encouraged to inform the researcher and contact the resources provided at the end of this sheet.

Are there any benefits to taking part?

There are no direct benefits for taking part. However, it is hoped that the experiences of people taking part in the study will help health services and researchers better understand how to help people living with dystonia. People taking part in the study may also have their travel expenses reimbursed up to the value of £20.

Who has reviewed the project?

This study has been reviewed by the Lancaster University Research Ethics Committee and approved by Lancaster University Research Ethics Committee.

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:

Andrew Morgan
Division of Health
Research Lancaster
University Lancaster
LA1 4YT

[Email: a.morgan2@lancaster.ac.uk](mailto:a.morgan2@lancaster.ac.uk)

What should I do if I want to take part?

If you would like to take part in the study, please send an email to a.morgan2@lancaster.ac.uk including information on the best way to contact you (email address or telephone number preferred) and the times you are available to talk. I will be in touch to arrange the time and location of the interview.

Project Supervisor:

Dr Peter Greasley
Teaching Fellow,
Doctorate in Clinical Psychology
Division of Health Research
Lancaster University
Lancaster
LA1 4YT

OMITTED

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 Bill Sellwood
Programme Director, Doctorate in Clinical
Psychology Division of Health Research
Lancaster University
Lancaster

LA1 4YT

OMITTED

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

Professor Roger Pickup
Associate Dean for Research (Division of Biomedical and Life Sciences)
Lancaster University
Lancaster
LA1 4YD

OMITTED

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

Resources for use 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. It may also be appropriate in such situations to speak with your GP.

The Dystonia

Society Help line:

0845 458 6322

Website address: <http://www.dystonia.org.uk/>

Mind

Info line: 0300 123 3393

Website address: <http://www.mind.org.uk/>

The Samaritans

Info line: 08457 90 90 90

Website address: <http://www.samaritans.org/>